Public health

CHANGES IN VOICE QUALITY OF THE CHILDREN WITH CLEFT/ LIP IN RELATION TO CHILDREN WITHOUT ANOMALY

Mira Jovanovska[1], Mirjana Petrovic-Lazic[2], Vesna Lazarovska[1]

[1] *Institute for Rehabilitation of Hearing, Speech and Voice, Skopje, Republic of North Macedonia*
[2] *University of Belgrade, Faculty of Special Education and Rehabilitation, Serbia*

Citation: Jovanovska M, Petrovic-Lazic M, Lazarovska V. Changes in voice quality of the children with cleft/lip in relation to children without anomaly. Arch Pub Health 2021; 13 (2)30-38.

doi.org/10.3889/aph.2021.6005

Key words: voice, cleft palate/lip, changes in voice quality, speech.

***Correspondence:** Mira Jovanovska, Hearing, Speech and Voice Rehabilitation Center, Skopje. E-mail: mira.jovanovska@yahoo.com

Received: 15-Apr-2021; **Revised:** 30-May-2021; **Accepted:** 5-Jun-2021; **Published:** 20-Noe-2021

Competing Interests: The author have declared that no competing interests

Abstract

Children with cleft palate/lip are exposed to risk for verbal communication disorders that include resonance, articulation, voice disorders, and expressive language. The aim of this paper was to evaluate the changes in the voice quality of the children with cleft palate in relation to children without anomaly. The study included 52 participants, 26 with previously corrected cleft palate / lip, and 26 respondents without anomaly who are between 3 and 6 years old. Subjective assessment of voice quality was performed by using the GRBAS scale. Perceptual scales are important in assessing the voice quality, determining the degree and severity of voice disturbance, and deciding on further clinical procedures. Although the instrumental approach to voice examination is relevant because it provides objectivity, the subjective impression in assessing voice quality is crucial. Statistical processing was performed by groups, group structure (gender and age), a certain highest value, as well as percentage participations. The results showed that 50% of the participants were diagnosed with certain changes in the voice. Children aged 3-5 years have a 2.25 times higher incidence of voice changes than children aged 6-8 years. The largest number of participants belong to group 0 - There is no change in voice quality which represents as much as 50% of the total number of patients in the first group. While *„3 - Pronounced changes in the voice"* were not identified in the respondents aged 3-5 years. In 50% of the total number of participants there is no change in voice quality and these are located in girls aged 3-5 years.

Јавно здравје

ПРОМЕНИ НА КВАЛИТЕТОТ НА ГЛАСОТ КАЈ ДЕЦА СО РАСЦЕП НА НЕПЦЕ/УСНА ВО ОДНОС НА ДЕЦАТА БЕЗ АНОМАЛИЈА

Мира Јовановска[1], Мирјана Петровиќ -Лазиќ[2], Весна Лазаровска[1]

[1] *Завод за рехабилитација на слух, говор и глас, Скопје, Република Северна Македонија*
[2] *Белградски универзитет, Факултет за специјална едукација и рехабилитација, Србија*

Цитирање: Јовановска М. Петровиќ –Лазиќ М, Лазаровска В. Промени на квалитетот на гласот кај деца со расцеп на непце/усна во однос на децата без аномалија. Арх Ј Здравје 2021;13(2)30-38..

doi.org/10.3889/aph.2021.6005

Клучни зборови: глас, расцеп на непце/усна, промени на квалитетот на гласот, говор.

***Кореспонденција:** Мира Јовановска, Завод за рехабилитација на слух, говор и глас – Скопје, Република Северна Македонија. E-mail: mira.jovanovska@yahoo.com

Примено: 15-апр-2021; **Ревидирано:** 30-мај-2021; **Прифатено:** 5-јун-2021; **Објавено:** 20-нео-2021

Конкурентски интереси: Авторот изјавува дека нема конкурентски интереси.

Извадок

Децата со расцеп на непце /усна се изложени на ризик од нарушувања во вербалната комуникација кои вклучуваат резонанца, артикулација, нарушувања на гласот и експресивниот јазик. Целта на овој труд беше да се согледаат промените на квалитетот на гласот кај децата со расцеп на непце во однос на децата без аномалија. Во истражувањето беа вклучени 52 испитаници од кои 26 сопретходно корегиран расцеп на непце/усна, а 26 испитаници без аномалија на возраст од 3 до 6 години. Субјективна проценка на квалитетот на гласот е извршена со примена на скалата Грбас. Перцептивните скали се значајни во проценка на квалитетот на гласот, одредување на степенот и тежината на пореметувањето на гласот, како и во одлучување за понатамошните клинички постапки. И покрај тоа што инструменталниот пристап во испитувањето на гласот е релевантен бидејќи обезбедува објективност, субјективниот впечаток во проценка на квалитетот на гласот е клучен. Извршена е статистичка обработка по групи, структура на групите (пол и возраст), одредена е највисока вредност, како и процентуални учества. Резултатите покажаа дека кај 50% од испитаниците се дијагностицирани одредени промени во гласот. Децата на возраст од 3-5 години имаат 2,25 пати поголема инциденца на промени на гласот во однос на деца на возраст од 6-8 години. Најголемиот број на испитаници спаѓаат во групата 0 - *Не постои промена во квалитетот на гласот* кој што претставува дури 50% од вкупниот број на пациенти во првата група. Додека „3 - *Изразени промени во гласот*",не се идентификувани кај испитаниците на возраст од 3-5 години. Кај 50% од вкупниот број на испитаниците не постои промена во квалитетот на гласот и овие се лоцирани кај девојчиња на возраст од 3-5 години.

Introduction

Correct, clear and aesthetically pleasing voice is a means of communication between people and through speech, as a perfect form,represents the most comprehensive human activity.[1]

Voice disturbance can be named as anything that reduces the effect of communication and makes the voice less pleasant whereby the speaker for an exemplary strong and pleasant voice consumes too much energy. Voice disorder exists when one or more aspects of the voice e.g., strength, height, quality or resonance deviates from the norms of age or gender of the interlocutor.[2,3]

When we talk about voice disorders in preschool children, we usually think of childhood hoarseness. Hoarseness or dysphonia is the medical and speech therapy name for any deviation from the normal features of pitch and volume of the voice.

Dysphonia can occur at any age, even at the earliest one, and often requires intervention by various professionals, not only phonologists and otolaryngologists, but also the speech therapist himself. Increased strain of the laryngeal musculature is possible to be replaced by intensity of the neck muscles during speech especially in boys. Prevention and early detection of voice disorders are very important so that they do not linger in later years. Changes in the voice of children are most often observed in the period from the third to the sixth year, which is associated with the specifics and limited possibilities of the child's voice in this period.[4]

Some research show that changes in voice quality, especially in the form of hoarseness, are present in more than 50% of preschool children. The most common cause is hyperkinetic dysphonia which most often occurs due to incorrect or inappropriate use of the voice.[5] The voice in hyperkinetic dysphonia is most often hoarse, with a lot of noise and hoarseness.

Children with cleft lip / palate are at risk for verbal communication disorders that include resonance, articulation, voice disorders, and expressive language.[6]

Many researchers note an increase in the symptom of hoarseness in patients with cleft palate compared to the normal population; however, the exact prevalence is currently unknown. Previous reports of dysphonia indicators for the cleft palate population are 12% to 43%.7, These are similar to, until present, rates of hoarseness in the normal paediatric population, estimated to be 6 to 34%.[8, 9,10,11]

Researchers have set up many different hypotheses to explain this possible increase in hoarseness, the most common being laryngeal compensation for abnormal velopharyngeal valve.[12,13]

Many patients with cleft palate also have velopharyngeal insufficiency (VPI), secondary to palatal muscle orientation. Velopharyngeal insufficiency, which may persist after surgical cleft correction, may cause speech hypernasality and difficulty articulating specific consonants due to the inability to build up sufficient air pressure in the oral cavity. Studies report an incidence of hypernasality in patients with cleft palate of 25% to 40%.[14]

Patients with cleft palate with VPI often use compensatory mechanisms to increase articulation. Some of these mechanisms include palatal stops, posterior nasal fricative, velar

fricative, pharyngeal stops, pharyngeal fricative, and glottal stops.14

Glottal stops, in particular, have been implicated in vocal cord abnormalities and voice disorders such as hoarseness.[15,16]

In fact, the Cleft Palate Foundation literature states: "Children with velopharyngeal disorder may also have voice disorders. In this case, your child's voice may sound "breathing" and he or she can get tired easily. This problem is usually caused by the strain he or she puts on the vocal cords while trying to build up the pressure necessary for normal speech.[11]

There are studies in which no link has been found between hoarseness in patients with cleft palate and VPI, which calls into question the long-standing theory of laryngeal compensation as a source of hoarseness.[17]

The aim of this paper is to evaluate the voice changes in children with cleft palate / lips in relation to children without such anomaly.

Material and methods

In the Institute for Rehabilitation of Hearing, Speech and Voice - Skopje 28 patients were diagnosed with cleft lip / palateduring the two years 2019 and 2020. 26 of them are covered by speech therapy. A total of 52 participants are included in the study. Inclusion criteria:

- Respondents with previously repaired cleft palate
- Age of 3-8 years

The participants are divided into 2 groups. The first group consists of 26 (n = 26) children with previously repaired cleft lip / lip, of which 18 are girls and 8 are boys at the age of 3-8 years. The second group - control consists of 26 (n = 26) children without anomaly. The groups matched by age and gender.

The participants were examined individually in a speech therapy office in which the impact of possible noise from the environment was minimized. Before the beginning of the examination, instructions for the course of the examination about the expected activities that are expected from the participantsare given. The examination was performed by three vocal therapists. The therapists were in direct contact with the patient at a distance of one metre and independently assessed the voice. The degree of agreement between the three vocal therapists using the Kappa coefficient was examined. The degree of agreement between the first and second therapist is good (kappa = 0.490, $p < 0.001$), as well as between the second and third (kappa = 0.610, $p < 0.001$) also between the first and third (kappa = 0.481, $p < 0.001$) which has been shown to have a high degree of agreement among vocal therapists.

Perceptual voice assessment was used as a method, which is a subjective method for assessing dysphonia. It is a non-invasive method that does not require additional equipment. The questionnaire for perceptual assessment of the voice GRBAS was used. The auditory-perceptual scale GRBAS is a focused phonetic component in assessing the quality of the voice and is most often used in practice. This scale, developed in Japan by the Japanese Society of Speech Therapists and Phoniatrics (Webb et al., 2004), is considered as the gold standard for perceptual voice assessment and is commonly used by experts around the world.[18]

It is used to analyse voice quality according to five parameters: G - overall grade of dysphonia or abnormality of the voice, R - roughness or psychoacoustic impression of improper vibrations of the extinguishers; B - breathiness - refers to the psychoacoustic sensation of air flow through the glottis; A - asthenia which is a weakness in the voice; S - strain (hyperfunction) in the voice during phonation.

Each parameter of the GRBAS scale is rated on a scale from 0 (normal voice) to 3 (severe pathology). That is, it is used in the evaluation where the quality of the voice is described as follows: when there is no change in the quality of the voice a score of 0 is determined, slight changes in the quality of the voice are marked with a score of 1, moderately changed voice with a score of 2, while changes in the voice with a grade 3. The score is calculated so that for each parameter is used the average value obtained by the therapist, i.e., the average grade that is determined for all three vocal therapists.[18]

In this research, statistical processing was performed by groups, group structure (gender and age), a certain highest value, as well as percentage participations.

Results

Out of a total of 52 children examined in 2019 and 2020, 15.4% are children - patients with cleft palate / lip at the age of 3-5 years, while 34.6% are children - respondents who have been diagnosed deformities at the age of 6 - 8 years.

The distribution of examined patients by years of research is 46 respondents in 2019 and only 6 in 2020, i.e., 88.5% and 11.5% by years, respectively. Hence, the results of the analysis of the obtained results as much as 88.5% refer to the diagnoses determined by the examinations in 2019, which is a dominant level of responsiveness. Due to the negligibly small number of respondents (patients) in 2020 (only 6), which refers to only one group of children aged 3-5 years, in this study the processing and analysis of test results, obtained by the examinations, refer to both years together.

Table 1: Distribution of covered patients by age and sex, shows the distribution of respondents who have been diagnosed with changes and who have not been diagnosed with any changes, by age and sex. It is evident from the numbers in the table that in the research, 50% of the respondents were diagnosed with some changes in the voice, while the other 50% of the respondents were not diagnosed with any deviations.

From the 26 diagnosed respondents who have a certain type of voice change, which are divided into 4 groups, listed above in the text below the table, the conclusion is that children aged 3-5 years have a 2.25 times higher incidence of voice changes related to children aged 6-8 years, of which girls in this group aged 3-5 years have 2 times higher incidence of voice changes than boys (ratio 6: 3 girls to boys).

In the group of participantsaged 6-8 years a decrease in diagnosed patients is evident, by 67% in boys aged 6-8 years compared to those aged 3-5 years, and by 50% in girls aged 6-8 years. 8 years compared to those of 3-5 years and, by 50% in girls.

The other participantsfrom the control group are of the same age and sex but certainly without cleft palate as an anomaly and without voice disorders.

Table 1. Distribution of covered patients by age and sex

Age	First group (n=26)		Control group (n=26)	
years	boys	girls	girls	girls
3-5	6	12	6	12
6-8	2	6	2	6
total	8	18	8	18
	26		26	

The following, Table 2, shows the distribution of diagnosed patients with voice changes, who are disaggregated into 4 groups, as follows: 0- There is no change in voice quality; 1- Slight changes in voice quality; 2- Moderately altered voice and 3- Pronounced voice changes.

Table 1. Distribution of diagnosed patients by categories of voice changes

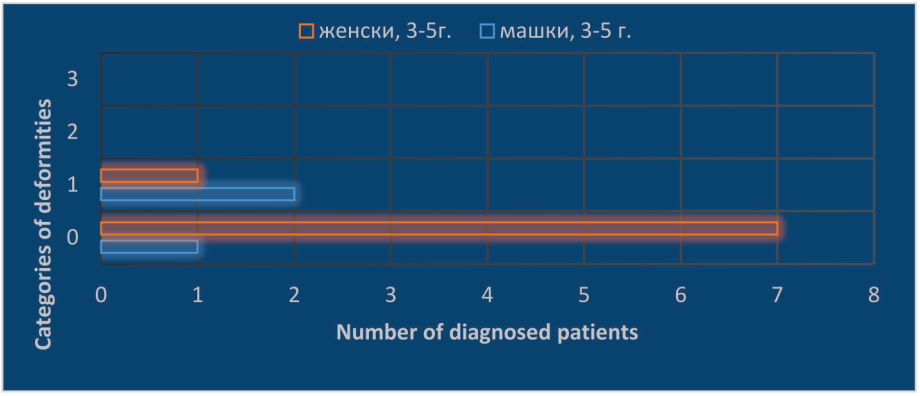
	First group				Control group			
	boys		girls		boys		girls	
	3-5 years	6-8 years	3-5 years	6-8 years	3-5 years	6-8 years	3-5 years	6-8 years
There is no change in voice quality 0	2		14		6	2	12	6
Slight changes in voice quality 1	4		2					
Moderately altered voice 2		2		2				
Pronounced voice changes 3								

The structural analysis of the obtained results in the first group - 26 patients with cleft palate / lip, shows that, analysed by categories of deformities, in the respondents aged 3-5 years, in the category "O - There is no change in voice quality" , 87.5% are girls, while only 12.5% are boys, out of the total of 16 diagnosed patients (participants). What is significant about this group is that it includes the largest number of respondents with diagnosed deviations in general, which represents as much as 54% of the total number of patients in this First group. These 54% are located in girls aged 3-5 years.

In the category "1 - Slight changes in voice quality", out of a total of 6 patients diagnosed aged 3-5 years, the ratio between boys and girls is

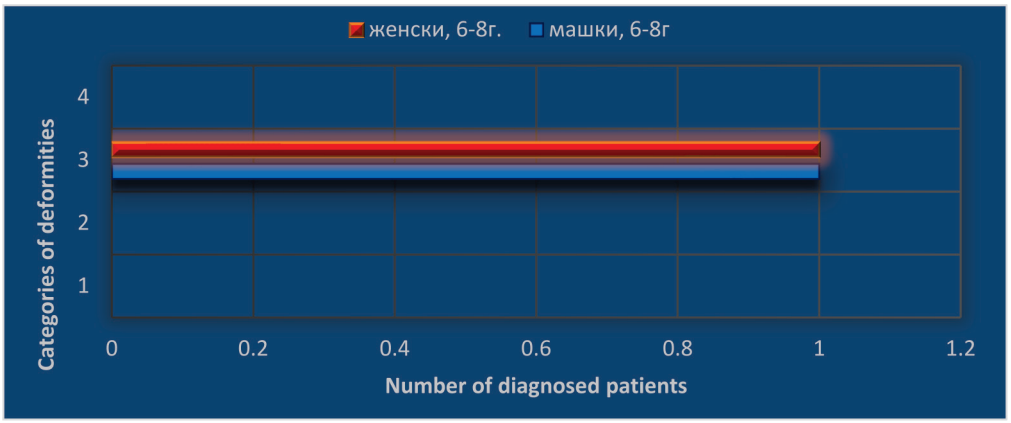
inversely proportional to the previous diagnosis (category), i.e., 1/3 are girls, while as much as 2/3 are boys, out of the total of 6 diagnosed patients (participants). In the categories "2 - Moderately altered voice" and "3 - Pronounced changes in the voice", during the study, no deviations were identified in the participants aged 3-5 years.

Picture 1. Changes in cleft palate patients voice according to categories of deformities 2019-2020



Among the participants aged 6-8 years, the analysis of the data shows that in children with cleft palate in the category "2 - Moderately altered voice", there is an even distribution in boys and girls (50-50%), out of a total of 2 diagnosed children aged 6-8 years in this category.

Picture 2. Changes in cleft palate patients voice according to categories of deformities 2019-2020



Discussion

The results obtained in this research show that 50% of the respondents were diagnosed with some changes in the voice, while the other 50% of the respondents were not diagnosed with any deviations. In the category "O - There is no change in voice quality", 87.5% are girls, while only 12.5% are boys, out of a total of 16 respondents. What is significant in this group is that even in 50% of the total number of respondents there is no change in voice quality and these are located in girls aged 3-5 years.

According to the results of the research, it can be seen that there are no registered respondents with cleft palate / lip in whom there are pronounced changes in the voice in relation to the control group.

From the 26 participants (patients) diagnosed with some kind of devia-

tion in voice change, which are divided into 4 groups, listed above in the text below the table, the conclusion is that children aged 3-5 years have a 2.25 times higher incidence of voice changes in relation to children aged 6-8 years, of which girls in this group aged 3-5 years have 2 times higher incidence of voice changes than boys (ratio 6: 3 girls to boys). Out of a total of 2 diagnosed children aged 6-8 years in this category are with equal distribution among boys and girls (50-50%), in the category "2 - Moderately altered voice".

The values of the parameters obtained from this research show that even in 50% of the total number of respondents there is no change in voice quality as shown by the research of other authors regarding voice disorders in children with cleft palate that may occur due to compensatory mechanisms of speech.[17]

The results showed that there is a change in the voice of children with cleft palate / lips who have ORL infections often resulting in occasional hearing loss recorded by audiometry.

Studies show that in addition to the characteristics of speech disorders in the form of articulatory features and increased nasal air emissions, voice disorders may occur in these patients, especially in boys.[13]

Although future research on voice disorders in patients with cleft lip / palateis needed, now it seems reasonable to include voice therapy strategies in the speech and language intervention plan for these patients with this type of anomaly.[19]

Subjective assessment of the voice quality is the analysis of the voice with its own hearing. Despite modern technology, the trained human ear is indispensable in the assessment of the voice and the most important "apparatus" in examining the subjective or psychoacoustic characteristics of the voice. Considering that subjective impression is crucial in communication, the role of perceptual voice assessment in clinical terms is extremely important. This instrument is characterized by simplicity in application and economy. Perceptual assessment is inevitable in the domain of interpretation of the objective ways of evaluating the quality of the voice, it is a mean of diagnosis and assessment of the success of the therapy of voice disorder. Perceptual assessment results may be influenced by different criteria of the examiner as well as differences in the experiences of the examiners.[20,21]

The reliability of the Grbas scale has been examined by De Bodt et al. (De Bodt, Wuyts, Van de Heyning, &Croux, 1997). The results of the research of these researchers show that the reliability of this scale is satisfactory. Aguero et al. (Agüero, Tulli, Moscardi, Gonzalez, &Uriz, 2011) examined the reliability of the Grbas scale and compared the results obtained with other methods of subjective assessment of voice quality. (The Consensus Auditory-Perceptual Evaluation of Voice - CAPE-V; Buffalo Voice Profile - BVP; Vocal Profile Analysis Scheme - VPA). The results of these studies show that the Grbas scale is the most reliable compared to other methods with good reliability for all parameters of the voice, except for the scale S whose certainty was moderate. When compared to GRBAS, CAPE-V showed slightly better assessor reliability. [22,.23]

The shortcomings of subjective voice assessment can be overcome by researching the correlation between acoustic and perceptual parameters of voice quality as well as reaching an agreement on terminology and using numerical rating scales. [24]

Ongoing work on more effective assessment focuses on a physio psychoacoustic model linking voice production, acoustics, and perception.[20,25]

Conclusion

In addition to the proper stimulation of speech and language development in children, it is extremely important to monitor possible changes in the state of the voice in childhood, especially in children with certain congenital anomalies.

When it comes to perceptual assessment of voice, previous research indicates some shortcomings, such as the competence and experience of examiners, as well as mutual disagreement in assessing the degree of expression of certain characteristics of voice quality.

The main disadvantage of perceptual voice evaluation is the problem of description. There are no reliable verbal terms describing vocal features, although there is a constant need to define descriptive terminology.

To get more accurate results, more research on voice changes in children with palate cleft palate and velopharyngeal dysfunctionshould be done in future, and also, at the same time, checking the state of hearing.

The obtained more accurate results give an accurate insight into the condition of the voice in children with this type of congenital anomaly. If there are changes in the voice, then the condition is more complex and accordingly the speech therapy will be the same. This is very important for its proper course and success.

References

1. Petrović-Lazić, M., Kosanović, R. Vokalna rehabilitacija glasa. Beograd: Nova naučna 2008
2. Bolfan Stošić, N, Zorić, A. Higiena dečjeg glasa – Uputstva za rad kod kuće i razvijanje higijene dečjeg glasa. Zagreb: Hrvatsko logopedsko društvo 1997.
3. Boone DR, McFarlane SC. The voice and voice therapy, 6th edition. Needham Heights: Pearson Education Co. 2000.
4. Dembitz A. Glasinje gove promjene. Znanstveno-stručni simpozij – Logopedija: jučer, danas,sutra. Zagreb: Hrvatsko logopedsko društvo 2012.
5. Parčina, M. Poremećaji glasa predškolskog uzrasta – dječja promuklost, U: Hrvatsko logopedsko društvo – Podružnica Splitsko-dalmatinske županije, Logopedski vodič Splitsko- dalmatinske županije (11-13). Split: HLD 2019
6. Peterson-Falzone S, Hardin-Jones M, Karnell M.. Cleft Palate Speech (3rd ed.) St. Louis (M): Mosby, Inc. 2001.
7. Brøndsted K, Liisberg WB, Orsted A, Prytz S, Fogh-Andersen P. Surgical and speech results following palatopharyngoplasty operations in Denmark 1959-1977. Cleft Palate J 1984;21(3):170-179.
8. Timmons MJ, Wyatt RA, Murphy T. Speech after repair of isolated cleft palate and cleft lip and palate. Br J Plast Surg 2011;54(5):377-384.-
9. Hocevar-Boltezar I, Jarc A, Kozelj V. Ear, nose and voice problems in children with orofacial clefts. J Laryngol Otol 2006;120(4):276-281.
10. McWilliams BJ, Lavorato AS,

Bluestone CD. Vocal cord abnormalities in children with velopharyngeal valving problems. Laryngoscope 1973;83(11):1745- 1753.

11. Seyfer AE, Simon CD. Long-term results following the repair of palatal clefts: a comparison of three different techniques. Plast Reconstr Surg 1989;83(5): 785-792.
12. Van Lierde KM, Claeys S, De Bodt M, Van Cauwenberge P. Vocal quality characteristics in children with cleft palate: a multiparameter approach. J Voice 2004; 18(3):354-362.
13. Lewis JR, Andreassen ML, Leeper HA, Macrae DL, Thomas J. Vocal characteristics of children with cleft lip/palate and associated velopharyngeal incompetence. J Otolaryngol 1993;22(2):113-117.
14. Grunwell P, Brondsted K, Henningsson G, et al. A six-centre international study of the outcome of treatment in patients with clefts of the lip and palate: the results of a cross-linguistic investigation of cleft palate speech. Scand J Plast Reconstr Surg Hand Surg 2000; 34(3):219-229.
15. D'Antonio LL, Muntz HR, Province MA, Marsh JL. Laryngeal/ voice findings in patients with velopharyngeal dysfunction. Laryngoscope 1988; 98(4):432- 438.
16. Leder SB, Lerman JW. Some acoustic evidence for vocal abuse in adult speakers with repaired cleft palate. Laryngoscope 1985; 95(7, pt 1):837-840.
17. Hamming KK, Finkelstein M, Sidman JD. Hoarseness in children with cleft palate. Otolaryngol Head Neck Surg 2009;140(6):902-906.
18. Webb AL, Carding PN, Deary IJ, MacKenzie K. The reliability of three perceptual evaluation scales for dysphonia. European Archives of Otorhinolaryngology 2004; 261:429-434
19. Villafuerte-Gonzalez R, Valadez-Jimenez VM, Hernandez-Lopez X, Ysunza PA. Acoustic analysis of voice in children with cleft palate and velopharyngeal insufficiency. Int J Pediatr Otorhinolaryngol 2015; 79(7):1073-6.
20. Bonetti A. Perceptivna procjena glasa. Hrvatska revija za rehabilitacijska istraživanja 2011; 47(1), 64-71
21. Chan K. & Yiu E M L. A comparison of two perceptual voice evaluation training programs for naive listeners. Journal of Voice 2006; 20(2): 229–241.
22. De Bodt MS, Wuyts FL, Van de Heyning PH, Croux C. Test-retest study of the GRBAS scale: influence of experience and professional background on perceptual rating of voice quality. J Voice 1997;11(1):74-80.
23. Zraick RI, Kempster GB, Connor NP, et al. Establishing validity of the Consensus Auditory-Perceptual Evaluation of Voice (CAPE-V). Am J Speech Lang Pathol 2011;20(1):14–22.
24. Baylor C R, Yorkston K M, Eadie T L, Strand E A, Duffy J. A systematic review of outcome measurement in unilateral vocal fold paralysis. Journal of Medical Speech-Language Pathology 2006; 14(1), 1–33.
25. Kreiman J, Gerratt B. Reconsidering the Nature of Voice. In: Frühholz S, Belin P, editors. The Oxford Handbook of Voice Perception. Oxford University Press; 2019.