

OSAHS

Evaluation of neurocognitive abilities in children affected by obstructive sleep apnea syndrome before and after adenotonsillectomy

Valutazione delle abilità neurocognitive in bambini affetti da sindrome delle apnee ostruttive in sonno prima e dopo adenotonsillectomia

Domenico Testa¹, Marco Carotenuto², Francesco Precenzano², Alessia Russo¹, Anna Donadio¹, Giuseppina Marcuccio¹, Gaetano Motta¹

¹ Otolaryngology, Head and Neck Surgery, Department of General and Specialistic Surgery, University of Campania "Luigi Vanvitelli", Italy; ² Clinic of Child and Adolescent Neuropsychiatry, Department of Mental Health, Physical and Preventive Medicine, University of Campania "Luigi Vanvitelli", Italy

SUMMARY

Obstructive sleep apnoea syndrome (OSAS) is the most severe form of sleep-related disordered breathing (SRDB) and is characterised by snoring, apnoeas, and/or hypopnoeas associated to hypoxia, hypercarbia, or repeated arousals from sleep. OSAS has three major categories of morbidities: neurobehavioural, cardiovascular and somatic growth failure. The gold standard for objective diagnosis of obstructive-SRDB severity is polysomnography (PSG). The indication for surgical treatment in children is moderate-severe OSAS (AHI, apnoea hypopnoea index > 5/h) and in mild OSAS (AHI 2-5/h) with complications or morbidity. The entire spectrum of PSG-defined SRDB (ranging from Primary Snoring to severe OSAS) may correlate with behavioural, attentional and executive function deficits relating to hypoxia and sleep disruption: in some cases, these alterations may mimic attention deficit hyperactivity disorder (ADHD). The aim of this research was to evaluate visuoperceptual and constructional abilities, paediatric sleep questionnaire and polysomnographic scores before and 6 months after adenotonsillectomy with objective and subjective information. We included 59 children who underwent neuropsychiatric and otolaryngologist clinical evaluation and the Beery-Buktenica Developmental Test of Visual-Motor Integration (VMI); children parents were asked to fill in the Paediatric Sleep Questionnaire (PSQ); each child underwent PSG. At 6 months after adenotonsillectomy, all patients were evaluated again. There is a significant difference in PSQ parameters, VMI standard, visual tests scores and PSG parameters before and after adenotonsillectomy in children affected by OSAS. These results showed the achievement of therapeutic benefits with improvement of the quality of life for both children and their parents.

KEY WORDS: OSAS, adenotonsillectomy, pediatric OSAS, neuropsychological non-verbal skills

RIASSUNTO

La Sindrome delle Apnee Ostruttive del Sonno (OSAS, Obstructive Sleep Apnea Syndrome) è la forma più severa di Disturbo Respiratorio Sonno-correlato (SRDB, Sleep-related Disordered Breathing) ed è caratterizzata da russamento, apnee, e/o ipopnee associate a ipossia, ipercapnia, o ripetuti risvegli dal sonno (arousals). L'OSAS include tre categorie di morbidità: neurocomportamentali, cardiovascolari, disturbi della crescita somatica. Il gold standard per la diagnosi oggettiva di severità del SRDB ostruttivo è rappresentato dalla polisonnografia (PSG). L'indicazione al trattamento chirurgico nei bambini è la diagnosi di OSAS moderata-severa (indice di apnea-ipopnea > 5/h) e di OSAS lieve (indice di apnea-ipopnea 2-5/h) se sono presenti complicanze o comorbidità. Lo spettro intero di SRDB definiti mediante PSG (dal Russamento Primario all'OSAS severa) potrebbe correlare con deficit comportamentali, attentive e delle funzioni esecutive dovuti all'ipossia ed alla frammentazione del sonno: in alcuni casi, queste alterazioni possono mimare il Disturbo da deficit di Attenzione con Iperattività (ADHD, Attention Deficit Hyperactivity Disorder). Lo

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Correspondence

Giuseppina Marcuccio

Via Fontana 7, 81010 Castel Campagnano (CE), Italy

E-mail: giuseppina_marcuccio@hotmail.it

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Conflict of interest

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scopo di questa ricerca è stato quello di valutare le abilità visuoperceptive e costruttive, i punteggi al Pediatric Sleep Questionnaire ed alla PSG prima e dopo l'intervento di adenotonsillectomia fornendo informazioni oggettive e soggettive. Abbiamo incluso in questo studio 59 bambini sottoposti a valutazione clinica otorinolaringoiatrica e neuropsichiatrica e poi al test di Beery-Buktenica Developmental Test of Visual-Motor Integration (VMI) ed ai genitori veniva chiesto di compilare il Pediatric Sleep Questionnaire (PSQ); ogni bambino veniva sottoposto a PSG. Dopo 6 mesi dall'intervento di adenotonsillectomia sono state ripetute tutte le valutazioni come al tempo 0. L'analisi statistica evidenzia risultati significativi al PSQ, VMI standard e visivo ed ai parametri polisomnografici se confrontati prima e dopo l'intervento chirurgico di adenotonsillectomia nei bambini affetti da OSAS moderata-severa. Questi risultati hanno mostrato il raggiungimento dei vantaggi terapeutici attraverso il miglioramento della qualità della vita dei bambini e dei loro genitori.

PAROLE CHIAVE: OSAS, adenotonsillectomia, OSAS pediatrica, abilità neuropsicologiche non verbali

Introduction

Sleep-related disordered breathing (SRDB), pathologic nocturnal respiratory functioning, includes clinical conditions that range from primary snoring (PS) to obstructive sleep apnoea syndrome (OSAS) and represents one of the most common sleep disorders in childhood affecting up to one-third of children (prevalence up to 34.5%)^{1,2}.

OSAS is believed to be present in about 1% to 3% in children aged 2 to 18 with no gender predominance; furthermore, chronic snoring may be present in more than 10% of children³⁻⁵. PS and OSAS are the two extreme conditions of a wide spectrum of increased resistance in the upper airways³⁻⁵.

PS is the mildest form of SRDB and is defined as habitual snoring without discrete respiratory events, gas exchange abnormalities, or evidence of sleep fragmentation; OSAS is the most severe form of SRDB and is characterised by snoring, apnoeas, and/or hypopnoeas associated with hypoxia, hypercarbia, or repeated arousals from sleep⁶.

OSAS has three major categories of morbidities: neurobehavioural, cardiovascular and somatic growth failure^{7,8}. Symptoms in children with OSAS include snoring, breathing difficulty and/or breathing pauses during sleep, excessive sweating and enuresis; daytime symptoms may include oral breathing, headaches and excessive sleepiness, behavioural and neurocognitive changes such as attention deficit, hyperactivity, irritability, learning difficulties, memory loss and intelligence alterations⁹⁻¹². In adults but less so in children, OSAS is associated with arterial systemic hypertension due to alterations in the renin-angiotensin axis secondary to hypoxia during sleep; left ventricular wall thickness; pulmonary vascular hypertension^{13,14}. The gold standard for diagnosing OSAS is overnight polysomnography (PSG) which gives information about the frequency and severity of respiratory events and associated blood gas changes¹⁵.

The European Respiratory Society (ERS) Task Force on diagnosis and management of obstructive SRDB in childhood (2 to 18 years old) analysed selected evidence

from 362 articles since prospective cohort studies describing its clinical topics and randomised, double-blind, placebo-controlled trials, regarding its treatments, are scarce¹⁶. Kaditis et al. summarised the conclusions of the ERS Task Force indicating seven steps for diagnosis and management of SRDB in children (Tab. I)^{16,17}.

Some clinical conditions can be considered as factors predicting long-term persistence of obstructive SRDB: obesity and increasing BMI percentile, male sex, severity of OSAS (AHI > 5 episodes/h), African-American Ethnicity and persistent tonsillar hypertrophy with a narrow mandible^{18,19}. The gold standard for objective diagnosis of obstructive-SRDB severity is PSG and is indicated in children candidate to adenotonsillectomy especially in the presence of obesity, craniofacial deformities, neuromuscular disorders, complex abnormalities such as Chiari malformation, Down syndrome and Prader-Willi syndrome or when the need for treatment is unclear^{20,21}. Furthermore, PSG is indicated before and after rapid maxillary expansion or application of oral appliances, continuous positive airway pressure (CPAP) or noninvasive positive pressure ventilation (NPPV) treatments and when symptoms of OSAS persist after therapy²⁰.

The AHI, or the number of mixed, obstructive or central apnoeas and hypopnoeas per hour of total sleep time, is the most commonly used parameter to describe the severity of SRDB²².

In children without SRDB symptoms or associated morbidities, the 90th percentile for the AHI according to the American Academy of Sleep Medicine (AASM 2007 scoring rules) is 3.2 episodes/h for the second year of life, up to 2.5 episodes/h for the ages > 2 and ≤ 6 years, and up to 2.1 episodes/h for the ages > 6 and < 18 years²³. OSAS is defined when AHI ≥ 2 episodes/h or in presence of SRDB symptoms when AHI ≥ 1 episode/h: mild OSAS if AHI 2-5 episodes/h; moderate-severe OSAS if AHI > 5 episodes/h (Tab. II)^{18,23}.

Indications for treatment in children is moderate-severe OSAS (AHI > 5/h) or mild OSAS (AHI 2-3/h) only when morbidities are present¹⁸. The need for combined treatments (adenotonsillectomy when needed, weight loss, CPAP,

Table 1. A stepwise approach to the diagnosis and management of obstructive SRDB in 2-18-year-old children (from Kaditis et al., 2016¹⁶; Kaditis et al., 2012¹⁷).

Step 1	Child at risk of SDB if (one or more) <ol style="list-style-type: none"> 1.1. Symptoms of upper airway obstruction (snoring, apnoea, restless sleep, oral breathing) 1.2. Finding 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 SRDB <ol style="list-style-type: none"> 2.1. Morbidity <ul style="list-style-type: none"> • Cardiovascular system: <ul style="list-style-type: none"> – elevated blood pressure – pulmonary hypertension and cor pulmonale • Central nervous system: <ul style="list-style-type: none"> – excessive daytime sleepiness – inattention/hyperactivity – cognitive deficits/academic difficulties – behavioural problems – enuresis and somatic growth delay or growth failure – decreased quality of life • Conditions coexisting with SRDB (probably common pathogenesis): <ul style="list-style-type: none"> – history of recurrent otitis media or tympanostomy tube placement – recurrent wheezing or asthma – metabolic syndrome – oral-motor dysfunction
Step 3	Recognition factors predicting long-term persistence of SDB <ol style="list-style-type: none"> 3.1. <ul style="list-style-type: none"> • Obesity and increasing BMI percentile • Male sex • Obstructive AHI > 5 episodes/h • African-American ethnicity • Untreated tonsillar hypertrophy, narrow mandible
Step 4	Objective diagnosis and assessment of SDB severity <ol style="list-style-type: none"> 4.1. PSG or polygraphy if child at risk for SDB (step 1 and 2) 4.2. <ul style="list-style-type: none"> • OSAS-definition 1: SDB symptoms in combination with obstructive AHI \geq 2 episodes/h or obstructive apnea index \geq 1 episodes/h • OSAS-definition 2: SDB symptoms and AHI \geq 1 episodes/h (including central events) 4.3. If AHI \geq 5 episodes/h 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	Indication for treatment of SDB <ol style="list-style-type: none"> 5.1. <ul style="list-style-type: none"> • AHI > 5 episodes/h irrespective of the presence of morbidity • Treatment may be beneficial if AHI 1-5 episodes/h especially in the presence of morbidity from the cardiovascular system (see 2.1.); 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 step 3) • If at risk for SDB and PSG or polygraphy not available, treatment is considered when positive oximetry or SDB questionnaire (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 <ol style="list-style-type: none"> 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 6.4. Adenotonsillectomy 6.5. Unclear whether adenoidectomy or tonsillectomy alone are adequate 6.6. Rapid maxillary expansion and orthodontic appliances 6.7. CPAP or NPPV (for nocturnal hypoventilation) 6.8. Craniofacial surgery 6.9. Tracheostomy

Continues

Table I. *Follows.*

Step 7	Recognition and management of persistent SDB
7.1.	<ul style="list-style-type: none"> • Outcomes monitored after intervention (6 weeks - 12 months): symptom, PSG, quality of life, cardiovascular or central nervous system morbidity, enuresis, growth rate • If PSG not available: polygraphy, oxymetry/captography • PSG after 6 weeks after adenotonsillectomy (persistent SDB symptom or at risk of persistent OSAS preoperatively); after 12 weeks of montelukast/nasal steroids • PSG after 12 months of rapid maxillary expansion (earlier if symptoms persist) and after 6 months with an oral appliance • PSG for titration of CPAP, NPPV and then annually; PSG as predictor of successful decannulation with tracheostomy • Airway re-evaluation by nasopharyngoscopy, drug-induced sleep endoscopy, MRI

MRI: magnetic resonance imaging; CT: computed tomography; BMI: body mass index; AHI: apnoea-hypopnoea index; PSG: polysomnography; OSAS: obstructive sleep apnoea syndrome; CPAP: continuous positive airway pressure; NPPV: noninvasive positive pressure ventilation.

Table II. Definition of OSAS from AHI parameter (from Marcus et al., 2013¹⁸; Iber et al., 2007²³).

Normal values or OSAS definition	AHI (episodes/h)
No SRDB symptoms or morbidity	3.2 AHI (2 years old) (90 th percentile) 2.5 AHI (2-6 years old) (90 th percentile) 2.1 AHI (6-18 years old) (90 th percentile)
SRDB symptoms and/or morbidity	
Mild OSAS	2-5 AHI
Moderate-severe OSAS	> 5 AHI

NPPV) is a priority in these clinical conditions: major craniofacial abnormalities, neuromuscular disorders (i.e. Duchenne muscular dystrophy), achondroplasia, Chiari malformation, Down syndrome, mucopolysaccharidoses, Prader-Willi syndrome²⁴⁻²⁶.

The entire spectrum of PSG-defined SRDB (ranging from PS to severe OSAS) may correlate with behavioural, attentional and executive function deficits relating to hypoxia and sleep disruption²⁷.

It is important to emphasise the risk of greater impairment of both behavioural and cognitive functions in children with milder forms of SRDB so that PS is not a universally benign condition²⁸. Preschool children with obstructive SRDB are more vulnerable in their adaptative and behavioural function vs. cognitive function: this age has a sort of ‘*window of opportunity*’ for early treatment by preventing cognitive deficits arising later in childhood²⁸.

Some authors analysed neurocognitive function in children with OSAS after CPAP-treatment (5-6 months): significant improvements in their abilities to analyse and synthesise abstract information, multitasking, simultaneous processing, divided attention and in recognition and recall for visual information were all shown²⁹.

Sleep disruption, even without respiratory compromise, may induce neurocognitive alterations, and for this reason the severity of SRDB, from PS to OSAS, does not correlate with neurocognitive deficits³⁰.

Cognitive functions are correlated with sleep fragmentation and thus to SRDB: arousals seem to be an important

defensive mechanism against sleep fragmentations induced by SRDB³¹. Children with high levels of arousal seem to present a high degree of protection against cognitive consequences of SRDB³².

The aim of the present study is to analyse changes in neurocognitive and behavioural functions in children 4 to 11 years old affected by moderate-severe OSAS, after surgical treatment with adenotonsillectomy to investigate the role of obstruction in SRDB symptoms.

We evaluated visuoperceptual and constructional abilities, PSQ and full overnight polysomnographic values, before and 6 months after adenotonsillectomy, giving objective and subjective information about sleep disturbances in OSAS.

Materials and methods

We enrolled 86 children with obstructive SRDB, aged between 4 to 11 years old, referred by primary care paediatricians for snoring and suspected apnoeas to the Clinic of Child and Adolescent Neuropsychiatry and then to the Ear Nose and Throat Unit of the University of Campania “Luigi Vanvitelli” between October 2015 and April 2018. Each child underwent neuropsychiatric and otolaryngologic clinical evaluation and VMI test, and parents were asked to fill in the PSQ.

Three overnight full sleep PSG were performed, and obstructive AHI were identified as normal, mild, moderate, or severe OSAS.

Inclusion criteria were: history of habitual snoring and/or apnea; frequent and continuous breathing pauses referred by parents; nocturnal difficult breathing; daytime hyperactivity; attentive deficit, poor school performance. Moreover, children with moderate (respiratory disturbance index of 5-10 episodes/h with average SaO₂ > 95%) or severe (respiratory disturbance index >10 episodes/h with an average < 95% SaO₂) OSAS were included; tonsillar and adenoidal grading 3-4, clinical febrile episodes (FE) and frequent acute pharyngotonsillitis²; age between 4 and 11 years.

Children were excluded if they had any of the following: sensorineural hearing loss; tube-tympanic alterations (OME

otitis media with effusion, acute otitis media recurrent AOM); nasal obstruction due to nasal septal deviation or hypertrophy of inferior and middle turbinates, obesity (BMI \geq 90th percentile); cardiac and pulmonary metabolic disorders; craniofacial anomalies; neuromuscular disorders and genetic syndromes.

During the first step of enrollment, we excluded 27 children:

- 15 did not meet inclusion criteria (4 obese children, BMI \geq 90th percentile; 2 affected by otitis media with effusions associated to conductive hearing loss; 1 affected by craniofacial anomalies, Pierre Robin Sequence and 8 children affected by mild OSA (AHI < 2);
- 10 children declined to participate;
- 2 children for other reasons.

At the end of this evaluation, we included 59 children affected by moderate-severe OSA (AHI > 5) with adenotonsillar hypertrophy.

The second step of treatment included adenotonsillectomy

performed in the Ear Nose and Throat Unit of University of Campania “Luigi Vanvitelli”.

The third step included follow-up at 6 months after surgery: children underwent sleep PSG, VMI test and their parents filled in the PSQ. During this step, 22 patients refused to continue.

The last step of the analysis of data included 37 children mean age 8.44 ± 2.26 (Fig. 1).

Paediatric Sleep Questionnaire

PSQ is a SRBD scale questionnaire for children parents containing 22 items, indicating the presence of apnoeas. These items regard frequency, loud snoring, observed apnoeas, alterations in breathing during sleep, daytime symptoms and signs such as sleepiness, inattentive and hyperactive behaviour. Responses are “yes” (= 1 score), “no” (= 0 score), and “do not know” (= missing). A cut-off value of 0.33, which would be most effective in identifying pediatric OSA, was used.

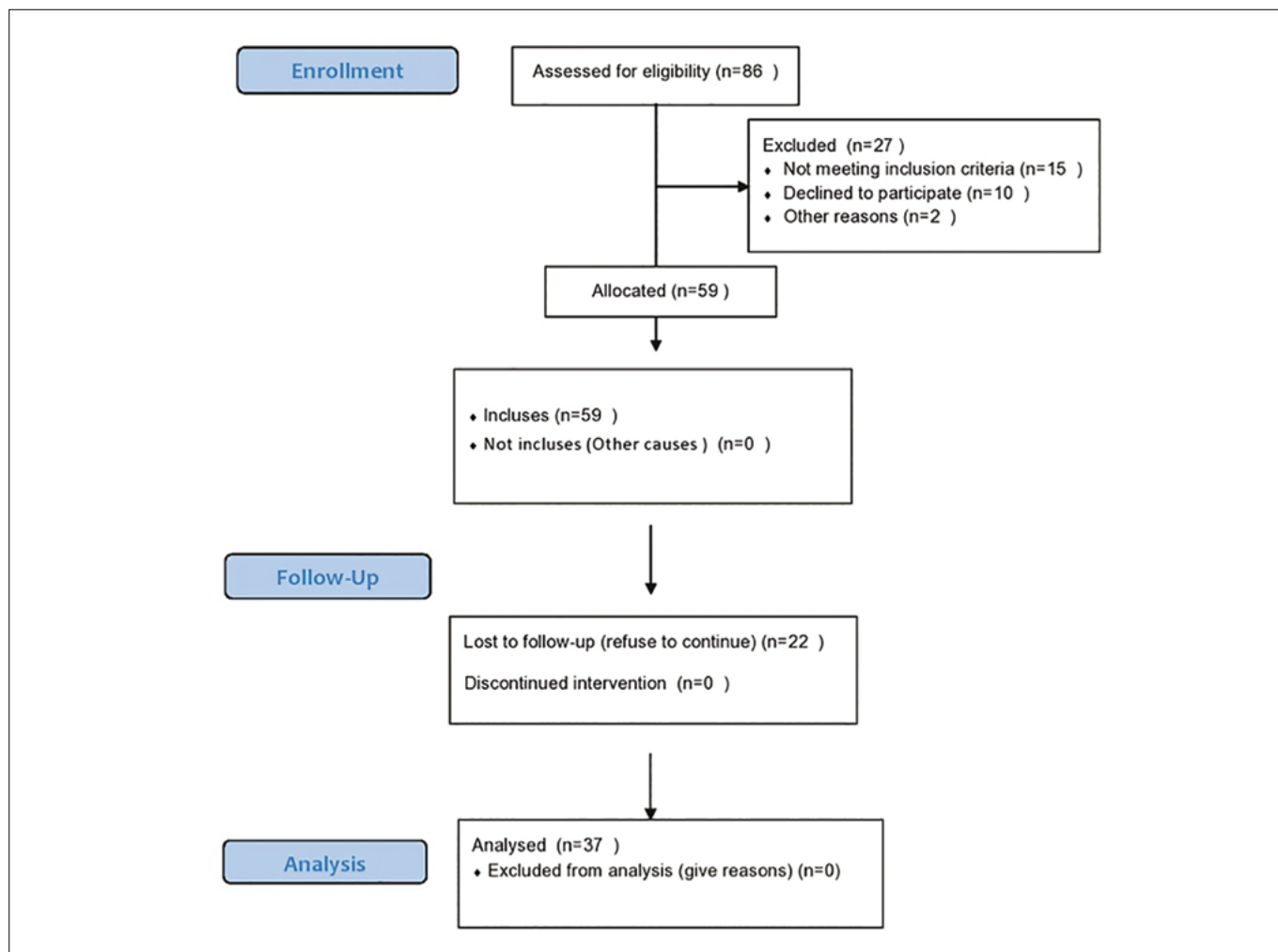


Figure 1. Enrollment and study flow.

Beery-Buktenica Developmental Test of Visual-Motor Integration (VMI)

The Beery-Buktenica Developmental Test of Visual-Motor Integration Performances in Children (VMI task) is a paper-and-pencil test in which children have to imitate or copy up to 27 geometric forms with increasing complexity using paper and pencil³². The test was stopped when a child made more than two errors in a row³². Copying errors were marked if they reflected problems in fine motor coordination and a pure visuospatial problem. The Beery VMI task is specifically designed for children and takes about 10 minutes to complete³². The Beery VMI scores were standardised for age and gender using normative data for the Italian general population³².

The percentile scores were used for diagnosing the visuomotor abnormalities in our sample. A value less than or equal to the 5th percentile was considered to indicate VMI impairment.

Overnight full polysomnography recordings (PSGs)

The polysomnographic recording was performed using a videorecorder connected to probes, electrodes and bands suitable to the patients' age and weight. The exam is computerised and uses the Embletta System, a system recognised and appointed by the American Academy of Sleep Medicine for the study of sleep apnoea. The recording time is 6-8 hours, and supervised by medical and technical staff, who interpreted the data collected. We analysed sleep and respiratory parameters:

- Sleep Latency, SL (min);
- REM Latency, RL (min);
- Total Sleep Time, TST (min);
- Sleep Period Time, SPT or TIB-SL (min);
- Wake After Sleep Onset, WASO (min);
- 1-2-3 NREM, N1-N2-N3 (%TST);
- REM (%TST);
- Sleep Efficiency, SE or TST/TIB% (%);
- Arousal Index, AI (n Arousal/h sleep);
- Periodic Limb Movement Index, PLMI (n PLM/ h Sleep);
- AHI (number of AH/h sleep)
- Nadir SaO₂ during sleep;
- Nadir SaO₂ mean during sleep;
- Oxygen Desaturation Index, ODI (n desaturation/h sleep);
- TST % with SaO₂ < 90%;
- TST % with SaO₂ < 80%.

ENT evaluation

Each child underwent ear, nose and throat evaluation characterised by otoscopy, anterior rhinoscopy and oropharyngoscopy with particular attention to febrile

episodes per year in the last 3 years. We evaluated haemochrome, antistreptolysin test, erythrocyte sedimentation rate, and creatine phosphokinase for all children. These clinical parameters are important to understand the indication for surgical treatment in children with adeno-tonsillar hypertrophy.

The tonsils were subjectively measured using a grading system. In grade I, the tonsils were hidden in the tonsillar fossa and were barely visible behind the anterior pillars. In grade II, the tonsils were visible behind the anterior pillars and occupied up to 50% of the pharyngeal space (the distance between the medial borders of the anterior pillars). In grade III, the tonsils occupied between 50 and 75% of the pharyngeal space. In grade IV, the tonsils occupied more than 75% of the pharyngeal space.

Adenoids were analysed during rhinofiberoptic evaluation: a grading system for adenoid hypertrophy was created based on the anatomical relationships between the adenoid tissue and the vomer, soft palate, and torus tubaris. The grading is based on the relationship of the adenoids to adjacent structures when the patient is at rest (i.e., when the soft palate is not elevated).

Adenotonsillectomy

Patients underwent general anaesthesia in oral intubation maintaining supine in the Rose position. Adenoidectomy was performed followed by extracapsular tonsillectomy using cold/hot technique for dissection and cold/hot for homeostasis^{33,34}.

This study was conducted according to the World Medical Association Declaration of Helsinki and was retrospectively registered with number 28/2018.

Statistical analysis

Nonparametric analyses using the Wilcoxon Test were used to evaluate the effects of treatment (pre- and post-adenotonsillectomy) in the variables examined: PSQ test scores, VMI test standard scores (VMI, test motor and visual test), PSGs parameters (TIB, SPT, TST, SOL, FRL, SS-h, Awn-h, SE%, WASO-min, N1-min, N2-min, N3-min, REM-min, WASO spt, N1-spt, N2-spt, N3-spt, REM spt, N1-tst, N2-tst, N3-tst, REM-tst, PSQ, AHI, ODI, OD%, PLMI).

Pearson's correlation was used to assess the mean preoperative and postoperative VMI scores with preoperative and postoperative polysomnographic parameters. The threshold for statistical significance was $p < 0.05$. All statistical analyses were performed with a statistical software package (STATISTICA 8.0, StatSoft Inc.)

Results

The data collected before and 6 months after adenotonsillectomy are indicated in Tables III and IV. Table III shows the results of PSQ and VMI tests. There was a significant difference for PSQ parameters, VMI standard and VMI visual test ($p < 0.05$), but not for the VMI motor test.

In Table IV PSG parameters before and after adenotonsillectomy are shown. There was a significant difference for macrostructural sleep and respiratory parameters TIB, SPT, TST, SOL, SS-h, AWN-h, SE%, WASO-min, N1-min, N2-min, N3-min, REM-min, WASO spt, N1-spt, N2-spt, N3-spt, N1-tst, N2-tst, N3-tst, PSQ, AHI, ODI, OD%, PLMI ($p < 0.05$).

Table III. Comparison of PSQ and VMI scores before and after adenotonsillectomy.

	Pre		Post		Wilcoxon Test		
	Mean (DS)		Mean (DS)		U	Z	p
PSQ	0.37 (0.15)		0.15 (0.07)		0.00	5.011926	0.000001
VMI st	106.42 (17.84)		111.24 (13.32)		61.50000	4.375023	0.000012
VMI motor test st	103.87 (25.39)		114.35 (17.67)		60.00000	1.679970	0.092964
VMI visual test st	97.78 (15.82)		107.43 (14.61)		24.00000	4.854568	0.000001

Table IV. Comparison of PSG parameters before and after adenotonsillectomy.

	Pre		Post		Wilcoxon Test		
	Mean	DS	Mean	DS	U	Z	p
TIB-min	386.4057	83.37142	589.1892	86.07090	0.00	5.302829	0.000000
SPT-min	342.3486	64.93097	555.4730	75.34052	0.00	5.302829	0.000000
TST-min	269.0622	56.76741	529.8784	70.22715	0.00	5.302829	0.000000
SOL-min	44.0570	43.74958	24.6757	18.74656	211.0000	2.119623	0.034039
FRL-min	104.2919	31.16452	130.0405	54.27404	220.0000	1.983847	0.047274
SS-h	9.7838	2.59235	7.5243	3.41121	162.0000	2.858851	0.004252
AWN-h	8.7324	2.60502	1.6486	1.86467	0.00	5.302829	0.000000
SE%	70.3054	9.19560	90.2514	5.60063	3.000000	5.257571	0.000000
WASO-min	73.2865	23.48791	25.5946	26.47479	23.50000	4.862423	0.000001
N1-min	57.1135	23.98776	17.3514	21.95572	46.00000	4.608860	0.000004
N2-min	88.5378	33.91517	231.5946	46.38110	0.00	5.302829	0.000000
N3-min	57.7568	25.12663	162.1622	69.83563	1.000000	5.287743	0.000000
REM-min	65.6541	23.08347	118.6892	32.43141	19.00000	5.016190	0.000001
WASO-spt	21.5716	6.21865	4.4541	4.44607	2.000000	5.272657	0.000000
N1-spt	16.7676	6.79174	3.1730	3.93211	9.000000	5.167053	0.000000
N2-spt	25.6962	8.50912	42.1000	7.81466	12.00000	5.121794	0.000000
N3-spt	16.8414	6.54840	28.6946	9.68389	31.00000	4.835155	0.000001
REM-spt	19.1227	6.26117	21.5595	5.89545	245.0000	1.606689	0.108124
N1-tst	21.7743	9.65647	3.4189	4.38209	8.000000	5.182139	0.000000
N2-tst	32.3411	9.17601	44.0432	7.74441	63.00000	4.352393	0.000013
N3-tst	21.4795	8.15687	30.0432	10.01681	106.0000	3.703683	0.000213
REM-tst	24.4041	7.89300	22.5108	5.83423	284.0000	1.018324	0.308525
AHI	10.3378	3.01786	5.9216	1.30110	11.00000	5.136880	0.000000
ODI	5.8270	1.81974	3.5027	0.55752	12.00000	5.043095	0.000000
Mean OD%	94.4865	1.97120	96.8000	0.73749	7.000000	5.197225	0.000000
Lowest OD%	89.3405	2.96287	94.3568	0.93290	0.00	5.231621	0.000000
Average OD%	5.1459	2.34338	2.4432	0.73543	5.000000	5.227398	0.000000
PLMI	6.5062	2.31283	3.1746	1.14721	29.00000	4.865327	0.000001

Clinical evaluation at PSQ six months after adenotonsillectomy showed a reduction in values compared with those before surgery (Fig. 2).

VMI standard scores, VMI motor test and VMI visual test scores showed improvements in performance at 6 months after surgery, which was significant for standard and visual scores (Figs. 3, 4) and not significant for motor scores (Fig. 5).

Spearman rank order correlation between preoperative

and postoperative VMI scores and preoperative and postoperative PSG parameters was statistically significant $p < 0.05$ (Tab. V): there was no linear correlation. There was a significant difference for PSQ parameters, VMI standard, visual tests scores and PSG parameters, but not for motor test scores before and after adenotonsillectomy in children affected by OSAS. When comparing VMI standard, motor and visual tests score with PSG parameters, the difference before and after surgery was statistically significant.

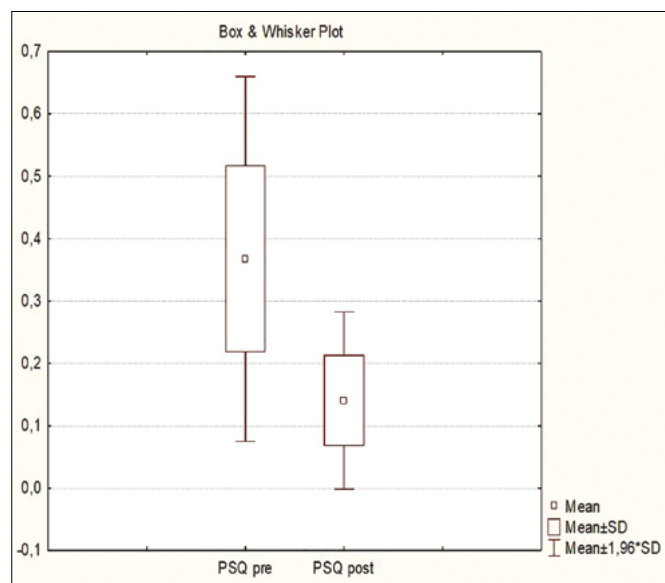


Figure 2. Comparison of PSQ scores before and six months after adenotonsillectomy.

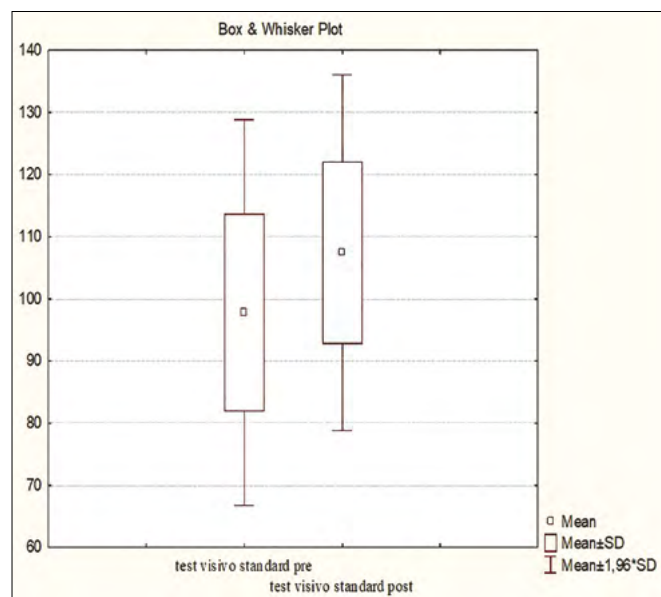


Figure 4. Comparison of VMI visual test scores before and six months after adenotonsillectomy (statistically significant).

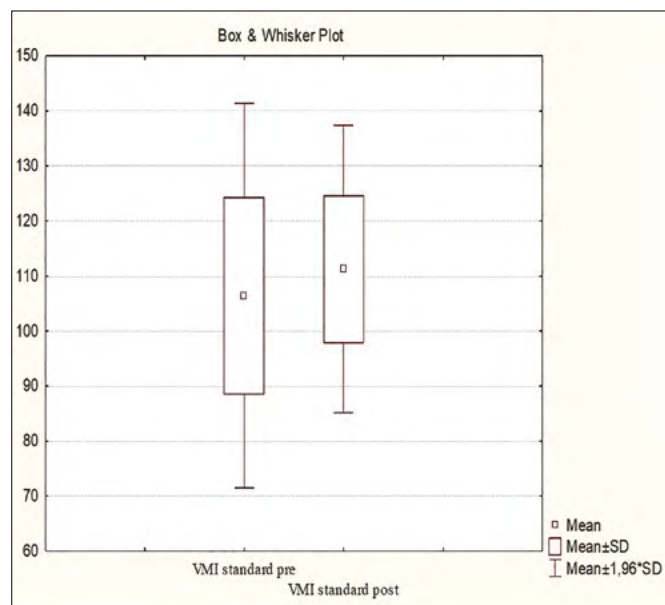


Figure 3. Comparison of VMI standard scores before and six months after adenotonsillectomy (statistically significant).

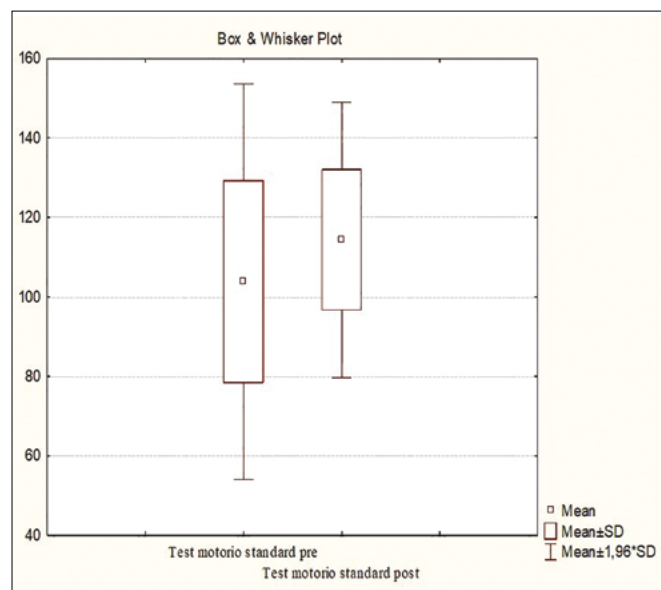


Figure 5. Comparison of VMI motor test scores before and six months after adenotonsillectomy (not statistically significant).

Table V. Correlation between VMI scores and PSG parameters before and 6 months after adenotonsillectomy.

	Spearman Rank Order Correlations MD pairwise deleted Marked correlations are significant at $p < 0.05000$		
	VMI st	VMI motor test st	VMI visual test st
TIB-min	0.144274	-0.097321	-0.116585
SPT-min	0.211697	-0.059812	-0.108225
TST-min	0.167254	-0.058115	-0.099394
SOL-min	-0.132756	-0.016344	-0.042546
FRL-min	-0.068619	-0.072612	0.044218
SS-h	0.097221	-0.171699	-0.155499
AWN-h	0.110438	-0.014303	-0.214789
SE%	0.063701	0.049596	0.152116
WASO-min	0.132909	-0.046434	-0.173408
N1-min	-0.018648	0.093144	-0.037976
N2-min	0.099488	0.025531	0.027575
N3-min	0.197135	-0.080507	-0.121304
REM-min	-0.039059	-0.070151	0.111569
WASO-spt	0.023792	-0.038759	-0.101054
N1-spt	-0.053335	0.119138	0.011890
N2-spt	-0.035937	0.064147	0.116147
N3-spt	0.170595	-0.085442	-0.132817
REM-spt	-0.203929	-0.072725	0.220432
N1-tst	-0.055982	0.096213	0.001882
N2-tst	-0.040447	0.066204	0.088461
N3-tst	0.194365	-0.082518	-0.158518
REM-tst	-0.173607	-0.081733	0.147272
PSQ	-0.121546	0.035007	-0.021625
AHI	-0.084110	0.144959	0.085684
ODI	-0.011723	0.134956	-0.076610
Mean OD%	-0.006353	0.062628	-0.027591
Lowest OD%	0.012973	-0.005733	0.034711
Average OD%	-0.019340	0.040488	-0.044393
PLMI	0.026528	-0.054086	-0.021249

Discussion

Diagnosis and treatment of suspected mechanical OSAS in children represent a highly controversial issue for the differences in terms of diagnostic resources and therapeutic approach. Only careful evaluation of medical history and clinical presentation together with polysomnography, can provide the right diagnostic classification and, consequently, the correct choice of treatment³⁵⁻³⁸. The resolution of mechanical obstruction must be as fast as possible in order to reduce the risk of cardiac, metabolic and neurological complications^{7,8}. Our attention is focused on the close relationship between OSAS and neurocognitive and neurobehavioral disorders

in very young children with a mean age of 8.44 ± 2.26 . The set of symptoms referred by patients or by children's parents together with respiratory distress is often characterised by attention deficit and hyperactivity, irritability and learning disorders⁹⁻¹². Recent studies have shown a clear relationship of cause and effect between nocturnal hypoxia leading to respiratory events (obstructive apnoeas and hypopnoeas), alteration of the structure of sleep with fragmentation and arousals and neurocognitive deficit⁹⁻¹². In particular, it has been established that in OSAS patients the brain area mostly involved in the hypo-anoxic stress is the prefrontal cortex^{9,38}. It presents a considerable reduction in activity in all stages of sleep and seems to be disconnected from other cortex areas^{9,37}. Many authors have shown considerable improvement after adenotonsillectomy in children's cognitive performance (memory, learning, IQ) and in quality of life (reduced daytime sleepiness, irritability and mood alteration)^{15,38}. These authors have used: Differential Ability Scales (similar to I.Q.), Non Verbal Cluster³⁶; Behavioral Assessment System for Children³⁹ (BASC), to evaluate the behaviour (mood, hyperactivity and somatisation); Epworth sleepiness Stairs and Osler Test (to evaluate semantic, episodic and work memory)¹⁵; the Standfort Binet Intelligence Scale 5th edition and the Developmental Neuropsychological Assessment (NEPSY) for neuropsychological skills⁴⁰.

VMI test has never been performed to evaluate visual-motor performances in children with OSAS.

The Beery-Buktenuica Developmental Test of Visual-Motor Integration (VMI; Beery & Beery, 2004) was developed to assess visuoperceptual and constructional abilities in children and adolescents and is among the most widely administered neuropsychological tests^{32,41,42}. This test has been used to evaluate children with traumatic brain injury and attention-deficit/hyperactivity disorder (ADHD)⁴².

We evaluated children with OSAS using VMI standard scores, motor and visual test scores, PSQ and PSG parameters before and at 6 months after adenotonsillectomy. Visuoperceptual and constructional performances were improved in all children after surgical treatment; analysis of sleep quality (respiratory parameters and neurological ones) showed better sleep macrostructural architecture and better respiratory scores after adenotonsillectomy than before.

The results showed the achievement of therapeutic benefits. Six months after the mechanical removal of the obstruction there was an improvement of cognitive performance and quality of life.

Conclusions

Dealing with a subject so controversial for the lack of homogeneity in terms of diagnostic and therapeutic

resources, such as OSAS in children, we chose a multidisciplinary approach. Nowadays there are no studies in the literature specifically correlating mechanical OSAS and visual-spatial skills. We first assessed the deficiencies and then the improvements in visual-spatial skills in children affected by OSAS before and after adenotonsillectomy. Our idea has been confirmed by the results obtained from the 6 month postoperative VMI tests. At any starting level, all children showed improvement in performance. Thus, at 6 months after adenotonsillectomy important therapeutic benefits have been demonstrated both in visual-motor performance and in the child's quality of life according to the objective data of PSG.

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