EMBJ.ORG FOR YOUNG DOCTORS

EUROMEDITERRANEAN BIOMEDICAL JOURNAL
2022,17 (8) 27-32
(*FORMERLY: CAPSULA EBURNEA*)

Original article

SWALLOWING, SPEECH AND OROFACIAL DISORDERS IN CHILDREN WITH ADENOTONSILLAR HYPERTROPHY

Rosalia Gargano, Donatella Marchese, Simone Oliva, Serena Gerardi, Carmelo Saraniti, Francesco Riggio, Francesco Dispenza, Francesco Lorusso, Gabriella Sampognaro, Federico Sireci, Salvatore Gallina

Department of Otorhinolaryngology, Department of Biomedicine, Neuroscience and Advanced Diagnostics , University of Palermo, Italy.

ARTICLE INFO

Article history:
Received 25 Nov2021
Accepted 18 Dec 2021
Published 10 Jan 2022

Keywords:
Adenotonsillectomy, Dysphagia, Phonological development, Dentofacial abnormality

ABSTRACT

A prospective observational study was conducted to evaluate the presence of swallowing, speech and orofacial muscle imbalance (OMI) disorders in patients with adenotonsillar hypertrophy and their regression after surgery. ENT, speech therapist and dentistry examination were conducted before and 12 months after surgery in 78 children between the ages of 2 and 12 with dysphagia, speech disorders and OMI adenotonsillar hypertrophy related with sleep-disordered breathing. Of the 78 patients enrolled, 62 underwent adenotonsillectomy, 9 adenoidectomy and 7 volume reduction of the palatine tonsils with a 12-month post-operative increase in SWAL-QOL score, weight gain, correction of ogival palate, dental crowding and tongue posture and consequent resolution of phono-articulatory disorders. All patients show an improvement in swallowing, phono -articulatory disorders' symptoms and OMI correction at 12-month evaluation.

1. Introduction

Swallowing is one of the most complex movements in the human body. It begins to structure itself in the 13th week of gestation and continues to evolve passing from infantile to adult swallowing.

Physiological swallowing can be divided into four phases: 1) oral phase (preparation and transport of the bolus towards the pharynx); 2) stimulation of the swallowing reflex; 3) pharyngeal phase; 4) esophageal phase. [1]

The evolution of infant swallowing can be divided into 4 age groups:

- 0-6 months: Suckling: suction by squeezing the (edentulous) arches on the nipple, antero-posterior lingual movements, hyoid-larynx complex fixed in an elevated position, suction-swallowing-breathing coordination in 1/1/1 ratio;
- 6 months - 1 year: Sucking: the lip musculature forms a suction cup on the nipple; reduced mandibular movements - superior-inferior lingual movements - negative pressure in the oral cavity - descent of the hyoid-larynx complex with protective elevation during the pharyngeal phase - spoon feeding with development of the swallowing phases;
- 1 - 6 years: beginning and maturation of chewing;
- 6 years: transition to adult swallowing with disappearance of the anterior lingual thrust.

By dysphagia we mean the dysregulation of the correct swallowing action that results in an impairment of adequate nutrition and sometimes in a reduced ability of the child to protect the airways during the ingestion of liquids and solids. [1]

Pediatric dysphagia is always a consequence of pathogenic noxa such as:

- prematurity (low birth weight, other comorbidities)
- congenital or acquired structural abnormalities (palatine clefts, esophageal malformations, tonsillar or adenotonsillar hypertrophy) [2] – [3]
- neurological diseases (including mental retardation and pervasive developmental disorders)[4]
-behavioral disorders
-cardio-respiratory disorders (osas, respiratory distress syndrome, laryngomalacia) [5] – [6]
- metabolic disorders (diabetes, jaundice)

* *Corresponding author*: Simone Oliva, *simoneoliva1991@gmail.com*
DOI: 10.3269/1970-5492.2022.17.8
 ISSN: 2279-7165 - Available on-line at www.embj.org

The surgical treatment of isolated adenotonsillar or tonsillar hypertrophy is fundamental in the resolution of obstructive sleep apnea syndrome but in the literature, it is possible to find some data, albeit rare, on the indication as a treatment for swallowing disorders or in the improvement of speech disorders and of the OMI. [3]

In particular, it is possible to find a correlation between OSAS and dysphagia in pediatric age [2] – [7] as a reduced muscle coordination is established with alteration of the processes of sucking, chewing, swallowing, nasal breathing and language.

Isolated adenotonsillar or tonsillar hypertrophy also compromises the other processes that occur in the oral cavity and oropharynx in addition to swallowing, such as the articulation of speech sounds and harmonious dental-skeletal development.

In the oral respirator, like the patient suffering from adenotonsillar hypertrophy, the tongue assumes a low and protruded position, exerting an abnormal thrust on the anterior dentition thus causing dental malocclusions, dental crowding, ogival palate and facial dysmorphisms with particular consequences on the processes of swallowing and articulation of language.

Dental malocclusion consists in an incorrect alignment of the upper with the lower dental arch for which the dental, muscular, articular and aesthetic requirements of a first class, according to the Angle classification, like a correct incisor overjet and a greater transverse diameter of the upper arch compared to the lower arch, symmetrical movements of the mandibular condyles, are not respected; therefore a condition of Retrognathism (2nd class occlusion) or Prognathism (3rd class occlusion) occurs.

Malocclusion contributes to the onset or persistence of joint disorders due to the presence of morphological alterations of the various organs involved in the production of sound. A condition of interdental Sigmatism, Zetacism, Rotacism, Kappacism or Gammacism can therefore be frequently encountered in addition to alterations in the timbre of the voice such as closed rhinolalia due to adenoid hypertrophy, if present.

The aim of our study is to evaluate the presence of swallowing and speech disorders and the presence of OMI in patients with adenoid or adenotonsillar hypertrophy and their eventual regression after surgery.

2. Material and methods

Study design

This study was approved by the Ethics Committee of the University Hospital "Paolo Giaccone" of Palermo and examined patients, aged between 2 and 12 years, who underwent adenotonsillectomy and adenoidectomy from January 2019 to January 2020 with a one-year follow-up. The indications for adenotonsillectomy in all children were obstructive respiratory disorders clinically characterized by loud and persistent snoring for at least four nights per week during the last month, predominant oral breathing, observed apnea, restless sleep, tonsillar hypertrophy on ENT examination and adenoids that occupy more than 80% of the rhinopharynx.

Patient selection

78 patients (47 male and 31 female) were examined and recruited.

The patients were divided into four age groups:

- 2-3 years = 12 patients.
- 3-6 years = 27 patients.
- 6-9 years = 31 patients.
- 9-12 years = 8 patients.

Of the 78 patients, 65 were born at term, 13 prematurely.

Data collection

For each of them, the pre- and post-operative clinical protocol provided for:

- Instrumental otolaryngological evaluation with a flexible fiber optic laryngoscope of tonsillar hypertrophy's (according to the Friedman classification) and adenoid's degree (according to the Cassano-Gelardi classification); evaluation of the palatal morphology.
- Speech therapy evaluation of phonetic and articulatory disorders, OMI and lingual posture through an analysis of the spontaneous speech and the administration of the test "The test of phonemes: correct pronunciation defects", carried out on denomination, to obtain a detailed pre- and post-operative description of children'articulatory skills and phonetic repertoire.
- Dental evaluation of the classes of dental occlusion, dental crowding, retrogratism and mandibular prognathism.
- Type of surgery.
- Body weight measurement.
- Administration of the SWAL-QOL questionnaire (Swallowing quality of life questionnaire) for swallowing disorders. With regard to the questionnaire proposed in Table 1, in the present study only the items related to the Swallowing Quality Life Questionnaire most significant for the final objective were considered. In particular, items related to Mental health and Social function were excluded from the original domains, focusing more on symptoms. Each survey response was linearly transformed into a metric between 0 and 100 points, where "100" represents the most favorable state and "0" the least favorable state of symptoms related to swallowing and quality of life. Individual answers were equally weighted and added together to generate an overall score.

Statistical analysis

The analysis of the sample was carried out through descriptive statistical techniques necessary to evaluate the mean, standard deviation, absolute frequency, and percentage frequency connected to each clinical survey. Furthermore, the Student's t-test for 2 dependent samples or for paired data with confidence interval of the mean of the differences was used to compare the Swallowing Quality of Life (SWAL-QOL) scores before and after.

Domains	Items
Burden	Dealing with my child's swallowing problem is very difficult My child's swallowing problem is a major distraction in our lives
Duration	It takes my child longer to eat than other children It takes my child forever to eat a meal
Desire	My child doesn't enjoy eating anymore My child loses interest in eating due to his/her swallowing difficulty
Selection	It is difficult to find foods that my child likes and can eat Figuring out what my child can eat is a problem for our family
Fear	I fear my child may start choking when eating solid food I worry about my child getting pneumonia I am afraid my child will choke when drinking liquids I never know when my child is going to choke
Mental health	Having to be so careful when my child eats and drinks annoys me I've been discouraged by my child's swallowing problem My child's swallowing problem frustrates me I get impatient dealing with my child's swallowing problem
Social function	My child's swallowing problem makes it difficult for him/her to socialize with other children Our family's usual work or leisure activities have changed because of my child's swallowing problem Social gatherings (like holidays and get-togethers) are not enjoyable because of my child's swallowing problem It is difficult to eat outside of the home because of my child's swallowing problem
Symptoms	Coughing Choking when eating solid food Choking when drinking liquid Having thick saliva or phlegm Having excess saliva or phlegm Gagging Drooling Problems chewing Having to clear his/her throat Food sticking in his/her throat Food sticking in his/her mouth Food or liquid dribbling out of his/her mouth Food or liquid coming out of his/her nose Coughing food or liquid out of his/her mouth when it gets stuck

Table 1. Items of the "Swallowing Quality of Life" Questionnaire Adapted for Use with Pediatric Patients.

Item 1 - Burden Dealing with my child's swallowing problem is very difficult	0 = fully agree	25 = quite agree	50 = neither agree nor disagree	75 = quite disagree	100 = fully disagree
Item 2 – Eating duration My child takes longer to eat than other children	0 = fully agree	25 = quite agree	50 = neither agree nor disagree	75 = quite disagree	100 = fully disagree
Item 3 - Desire My child doesn't enjoy eating due to swallowing problems	0 = fully agree	25 = quite agree	50 = neither agree nor disagree	75 = quite disagree	100 = fully disagree
Item 4 – Food selection It's difficult to find foods that my child likes and can eat	0 = fully agree	25 = quite agree	50 = neither agree nor disagree	75 = quite disagree	100 = fully disagree
Item 5 - Fear I fear my child may start choking when eating solid food	0 = fully agree	25 = quite agree	50 = neither agree nor disagree	75 = quite disagree	100 = fully disagree
Item 6 - Symptoms a) My child's swallowing causes breathing problems	0 = fully agree	25 = quite agree	50 = neither agree nor disagree	75 = quite disagree	100 = fully disagree
Item 7- Symptoms b) My child stops breathing and refuses food	0 = fully agree	25 = quite agree	50 = neither agree nor disagree	75 = quite disagree	100 = fully disagree
Item 8 - Symptoms c) After tonsillitis, my child began to have swallowing problems	0 = fully agree	25 = quite agree	50 = neither agree nor disagree	75 = quite disagree	100 = fully disagree
Item 9 - Symptoms d) My child gags when eating or drinking	0 = fully agree	25 = quite agree	50 = neither agree nor disagree	75 = quite disagree	100 = fully disagree
Item 10 - Symptoms e) My child has choking symptoms when eating or drinking	0 = fully agree	25 = quite agree	50 = neither agree nor disagree	75 = quite disagree	100 = fully disagree

Table 2. Items of the "Swallowing Quality of Life" Questionnaire adapted to our study.

3. Results

In the ENT evaluation, particular attention was paid to the degree of tonsillar and adenoid hypertrophy. It was found that the majority of patients (40%) had grade 3 tonsillar hypertrophy according to Friedman's classification, 21.79% grade 4 hypertrophy and 15.38% grade 2 hypertrophy. Only 9 children had hypertrophy isolated adenoid with percentages between 70-80% of occupation of the nasopharyngeal cavity.

The surgical interventions conducted in the present study, with reference to the distribution described in Table 3, were:

- Adenotonsillectomy.
- Adenoidectomy.
- Volumetric reduction of the palatine tonsils.

We also analysed the data relating to the weight of children undergoing surgery, dividing the sample studied into four age groups and evaluating the average pre-operative weight and the average post-operative increase. For children between 0 and 3 years of age, we recorded an average weight increase of 1.63 kg and an average percentage increase of 12.84% compared to the average pre-intervention body weight of 13.3 kg. In the age group 3- 6 years, we report average pre-intervention weight values of 22.2 kg, average weight increase of 1.38 kg and average percentage increase of 6.4%. In the age group between 6 and 9 years, we recorded average pre-surgery weight values of 26.9 kg, average increase of 1.41 kg and percentage increase of 5.30%. Finally, in the last category, children aged between 9 and 12 years, the following values: average pre-surgery weight of 36.43 kg, 1.55 kg average weight increase and 4.32% weight increase in percentage.

47.44% of patients (37 children) had a weight increase between 1 and 1.5 kg compared to the values prior to surgery, analyzed 1 year after the procedure; 24.36% (19 children) a body increase between 1.5 and 2 kg; 8% of the population under consideration show an increase in weight of more than 2 kg, while 19.2% an increase of less than 1 kg, of which only 1 case did not register a change in weight.

Patients undergoing surgery, in the post-operative period, show a significant improvement due to the increase in weight correlated both to the improvement of symptoms related to swallowing and to the quality of life. The evidence is provided through the corresponding increase in the mean value in all domains of the SWAL-QOL at 1 year after surgery (Table 4).

In fact, comparing the post-operative scores of the SWAL-QOL with the pre-operative ones, an overall increase in the average score from 15.13 (sd 6.00) to 66.67 (sd 5.22) is observed. In particular, as can be seen from the table above, there is a significant increase in the score in the items that correlate the problem of dysphagia to that of respiratory disorders such as tonsillitis and apnea.

After surgery, the distribution range of the SWAL-QOL scores appears to be closer to the ideal score of 100 (Figure 1).

The results obtained by evaluating the weight variation in the pre-operative and in the post-operative related to the score obtained on the SWAL-QOL test, showed a positive correlation (R2 = 0.21). (Figure 2)

It is observed that the data are in favor of a greater benefit from the respiratory and swallowing point of view for patients in the age group between 2 and 3 years as the growth of the bones of the skull and face (craniofacial growth) is of great importance by significantly influencing the development of dental occlusion.

The following characteristics were highlighted from the pre-operative morphological observation of the extra - oral and intra - oral district at rest:

- Ogival palate: in 66 patients (84.6%);
- Low and anteriorized tongue posture in 66 patients (84.6%);
- Dental crowding: present in 20 patients (25.6%);

Based on Angle's classes for malocclusion we observed:

- Retrognathism in 39 patients (50%);
- Prognathism in 23 patients (29.5%)

From the post-operative morphological observation at 9 months of the extra - oral and intra - oral region at rest, the following characteristics were highlighted:

- Ogival palate: improved in 49 of 66 patients with this alteration (74.24%);
- Low and anteriorized tongue posture: was absent in 57 of 66 patients (86.3%) with this alteration;
- Dental crowding: it was improved in 15 of the 20 patients with this alteration (75%);
- Retrognathism: it was improved in 11 of 39 patients with this alteration (28.2%);
- Prognathism: it was improved in 3 of 23 patients with this alteration (13%)

Motor development and language development were impaired for 25% of the children evaluated. It emerged that, in almost all the patients, there was no familiarity with bone alterations in the orofacial area.

Through an accurate pre-operative assessment of the patients' spontaneous speech and thanks to the administration of the test "The phoneme test: correcting speech defects"[8] it emerged that 40% of the patients observed presented phonetic-articulatory disorders, and therefore a production of linguistic sounds not adequate to age. In particular, it was found:

- Interdental sigmatism: 75% of the patients observed presented this disorder;
- Zetacism: this disorder emerged in 75% of the patients evaluated;
- Rotacism: 25% of the observed patients showed this disorder.
- Kappacism: 25% of the children examined presented this disorder;
- Gammacism: 25% of patients had this disorder

In addition, in all the patients evaluated, we found hyporrinophony, an alteration of the timbre of the voice due to the reduction of the nasopharyngeal lumen.

At the post-operative re-evaluation at 9 months, a clear improvement of these articulatory-phonetic and vocal resonance disorders was found, with a persistence of these disorders in the following percentages:

- Interdental sigmatism: 25% of the patients observed presented this disorder;
- Zetacism: this disorder emerged in 28% of the patients evaluated;
- Rotacism: 15% of the patients observed showed this disorder.
- Kappacism: 18% of the children examined presented this disorder;
- Gammacism: 13% of patients had this disorder
- Hyporrinophony: 0%

The latter patients are continuing a speech therapy rehabilitation process.

	n	%
Adenotonsillectomy	62	79.49
Adenoidectomy	9	11.54
Volumetric reduction of the palatine tonsils	7	8.97
Total	78	100.00

Table 3. Absolute and percentage frequencies of the clinical data "Type of surgical interventions".

Domain	Before Surgery Average (Standard Deviation) (n = 78)	After Surgery (1 year later) Average (Standard Deviation) (n = 78)	α = 0.05 (significance level) P - Value Comparison before and after surgery
Item 1 - Burden	16.35 (14.98)	34.62 (12.24)	< 0.0001
Item 2 – Eating duration	25.32 (17.33)	63.46 (19.61)	< 0.0001
Item 3 - Desire	11.54 (13.78)	70.83 (18.21)	< 0.0001
Item 4 – Food selection	24.36 (13.94)	69.87 (12.97)	< 0.0001
Item 5 - Fear	9.94 (12.96)	26.60 (12.96)	< 0.0001
Item 6 - Symptoms a)	17.95 (12.02)	75.32 (13.05)	< 0.0001
Item 7- Symptoms b)	14.42 (12.43)	75.32 (18.24)	< 0.0001
Item 8 - Symptoms c)	14.42 (13.67)	84.94 (13.57)	< 0.0001
Item 9 - Symptoms d)	15.38 (13.50)	82.05 (12.02)	< 0.0001
Item 10 - Symptoms e)	1.60 (0.57)	83.65 (11.97)	< 0.0001
Overall	15.13 (6.00)	66.67 (5.22)	< 0.0001

Table 4. Swallowing Quality of Life Questionnaire score and domains adapted for the case study.

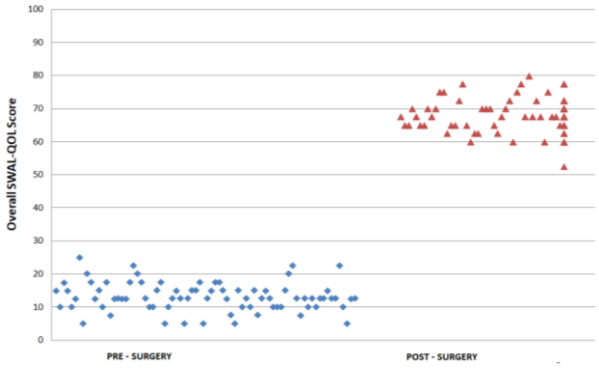

Figure 1. Distribution of the "Swallowing Quality of Life" Questionnaire pre-surgery and post-surgery.

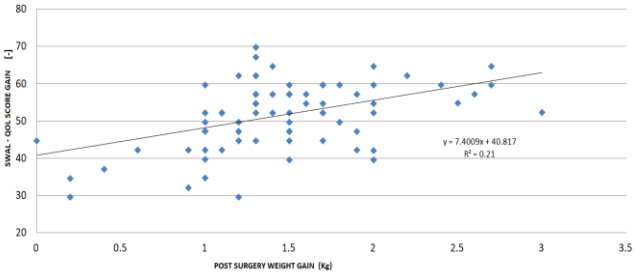

Figure 2. Correlation between SWAL-QOL score increase and pre-post surgery weight gain.

4. Discussion

The biological function of the structures of the upper airways is to facilitate the passage of air and food from the oral cavity to the pharynx. This function can be compromised when obstructive factors are present in the upper airways causing dysphagia by altering the coordination between breathing and swallowing especially during the pharyngeal phase of swallowing [2].

Dysphagia, as difficulty in swallowing solid and liquid foods, is a very common symptom in patients with adenotonsillar hypertrophy, but a completely different story is the real pediatric dysphagia which is often multifactorial, especially related to neurological and genetic diseases (not subjects of our study).

Our study aims to fill, albeit with the limitations found, a gap in the literature, as few studies have examined the role of tonsillectomy as a treatment of dysphagia and orofacial muscle imbalance with the consequent language alterations.

According to the study conducted, almost all patients have increased their body weight in the twelfth month following surgery and have had net improvements in the SWAL-QOL score, demonstrating how the anatomical encumbrance can affect not only breathing with related OSAS problems, but also swallowing.

Our data agree with those obtained by Cleyburg et al., who showed that there is an increase in the SWAL-QOL score at 1 month after surgery that lasts up to 6 months post-operative, associated with an improvement in the quality of life and greater compliance with regular diets with freeing of liquid and semi-liquid diets [3].

As described by Dong-Kyu Kim [9] and Dudley J. Weidera [10], we found a correlation between dentofacial abnormalities and adenotonsillar hypertrophy in pediatric age, highlighting how early surgery can play a decisive role in resolving these problems. Orofacial muscle imbalance can go against resolution or improvement following surgical treatment due to the readjustment of the muscle fibers of the tongue and lips, with a consequent increase in strength and contractility, as well as movements in space of these muscles that act by remodelling the facial structure, as Fernanda Bastos de Andrade-Balieiro [11] states.

In our study we found a complete post-surgical resolution of hyporrinophony in accordance with the results that can be seen in the literature [12-15]. Our improvement after surgical treatment of phonoarticulation and the problems of gammacism, kappacism, sigmatism, zetacism and rotaxism are confirmed by Youri Maryna [12] and by Silvia F Hitos [15] who recognize the responsibility of tonsillectomy to create the necessary space so that the tongue is not protruded anteriorly or laterally from the hypertrophic tonsils resulting in such articulatory disorders.

The limitations of the study are, first of all, the small number of patients; moreover, since it is a surgical study which involves the transition between the pre-operative of the patients and the post-operative phase, it was impossible to have a group of "control" patients.

Since our sample is made up of children, it was not possible to carry out videofluorography, the Gold standard in the study of dysphagia in adult patients; therefore, we relied exclusively on endoscopic diagnoses and on the data extracted from the administration of the questionnaire. A further limitation to the diagnostic investigation through the execution of the FEES was represented by the onset of the COVID-19 pandemic.

The positive correlation between the optimal score of the questionnaire administered to the parents of pediatric patients and the post-operative weight gain could have the following reasons:

- Post tonsillectomy there is an increase in insulin-like growth factors which can lead to weight gain [16];
- The resolution of the anatomical obstruction improves breathing [17] and therefore basically promotes optimal nutrition;
- The reduction of the levels of the typical markers of chronic inflammation and of TNF-α [18].

5. Conclusions

This study shows how the surgery improves the swallowing of patients with adenotonsillar hypertrophy, eliminating the mechanical encumbrance and causing an increase in weight and an improvement in the patient's oral functions and in the early stages of swallowing. There is also an improvement in phono-articulatory performance and OMI in those patients with speech disorders related to adenotonsillar hypertrophy.

The importance of an integrated multidisciplinary approach which includes the speech therapist, the dentist and the orthodontist, the otolaryngologist, and others if necessary, so that the intervention is more effective and appropriate, is emphasized [19-20]. Furthermore, it would be advisable to maintain a late follow-up to better evaluate the complete recovery of the respiratory, swallowing, and phonetic oral functions.

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