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REVIEW ARTICLE

Obstructive sleep disordered breathing in children – an important problem in the light of current European guidelines

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ABSTRACT:

Obstructive sleep disordered breathing (SDB) is a common clinical problem. An unrecognized and untreated SDB is a serious threat for an intensively developing organism of a child. The consequences of SDB include cardiovascular and neurological complications, growth disorders and enuresis. Therefore, SDB in children becomes an important subject of many scientific investigations, publications and congresses.

In 2015 the European Respiratory Society Task Force published a document concerning the conclusions about the diagnostics and treatment of SDB in children and youth from 2 to 18 years of age (Fig. 1). The scientific data from 362 publications were presented in a condensed form of "seven steps", very useful in diagnosing and treatment planning (1). The authors underline the limited number of the reliable evidence about SDB: prospective studies, randomized double-blinded studies with placebo. The presented evidence was categorized depending on their quality according to the classification of the American Academy of Neurology (ANN) into classes I – IV.

Previously, in 2012, the guidelines of the American Academy of Pediatrics on obstructive sleep apnea syndrome (OSAS) in children with tonsillar hypertrophy and/or obesity were published (2, 3) and they were a valuable diagnostic and therapeutic compendium. The European guidelines discussed in this article result from the progress of knowledge in the recent years, they cover the subject broadly, consider rare and difficult cases and present the spectrum of potential therapeutic actions. The aim of the guidelines is a better recognition of SDB, a systematization of diagnosis and treatment at every stage of medical care, including the causes of this disorder and its complications.

KEYWORDS: snore, obstructive sleep disordered breathing, polysomnography, obstructive sleep apnea syndrome

INTRODUCTION

Sleep is indispensable in a normal physical, mental and cognitive development of an organism, therefore children sleep longer than adults [4]. The pathomechanism of sleeping disorders in adults and in children does not significantly differ but the clinical manifestations of these disorders can be extremely different. For example, sleep apnea in adults occurs more often in men than in women, it occurs in snoring obese patients with pathological somnolence. Sleep apnea in children before puberty occurs with the same frequency in both sexes, it correlates with the growth of the lymphatic tissue of the pharynx more than with obesity and is accompanied rather by attention disorders and problems in learning than by pathological somnolence [5].

Sleeping disorders in children are an increasingly often diagnosed problem. They occur most often in a form of obstructive sleep apnea syndrome, described in the literature for the first time in 1976 [4]. Recently, in 2002, the American Academy of Pediatrics (AAP) recognized snoring in children as a major medical problem in this age group and published the first report concerning sleep apnea diagnostics and treatment in children [6]. Ten years later, in 2012, the Academy amended these guidelines [2, 3].

In 2015 the European Respiratory Society Task Force published a document summarizing the conclusions about obstructive sleep disordered breathing diagnostics and treatment in children and youth form 2 to 18 years old [1]. It emphasizes that SDB is

not a separate disease but a syndrome / a set of symptoms of respiratory tract dysfunction during sleep, which is characterized by snoring and/or increased effort to breath caused by an increased airway resistance (upper airway resistance syndrome, UARS) and a narrowing of the pharynx. The concept of obstructive sleep disordered breathing concerns multiple clinical units varying with the level of advancement of upper airway obstruction (Table I.). It is used when the symptoms of upper airway obstruction while sleeping are observed but the level of advancement was not defined by objective methods such as polysomnography. The frequency of habitual snoring/simple snoring occurrence was estimated at 7.45% and the frequency of obstructive sleep apnea syndrome according to available studies is 1-4%. The apnea-hypopnea index (AHI) is the most commonly used polysomnographic parameter describing the SDB advancement level.

In the Polish literature there is no unified nomenclature concerning obstructive sleep apnea syndrome. The most often used terms are "sleep apnea syndrome", "obstructive apnea", "obstructive sleep apnea syndrome", "sleep apnea" and abbreviations: OSA, OSAS, OSAHS, OBPS [6].

The unique character of the ERS Task Force document in comparison to previously published guidelines manifests itself in a broader view of a problem. The authors consider the whole spectrum of obstructive SDB advancement in children and youth from 2 to 18 years old, they refer to the obstructive SDB predisposing factors other than hypertrophy of the lymphatic tissue of the pharynx and obesity, they consider other diagnostic methods in situations when polysomnogaphy is unavailable, and present diagnostic and therapeutic approach "step by step". There are seven steps. Each of them contains important clinical questions posed during ERS Task Force meetings, the answers to these questions in the form of useful guidelines and the references on the basis of which the answers/the guidelines were created with the evidence quality evaluation according to the classification of the American Academy of Neurology (Figure 1).

Step 1. The risk of obstructive sleep disordered breathing

Frequent and loud snoring, recognizable apnea, restless sleep and breathing through the mouth are the symptoms of upper airway obstruction reported by parents/guardians. Physical examination can reveal hypertrophy of palatine tonsils, obesity, anomalies of the central part of the face (Apert, Crouzon, Pfeiffer syndromes, the cleft of the soft palate), micrognathia/micrognathism (Pierre Robin sequence, Treacher, Collins, Nager, Stickler syndromes, juvenile idiopathic arthritis), neuromuscular disorders (cerebral palsy, muscular Duchenne/Becker dystrophy, juvenile dystrophy), poorly controlled epilepsy, some congenital malformation syndromes (Arnold-Chiari, Down, Ehlers-Danlos, Prades-Willi syndromes and mucopolysaccharidosis). The useful imaging test for evaluation of obstructive SDB predisposing factors is a lateral neck radiography showing the hypertrophy of lymphatic tissue of the pharynx and tonsils (high sensitivity, low specificity for OSAS); fiberoptic nasopharyngoscopy under local anesthesia instead of radiography to avoid radiation. Prematurity, OSAS and adenotonsillectomy in parents, OSAS and hypertrophy of the lymphatic tissue of the pharynx in siblings can be found in patients' medical history.

Step 2. Complications and comorbidities

The most significant cardiovascular complication is increased blood pressure (by about 3-5 mmHg, both systolic and diastolic). OSAS should be excluded in children with blood pressure higher than the 95th percentile. Children with severe OSAS can suffer from pulmonary hypertension and pulmonary heart disease, especially with concomitant congenital malformation syndromes.

Complications related to the nervous system are somnolence during the day (especially in obese patients), attention disorders, hyperactivity (regardless of the stage of obstructive SDB), deterioration of cognitive functions, general intelligence, eloquence, learning ability, memory, visual spatial orientation, analytical and mathematical abilities (mild obstructive SDB does not significantly influence these parameters). Connections between SDB and behavioral disorders such as aggression, worse social, adaptation, and communication skills, as well as depression, were also described.

There is a strong correlation between nocturnal enuresis and obstructive SDB exacerbation. The influence of SDB on growth inhibition below the 5th percentile for age and sex in terms of height and weight is also underlined.

The authors also describe the SBD comorbidities probably having the same pathomechanism. SDB treatment improves the course of comorbidities such as otitis media and/or insertion of a tympanostomy tube, recurrent episodes of wheezing, metabolic syndrome, oral motor dysfunctions accompanying hypertrophy of the lymphatic tissue of the pharynx (chewing, swallowing, articulation disorders).

Step 3. Predisposing factors of persistent SDB

These factors are: obesity and increased BMI values (calculated in percentiles), male sex, OSAS severity (AHI rate of more than 5 per hour), African-American origin, narrow mandible.

Tab. I. Definition of obstructive sleep disordered breathing and clinical units.

DEFINITIONS	CHARACTERISTICS OF CLINICAL UNITS
Obstructive sleep disordered breathing (SDB)	Upper airway sleep dysfunction syndrome with snoring and/or increased effort to breath caused by an increased airway resistance and a narrowing of the pharynx
Obstructive SDB – clinical units	
• Primary snoring	Habitual snoring (more than 3 nights a week) without apnea, shallow breathing, frequent awakening or gas exchange disorders.
• Upper airway resistance syndrome (UARS)	Snoring, increased effort to breath, frequent awakening without recognizable incidents of obstruction or gas exchange disorders.
• Obstructive hypoventilation	Snoring and incorrect increase of end expiratory partial CO_2 pressure without recognizable incidents of obstruction
• Obstructive sleep apnea syndrome (OSAS)	Recurrent incident of partial or complete upper airway obstruction (shallow breathing, obstructive or mixed apnea) with concomitant impaired oxygenation, ventilation and sleep architecture.

Step 4. Objective diagnosis and assessment of severity of SDB

Polysomnography or polygraphy is a test recommended in children with obstructive SDB before performing adenotonsillectomy. It can be applied in obese children, with cranial and facial deformities, neuromuscular disorders, congenital malformation syndromes and when clear indications for treatment cannot be found. Polysomnography or polygraphy is also recommended after adenotonsillectomy in patients with persistent OSAS symptoms (despite surgical treatment), with preoperative moderate or severe OSAS, obesity, with cranial and facial deformities, nauromuscular disorders and congenital malformation syndromes. Polysomnography or polygraphy should be also performed before and after orthodontic treatment (jaw expansion, dental braces), continuous positive airway pressure (CPAP) or noninvasive positive pressure ventilation (NPPV) treatment.

The authors present two definitions of OSAS: 1) The symptoms of SDB with 2 or more obstructive AHI per hour or with 1 or more obstructive apnea index (calculated on a total sleep time). 2) The symptoms of SDB with 1 or more obstructive or central AHI per hour (definition often occurring in the literature).

Clinical significance of distinguishing primary snoring from upper airway resistance syndrome (UARS) remains unknown. The effects of treatment are also unclear. A positive correlation between SDB severity and increased blood pressure and enuresis is observed but SDB severity does not correlate with somnolence, behavioral and cognitive disorders. The chances of resolution of moderate and severe OSAS without treatment are smaller than of mild OSAS. The probability of developing persistent OSAS after adenotonsillectomy increases with preoperative severity of the disease. The risk of postoperative respiratory complications also increases with the severity of the disease. It has been well proven that recovering from SDB with AHI > 5 is unlikely.

The best way to diagnose obstructive SBD is polysomnography. If it is not possible to perform polysomnography, outpatient polysomnography or (more frequently performed) polygraphy during sleep at patient's home (without the supervision of qualified staff) should be carried out. Sensitivity of these methods in OSAS diagnosing is estimated at 90%. Nocturnal pulse oximetry can also be used but it gives a lot of false negative or unclear results. Pediatric sleep questionnaire (PSQ) is a useful prognostic tool for obstructive AHI > 5, neurological complications and behavioral disorders associated with obstructive OSAS and to observe the improvement after adenotonsillectomy (Table II.). Its sensitivity and specificity in OSAS diagnostics is estimated at respectively 78% and 72%.

Step 5. Indications for treatment

AHI > 5 is an indication for treatment regardless of the complications. However, more restrictive values may also be considered as such an indication (obstructive AHI ≥ 2 and/or obstructive apnea index > 1). Based on the analysis of the literature the authors admitted that the effectiveness of adenotonsillectomy and polysomnographic parameters amelioration is better in patients with AHI > 5. The treatment can be beneficial in patients with AHI 1-5 % (less frequent nocturnal enuresis, acceleration of growth, weight gain also in obese patients) who also show cardiovascular and neurological complications, enuresis, inhibited or abnormal growth, worse quality of life, risk factors of persistent SDB.

In case of obstructive SDB suspicion when polysomnography is not available, the treatment should be considered if alternative diagnostic methods confirm OSAS or complications of SDB are found.

Tab. II. Pediatric sleep questionnaire (PSQ) (7).

WHILE SLEEPING, DOES YOUR CHILD...
· snore more than half of the time?
· always snore?
· snore loudly?
· have "heavy" or loud breathing?
· have troble breathing or struggle to breathe?
HAVE YOU EVER...
· seen your child stop breathing during the night?
DOES YOUR CHILD...
· tend to breathe through the mouth during the day?
· have a dry mouth on waking up in the morning?
· occasionally wet the bed?
· wake up feeling un-refreshed in the morning?
· have a problem with sleepiness during the day?
· Has a teacher or other supervisor commented that your child appears sleepy during the day?
· Is it hard to wake your child up in the morning?
· Does your child wake up with headaches in the morning?
· Did your child stop growing at a normal rate at any time since birth?
· Is your child overweight?
THE CHILD OFTEN...
· does not seem to listen when spoken to directly
· has difficulty organizing tasks
· is easily distracted by extraneous stimuli
· fidgets with hands or feet or squirms in the seat
· is "on the go" or often acts as if "driven by a motor"
· interrupts or intrudes on others (e.g. butts into conversation or games)

If eight or more statements are answered "yes", the result is positive.

The treatment of obstructive SDB should be applied especially in children with predispositions to upper airway obstruction: large craniofacial abnormalities, neuromuscular disorders, achondroplasia, Chiari and Down syndromes, mucopolysaccharidosis, Prader-Willi syndrome. There are different therapeutic approaches in different stages of the disease. The risk of developing pulmonary hypertension in these patients should be noted.

Some children with primary snoring can develop OSAS in a longer period, so they require annual assessment.

Step 6. Therapeutic approach

The authors describe multiple treatment options to completely solve the problem of SDB. Described below therapeutic methods can be used together depending on the severity and the causes of upper airway obstruction.

Significant weight reduction is an effective way of OSAS treatment in obese adolescents but the effectiveness of this approach has not been proven in children (obese or overweight). Intranasal application of glucocorticosteroids or montelukast for 6-12 weeks reduces the severity of mild and moderate OSAS. The response to this treatment is worse in obese and those over 6 years old. There are not sufficient data on this approach in SDB complications.

Adenotonsillectomy is indicated in children with OSAS and hypertrophy of the lymphatic tissue of the pharynx. This procedure improves the quality of life, relieves the symptoms of SDB and its complications. The biggest improvement in polysomnographic parameters is observed when AHI is larger than 5. The increased risk of persistent OSAS after tonsillectomy is observed in children with a severe form of the disease, obesity, bronchial asthma, inferior nasal turbinate hypertrophy, curvature of the nasal septum, mandibular retrognathia, 3rd and 4th class in Mallampati score, craniosynostosis, Down syndrome, achondroplasia, cerebral palsy, Prader-Willi syndrome. Local bleeding and upper airway occlusion are the most serious complications of the procedure. The patients with the highest risk of complications (AHI > 26; 3 or more desaturation episodes, obesity or underweight, age < 3 years old, neuromuscular disorders, craniofacial and genetic abnormalities) should stay in hospital under observation for at least one night after adenotonsillectomy. There are not sufficient data to compare the effectiveness of adenotonsillectomy and selective adenoidectomy or tonsillectomy.

Orthodontic treatment and jaw expansion can be performed in children with OSAS and jaw narrowing. Individually adjusted dental braces are indicated in malocclusion and retrognathia.

The indication for CPAP is persistent OSAS after adenotonsillectomy (AHI > 5), OSAS associated with obesity, craniofacial abnormalities and neuromuscular disorders. In case of nocturnal hypoventilation NPPV is preferred. The available data show the improvement of the quality of life and school achievements in children treated with positive pressure ventilation. The compliance with medical recommendations can be difficult therefore electronic monitoring of compliance is required. The side effects of recommended treatment are nasal congestion, nasal bleeding, facial erythema (skin contact with the mask), changes in the shape of the face.

In case of bostructive SDB in children with craniofacial deformities, surgical expansion of the upper airway is indicated

STEP 1: CHILD AT RISK FOR SDB IF (ONE OR MORE):
1.1 Symptoms of upper airway obstruction (snoring, apnoea, restless sleep, oral breathing)
1.2 Findings on exam (tonsillar hypertrophy, obesity, midface deficiency, mandibular hypoplasia, neuromuscular disorders, Down syndrome, Prader–Willi syndrome)
1.3 Objective findings related to SDB (lateral neck radiography, flexible nasopharyngoscopy, cephalometry, upper airway MRI or CT)
1.4 Prematurity or family history of SDB

STEP 2: RECOGNITION OF MORBIDITY AND CONDITIONS COEXISTING WITH SDB:
2.1 Morbidity
Cardiovascular system
a) Elevated blood pressure
b) Pulmonary hypertension and cor pulmonale
Central nervous system
a) Excessive daytime sleepiness
b) Inattention/hyperactivity
c) Cognitive deficits/academic difficulties
d) Behavioural problems
Enuresis and somatic growth delay or growth failure
Decreased quality of life
2.2 Conditions coexisting with SDB (probably common pathogenesis)
a) History of recurrent otitis media or tympanostomy tube placement
b) Recurrent wheezing or asthma
c) Metabolic syndrome
d) Oral-motor dysfunction

STEP 3: RECOGNITION OF FACTORS PREDICTING LONG-TERM PERSISTENCE OF SBD:
3.1
a) Obesity and increasing BMI percentile
b) Male sex
c) Obstructive AHI >5 episodes·h–1
d) African-American ethnicity
e) Untreated tonsillar hypertrophy, narrow mandible

STEP 4: OBJECTIVE DIAGNOSIS AND ASSESSMENT OF SDB SEVERITY:
4.1 PSG or polygraphy if child at risk for SDB (see steps 1 and 2)
4.2 OSAS-definition 1: SBD symptoms in combination with obstructive AHI ≥2 episodes·h–1 or obstructive apnoea index ≥1 episode·h–1; OSAS-Definition 2: SDB symptoms and AHI ≥1 episode·h–1(including central events)
4.3 If AHI >5 episodes·h–1 SDB unlikely to resolve spontaneously and child at risk for morbidity
4.4 If PSG or polygraphy not available: ambulatory PSG or polygraphy, nocturnal oximetry, Paediatric Sleep Questionnaire or Sleep Clinical Record

STEP 5: INDICATIONS FOR TREATMENT OF SDB:
5.1
a) AHI >5 episodes·h–1 irrespective of the presence of morbidity
b) Treatment may be beneficial if AHI 1–5 episodes·h–1 especially in the presence of: morbidity from the cardiovascular system (see 2.1); morbidity from the central nervous system (see 2.1); enuresis; somatic growth delay or growth failure; decreased quality of life; risk factors for SDB persistence (see 3)
c) If at risk for SDB and PSG or polygraphy not available, treatment is considered when positive oximetry or SDB questionnaires (see 4.4) or morbidity present
5.2 Unclear whether should treat primary snoring (evaluation annually)
5.3 OSAS treatment is a priority in the presence of: major craniofacial abnormalities; neuromuscular disorders; achondroplasia; Chiari malformation; Down syndrome; mucopolysaccharidoses; Prader–Willi syndrome

STEP 6: STEPWISE TREATMENT APPROACH TO SDB#:
6.1 A stepwise treatment approach (from 6.2 to 6.9) is usually implemented until complete resolution of SDB
6.2 Weight loss if the child is overweight or obese
6.3 Nasal corticosteroids and/or montelukast p.o.
6.4 Adenotonsillectomy
6.5 Unclear whether adenoidectomy or tonsillectomy alone are adequate
6.6 Rapid maxillary expansion or orthodontic appliances
6.7 CPAP or NPPV (for nocturnal hypoventilation)
6.8 Craniofacial surgery
6.9 Tracheostomy

STEP 7: RECOGNITION AND MANAGEMENT OF PERSISTENT SDB:
7.1
a) Outcomes monitored after intervention (6 weeks-12 months): symptoms, PSG, quality of life, cardiovascular or central nervous system morbidity, enuresis, growth rate
b) If PSG not available: polygraphy, oximetry/capnography
c) PSG ≥6 weeks after adenotonsillectomy (persistent SDB symptoms or at risk of persistent OSAS preoperatively); after 12 weeks of montelukast/nasal steroid
d) PSG after 12 months of rapid maxillary expansion (earlier if symptoms persist) and after 6 months with an oral appliance
e) PSG for titration of CPAP, NPPV and then annually; PSG as predictor of successful decannulation with tracheostomy
f) Airway re-evaluation by nasopharyngoscopy, drug-induced sleep endoscopy, MRI

Fig. 1. Gradual diagnosis and approach in obstructive sleep disordered breathing in children from 2 to 18 years old; the guidelines of the European Respiratory Society Task Force (1).

to improve polysomnographic parameters, the quality of life, to avoid tracheostomy or to remove tracheostomy tube. The complications of the procedure are rare but very severe. There can be listed: cerebrospinal fluid leak, infections, perforation of the palate, open bite malocclusion, nerve damage. The most effective method of obstructive SDB treatment is tracheostomy. It should be performed only in the most severe OSAS cases, when other non-surgical and surgical methods of treatment have failed or are contraindicated. Unfortunately, tracheostomy worsens the quality of life and influences negatively psychosocial development. Early complications of the procedure such as pneumothorax, penumomediastinum, infection, bleeding occur in 5% of patients. Late complications such as granulation tissue, tracheocutaneous fistulas, larynx and/or trachea occlusion occur in 40% of patients. The blockage (e.g. by mucosa) or accidental protrusion of tracheostomy tube may lead to a life-threatening condition. The mortality associated with tracheostomy is estimated at 3% or less. Respiratory infections are very frequent.

Step 7. Further proceeding

Six weeks after treatment, a 12-month follow-up is usually required, and the following parameters are evaluated: symptoms, polysomnography results (objective assessment of SDB severity), quality of life, cardiovascular and neurological complications, enuresis, growth. If polysomnography is unavailable, polygraphy, oximetry or capnography should be performed. The majority of the studies suggest repeating polysomnography or polygraphy 6 weeks after adenotonsillectomy (if symptoms persist or in case of preoperative risk of developing persistent SDB), 12 weeks after montelukast /intranasal glucocorticosteroid treatment and also 12 months after jaw expansion (earlier if symptoms of SDB persist), 6 months after using dental braces, during CPAP, NPPV regulation (subsequently once a year) to evaluate the probability of successful removal of the tracheostomy tube. Nasopharyngoscopy, drug-induced sleep endoscopy or magnetic resonance imaging can be performed in persistent SDB to diagnose upper airway disorders.

SUMMARY

Obstructive sleep disordered breathing occurs in a few percent of pediatric population. Considering this epidemiology, every pediatrician has a child with DSB among his/her patients. SDB can cause multiple complications influencing physical and psychological development. The anticipatory approach preferred so far can lead to huge damage of a young organism. Moreover, clinicians' awareness of the disease in Poland is very small [8]. European Respiratory Society Task Force published a document in a form of seven steps referring to the obstructive sleep disordered breathing and detailed therapeutic approach. If eight or more statements are answered "yes", the result is positive.

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