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Practice guidelines

Management of obstructive sleep apnea syndrome type 1 in children and adolescents – A French consensus

G. Aubertin^{a,b,c,*}, M. Akkari^d, A. Andrieux^{e,f,g}, C. Colas des Francs^{h,i}, B. Fauroux^{j,k},
 P. Franco^{h,l}, F. Gagnadoux^{m,n}, O. Gallet de Santerre^o, B. Grollemund^p, S. Hartley^{i,q}, D. Jaffuel^{r,s},
 L. Lafond^t, C.M. Schröder^{u,v,w}, C. Schweitzer^{x,y}, C. Charley-Monaca^z

^a Pediatric Pulmonology, Armand Trousseau Hospital, AP-HP Hospital, Sorbonne University, Paris, 75012, France

^b Centre de recherche Saint Antoine (CRSA), INSERM UMR-S 938, Paris, 75012, France

^c Centre de pneumologie de l'enfant, Ramsay Générale de Santé, Boulogne-Billancourt, 92100, France

^d Ear, Nose and Throat & Head and Neck Surgery, University Hospital Gui de Chauliac, University of Montpellier, Montpellier, 34000, France

^e Cabinet de pneumo-pédiatrie et somnologie pédiatrique, Mérignac, 33700, France

^f Pôle d'Exploration des Apnées du Sommeil (PEAS), Nouvelle Clinique Bel Air, Bordeaux, 33200, France

^g Pediatric Pulmonology Unit, University Hospital Pellegrin – Enfants, University of Bordeaux, Bordeaux, 33000, France

^h Pediatric Sleep Unit, Hôpital Femme-Mère-Enfant, Hospices Civils de Lyon, Bron, 69500, France

ⁱ Réseau Morphée, Garches, 92380, France

^j Pediatric Noninvasive Ventilation and Sleep Unit, AP-HP, Hôpital Necker-Enfants Malades, Paris, 75015, France

^k VIFASOM, University of Paris Cité, Paris, 75004, France

^l Integrative Physiology of the Brain Arousal Systems, CRNL, INSERM U1028, CNRS UMR5292 University of Lyon 1, Bron, 69675, France

^m Department of Pulmonary and Sleep Medicine, University Hospital of Angers, Angers, 49000, France

ⁿ INSERM 1083, UMR CNRS 6015, MITOVASC, Equipe CarME, SFR ICAT, University of Angers, Angers, 49000, France

^o ENT surgeon, Montpellier, 33000, France

^p Department of Dental-Facial Orthopedics, Pole of Bucco Dentaries' Medicine and Surgery, Cleft Competence Center, Strasbourg University Hospital, Strasbourg, 67000, France

^q Sleep Unit, Department of Physiology, Raymond Poincaré Hospital, AP-HP, Garches, 92380, France

^r Department of Respiratory Diseases, University Hospital of Montpellier, Montpellier, 34000, France

^s PhyMedExp, CNRS, INSERM, Montpellier University, Montpellier, 34000, France

^t Oro-myofunctional Therapy Office, Bordeaux, 33000, France

^u Department of Child and Adolescent Psychiatry, Strasbourg University and Strasbourg University Hospitals, Strasbourg, 67000, France

^v CNRS UPR 3212, Institute for Cellular and Integrative Neurosciences, Strasbourg, 67000, France

^w Sleep Disorders Center & CIRCSom (International Research Center for ChronoSomnology), Strasbourg University Hospitals, Strasbourg, 67000, France

^x Children's Medicine, Department of Pediatric Lung Function Testing, Children's Hospital, University Hospital of Nancy, Vandoeuvre les Nancy, 54501, France

^y EA3450 Développement Adaptation et Handicap (DevAH), University of Lorraine, Vandoeuvre les Nancy, 54505, France

^z Department of Clinical Neurophysiology-Sleep Disorders Unit, University of Lille, University Hospital of Lille, and U1172 – LilnCog – Lille Neurosciences & Cognition, Lille, 59000, France

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ABSTRACT

This document is the outcome of a group of experts brought together at the request of the French Society of Sleep Research and Medicine to provide recommendations for the management of obstructive sleep apnea syndrome type 1 (OSA₁) in children.

The recommendations are based on shared experience and published literature. OSA₁ is suspected when several nighttime respiratory symptoms related to upper airway obstruction are identified on clinical history taking. A specialist otolaryngologist examination, including nasofibroscope, is essential during diagnosis. A sleep study for OSA₁ is not mandatory when at least two nighttime symptoms (including snoring) are noted. Therapeutic management must be individualized according to the location of the obstruction. Ear, nose, and throat (ENT) surgery is often required, as hypertrophy of the lymphoid tissues is the main cause of OSA₁ in children. According to clinical findings, orthodontic treatment generally associated with specialized orofacial–myofunctional therapy might also be indicated. Whatever treatment is chosen, follow-up must be continuous and multidisciplinary, in a network of trained specialists.

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* Corresponding author at: Department of Pediatric Pulmonology, Hôpital Armand Trousseau, 26 avenue du Dr Arnold Netter, 75012, Paris, France.

E-mail address: guillaume.aubertin@aphp.fr (G. Aubertin).

1. Introduction

Obstructive sleep-disordered breathing (OSDB) is the consequence of an anatomical and/or functional reduction in the caliber of the upper airways (UA) during sleep. This is characterized by snoring and/or increased respiratory effort due to greater resistance in the UA and pharyngeal compliance. Depending on the degree of obstruction of the airways and its consequences, four clinical–functional entities can be distinguished: primary snoring, high upper airway resistance syndrome (UARS), obstructive hypoventilation syndrome, and obstructive sleep apnea–hypopnea syndrome (OSA). OSA is defined by the French National Authority for Health (*Haute Autorité de Santé*) as the occurrence of abnormally frequent episodes of ventilatory pauses (apnea) or a significant reduction in ventilation (hypopnea) during sleep leading to hypoxemia and/or microarousals [1–3].

The prevalence of primary snoring in children is approximately 8–10% [4]. OSA is also frequent in children, with a peak prevalence of approximately 3% between the age of 3 and 8 years [4] corresponding with a period of adenoidal-tonsil growth in conjunction with poor facial bone growth. The obstructive apnea–hypopnea index (OAH) is the poly(somno)graphic parameter most commonly used to determine the severity of OSA. Assessment of alveolar hypoventilation requires continuous measurement of percutaneous carbon dioxide during sleep.

There are three types of OSA in children [5]. OSA type 1 (OSA₁) is most often found in young, non-obese, children with no associated comorbidity, presenting with a nose and/or throat obstruction, generally a hypertrophy of the lymphoid tissue (tonsils and/or adenoids). This is distinguished from OSA type 2, which is predominant in obese children, usually without significant lymphoid hypertrophy, and type 3, which is characteristic of children with complex and/or syndromic disease. In 2015 and 2016, the French Society for Sleep Research and Medicine (SFRMS) brought together French experts from all disciplines to review the current situation and provide recommendations (a) on the clinical criteria for OSA in children aged 3–8 years without comorbidity [6], (b) on the use of polysomnography (PSG) and respiratory polygraphy (RPG) for the diagnosis of OSA in children [7] and in adolescents [8], and (c) on their management [9,10]. European recommendations were also published in 2016 [2,11].

It is clear that these recommendations are not followed fully in practice, and it would appear necessary to reiterate and update them. This is the purpose of this updated guideline, which aims to define the management of a child with OSA₁.

2. Method

An advisory group was set up in the summer of 2021 following a proposal from the SFRMS scientific council. This group comprised 14 members with a coordinating member (GA). A range of medical and paramedical specialties were represented: general practitioner (CCDF), pediatric pulmonologist (GA, AA, BF, CSchw), neuro-pediatrician (PF), child and adolescent psychiatrist (CSchr), sleep doctor (CCDF, PF, SH, CM), pulmonologist (FG, DJ), ENT surgeon (MA, OGDS), orthodontist (BG), and physiotherapist (LL) specialized in orofacial–myofunctional therapy (OMT). The participants all worked in private practice and/or a hospital.

The theme defined was *the management of a child suspected of having or affected by OSA₁*.

During the first meeting, each member explained the problems encountered in clinical management on a daily basis and their view of the objectives of the advisory groups. The coordinator subsequently drafted four questionnaires on clinical diagnosis, sleep studies, treatment, and monitoring. A first series of questionnaires was shared with the members. Responses with more than 80% approval were considered validated. Other questionnaires were then circulated to clarify and refine responses, always aiming for at least 80% approval. Propositions and conclusions were subsequently reformulated over two meetings with the whole advisory group. The coordinator then drafted the present recommendations, which were subsequently modified and validated by the entire advisory group.

3. Clinical diagnosis of OSA₁ in children

It should be emphasized that the advisory group only considered the management of children suspected of having or affected by OSA₁.

In the opinion of the working group experts, the SFRMS 2016 recommendations for diagnostic management, especially the indications for sleep studies of children with suspected OSA, have not always been applied in clinical practice. The 2016 recommendations distinguished between daytime and nighttime as well as major and minor clinical symptoms, and established the need for a systematic ENT specialist consultation with nasofibroscopy [6].

The experts felt it important to clarify a few points about symptoms from the 2016 recommendations that they still consider both appropriate and valid (Table 1).

Accurate clinical history taking is essential to the diagnosis. Snoring remains a major symptom for suspecting OSA₁. The defining criteria for pathological snoring are listed in Table 1. The clinical history

Table 1

Major and minor criteria for the diagnosis of obstructive sleep apnea syndrome (OSA) in children based on anamnestic, ENT, and maxillofacial findings (adapted from [6]). Apart from clinical signs, prematurity, parental smoking, and a history of OSA in the parents are risk factors for OSA in the child.

	Nighttime symptoms	Daytime symptoms	ENT and dentofacial signs
Major criteria	<p>Snoring</p> <ul style="list-style-type: none"> - frequent (>3 nights / week) - loud (heard through a closed door) - duration (≥3 months) <p>Respiratory irregularities or apneas</p> <p>Noisy inspiratory recovery</p> <p>Child needs to be shaken to make him/her breathe again when asleep</p>	<p>Behavioral disorders</p> <ul style="list-style-type: none"> - restlessness - irritability <p>Attention disorders</p> <p>Height and/or weight growth disorders</p>	<p>ENT examination with nasofibroscopy:</p> <ul style="list-style-type: none"> - adenoid hypertrophy - tonsil hypertrophy <p>Long face, adenoidal face</p> <p>Disharmony in the 3 thirds of the face</p>
Minor criteria	<p>Noisy or struggled breathing</p> <p>Mouth breathing</p> <p>Restless sleep</p> <p>Repeated and brief nocturnal awakenings</p> <p>Parasomnias</p> <p>Excessive sweating</p> <p>Abnormal sleeping position</p> <p>Bed wetting</p>	<p>Learning disorders</p> <p>Poor academic performance</p> <p>Difficult morning awakenings or fatigue</p> <p>Morning headaches</p> <p>Daytime sleepiness</p> <p>Mouth breathing</p> <p>Chronic rhinitis, nasal obstruction</p> <p>Dark circles under eyes</p> <p>Abnormal posture</p>	<p>Retromaxillia, retromandibulia</p> <p>Hypoplasia of the middle face</p> <p>Mouth breathing</p> <p>Narrow palate</p> <p>Dental malposition</p> <p>Macroglossia</p> <p>Abnormal tongue position</p> <p>Short tongue tie</p> <p>Deviation of the nasal septum</p>

ENT: ear, nose, and throat

and examination are the keys to the identification of signs of OSA, which in the experts' opinion are divided into major and minor criteria (Table 1). The experts underline that the diagnosis is above all clinical and that the OSA severity should also be evaluated on the basis of all of the clinical signs.

A screening questionnaire can be used in routine practice, especially by those not specialized in child sleep disorders. Many questionnaires exist but none is sensitive or specific enough to make a diagnosis with certainty. Three questionnaires that have been validated in French were retained by the advisory group:

- The sleep disturbance scale for children from 6 months to 4 years [12] and from 4 to 16 years [13], adapted from the Sleep Disturbance Scale for Children (SDSC) [14], provides screening for different sleep disorders in children. Certain questions are more specific to OSDB. The score, based on 25 items each rated from 1 to 5 according to the severity of the disorder, evaluates insomnia, sleep-disordered breathing, parasomnias, sleep inefficiency, and excessive daytime sleepiness (Supplementary appendix S1 and S2).
- The sleep-disordered scale, derived from the pediatric sleep questionnaire (PSQ). When this score is not pathological (meaning <0.3 , which corresponds to fewer than 8 positive responses out of 22), it has good predictive value for spontaneous normalization of mild OSA only [15–17] (Supplementary appendix S3).
- The severity score derived from the Spruyt and Gozal questionnaire. If this score is >2.72 , the child very likely has OSA with an OAHI >5 (sensitivity = 82%, specificity = 81%, and negative predictive value = 92%) [18] (Supplementary appendix S4).

An allergy test is essential (blood test or skin prick test). An assessment of iron deficiency, by measuring ferritin levels, is also recommended [19].

If OSA is suspected, with or without a visible obstruction on medical examination, the child should initially be referred to a pediatric ENT surgeon to undergo nasofibroscope. Some ENT surgeons may not perform nasofibroscope if the child is distressed or if ENT surgery is clearly indicated (see below). In this case, the adenoidal volume should be checked during the surgical procedure performed under general anesthesia. An X-ray of the cavum is not indicated in this context.

In summary, at this initial stage, a clinical history, clinical examination, and specialist ENT examination are essential and determine further management, in particular the indication of a sleep study (Fig. 1).

4. Sleep study

In the case of isolated hypertrophy of the adenoids responsible for clinical symptomatology, a sleep study is not recommended.

In the case of significant tonsil hypertrophy (Brody grade 3 or 4) (Fig. 2.a and b) [20] and if at least two major nighttime clinical signs are found on examination, regardless of daytime symptomatology (Table 1), a sleep study is not recommended in children ≥ 2 years old without comorbidity [6,21]. ENT surgery is, in this case, the first line of treatment. Pathological but isolated snoring is not considered sufficient to indicate first-line ENT surgery.

In the case of absence of lymphoid tissue hypertrophy or only moderate tonsil hypertrophy (Brody grade 1 or 2), a sleep study is recommended (Fig. 2.a and b) [20].

A hospital PSG is the reference examination (gold standard), but an ambulatory PSG or an RPG (in hospital or ambulatory) can be used provided the recording is of good quality. Ambulatory sleep studies may not be as reliable as studies performed in a sleep center, with a risk of underestimating respiratory event indexes. The type of study

and the place of realization must be adapted to each child, their age, and their family environment.

A child with OSA₁ very often breathes through the mouth. The advisory group strongly recommends performing the sleep study (PSG or RPG) with an objective evaluation of nasal breathing (nasal cannula pressure sensor) and mouth breathing (e.g., mouth thermistor, tracheal sound sensor, respiratory inductance plethysmography by measuring the sum of thoracic and abdominal movements). These may be combined with a mandibular movement sensor, which can measure mouth openings and increased respiratory effort by monitoring mandibular position and, consequently, enhance the detection of respiratory events [22]. However, no study other than PSG or RPG is currently recommended for the diagnosis of OSA.

The recording must cover at least 6 h of sleep (and 20% of REM sleep in the case of a PSG reading). The rules for coding respiratory events (RE) and, in the case of PSG, sleep stages, should follow the international rules of the American Academy of Sleep Medicine (AASM) [23]. Automatic analysis of sleep studies is inaccurate and not acceptable. Tests must be read and validated by physicians trained to interpret PSG/RPG in children. These physicians must be able to prove both initial training and ongoing relevant experience in pediatric sleep medicine.

According to the AASM, an RE corresponds to the absence or the significant decrease of air flow over a period of at least two respiratory cycles. Obstructive apnea is defined as a decrease in the amplitude of naso-buccal flow by $\geq 90\%$, with persistent respiratory effort. Coding of respiratory efforts inducing microarousal is considered optional by the AASM. Hypopnea is identified when there is a decrease in the signal amplitude of $\geq 30\%$ associated with a microarousal, an arousal, or a drop in saturation of $\geq 3\%$. The obstructive character of hypopnea is characterized by the presence of snoring during the RE, an accentuation of the inspiratory plateau of nasal pressure relative to baseline breathing, or phase opposed thoracoabdominal movements. Standards for both types of RE exist. Other REs are distinguished such as hypopnea without microarousal or a drop in saturation of $\geq 3\%$ or hypopneas with less than 30% drop in amplitude. The presence and frequency of flow limitations (increase in respiratory effort, flattening of nasal pressure on inspiration, or increased partial transcutaneous carbon dioxide [ptCO₂] leading to microarousal) should also be noted.

Central apnea (CA) is identified if there is no respiratory effort throughout the duration of the event, which must last at least 20 s or have a minimum duration of two respiratory cycles, and be associated with microarousal, an arousal, or a drop in saturation of $\geq 3\%$. Hypopnea is coded as central in the absence of snoring, no accentuation of the inspiratory plateau of the nasal pressure, and no phase opposed thoraco-abdominal movements during the RE. It is recommended to distinguish obstructive RE by specifying the obstructive and the central apnea index included in the overall AHI, as children physiologically present with more central REs than adults.

A recording of transcutaneous or exhaled carbon dioxide is recommended to screen for hypoventilation.

In children >13 years old, PSG can be analyzed based on adult criteria. The adoption of adult criteria underestimates the RE indexes in particular, notably the number of hypopneas. The advisory group recommends systematically using pediatric criteria until the end of the adolescent growth period.

The sleep study report should be detailed. It should include the indication for the study, the patient's symptomatology and clinical examination findings, information about the quality of sleep during the child's usual nights, information about the overall quality of the study, information on the quality of nasal flow and/or presence of mouth breathing, the analysis of the different sleep and respiratory parameters, a conclusion about the severity of the OSA, and a management proposal. The standards used to code events must be listed in the report.

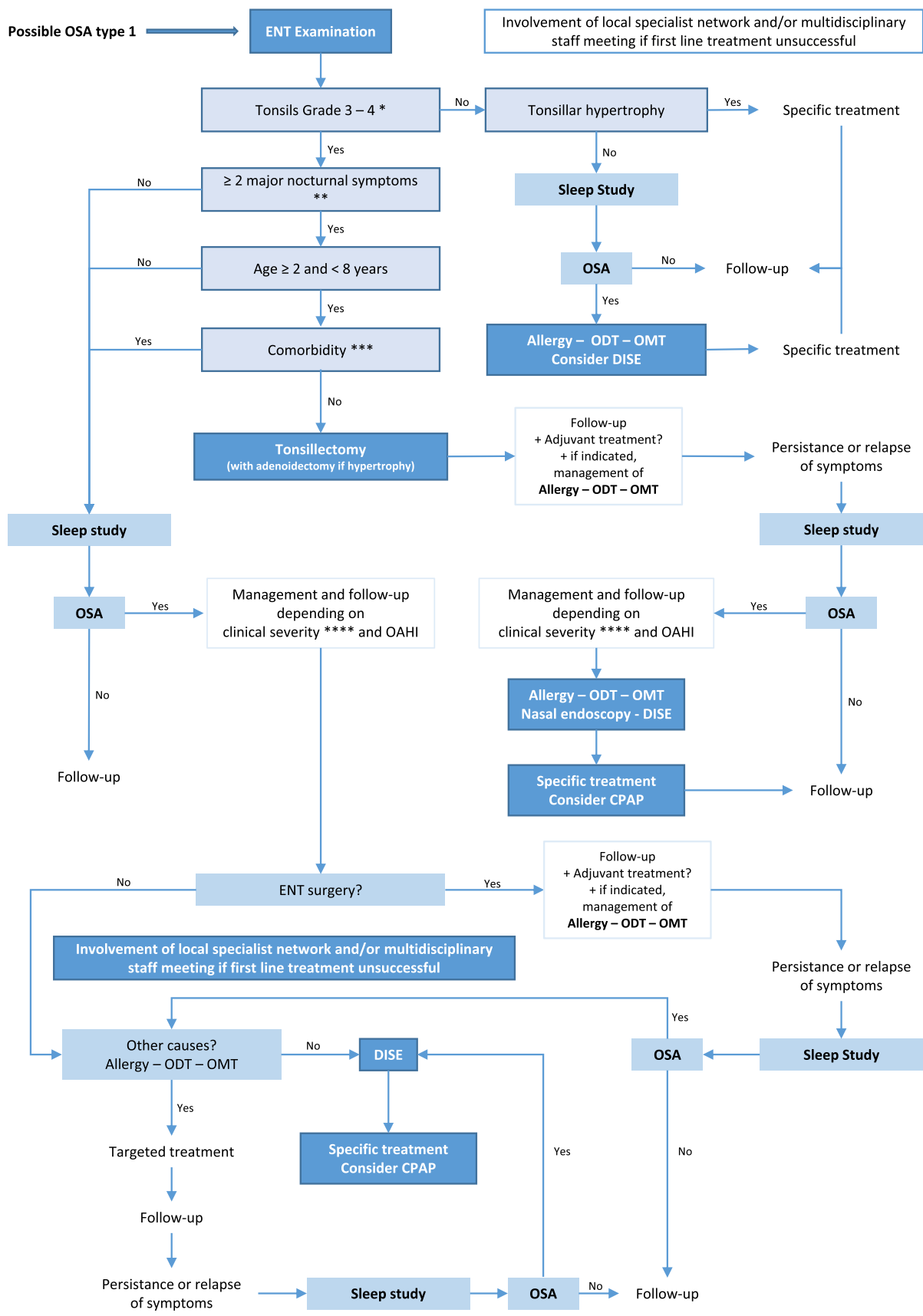


Fig. 1. Management of children with suspected obstructive sleep apnea syndrome type 1 (OSA₁).

*Tonsil volume, evaluated based on Brodsky's classification (Fig. 2) [20].

**According to Table 1, adapted from [6].

***Severe comorbidity (mainly cardiopathy, hemostasis disorders) or uncontrolled comorbidity (e.g., severe uncontrolled asthma).

****Clinical severity: all major and minor signs (Table 1), including potential complications of OSD (height and weight, neurocognitive and/or behavioral repercussions).

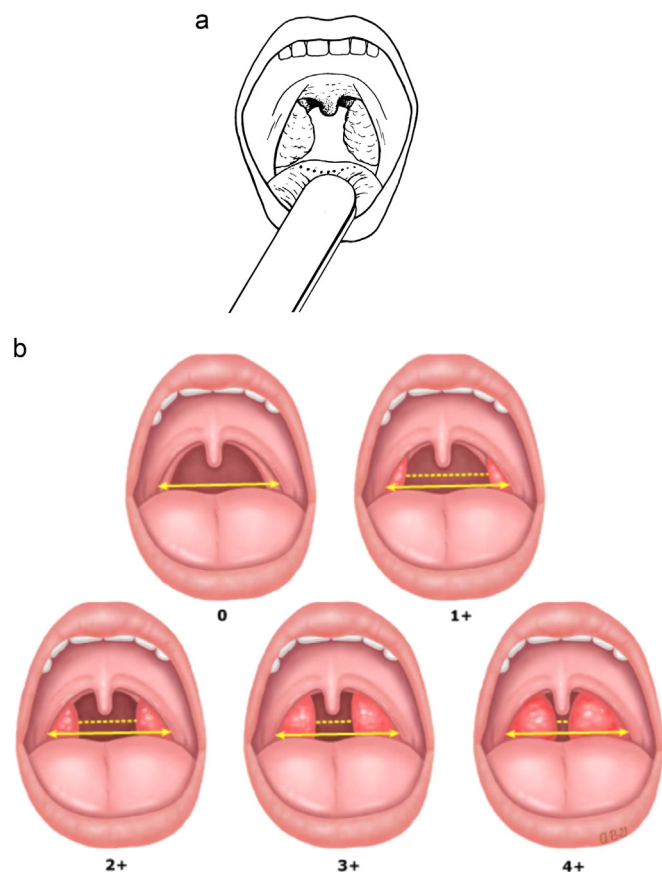


Fig. 2. Evaluation of tonsil volume according to Brodsky [20].

Fig. 2.a. According to Brodsky, physical examination of the tonsils is accomplished by placing two tongue depressors gently on the body (anterior two thirds) of the tongue.

Fig. 2.b. Standardized evaluation of the size of the tonsils.

Grade 0: The uvula and pillars of the tonsil compartment are visible.

Grade 1: The tonsils emerge slightly from the tonsil compartment, airway obstruction is less than 25%.

Grade 2: The tonsils protrude from the tonsil compartment, airway obstruction is 25–50%.

Grade 3: The tonsils protrude well beyond the compartment, airway obstruction is 50–75%.

Grade 4: The tonsils are almost touching, airway obstruction is more than 75%.

Concerning the severity of the OSA, the advisory group underline that this is assessed on the basis of the OAHl values (OSA is considered mild when the OAHl is ≥ 2 and < 5 , moderate when the OAHl is ≥ 5 and < 10 , and severe if the OAHl is ≥ 10) [24] but also on the basis of the whole clinical picture including repercussions on the child (schooling, behavior, and/or growth) and on the basis of all the sleep study data (see above).

5. Treatment

Management of OSA₁ in children should be multidisciplinary, within a local network of physicians in primary and secondary care trained in the management of OSA₁ in children and able to communicate easily to optimize the management of each child. It is not essential for the initial management to be based in hospital. Complex cases (comorbidities, failure of first-line treatments) should be discussed in a multidisciplinary meeting in a hospital and/or reference center. With the exception of OSA₁, all patients must be referred to a

reference center or to a physician specialized in pediatrics and sleep in children.

In the case of lymphoid tissue hypertrophy, tonsillar-adenoid surgery is the first-line treatment according to current ENT guidelines [25,26].

In the case of absence of important hypertrophy of the tonsils and in the case of mild OSA₁, drug treatment can be proposed in addition to specific allergy and weight control treatments. This drug treatment must be reassessed at least clinically at 3 months. Nasal hygiene must be appropriate and nose cleaning with saline solution is essential. Medical treatment is based on nasal corticosteroids and second-generation oral anti-histamines (even in the absence of allergies) alone or in combination. Montelukast can be used under supervision due to its adverse effects on sleep (e.g., nightmares, insomnia), mood (depressive syndrome, possible suicidal ideation), and behavior (e.g., aggressiveness). None of these drugs has a Marketing Authorization for this indication.

In the absence of tonsil hypertrophy and in cases of moderate OSA₁, the same drugs can be used in addition to ENT management guided by drug-induced sleep endoscopy (DISE) but cannot replace it.

In the case of mild-to-moderate OSA₁, a conservative medical approach (drug treatments, and/or orthodontics, and/or OMT without surgery) can be proposed. In 54% of non-obese children with tonsil hypertrophy, the AHI normalizes at 7 months and symptoms disappear without surgery, as shown by Marcus et al. in the CHAT Study. This percentage drops to 29% in the case of obesity. In the same study population, the normalization of the AHI is more frequent after adenoid-tonsillectomy: 85% in non-obese children and 67% in obese children [27].

A specialized consultation with an orthodontist and/or orofacial-myofunctional therapist (trained and specialized in OSA) is essential, even in the case of ENT causes requiring surgery, especially if on clinical examination there is mouth breathing, a dental occlusion disorder, a lingual apraxia, or swallowing or postural anomaly. A consultation with these specialists is essential too in the case of failure of the initial surgical treatment and/or persistent mouth breathing. The assessment and treatment by a physiotherapist trained in OMT should include postural evaluation. If OMT is performed by a speech therapist, morpho-dynamic assessment of postural positioning must be carried out by a physiotherapist.

As in adults, the practice of DISE is also growing in children. It enables visualization of the location(s) of the obstruction thereby enabling targeted therapeutic management. However, issues of access and the lack of national recommendations for children (which exist in adults [28]) limit its use. There are a few publications on the practice [29–31] but there is currently no international consensus on the use of DISE for children. That said, the French Society of ENT Surgery (SFORL) and the International Pediatric Otolaryngology Group (IPOG) have established some indications in recent publications [21,25]. In OSA₁, DISE is recommended (after confirmation of OSA via a sleep study) in the case of failure of an adenoid-tonsillectomy [25] or absence of an obvious obstacle on nasofibroscope performed on the awake child, particularly in the case of small tonsils and adenoids [21,25].

6. Follow-up

A reference physician trained in OSA in children (who can be their pediatrician) must coordinate the management.

Whatever treatment is chosen, patient follow-up must be regular and prolonged to verify the effectiveness of the treatment, the resolution of symptoms, the absence of their recurrence, and the recovery of effective nasal breathing. The child and family must be made aware of the risk of recurrence and the need to consult again if necessary.

An ENT consultation 2–6 months after ENT surgery is recommended [24]. This enables the complete resolution of symptoms and reappearance of nasal breathing (if initially absent) to be verified. In the case of persistent mouth breathing, a specialized assessment by an orofacial-myofunctional therapist and, if needed, a consultation with an orthodontist are necessary to optimize management. A second ENT consultation is recommended 1 year after surgery, especially in the case of a partial tonsillectomy [26].

After an adeno-tonsillectomy, it is not recommended to systematically redo a sleep study. Indications are listed in the AASM 2011 recommendations [32], the French National Authority for Health (HAS) 2012 report [3], and the SFORL recommendations on OSA [24] and tonsillectomy in children [26]: persistence or reappearance of obstructive symptoms, existence of one or several risk factors for residual OSA (pre-surgery OAH1 >10/h, age >7 years, asthma associated – in this case severe and/or uncontrolled asthma – with OSA, obesity, OSA type 2 or 3).

In the case of persistent or recurrent symptoms, the child should undergo a new ENT assessment with nasofibroscope and a sleep study (preferably a PSG if a differential diagnosis is suspected). If OSA is confirmed in the absence of an ENT obstruction, the child should be treated in a multidisciplinary network with an orthodontist, an OMT specialist, a sleep doctor, a pediatric pulmonologist, or any other specialist if necessary. The different treatments should then be discussed to individualize care: DISE, orthodontics, OMT, allergy treatment, or even continuous positive airway pressure (CPAP) as a last resort.

The prescribing of CPAP is restricted to some pediatricians in France (rules of the decree of 16 December 2017 [33] (Supplementary appendix S5)). In the case of OSA₁, the cause and level of the airway obstruction must be known in order to evaluate other suitable therapies. For OSA₁, CPAP should only be used in the absence of another alternative. If CPAP is necessary, its usefulness should be regularly re-evaluated, throughout follow-up, in order to attempt withdrawal as soon as the OSA has regressed sufficiently, confirmed by a sleep study conducted without CPAP.

Follow-up is therefore essential to ensure that the clinical symptoms disappear and, eventually, effective nasal breathing is restored. A functional ventilatory assessment performed by a trained physiotherapist at the end of treatment is considered useful.

7. Conclusion

Any child with OSA₁ needs management coordinated by a reference physician and carried out by trained specialists, working within a local multidisciplinary network. The relative frequency of OSA₁ requires increased awareness and training of practitioners to screen and optimally manage children with OSA₁. These expert recommendations outline the management for these children (Fig. 1).

Declaration of Competing Interest

G.A. Clinical trials: as co-investigator for Orkyn. Conferences: invitations as a speaker for Elivie, Cidelec. Conferences: invitations as an auditor (travel expenses paid by a company) for ASV Santé, ADEP Assistance, Elivie.

M.A. Conferences: invitations as an auditor (travel expenses paid by a company) for Resmed.

A.A. Conferences: invitations as an auditor (travel expenses paid by a company) for SOS Oxygène, Isis Médical and Domair Santé.

C.CDF. Conflict of interest: none

B.F. Conflict of interest: none

P.F. Conferences: invitations as a speaker for Elivie.

F.G. Conferences: invitations as a speaker for Sefam, Cidelec, Resmed. Conferences: invitations as an auditor (travel expenses paid by a company) for Sefam, Asten Santé. Occasional consultancies: expert reports for Resmed, Air Liquide.

O.GDS. Conflict of interest: none

B.G. Conflict of interest: none

S.H. Conflict of interest: none

D.J. Clinical trials: principal investigator, coordinator or main researcher for Nomics. Conferences: invitations as a speaker for Nomics. Conferences: invitations as an auditor (travel expenses paid by a company) for Philips Respironics, Sefam, Lowenstein. Occasional consultancies: expert reports for Nomics, Sefam.

L.L. Conflict of interest: none

C.Schr. Conflict of interest: none

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C.M. Conferences: invitations as an auditor (travel expenses paid by a company) for Resmed. Occasional consultancies: expert reports for Resmed.

Supplementary materials

Supplementary material associated with this article can be found in the online version at doi:10.1016/j.arcped.2023.06.009.

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