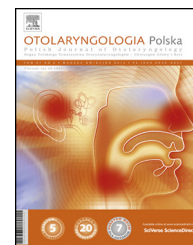


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Original research article/Artykuł oryginalny

Role of adenotonsillectomy in OSAS children and behavioural disturbance

Rola adenotonsilektomii u dzieci z zespołem obturacyjnego bezdechu sennego (OSAS) i związane z nią zaburzenia behawioralne

Desiderio Passali^{1,*}, Francesco Maria Passali², Jacopo Cambi¹, Luisa Bellussi¹

¹ENT Department, University of Siena, Siena, Italy

²ENT Department, University of Rome, Tor Vergata, Italy

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ABSTRACT

Aim: The main aim of this study was to assess the presence of behavioural disturbances in child with OSAS before and after adenotonsillectomy (AT). **Background:** In children adenotonsillar hypertrophy is associated with increased probability of OSAS. Children with OSAS present neurobehavioral disorders like attention deficit and hyperactivity, learning disabilities and daily attitudes due to excessive sleepiness. **Materials and methods:** 195 consecutive young patients suffering from OSAS and recurrent throat infections (control group) underwent AT. All underwent clinical evaluation, polysomnography, Behaviour Assessment System for Children questionnaire (BASC-2), for parents evaluation of behavioural disturbances and nasal functionality tests (before and 6 months after surgery). **Results:** Snoring and nocturnal apnoea were no more present in almost all. In OSAS group before AT 12 children were normal, 4 children were borderline and 2 were clinically significant at the BASC-2. After AT 16 children were normal, 2 children were borderline and none was clinically significant according to the same questionnaire. In the control group 9 children were normal and 1 was borderline both before and after AT. **Conclusion:** Adenoids/tonsils hypertrophy and nasal hypoventilation are frequent causes of snoring and OSAS. AT improves significantly both snoring/apnoeas and OSAS children's behavioural disturbances. Polysomnography cannot be carried out routinely due to the lack of specialised centres and because of its excessive cost. Nasal functionality tests can be useful for the differential diagnosis between sleep apnoea syndrome and other noises.

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Introduction

The pathophysiology of the obstructive sleep apnoea syndrome (OSAS) is still object of debate. Both anatomic airway

narrowing and abnormal upper airway neuromotor tone are likely to give their contribution to the syndrome as combined or isolated factors. The airway collapse during sleep is linked to smaller upper airways, but a dysregulation of the

* Corresponding author at: Dipartimento di Scienze neurologiche e sensoriali, Università di Siena, Viale Bracci, 11 53100, Siena, Italy. Tel.: +39 335 6102667.

E-mail address: d.passali@virgilio.it (D. Passali).

neuromotor tone could play an important role in the aetiology of OSAS too [1].

Other anatomical and pathological factors could influence the respiratory disturbances. For example, an important factor predisposing to OSAS is a congenital little pharynx, typical of these subjects. In other words, an unsuccessful surgical treatment can be due to alterations of the cranio-facial morphology, that is to say a narrow rhino-pharyngeal cavity or a poor development of maxilla and mandible [2, 3].

On the other hand it is clear that adenotonsillectomy (AT) gives good results, as it is confirmed by the improvement of the apnoea-hypopnoea index (AHI) and by the values of oxygen saturation in blood (SaO₂) in 78% of patients [3].

In children primarily the presence of adenotonsillar hypertrophy is associated with increased probability of OSA. The enlargement of lymphoid tissues exponentially increase the airflow resistance and determinate recurring upper airway collapse [4].

Children with OSAS present neurobehavioral disorders like attention deficit and hyperactivity, learning disabilities and daily attitudes due to excessive sleepiness. The behavioural and cognitive consequences of sleep breathing disorder are also present in children with referred snoring and care early can prevent long-term cognitive consequences [4, 5].

The Behaviour Assessment Scale for Children (BASC-2) is commonly used to assess the behaviour problems and child personality by a parent version that contains question on adaptability, social skills, leadership, activities of daily living and functional communication [6].

Italian guidelines for AT in child consider OSAS a primary indication for surgery, the other main indication to AT are recurrent throat infections [7-9].

The main aim of this study was to assess the presence of behavioural disturbances in child with OSAS before and after AT, the effect of AT on OSAS and chronic snoring was investigated. Finally the contribution of nasal functions tests in the evaluation of ethiology and pathogenesis of OSAS was examined.

Subjects and method

One hundred ninety-five consecutive children were recruited for AT in the ENT Department of the University of Siena between January 2012 and January 2013 because suffering from OSAS or recurrent throat infections.

All the children underwent to a clinical evaluation with rhinofibroscopy of the upper airways for grading the stenosis due to tonsils and adenoid (before and 2, 4, 6 months after AT), polysomnography (before and 6 months after surgery), a validated in Italian BASC-2 questionnaire for parents evaluation of behavioural disturbances (before and 6 months after surgery), and nasal functionality tests as rhinomanometry and mucociliary transport time (before and 6 months after surgery). Hypertrophy of pharyngeal tonsils were classified according Brodsky grading scale [10] as adenoid tissue were classified in classes of obstruction according Cassano classes of obstructions [11].

According to polysomnography findings OSAS in childhood was defined as slightest, mild, moderate and severe

considering the SaO₂ (>97%, >97%, >95%, <95% respectively), the sum of apneic and hypopneic events per hour (1-3, 3-5, 5-10, >10 respectively) and the presence of snoring associated with desaturation greater than 4% in slightest OSAS. An obstructive apnoic event in child was defined as a drop in thermal sensor amplitude by $\geq 90\%$ baseline, a duration ≥ 2 missed breaths and the continued or increased inspiratory effort during reduced airflow [12].

According to BASC-2 manual, preschool (ages 2 to 5) and child (ages 6 to 11) forms are used these questionnaire contains 134-160 items and uses a four-choice response format. T-scores of 40 or below indicate children "at risk", while scores of 30 or below indicate clinically significant behaviour impairment in areas of hyperactivity, aggression, anxiety, attention problems, atypically, depression, somatization, withdrawal.

Anterior Active Rhinomanometry (AAR) gives an objective measurement of the ventilatory function of the nose, when performed according to the rules suggested by the Committee on Standardization [13]. If the rhino-manometric values prove to be lower than normal ranges for nasal conductance, we carry out the nasal decongestion test (NDT). This test is performed following three steps: 1) Basal AAR; 2) Nasal decongestant: 2 sprays in each nostril; 3) After 10' control AAR.

The measure of the Muco-Ciliary Transport time (MCTt) can be performed by several methods: radio-active tracers, coloured tracer elements and radio-opaque elements, which give comparable results [14]. We usually use a mixture of charcoal powder and 3% saccharine: as it is an insoluble tracer, charcoal powder gives a monitoring of the transport of the particles entrapped into the outer gel layer; while saccharine is a soluble marker, and give us the time of clearance into the inner sol layer. The normal values in children for the charcoal powder and saccharine are 8 ± 3 and 11 ± 6 min respectively [15].

All the patients underwent AT under anesthesia after parents given their informed consent for surgery procedure and for all performed tests.

The study protocol was approved by the local Ethics Committee. Comparisons between groups were assessed by the paired t-test, as appropriate, at a significance level of $p < 0.05$. Statistical analysis was performed with SPSS software (SPSS, Inc., Chicago, IL, USA).

Results

Between January 2012 and January 2013, 195 consecutive children were subjected at AT for OSAS or recurrent throat infections. Demographic and clinical data of this population are reported in Table I. The children had a mean age of 5.3 ± 1.6 years with a male/female ratio of 2.2. The past medical history of these patients included episodic sleep apnea, chronic snoring and recurrent throat infections. Night and day disturbances like nocturnal awakening, enuresis and sleepiness, hyperactivity, aggressiveness, distraction, scarce school outcomes were also reported by parents during the anamnesis.

At first clinical examination adenotonsillar hypertrophy was found in 100% of patients: 190 had hypertrophy of

Table I – Demographic and clinical characteristics

		Number (%)
Number of subjects		195 (100%)
Sex (M/F)		134/61
Age (years)		5.3 ± 1.6 (range 3-8)
Main diagnosis	Recurrent throat infections	126 (65%)
	Snoring	195 (100%)
	OSAS	69 (35%)
Reported behavioural disturbances	Nocturnal awakening	12 (6%)
	Enuresis	7 (4%)
	Sleepiness	32 (16%)
	Hyperactivity	14 (7%)
	Aggressiveness	5 (3%)
	Distraction	26 (13%)
	Scarce school outcomes	10 (5%)

pharyngeal tonsils (Brodsky grading scale 2+ in 65, 3+ in 80, 4+ in 45) and 80 had adenoid tissue during rhinofibroscopy (Cassano classes of obstructions III in 55, VI in 25). Five children had recurrent throat infections with Brodsky grading scale 1+. In 34 children was not possible to perform rhinofibroscopy for nasal congestion or intolerance at this examination.

Polysomnography reported OSAS in 69 (35%) children and was defined as slightest in 18 (26%), mild in 16 (23%), moderate in 12 (17%) and severe in 23 (33%) considering the guidelines classification criteria. After AT polysomnography was performed in 45 of 69 OSAS children and resulted as normal in 35 (77%), slightest in 9 (20%), mild in 1 (3%).

All parents were asked to complete the BASC-2 questionnaire before and after six months from AT, incomplete questionnaire were excluded from the study and statistical analysis.

BASC-2 questionnaire for parents classified normal, T-score > 40, 81 (87%) children, at risk, 40 > T-score > 30, 10 (10%) and with behavioural impairment, T-score <30, 3 (3%) patients, 97 questionnaire was incomplete and were excluded.

In OSAS group the BASC-2 questionnaire classified normal, T-score > 40, 12 (67%) children, at risk, 40 > T-score > 30, 4 (22%) and with behavioural impairment, T-score < 30, 2 (11%) patients.

After six months BASC-2 questionnaire classified normal, T-score > 40, 25 (88%) children, at risk, 40 > T-score > 30, 3 (12%) and with behavioural impairment, T-score < 30, 0 (0%) patients, 69 questionnaire was incomplete, not compiled from parents or lost and were excluded.

In OSAS group after six months the BASC-2 questionnaire classified normal, T-score > 40, 16 (89%) children, at risk, 40 > T-score > 30, 2 (11%) and with behavioural impairment, T-score < 30, 0 (0%) patients.

There were statistically significant differences between the OSAS group T-score pre and after AT ($p = 0.0438$, t-test).

Specific data on impairment areas of hyperactivity, aggression, anxiety, attention problems, atypically, depression, somatization and withdrawal were reported in Table II. Statistically significant differences at t-test between single areas of impairment before and after AT in OSAS group were founded in aggression, attention problems, atypically, somatization ($p = 0.0038$, $p = 0.0002$, $p = 0.0158$, $p = 0.0067$, respectively).

Before AT total nasal airway resistance mean value at AAR was 0.373 ± 0.16 Pa/cm³/s at 150 Pa, six months after AT the mean value was increased at 0.271 ± 0.10 Pa/cm³/s ($p = 0.0001$, t-test). Nasal flow before AT was 685.97 ± 162.23 ml/s, after 765.41 ± 177.97 ml/s ($p = 0.0001$, t-test).

Before AT MCTt mean value was 39.15 ± 45.84 min, after was 10.79 ± 2.17 min ($p = 0.0001$, t-test).

Discussion

As respiratory alterations in children are mainly due to severe adenotonsillar hypertrophy, about 80% of children benefit from surgery [16]. A severe adenotonsillar hypertrophy could be the only responsible for the nocturnal respiratory obstruction [17]: as it is confirmed by the improvement of symptoms and of the “respiratory disturbance index” after AT. In our study AT improves significantly both snoring and nocturnal apnoeas.

The relationship between the size of adenoids/tonsils and apnoea indexes is still controversial, so that adenotonsillar hypertrophy is not always accompanied by nocturnal obstructive disturbances.

As regards the size of adenoids and tonsils, the obstruction in condition of “rest” doesn't allows to foresee the degree of the obstruction during the sleep when the muscular tone decreases, especially of the genioglossus muscle. This means that hypotony, alterations of the neural control of the pharyngeal muscles, or a high level of laxity of the ligaments could be responsible for the pharyngeal obstruction.

Most of the cases of OSAS are responsive to adenoid-tonsillectomy, nevertheless other risk factors and coexisting conditions should be investigated and treated prior to consider the surgical treatment.

Table II – Variation in BASC-2 impairment areas

	Pre AT OSAS group T-score	Post AT OSAS group T-score	Statistical analysis (t-test)
Hyperactivity	45.83 ± 13.95	52.77 ± 11.17	0.1145
Aggression	45.22 ± 12.07	57.61 ± 13.59	0.0038*
Anxiety	46.27 ± 13.29	54.77 ± 12.50	0.0507
Attention problems	45.72 ± 12.79	62.16 ± 13.85	0.0002*
Atypically	60.61 ± 23.25	69.38 ± 18.29	0.0158*
Depression	50.22 ± 15.29	57.27 ± 13.07	0.0615
Somatization	47.83 ± 13.94	58.72 ± 14.40	0.0067*
Withdrawal	47.55 ± 13.63	57.16 ± 12.48	0.0501

A common condition, which needs to be excluded, is allergic rhinitis (AR), which affects approximately 40% of children: OSAS occurs in 2% of them [18].

As AR is associated with nasal obstruction, enlargement of tonsils and adenoids, and an elongated face, it can be considered a risk factor for OSAS. The treatment of AR is helpful to decrease the severity of OSAS and prevent emergence of an elongated face, which is responsible for a smaller upper airway size.

Another point of view to study the clinical consequences of OSAS and their link to AT is to measure the changes of position during sleep, coming from the observation that the nocturnal iper-kynesia could be a manifestation of hypo-oxygenation. After AT positional changes during sleep and positional change index significantly decreased. The proportion of sleep time spent in the supine position was significantly increased, and the proportions spent in lateral and up positions were significantly decreased [19].

Sleep fragmentation should direct to unfavourable consequences on daytime performance. It is known that children who are poor sleepers manifest improved incidence and severity of behavioural issues compared to children without sleep trouble. The severity of behavioural changes like daytime hyperactivity, anxiety, and depressive symptoms and OSAS severity are strongly linked. Improvements in sleep are associated with improvement in daytime behaviour.

Evaluating the behavioural consequences of chronic hypo-oxygenation we pointed out that in the OSAS group before AT 33% of the children was borderline or clinically significant according to BASC-2. After AT only 11% of them was borderline according to the same questionnaire. In the control group (non OSAS children) 10% was borderline both before and after AT.

In addition to the traditional clinical diagnostic tools, such as history, physical examination and polysomnography, a significant contribution can be given by some tests, which are specific for evaluating the nasal physiopathology.

AAR gives an objective measurement of the ventilatory function of the nose, in adenoid hypertrophy the mean values of nasal resistances were increased.

Furthermore the nasal functionality is due to the efficiency of the nasal muco-ciliary transport, which is responsible for the mechanical defence of the airways from physical, chemical and biological attacks. Functional and/or morphological alterations of the nasal cavities give rise to muco-ciliary dysfunctions.

Alterations of the ciliary structure or metaplastic changes of the respiratory mucosa into squamous epithelium are usually pointed out in perennial nasal allergy. As a consequence of increased glandular secretion, the ratio between the sol and gel phase of the mucus is altered and the efficiency of muco-ciliary clearance is impaired in seasonal allergic rhinopathies.

The improvement both in total nasal resistance value and muco-ciliary time after AT demonstrates the role of adenoid tissue as a nasopharyngeal obstacle.

The above mentioned methods are suitable for the differential diagnosis between OSAS and other noises caused by the contact of the inspired air with the walls of the nose and the pharynx.

All the tests are sensitive tools, even though relatively aspecific.

Conclusion

According to our experience, hypertrophy of adenoids/tonsils and nasal hypoventilation are some of the most frequent causes of snoring, that sometimes can be followed by a real OSAS. Referring to the above mentioned results we are allowed to conclude that AT improves significantly both snoring and OSAS and determines an improvement in behavioural disturbances. Primary snoring and OSAS are a continuum, and surgery may be defined as a form of prevention against pathology with potential complications.

Polysomnography cannot be carried out routinely due to the lack of specialised centres and because of its excessive cost [16]. It could be prescribed only in unsuccessful AT and for those children who have complicated presentations from the outset.

Some tests, which are specific for evaluating the nasal physiology and pathology, such as AAR, and MCTt, can be useful for the differential diagnosis between sleep apnea syndrome and other noises caused by the contact of the inspired air with the walls of the nose and the pharynx.

In any case OSAS cannot be left untreated: the clinical condition must be individually evaluated at the aim of making the best choice between medical and surgical treatment to restore the upper respiratory ventilation because sleep fragmentation and intermittent hypoxia contribute to the neurobehavioral morbidity, and reversibility is achievable when treatment is realized early and effectively.

Authors' contributions/Wkład autorów

DP, FMP, JC and LB were responsible for study conception and design. DP and FMP acquired the data. JC analysed the data. DP, JC and LB drafted the manuscript. DP is guarantor. All authors critically revised the manuscript and approved the final version.

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Conflict of interest/Konflikt interesu

None declared.

Ethics/Etyka

The work described in this article have been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; EU Directive 2010/63/EU for animal

experiments; Uniform Requirements for manuscripts submitted to Biomedical journals.

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