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ASSESSMENT OF THE VELOPHARYNGEAL SPHINCTER AND ITS ROLE IN THE SPEECH OF PATIENTS WITH CLEFT LIP AND PALATE

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Abstract

The cleft lip and palate are congenital anomalies with different dimensions; their existence alters the facial aesthetics and many orofacial functions as well. They characterize with a discontinuity of the lip, hard and soft palate and the alveolar ridge. The gravity of the clinical picture can be determined by the lack of continuity in the skin tissue, muscles and bones, which can be manifested with a mild form of lip distortion, severe bilateral cleft lip, palate, and alveolar bone and the most severe - oblique facial cleft. The clefts can be of different dimensions; still, regardless of the size, the articulatory speech is rendered more difficult due to the constant communication between the oral and nasal cavity. Every child born with a cleft palate has dysfunction of the velopharyngeal sphincter leading to pathology of the verbal communication.
The velopharyngeal sphincter is a tridimensional muscle area between the oral and nasal cavity. It is built by the lateral and posterior pharyngeal wall and the soft palate. The role of the velopharyngeal sphincter is to separate the oral from the nasal cavity, and, in that way, enable normal speech and swallowing, that is, prevent the air from escaping through the nose instead of the mouth.
The speech of children with cleft is usually accompanied by a nasal sound that is associated with the velopharyngeal insufficiency (weak function of the soft palate). When the soft palate does not close completely the airflow through the nose, air passes through the thin passage and nasal speech is heard. Today, one of the techniques used to assess the articulatory function of the oropharyngeal region, both in normal and pathological cases, is the magnetic resonance. Compared to other techniques, it has more advantages due to its non-invasiveness, non-radioactivity and the fact that it is video fluoroscopic. It is used for obtaining dynamic pictures while in motion, the so-called, magnetic resonance imaging.

Keywords: cleft lip, cleft palate, velopharyngeal sphincter, velopharyngeal closure, magnetic resonance imaging

ПРОЦЕНКА НА ВЕЛОФАРИНГЕАЛНИОТ СФИНКТЕР И НЕГОВАТА УЛОГА ВО ГОВОРОТ КАЈ ПАЦИЕНТИТЕ СО РАСЦЕПИ НА УСНИ И НЕПЦЕ

Апстракт
Расцепите на усната и непцето се конгенитални аномалии со повеќе димензии, нарушен изглед на лицето, но и нарушени орофацијални функции. Карактеристика со која се одликуваат е прекин на континуитетот на усната, тврдото и мекото непце и алвеоларниот гребен. Тежината на клиничката слика се одредува со недостатокот на континуитет на ткивото на кожата, мускули и коски кои може да се манифестираат со благ облик во вид на оштетување на усната, тежок обострaн расцеп на усните, непцето и алвеоларната коска и најтежок - кос расцеп на лице. Расцепите може да бидат со различна големина, но без разлика на големината артикулаторниот говор е отежнат поради постојаната комуникација на усната и носната шуплина. Кај секое родено дете со расцеп на непце постои дисфункција на велофарингеалниот сфинктер, а тоа дава патологија во вербалната комуникација.
Велофарингеалниот сфинктер е тродимензионален мускулен простор кој се наоѓа помеѓу усната и носната шуплина. Го градат латералниот и постериорниот фарингеален ѕид и мекото непце. Улогата на велофарингеалниот сфинктер е да ги оддели усната и носната шуплина, а со тоа да овозможи нормален говор и голтање односно да не дозволи воздухот да избега преку носот наместо низ устата.
Говорот кај децата со расцеп често го прати назален призвук кој се поврзува со велофарингеалната инсуфициенција (слаба функција на мекото непце). Кога мекото непце не го затвара потполно протокот на воздух низ нос, низ тесниот простор поминува воздух и се слуша назален говор. Денес, една од техниките која се користи за испитување на артикулаторната функција во орофарингеалниот регион, и кај нормални и кај патолошки состојби, е магнетната резонанца. Таа во споредба со другите техники е во предност, поради нејзината неинвазивност, нерадиоактивност, а и поради фактот дека е видеофлуороскопска. Се користи за добивање на динамични слики за време на движење т.н. филмови на магнетна резонанца.
Клучни зборови: расцеп на непце, расцеп на усна, велофарингеален сфинктер, велофарингеално затварање, магнетна резонанца.

Introduction
The cleft lip and palate are one of the most common human congenital anomalies with numerous morphological variations and multiplicities. What singles them out is the fact that they are serious health issues affecting the individual for the rest of their lives. They mainly occur like isolated anomalies, usually 1 in 1700 babies, but in 11%, the impaired palatogenesis is accompanied by other congenital impairments of the extremities, heart, blood vessels, eyes, ears, etc. Also, there are hundreds of congenital syndromes where there is a presence of cleft. (1)
In the literature, they are known as cheilo-gnatho-palatoschisis, Lipper-Kiefergaumenspalten, cleft lip and palate, bec-de-lievre et de la division palatine, while among the people as rabbit mouth. The cleft is almost twice more common among boys than girls, and in cases of unilateral clefts, the left side is more affected.
They anatomically occur due to the partial or complete failure for a connection to be established between the maxillary process (processus maxillaris) and frontonasal process (processus frontonasalis) in weeks 4-12 of the intrauterine growth because of

the insufficient migration of the mesoderm from one processus to another. The etiology has not been uncovered yet, but what is clear is that it occurs as a result of the synergic action of more unfavorable factors. The core etiological cause is the heredity, while other possible causes include malnutrition, chemically teratogenic agents (alkaline agents, antimetabolites, kinins, thalomid, salicylates, corticosteroid), infective diseases (rubeola, toxoplasmosis, viral hepatitis), radiation, serious psychological stress, etc.
The cleft lip and palate, depending on which form they appear, generate more problems, such as difficulties in feeding, hearing loss, orthodontic anomalies, impaired speech, impaired resonance, aesthetic and psychosocial disorders, social and professional isolation, etc. (2)

The role of the velopharyngeal sphincter in speech
The impairments in the velopharyngeal sphincter especially affect speech - the most developed means for communication, an essential tool for communicating and expressing thoughts. Impaired speech disables the individual and deeply disturbs their emotions.
The core problem of impaired speech lies in hypernasality, that is, part of the airflow "escapes" through the nose during speech due to the incomplete closure of the palatopharyngeal sphincter, or the insufficient contact between the soft palate and the posterior wall of the pharynx. The situation can be more severe and with impaired articulation. Hypernasality is particularly noticeable when consonants are being produced, especially those with nasal resonance. The children with cleft develop an intuitive attempt to close the nostrils with contraction of the nasal ala muscles, or they produce a sound in the pharynx. The speech in children with cleft develops like in other children, with the exception to one element missing from the picture: the apparatus for normal articulation. Due to the inability to separate the oral from the nasal cavity, the speech is nasalized.
D'Antonio and Scherer (3) outline a few important communication factors: type and gravity of cleft; age, time and efficiency of correction; persistent remainder of the cleft; the status of the velopharyngeal sphincter; hearing status and socio-economic and linguistic status.
The main role in speech is played by the velopharyngeal sphincter. The velopharyngeal mechanism is constituted with the aid of the muscles of the soft palate, the posterior and lateral pharyngeal walls (4), separating the oropharynx from the nasopharynx. All these structures, especially the soft palate, play a crucial role in the velopharyngeal closure. (5)
In velopharyngeal closure, there are a few patterns: coronary (dominant is the movement of the soft palate towards the pharyngeal wall), sagittal (dominant is the movement of the lateral pharyngeal walls towards the middle line of the pharynx), circular (there are balanced movements of the lateral pharyngeal walls), circular with Passavant's ridge (the circular closure is connected with the mucous called Passavant which actually is the ridge of the posterior pharyngeal wall). (6)

The role of soft palate in speech
While researching the morphology and function of the soft palate, Skolnick et al. (6) have proved that the soft palate enables the nasal and oral cavity to be separated during activities such as sound emission, blowing, whistling, swallowing, sucking and vomiting reflex. (7,8)

The role of the soft palate is especially important for speech. The velopharyngeal mechanism allows expiratory flow of air coming from the lungs and sound produced by the vocal cords, directed towards the oral cavity in the class of oral sounds and the nasal cavity in the class of nasal sounds. It also aids the oronasal resonance; it restores balance and secures the necessary intraoral pressure. In case of dysfunction of the velopharyngeal mechanism, space called velopharyngeal closure is created inducing velopharyngeal dysfunction or incompetency. One of the causes for such a closure is the presence of short soft palate. This dysfunction is a type of velopharyngeal insufficiency that can be corrected surgically or with prosthetic correction followed by a speech therapy.

In cases when dysfunctions of this type occur as a result of a failure to move velopharyngeal structures, that is, there is physiological or neuromotor impairment; there is a case of velopharyngeal insufficiency, which can partially be removed with speech therapy. (5,9) On the other hand, if the dysfunctions are a result of the compensatory articulation or can be attributed to other mistakes in speech and are not reflected on the physical and neural changes, then they indicate towards speech therapy. (10)

People with velopharyngeal dysfunction have a nasalized speech, i.e. the nasal emission, poor intraoral pressure are the oro-nasal connection with consequences on the pronunciation of oral consonants. (11,12)

Leow (13) emphasizes that the cleft lip and palate affect other oral functions, while the consequences are especially evident in speech.

Diagnostics of the velopharyngeal dysfunction

Of all the subtypes of clefts, only palatal fractures can cause velopharyngeal dysfunction; therefore, it is essential to determine the level of dysfunction of the velopharyngeal sphincter for diagnostic and therapeutic purposes. One of the most commonly used methods for diagnostic and therapeutic purposes is the magnet resonance, a powerful tool used in the oropharyngeal region to examine the articulatory function of normal (14-18) and pathological (14,19) conditions. It has been proved that the magnetic resonance offers more potential advantages compared to other dynamic techniques for recording including the video nasal endoscopy (20-23), because of it non-invasiveness, and video fluoroscopy (20,23), due to its non-radioactivity. The magnetic resonance has been used so far to obtain static and continuous pictures of different parts of the oropharyngeal structures. (24, 25)

Masaki et al. (26) obtained the first dynamic pictures of magnetic resonance while in motion, the so-called magnetic resonance imaging. They used a synchronized method of a representative sample to record the articulatory movements while a healthy patient repeated short words synchronized with sequences of scans of magnetic resonance. The research confirmed substantial differences in the shape of the tongue, as well as the spatial difference between the tongue and palate when the picture of magnetic resonance were obtained either by a synchronized technique of a sample (i.e. dynamic pictures) or by a turbo-flash technique (i.e. continuous pictures). The applied technique has proved as more efficient since, in that way, different diagnoses can be established on the basis of the pictures obtained by these two methods.

Later Kane et al. (27) used an echo in patients with cleft lip and palate. Together they used axial view of the dynamic pictures of the magnetic resonance to quantify the temporary changes in the velopharyngeal structures. It is important that the tridimensional movement of the oropharyngeal structures included in the articulation

(lips, jaws, tongue, soft palate, oesophagus, vocal cords) is evaluated simultaneously in order to establish the damaged special structures of the articulators. No other study before has used the technique of video magnetic resonance in sagittal view in which the articulators can be seen perfectly. Theoretically, the pictures of magnetic resonance in sagittal view are more useful than the other dimensions (eg. axial) if we want to analyze the dynamic movements of the oropharyngeal structures while in function, since the reconstruction of the pictures in sagittal view, unlike the pictures from the other dimensions, gives low spatial resolution. Video fluoroscopy and video nasal endoscopy show dynamic articulatory movements of the velopharyngeal structures.

However, the video magnetic resonance outperforms these two video techniques from two aspects. First, it is not radioactive for the patient, unlike the video fluoroscopy which has this effect. Moreover, the magnetic resonance enables to obtain pictures from more dimensions for more situations, which is not the case with the video fluoroscopy. Due to its non-radioactive nature, the magnetic resonance allows another taking of detailed pictures together with the changes in articulation, such as the movement of the lower jaw.

Secondly, the video magnetic resonance allows performing research of a wider oropharyngeal space compared to the video nasal endoscopy, which allows only localized pictures to be made.

Shinagawa (28) used video magnetic resonance in patients with cleft lip and palate to obtain spatial information connected with the dynamic changes in the shape and interaction of the oropharyngeal structures involved in the articulation. In the formation of the bilabial, alveolar and velar consonants, the posterior movements of the tongue and frontal movement of the posterior pharyngeal wall were clearly perceivable in patients with cleft lip and palate compared to patients with no cleft lip or palate. It is not clear why the velopharyngeal closure was not noticed in the articulation of /pa/, /ta/ and /ka/ among patients with cleft lip and palate despite not showing hypernasality in the pronunciation of the separate sounds /pa/, /ta/ and /ka/. The author suggests that the findings can be attributed to the fact that separate sounds become words (eg. papapapapapa) than a single pronunciation of /pa/. This presumption can be a consequence of the existence of average hypernasality while speaking.

It is known that the posterior pharyngeal wall is immovable in articulation. (29) Still, the movement of the posterior pharyngeal wall, far below the Passavant's ridge, in patients with cleft lip and palate is interesting because of the functional compensation of the anatomic abnormality of the orophryngeal structures. The overall movements of the posterior pharyngeal wall are central to the relationship with the Passavant's ridge in patients with cleft lip and palate. These dynamic movements can be seen with video magnetic resonance and imaging, but no with other methods. (14-18)

The shape of the tongue and the spatial relation between the tongue and the palate are completely different in the dynamic pictures made with the video magnetic resonance and those obtained with the turbo-flash magnetic resonance. (26) This means that the synchronized method of a sample can produce more realistic movements of the oropharyngeal structures participating in the dynamic articulation.

The velopharyngeal closure is achieved with the combination of tridimensional changes of more oropharyngeal structures: posterior movements of the soft palate, mesial movements of the lateral pharyngeal walls and anterior movements of the posterior pharyngeal wall. (30) There are a few methods and locations of velopharyngeal closure (20,31), which point to the existence of individual variations in cases of functional adaption of the anatomic differences. Since congenital loss or impaired palatal

musculature appear in patients with cleft lip and palate (32), the movement of the posterior pharyngeal wall among these patients can lead to hyperfunction of the palatopharyngeal and palatoglossal muscles in order to compensate for the possible hypofunction of m.levator veli palatine, prompted by the anatomic abnormality. (31,33,34)

Two other factors can also explain the unusual movements of the posterior pharyngeal wall. One is the reclining pose. Shinagawa (28) cannot take into consideration the probability for increased compensation in this region during the articulation in patients with cleft lip and palate. Other possibility is the position of the head, which is not standardized with the apparatus for magnetic resonance. Since this recording asks for natural and comfortable head position, differences in the neck angle can contribute to differences in the movement of the posterior pharyngeal wall in two patients. Also, it is important for the temporal points for each of the patients to be established so as to determine the timing of the articulatory movements and constructions. This is essential since the patients with cleft lip and palate can speak more slowly and extend their words. The damage of the upper lip as a result of a post-operative scar, or disproportionate positioning of the upper and lower lip in sagittal position due to retruded maxilla can predispose these speech characteristics in patients with cleft.

Naturally, diagnostics is not the last link in the chain of solving the velopharyngeal impairments. The defining correction from an anatomical, morphological or functional point of view is central. The efficiency of the particular undertaken therapy will determine the success with which the existing problem will be solved. Therefore, palatoplastics comes here first.

Possible therapeutic procedures for treating cleft

Palatoplastics is indicated in cases where the cleft includes the palate. This intervention consists of anatomic and functional reconstruction of the cleft, enabling separation between the oral and nasal cavity. (35) The intervention can be carried out in around the 12^{th} month of life because better functional results have been noticed in cases when it was performed preventively. (36,37) Still, surgery does not always offer satisfactory results regarding the signs and symptoms caused by the velopharyngeal dysfunction and insufficiency. The velopharyngeal rehabilitation is the main aim of every performed surgical technique. The outlined aims also include correction of hypernasality and removal of the nasal cavity, as well as improvement of the conditions for intraoral pressure and airflow. (38,39) However, surgical interventions do not necessarily mean improved speech; thus, an additional post-surgical attempt would be speech therapy in most of the cases.

Wearing palatine braces, either temporary or permanent, can be another therapeutic possibility in cases when the surgery has been unsuccessful, or simply it cannot by performed due to the abovementioned aims.

Shinagawa (28) suggests perceptive and instrumental procedures for evaluating the velopharyngeal function and approaching the solution of the same by accentuating the instrumental video fluoroscopy and nasopharyngoscopy as the main tools for assessing the model of velopharyngeal closure (40-42) and the methods of choice. Nasopharyngoscopy is in fact a direct method for assessing the velopharyngeal function by allowing visualization of the velopharyngeal structures, even during speech. The Brazilian literature includes some studies which describe models of velopharyngeal closure in vitro (43,44), but only a few in vivo which can be used as a reference to further studies.

In the study by Silveira Di Ninno (45), the differences in velopharyngeal structure are observed in relation with the patients' age. According to the literature, the physiology of the velopharyngeal mechanism shows differences according to age with the appropriate changes that occur in the first year of life, and as the orofacial system grows and matures so do the functions of the orofacial muscles change. Around the fourth year of life, along with the teeth occlusion and facial growth, the tongue develops in the intraoral space and, in turn, reduces the anterior projection; it does not get involved directly in the oropharyngeal space and the movement of the soft palate, which actually proves the absence of significant correlation between the velohparyngeal closure and age. (46) Despite the fact that in most of the female patients there is a velopharyngeal coronary closure, while the males patients are characterized with circular one, the gender differences are not wide. In studies that used radiological methods of evaluation, such differences between the genders have not been noticed in the physiology of velopharyngeal mechanism for all types of closure and levels of constriction. (47)

No dependence between the type of the cleft and type of velopharyngeal closure has been noticed. In the literature, there has not been a study with such correlation, but the present results are expected since the cleft palate, either isolated or together with the cleft lip, affects the velopharyngeal function equally.

There has been some data in the literature which point to female patients being mainly diagnosed with cleft of the post foramen incisivum, while male with cleft of transforamenal one.

Among patients with adequate velopharyngeal closure, the sagittal is mainly present, while the coronary closure is less present. The dominant closure in patients with inadequate velopharyngeal closure is the coronary one. That is also the most present in the population without clefts.(20, 43, 44, 47)

The patients having undergone surgery have a better mobility of the velopharyngeal wall, and the velopharyngeal closure can be easily achieved due to the compensation with the insufficiency of the soft palate.

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