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The Role of Endoscopy in the Assessment of the Vocal Tract and Proper Speech Acquisition in Children with Cleft Palate

SUMMARY

Speech reception is a perceptual phenomenon. Hence in the assessment of cleft palate speech its auditory evaluation is a basic golden standard with respect to identification of speech disorder and defining the degree of its intensity. Yet auditory evaluation does not provide information about the cause of this disorder. And here, with a good knowledge of the cleft palate issues and perfect skills in using the endoscopic technique, nasofiberoscopy, i.e. endoscopy using flexible optics, is irreplaceable. It enables direct visualization of the velopharyngeal sphincter at rest and during speech, allowing the simultaneous assessment of its structure and function. It also allows simultaneous endoscopic evaluation of other elements of the vocal tract that exert a direct effect on speech resonance. Thus it has fundamental significance for the choice of the treatment procedures and for the evaluation of the successive stages of treatment and rehabilitation.

Key words: cleft palate, speech assessment, hypernasality, velopharyngeal insufficiency, nasofiberoscopy, pharyngoplasty.

The basic problem and the starting point with regard to the possibilities of proper speech acquisition in children with cleft lip and/or palate is a defect in the peripheral speech apparatus. The presence of orofacial clefting creates a pathological opening between the oral and nasal cavities, and the inability to achieve the velopharyngeal closure. This physical impossibility to create an appropriately high pressure in the oral cavity that in fact forms one common space with the nasal cavity, or even opens up at the front to the outside world through the split in the alveolus and the clefted lip, is responsible for the impossibility to implement oral consonants whose articulation requires the c reation of pressure in the oral cavity. Moreover, cleft muscles of the velum, instead of joining medially, and synergically drawing together in speech, guaranteeing its correct resonance, are attached alongside the cleft margins. The impossibility to implement the velopha-

ryngeal closure results in the escape of the air through the nose at the realization of oral sounds, giving the speech a characteristic tone called open nasalisation.

Cleft lip and/or palate is the most frequent developmental craniofacial defect. In Poland, about 600-800 children with this developmental defect are born every year, with an average frequency of occurrence equalling 1,6-2/1000 live births. The defect is constituted by partial or total lack of fusion of facial and palatal outgrowths of the maxilla in the midline, normally occurring in the first months of fetal life. Depending on the site, or sites, in which this lack of fusion has occurred. one has to do with cleft lip, alveolus or palate, uni- or bilaterally. The extent of the defect within a given site can differ with regard to the velum from the marginal form, e.g. the uvular cleft without the involvement of other muscles of the velum to a total clefting of the hard palate and the soft palate that reaches the foramen incisivum. A particularly difficult to diagnose form of the palatal clefting is the submucous cleft palate, especially in its occult form (Hortis-Dzierzbicka, 2011). It also seems worth noting that about 30% of the submucous cleft palate are revealed following a careless adenotomy (Witzel, 1986). In this context it must be remembered that it is not only the extent of the cleft on the Veau or Kriens scale that decides on the gravity of the defect, but also the cleft width of the and the degree of the development of the muscles of the velum.

The delay in the development of active speech in children born with cleft palate becomes apparent very early. As soon as at the period of babbling, the speech of infants with palates not treated surgically is characterized with lack or limited number of anterior consonants, a high frequency of retractions and glottal compensations. Thus, early surgical closing of the cleft palate crevice, i.e. in the first year of life, has a fundamental significance for the development of speech in children born with that developmental defect, simultaneously conditioning early rehabilitation on the basis of the reconstruction of the correct anatomical structures. It simultaneously decreases the impact of the defect upon the acquisition of the ability of correct articulation and upon language competence in children born with cleft lip and/or palate. This happens because pronunciation errors occur in those children as a direct result of the anatomical defect. Starting as soon as at the earliest years, over time they are incorporated into the phonological system of a given child (Chapman, 1993), thereby, in the long run, they adversely affect the communication of the child with the social environment and the chances of obtaining proper education. It is within the first three years of the child's life that most dynamic acquisition of the phonological system takes place (Łobacz, 1996). Hence, as correctly observed by Pluta-Wojciechowska (2011), the impact of the palatal cleft on the speech acquisition should be looked upon in an integral way, not only with regard to distortions in the realizations of phonemes.

According to the data in the literature of the subject, velopharyngeal insufficiency occurs on the average in about 25-30% of the cases, following the primary palatal closure. (Peterson-Falzone, Karnell, Hardin-Jones, 2001). (Fig. 1-2). Another frequent complication of primary palatoplasty are palatal fistulas, i.e. unintended communication between the oral and nasal cavities in the form of resting openings that have remained despite the surgery on the palate. (Fig. 3). The effect of the fistulas in the palate exerted upon speech depends on their localization and size, but generally it is similar to the symptoms of velopharyngeal inssuficiency, caused by insufficient length or dysfunction of the surgically joined palate. Hence

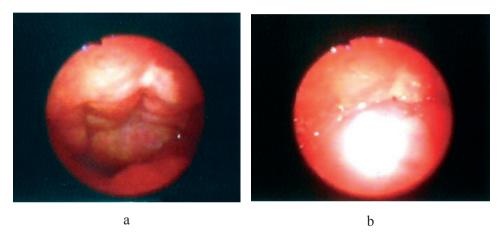


Fig. 1. Endoscopic view of the velopharyngeal sphincter function after surgical closure of the cleft palate: a good result of the surgery: (a) breathing; (b) phonation – complete velopharyngeal closure, correct speech resonance, open nasalization (-).

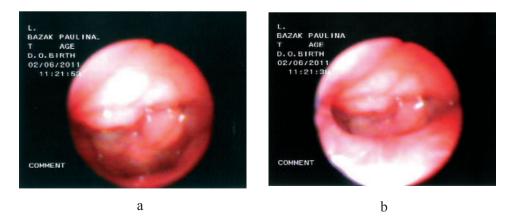


Fig. 2. Endoscopic view of the velopharyngeal sphincter after surgical closure of the cleft palate: an unsatisfactory result of the surgery: (a) breathing; (b) phonation – incomplete velopharyngeal closure, open nasalization in speech (3+).

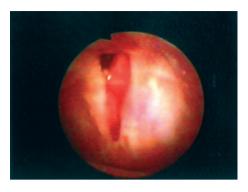




Fig. 3. The most frequent complication remaining after primary palatal repair – the so-called oro-nasal fistulas: (a) anterior (in the hard palate); (b) a fistula in the soft palate, an intraoral view.

it is crucial to realize that not only the moment at which the operation is performed is important, but also the skill and experience of the surgeon. The unsatisfactory result of the surgery consisting in the failure to obtain full air-tightness of the palate (oro-nasal fistula) or a failure to obtain tightness of the velopharyngeal closure (short palate, extensive scars, wrongly joined muscles) is more or less equivocal with an operation that was either late/delayed or for some reasons abandoned. Regardless of the cause, a common clinical sign of both velopharyngeal insufficiency and the presence of fistulas in the palate is air escape through the nose accompanying the articulation of oral sounds, manifested by open nasalization and/or specific air escape produced by the so-called anterior fistulas or a marginal velopharyngeal closure. These symptoms can be accompanied by a delay or complete inhibition of the development of articulation of oral consonants and the development of the so-called vowel speech, shifting of places of articulation of certain sounds to other places in the oral cavity (palatalizations, retractions, coarticulations) and outside it (compensatory pharyngeal and glottal closures). Secondary operations aiming at the repair of the above-mentioned effects of primary operations are as a rule performed later than at the 5th year of age and their result is often dubious, both in terms of surgery for velopharyngeal insufficiency and fistula closures. In the world literature, the effectiveness of pharyngoplasty procedures with regard to the elimination or significant reduction of hypernasality is evaluated as 70%, while the effectiveness of palatal fistula closures is about 30% and these operations must often be repeated several times.

Previously, the researchers dealing with open nasalization, the main cleft speech feature, wanting to explain its nature, focused their attention exclusively on the role played by the soft palate and its ability to approach the posterior pharyngeal wall during speech. It had a direct relation with the fact that the first meth-

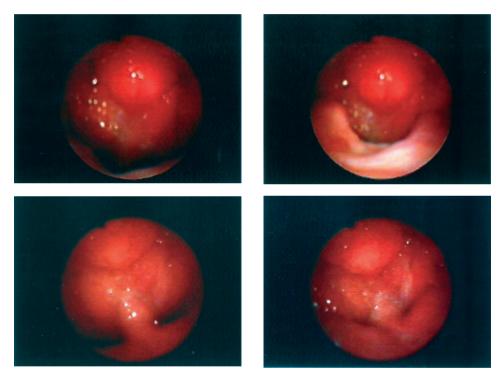
od of the evaluation of soft palate activity during speech was cephalometry with the use of lateral radiograms. However, it enables evaluation exclusively in the sagittal plane, that is, of the length of the palate and its position and distance from the posterior pharyngeal wall at rest and speech – pronouncing isolated sounds, most often, [a] or [e] or [s]. In the 1960's, this method evolved into the direction of cineradiography and, later, videofluoroscopy, a technique commonly used at present, especially in the USA and Great Britain (Hortis-Dzierzbicka and Komorowska, 1994). These methods allow one to analyze the activity of the velopharyngeal sphincter during speech using not only isolated sounds, but also consonant-vowel sequences and short utterances, both in the sagittal and horizontal planes.

Another method of the assessment of soft palate and pharyngeal activity during speech is nasopharyngoscopy, i.e. the assessment of the degree of velopharyngeal closure during speech with the use of fiberoptics, preferably using standardized speech tests (Heningson, 1988, Hortis-Dzierzbicka, 2004). Undoubtedly, the greatest advantage of nasofiberoscopy in the assessment of velopharyngeal closure lies in the fact that it allows a direct simultaneous and multiplane visualization of all elements of the velopharyngeal sphincter, i.e. the soft palate, posterior and lateral walls of the pharvnx. When performing nasopharvngoscopy, the clinician observes the soft palate from the top, having simultaneously a detailed view of the velopharyngeal sphincter area, both at rest and during speech. Using this method one can diagnose even the minute details such as e.g. the missing uvula muscle and irregularities caused by the presence of the adenoidal tissue. Joining the endoscope with the vision track and a simultaneous connection of a high quality microphone allows to register on durable carrier media the velopharyngeal activity during speech, which allows not only a direct assessment, but also archivization of the data for retrospective and comparative analyses. This has a fundamental significance for both final and periodic, often far removed in time, assessments of the effects of treatment and rehabilitation.

A significant argument for the endoscopic technique can be the fact that the velopharyngeal sphincter function assessment should not be removed from its context, that is, it should not be done without a simultaneous thorough assessment of other component elements of the vocal tract. This is because the efficiency of the vocal apparatus in children born with cleft palate is determined not only by the reconstructed palate, but also by the patency and air tightness of the nasal passages, the size of the palatine and pharyngeal tonsils, the state of the larynx, and the correct functioning of the Eustachian tubes that enables ventilation of the middle ear. All these organs exert an influence on the ultimate speech resonance, and direct visualization by means of nasofiberoscopy is indispensable in their complex assessment (Hortis- Dzierzbicka, 2001).

Sole rehabilitation of speech by a speech therapist following cleft palate surgery in patients with significant tissue deficit or restricted mobility of the palate

is often ineffective. It must then be supported by surgery. Surgical treatment of the secondary velopharyngeal inefficiency is aimed at the improvement of the velopharyngeal closure. These operations divide into two basic types: carried out with the involvement of the palatopharyngeal muscles (Sloan 2000). In cases of suspected wrong muscle fusion following primary surgery, the re-operation on the palate can be performed and the correct direction of the muscles of the palate can be then reconstructed. However, when the newly joined palate is too short, this operation can be connected with posterior pharyngeal flap pharyngoplasty procedure (Fig.4). The preoperative assessment with the use of the nasofiberoscopy



Ryc. 4. Severe velopharyngeal insufficiency following palatal surgery before and after secondary surgery for speech:

- a) Endoscopic view of the velopharyngeal sphincter before a palatal re-repair and pharyngoplasty the breathing phase
- b) Endoscopic view of the velopharyngeal sphincter function prior to a palatal re-repair and pharyngoplasty phonation incomplete velopharyngeal closure with a large centrally located velopharyngeal gap; open nasalization in speech (3+)
- c) Endoscopic view following palate re-repair with concurrent posterior pharyngeal flap pharyngoplasty the breathing phase; the pharyngeal flap is seen in the midline.
- d) Endoscopic view following palate palatal re-repair with concurrent posterior pharyngeal flap pharyngoplasty- phonation complete velopharyngeal closure with the flap at the bottom; correct speech resonance, open nasalization. (-)

permits the estimation of the degree of the velopharyngeal sphincter inefficiency and evaluation of the movement of its individual elements at speech (palate, the posterior and lateral walls of the pharynx), which has a fundamental significance for the choice of the method of the operation. Post-operatively, nasofiberoscopic assessment of velopharyngeal sphincter function during speech enables to determine the cause of its possible failure in the form of a persistent velopharyngeal inefficiency and a reflection of this state in speech as a persisting hypernasality or velopharyngeal snort (Hortis-Dzierzbicka, 2004). As was mentioned above, the results of various reparative operations that aim at an improvement of the functioning of the velopharyngeal sphincter are controversial, although they are regarded as satisfactory more often than in the case of palatal fistulas.

Next, the effect of the oro-nasal fistulas is to a considerable degree conditioned by their localization and size. Their most frequent location is the postalveolar region and the anterior part of the hard palate (i.e. the so-called anterior fistulas), and the border between the hard and soft palate. The fistulas in the soft palate are relatively least frequent. As a rule, larger fistulas, of a diameter exceeding 5 mm, cause open nasalization, and smaller than that, especially retroalveolar fistulas or small fistulas in the hard palate cause characteristic noises accompanying speech. In the Polish language, these noises usually accompany sibilants spelled as {s} and {sz}, because of particularly frequent occurrence of fistulas in the articulatory region of these consonants. Yet the hoarse or metallic nasal noises that sound definitely different and are often unpleasant to the ear are associated with the borderline, or small velopharyngeal inefficiency (Kummer at el., 1992). When one is not sufficiently acquainted with the cleft palate issues and has an untrained ear, the fistula noises are often confused with velopharyngeal insufficiency. The same can be said of the compensatory grimaces, often very intense, of the facial muscles, which is an unconscious defence of the organism against the escape of air through the nose during speech, independently on the cause. Not without significance is also the fact that the sole presence of oronasal fistulas is sufficient to disturb the functioning of the velopharyngeal sphincter, because as a result of a fistula presence the intraoral pressure can decrease, causing insufficient stimulation of the sphincter during speech (Henningson and Isberg, 1987). It also is important in planning and choice of further secondary operations aimed at improving the quality of speech. The integrity of the palate should be the restored first, and then, according to need, other reparatory operations should be performed, aiming at the improvement of the velopharyngeal sphincter. However, every case requires careful consideration of the choice of the method if treatment, as well as individual approach, that is, extensive experience in complex treatment of the defect.

The fact that the symptomatology of oronasal fistulas and velopharyngeal inefficiency is often similar is the reason why lack of adequate knowledge and

lack of the possibility of establishing correct differential diagnosis makes that the choice of the surgical method is often erroneous and leads to an unnecessary operation that does not remove the cause of the speech defect, not giving a chance for its improvement. Thus the endoscope is indispensable for correct differentiation of the participation of palatal fistulas and the velopharyngeal dysfunction in the persistence of the described features of cleft speech, both pre- and post-operatively.

In the literature on complications following pharyngoplasty, the issue of the occurrence of obturation of the upper respiratory tract following the use of a posterior pharyngeal flap is often raised (Shprintzen, 1988; Fraulin at al., 1998). Its extreme form leads to the development of the obturative apnea syndrome (Wells at al., 1999; Liao et al., 2002). It seems that the conception of the use of extra large flaps, accepted by many surgeons, also known as 'overcorrecton' is erroneous. This procedure is applied to patients with weak movements of the lateral walls of the pharynx in speech; the condition is usually diagnosed prior to surgery by means of nasofiberoscopy or vieofluoroscopy. Another cause of the sleep apnea syndrome following posterior pharyngeal flap pharyngoplasty is leaving intact hypertrophied palatine tonsils. As a rule, they should be removed as early as several months before pharyngoplasty (Witt, 2000), or as a concurrent procedure (Eufinger i Eggeling, 1994). Otherwise, the tonsils can obstruct, partially or fully, the bottom side of lateral portals, obstructing the patency of the respiratory tract at this level. With regard to the pharyngeal tonsil, because of the mentioned large percentage of velopharyngeal closures with its indisputable participation, also following pharyngofixation, especially in young patients, qualifying for its removal in cleft palate patients should be performed with extreme caution.

It should be emphasized again that in all these classifications for additional or complementary surgeries, pre-operative nasofiberoscopic assessment is necessary, as only it enables a targeted therapy rather than a blind treatment. It is presently regarded as an instrumental "golden standard" in establishing the diagnosis of speech disorders in cleft palate patients.

Identification of the adenoid hypertrophy, its involvement in the velopharyngeal closure, assessment of tonsillar hypertrophy, especially posteriorly directed, a resulting obturation of the upper respiratory tract, identification of extra large pharyngeal flap or flap retraction, and its involvement in the velopharyngeal closure can be performed only with the use of an endoscopic examination. Thus fiberoptic endoscopy, enabling a direct and complete assessment of the velopharyngeal sphincter shape and function, as well as of those other elements of the vocal tract that exert a direct effect on the nasal resonance, is indispensable at every stage of speech pathology treatment in cleft palate patients. What is absolutely significant, this technological possibility must be backed up by excellent knowledge of the

cleft issues and wide experience in the evaluation of cleft speech, as well as in assessment of the velopharyngeal sphincter function.

Nasofiberoscopy has also a fundamental significance for diagnosing and applying appropriate treatment in submucous cleft palate. As was widely discussed before (Hortis-Dzierzbicka, 2011), so-called occult submucous cleft palate that defies recognition even in a detailed intraoral examination, constitutes a particular diagnostic difficulty. It does not manifest the triad characteristic of the overt submucous cleft palate: a cleft uvula, "zona pellucida", a notch in the hard palate in place of a posterior nasal spine at palpation. Sometimes it involves only a part of the palate muscles and can be diagnosed only endoscopically. The contrast between the degree of velopharyngeal insufficiency and the image of the palate in endoscopy, vs. the image obtained at intraoral examination can often be amazingly strong, pointing clearly at the need of operative treatment. Unfortunately, in Poland this method is not used besides the authors' home institution. Moreover, the concept of occult submucous cleft is little known, which is reflected negatively in the possibility of appropriate management in such cases. More often than not, children with not only with occult submucous cleft palate, but also with its overt type (the symptom triad mentioned above) and with severe hypernasality and strongly impaired speech intelligibility remain without proper diagnosis throughout years. Moreover, what we particularly emphasize, having an exceptional experience in this field at the country scale, even if the presence of the symptom triad that is easy to recognize, and if the diagnosis of the submucous cleft palate is finally be established, speech therapists for their own or by order of a laryngologist or a phoniatrician, spend many years on "rehabilitation" of these children, in fact without any chance to obtain a favourable result. Reaching a good result is impossible because in such cases what is needed is surgical closure of the clefted palatal musculature. Speech rehabilitation should be implemented no sooner than after the operation, and not before it or in place of it.

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