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SFORL Guidelines

Guidelines of the French Society of Otorhinolaryngology. Role of the ENT specialist in the diagnosis of obstructive sleep apnea-hypopnea syndrome (OSAHS) in children. Part 2: Diagnostic investigations apart from sleep studies



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ABSTRACT

Objectives: To present the 2017 Clinical Practice Guidelines of the French Society of Otorhinolaryngology concerning the role of the ENT specialist in the diagnosis of pediatric obstructive sleep apnea-hypopnea syndrome. This manuscript specifically focuses on diagnostic investigations apart from sleep studies.

Methods: A multidisciplinary work-group drew up a first version of the guidelines, graded according to level of evidence following the GRADE grading system. The final version was obtained by including the suggestions and comments from the editorial group.

Results: At the end of the process, guidelines were established and graded regarding diagnostic investigations apart from sleep studies, in particular respiratory functional tests, biological markers, and morphologic assessment under induced sleep (drug-induced sleep endoscopy (DISE) and cine-MRI).

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1. Introduction

Clinical and complementary examination aims to identify obstruction sites and associated pathologies implicated in obstructive sleep apnea-hypopnea syndrome (OSAHS), risk factors affecting treatment, and individuals at risk of future complications.

2. Material and methods

2.1. Literature search

Relevant articles were retrieved from the PubMed database of the American National Center for Biotechnology Information, hosted by the National Institutes of Health National Library of

Medicine, for the period up to May 2018. Articles were graded by levels of evidence 1 to 4, and the guidelines were graded A, B, C, D or expert opinion by decreasing level of evidence, following the French Health Authority's 2000 guide to literature analysis and recommendations grading (available at <https://www.has-sante.fr>; see Appendix A).

2.2. Writing stages

The organizational committee:

- defined the guidelines objectives and plan;
- chose the work-group members so as to ensure that target specialties were well represented and that the group covered the national territory.

The work-group drew up a first draft of the guidelines, which was submitted to an editorial group for comments and suggestions.

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The final version was drawn up by the work-group taking account of these comments. It is available on the ORL France website: <https://www.orlfrance.org>. The present article summarizes this final version, keeping the sense while adding certain recent data from the literature: the original search stopped at 2016, whereas the present article extends to 2018.

3. Results

3.1. Functional exploration

The contribution of non-invasive upper-airway functional exploration (rhinomanometry, acoustic rhinometry, optical coherence tomography, etc.) in childhood apnea remains to be proven.

Respiratory function tests can be useful in certain laryngo-tracheo-esophageal and broncho-pulmonary pathologies.

3.2. Biological markers

Elevated levels of CRP, certain interleukins and other inflammatory syndrome markers are associated with increased risk of cognitive impairment and residual OSAHS following adenotonsillectomy [1,2] (level of evidence, 3).

Urine epinephrine, norepinephrine, GABA and taurine concentrations are predictive of cognitive deficit in OSAHS [3] (level of evidence, 1).

Assessment of these markers in childhood OSAHS, however, is still at the clinical research stage.

Guideline 5 (Grade C). Non-invasive exploration of upper-airway permeability and biological marker assay are not recommended in childhood OSAHS.

3.3. Drug-induced sleep endoscopy (DISE)

Drug-Induced Sleep Endoscopy (DISE) is one of the two forms of morphologic assessment under drug-induced sleep, alongside cine-MRI (dealt with in section 4 below). It consists in anatomic assessment of the upper airway from nasal cavity to larynx, using a flexible endoscope, under induced sleep with spontaneous respiration. It may in some cases be useful to associate visualization of the tracheobronchial tree [4].

3.3.1.1. Implementation

The procedure is performed in an operating room, endoscopy room or other room dedicated to procedures under general anesthesia in children. The environment should be suitably calm and dark (level of evidence, 3) [5].

Management may be on an in- or out-patient basis, depending on contraindications to out-patient care (care structure, history, family characteristics) and any associated procedure (expert opinion).

Endoscope diameter should be adapted to patient age (expert opinion). A video column with recording optimizes examination, enabling images to be read over and proportionality and relations of anatomic structures to be measured (expert opinion), which can be contributive to quantifying airway obstruction.

The patient is positioned supine, with the head in neutral position (expert opinion). Although no studies have been performed in children, it seems reasonable to suppose that head and body position impact observations, as in adults (level of evidence, 2) [6].

Local anesthesia (lidocaine) is to be eschewed, as loss of pharyngolaryngeal mucosal sensitivity would bias results (level of evidence, 4) [7]. Vasoconstrictors are contraindicated before 15 years of age, and thus cannot be used in children. Atropine to reduce salivation is not recommended, due to cardiovascular effects liable to impact examination (level of evidence, 4) [5,7].

Intra-procedural cardio-respiratory monitoring follows the guidelines for pediatric anesthesia, and should comprise at least one recording of heart rate, saturation, blood pressure and expired PCO₂ [7,8]. In children, it is generally recommended to set up a venous line before or during induction (level of evidence, 4) [9,10].

There is no consensus on sedation modalities. Inhalation requires a specific mask allowing for the endoscope, with consequently poorer control of sedation level [11]. Pediatric studies more often advise intravenous anesthesia, using a range of molecules and protocols (level of evidence, 3) [5,7,10,12–16]. Midazolam 0.5 mg/kg is particularly used for premedication or induction (level of evidence, 4) [17], relayed either by propofol (level of evidence, 4) [5,7,12,14–16] or by dexmedetomidine. The latter shows lower risk of respiratory depression and hypoxia-hypercapnia (level of evidence, 4) [9,17], but does not as yet have market authorization in this indication in France, being reserved to adult sedation in intensive care. Associating remifentanyl to propofol or dexmedetomidine seems not to improve sedation quality, while increasing the risk of desaturation (level of evidence, 4) [16]. Anesthesia is ideally adjusted to maintain effective spontaneous respiration without response of to normal-intensity vocal stimuli (level of evidence, 4) [12]. Depending on respiratory tolerance, oxygen enrichment may be associated (level of evidence, 2) [14,16], either by a nasal cannula close to the airway with flow and position set so as not to induce positive expiratory pressure, or by a specific mask during endoscopy, the positioning of which on the face must not modify anatomic structures. More exact assessment of sedation level on EEG-based bispectral index (BIS) has been suggested, but interpretation is more difficult in children than in adults, as EEG characteristics change with age, and its contribution in pediatric DISE remains to be shown (level of evidence, 2) [15,16,18–20].

3.3.1.2. Results reporting

In childhood apnea, DISE finds various obstruction sites, according to age and medical-surgical history [4]. They range from nasal cavity to larynx, and may be fixed (e.g., narrow nasal cavity, septal deviation, constitutionally narrow nasopharynx, or pharyngeal, palatine or basalingual tonsil hypertrophy) or dynamic (e.g., nasal valve collapse, lateral pharyngeal wall collapse, sleep-dependent laryngomalacia, or glossoptosis). Obstacles are often multiple, especially in type II and III OSAHS [4].

Reporting should include patient positioning, sedation protocol, any local anesthesia, vasoconstrictors or atropine (bearing in mind that atropine is not recommended for this examination in children and that vasoconstrictors are contraindicated before 15 years of age) (expert opinion).

A semi-quantitative classification of degree of obstruction per level is useful for before-and-after comparison in a given patients or comparison between patients in a given cohort (level of evidence, 3) [4]. Table 1 presents the Chan classification [21], but there also exist VOTE (Velum, Oropharynx, Tongue-base, Epiglottis), SERS (Sleep Endoscopy Rating Scale), Bachar, Fishman, and Boudewyns scores; none have as yet demonstrated superiority or been validated in French versions [13,14,22–24]. Given the frequency of obstacles detected on DISE in children, it seems preferable to use classifications based on rhinopharyngeal and supraglottic levels, such as those of Chan [21] and Boudewyns [14] (expert opinion). Some classifications advocate maneuvers such as mandibular propulsion or

Table 1
Drug-induced sleep endoscopy (DISE): Chan-Parikh classification [10].

	0	1	2	3
Adenoids	Absent	0–50% choanal obstruction	50–99% choanal obstruction	Complete choanal obstruction
Velum	No obstruction (complete view of tongue-base and larynx)	0–50% anteroposterior closure (partial view of tongue-base and larynx)	50–99% anteroposterior closure (some views of tongue-base and larynx)	Complete closure against posterior pharyngeal wall
Lateral pharyngeal wall/palatine tonsils	No obstruction	0–50% lateral obstruction	50–99% lateral obstruction	Complete obstruction
Tongue base	No obstruction (complete view of vallecula)	0–50% obstruction (vallecula not visible)	50–99% obstruction (epiglottis not contacting posterior pharyngeal wall)	Complete obstruction (epiglottis against posterior pharyngeal wall)
Lingual tonsils	Present		Absent	
Laryngeal vestibule	No obstruction (full view of vocal cords)	0–50% obstruction (but more than half of vocal cords still visible)	50–99% obstruction (less than half of vocal cords still visible)	Complete obstruction (vocal cords not visible)

raising the chin during DISE to predict the efficacy of removing certain obstacles in terms of airway permeability [4].

3.3.1.3. Indications

DISE is an invasive examination, in which indications remain to be standardized (level of evidence, 4) [7,10]. It is a heavy examination, and should be preceded by sleep study to check the existence of OSAHS (expert opinion).

There is, however, relative consensus on three indications (level of evidence, 3) [4,10]:

- failure of adenotonsillectomy, whatever the history. Incidence ranges between 10% and 15%, depending on whether the criteria are clinical or polysomnographic (level of evidence, 1) [25–27]. It usually involves other airway obstruction sites and is more frequent in case of small tonsils (level of evidence, 2) [28] and comorbidity (obesity, craniofacial or airway deformity, neuromuscular disease with pharyngeal hypotonia, metabolic disease with airway submucosal conjunctive tissue infiltration) (level of evidence, 2) [28,29]. Awake endoscopy performed in consultation can diagnose residual obstacles following adenotonsillectomy, but is less sensitive than DISE, which can approximately reproduce the pharyngolaryngeal hypotonia found during sleep. Several studies reported the clinical and polysomnographic efficacy of surgical resection of obstacles found on DISE (level of evidence, 3) [15,30];
- no obvious obstacle on clinical examination including awake endoscopy, notably with small tonsils and pharyngeal adenoids (level of evidence, 4) [14,31,32]. Small tonsil volume on awake clinical examination correlates with absence of tonsillar obstruction on DISE and more frequent lateral pharyngeal wall collapse [32];

OSAHS type II and III. These are often associated with multi-level obstacles, which DISE detects with greater sensitivity, better guiding treatment. It is notably often indicated in Down syndrome [33].

Whether DISE should be systematic ahead of adenotonsillectomy for OSAHS in children without particular history or previous surgery but presenting with tonsillar hypertrophy is, on the other hand, uncertain [14]. Some studies reported 35–60% rates of therapeutic modification in the light of DISE [34,35], but there are several arguments against such an attitude:

- adenotonsillectomy shows great efficacy in type I OSAHS with adenoid and/or tonsillar hypertrophy [25–27];

- systematic DISE would incur significant extra costs and risk of complications, notably related to general anesthesia;
- DISE interpretation is less reliable in case of adenotonsillar hypertrophy, which modifies local airflow (level of evidence, 4) [10];
- one-step association of adenotonsillectomy to other airway procedures according to intraoperative DISE findings raises the problem of parental informed consent.

When DISE reveals multiple obstacles, optimal strategy remains to be defined, between beginning with the main obstacle (notably, adenotonsillar) and simultaneously treating all [31,36]. Assessment error for secondary obstacles due to local aerodynamic disturbance caused by the main obstacle [10], and increased risk of complications (dyspnea, hemorrhage) due to simultaneous obstacles at multiple levels, argue for step-by-step surgery, although this is to be considered on a case-by-case basis (expert opinion).

3.4. Imaging

3.4.1. Airway CT and MRI

CT and MRI may be indicated in some very rare cases in which the culprit obstacle is unclear (small tonsils and adenoids, comorbidity with risk of multi-level obstacles) and neither awake endoscopy nor DISE are feasible (awake endoscopy: refusal by child, endoscope unable to pass through nasal cavity; DISE: no team experience, comorbidity precluding maintenance of spontaneous respiration under general anesthesia) (expert opinion). Contrast enhancement is not required. In children, MRI is harder to conduct than CT, being less widely available and requiring prolonged immobility sometimes necessitating sedation, but is to be preferred as radiation dose is lower. 3D slices are recommended.

3.4.2. Cine-MRI

Cine-MRI complements DISE in morphologic assessment under induced sleep. Like DISE, it assesses airway morphology during sleep, with the same indications. It comprises ultra-rapid MRI sequences providing dynamic 3D images during sleep. The technique was first described in 1992, yet few pediatric studies have been published [37]. The anesthesia protocol is similar to that in DISE, inducing sleep while maintaining good spontaneous respiration. There is a single study comparing the two techniques in children: it found no significant difference in observations (level of evidence, 4) [38]. Cine-MRI, however, is much less widely used than DISE (level of evidence, 4) [10], for the following reasons:

Guideline n° 6 (DISE)

- DISE requires general anesthesia under spontaneous respiration, and should be implemented in an adapted environment, respecting the safety recommendations for pediatric anesthesia (Grade C). It may be performed on an out-patient or in-patient basis (expert opinion).
- As the examination is heavy, it should be preceded by a sleep study confirming the existence of OSAHS (expert opinion).
- Local treatments (local anesthesia, vasoconstrictors, atropine) are not indicated (Grade C).
- The patient is positioned supine with the head in neutral position (expert opinion).
- There is no consensus on sedation modalities, but sedation should be maintained by venous rather than inhalational route (Grade C).
- Depending on respiratory tolerance, oxygen enrichment may be associated, without inducing positive airway pressure (Grade B).
- If a mask is used for anesthesia or oxygen enrichment, care must be taken not to distort facial and airway morphology (expert opinion).
- The contribution of sleep quality analysis on bispectral index (BIS) during pediatric DISE remains to be proven (Grade B).
- Endoscope diameter should be adapted to the patient's age (expert opinion).
- A video column with recording optimizes results analysis (expert opinion).
- DISE explores the airway from nasal cavity to larynx (Grade C). It may be useful to associate tracheobronchial endoscopy, depending on the context (expert opinion).
- Certain maneuvers, such as mandibular propulsion and chin raising, may help in assessing the individual obstacle contributions to OSAHS and the probable impact of surgical resection (Grade D).
- Reporting should specify patient positioning, sedation protocol, and any local anesthesia, vasoconstrictors or atropine (expert opinion).
- Obstacles observed should be semi-quantitatively assessed on any of the 6 scores found in the literature: Chan, Boudewyns, VOTE, SERS, Bachar or Fishman (Grade C).
- No particular classification has shown superiority or been validated in French, but those assessing the nasopharynx and supraglottic larynx should be preferred (expert opinion).
- Systematic DISE is not justifiable ahead of adenotonsillectomy for OSAHS in children without relevant history or previous surgery but presenting with tonsillar hypertrophy (Grade B).
- Current indications for DISE comprise: failure of adenotonsillectomy (Grade C), absence of obvious adenotonsillar obstacle on clinical examination (GRADE D) and type II and III OSAHS (Grade D).
- When several obstacles are identified on DISE, optimal surgical strategy (beginning with main obstacle, notably when adenotonsillar, or treating all simultaneously) is determined on a case-by-case basis (expert opinion).

- it is less precise than endoscopy at the laryngeal level, especially for diagnosis of sleep-dependent laryngomalacia;
- it is less widely available than DISE;
- DISE allows treatment in theater in the same step as the diagnostic examination.

Disclosure of interest

The authors declare that they have no competing interest.

Guideline 7

In childhood OSAHS:

- airway CT or MRI without contrast enhancement are seldom indicated, unless for absence of obvious obstacle or non-feasibility of awake endoscopy and DISE (expert opinion). MRI is to be preferred, involving lower radiation dose;
- cine-MRI is much less widely used than DISE, and there are few studies of its contribution (Grade C).

Appendix A. GRADE classification of recommendations.

Level of evidence provided by the literature	Recommendation grade
Level 1 - High-power randomized comparative trial - Randomized comparative meta-analysis - Decision tree based on well-conducted studies	GRADE A Established scientific proof
Level 2 - Low-power randomized comparative trial - Well-conducted non-randomized comparative study - Cohort study	GRADE B Scientific presumption
Level 3 - Case-control study	GRADE C Low level of evidence
Level 4 - Comparative study with serious bias - Retrospective study - Case series - Descriptive (transverse or longitudinal) epidemiological study	GRADE D Expert opinion

From ANES (Agence Nationale d'Accréditation et d'Evaluation en Santé) Guide to literature analysis and recommendations grading, January 2000 (available at the Health Authority website: <https://www.has-sante.fr>).

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