JURNAL PLASTIK REKONSTRUKSI

(Jurnal Plastik Rekonstruksi 2018; 1: 160:170)

CRANIOFACIAL

SPEECH OUTCOME EVALUATION OF CLEFT PALATE PATIENTS UNDERWENT PALATOPLASTY IN PLASTIC SURGERY DIVISION CIPTO MANGUNKUSUMO HOSPITAL INDONESIA

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ABSTRACT

Background : Speech is the primary goal of palatoplasty, however, there is no current data available about the long term speech evaluation after palatoplasty in our hospital which is the national referral hospital that has the only cleft craniofacial center in Indonesia. The initial data of speech outcome is required for further research which assessment should be standardized and applicable to Indonesian children that mostly speak bahasa. This study aims to get initial data by evaluating speech outcome of patients that underwent palatoplasty with adapted perceptual assessment words in Indonesian language, and describe factors influencing speech.

Method : This research is a cross-sectional study to evaluate speech outcome of patients underwent palatoplasty in Cipto Mangunkusumo Hospital from October 2010–December 2012 conducted from December 2017 – July 2018.

Result : Total 23 samples were measured for articulation rating where 17 (74%) patients had normal production of majority of phonemes, while there were 6 (26%) patients had predominantly distortion of phonemes. The hypernasality rating were normal in 12 (52%) patients, mild in 5 (22%) patients and moderate in 6 (26%) patients. The speech intelligibility rating were dominantly normal which all speech is understood in 17 (74%) patients and the rest of 6 (26%) patients were listeners attention needed. The velopharyngeal competence were good in 16 (70%) patients, fair in 1 (4%) patients and poor in 6 (26%) patients.

Conclusion: Management of cleft palate patients will be achieved by well integrated services including speech pathologist and orthodontist. By giving the long term follow up to the patients, the optimal outcomes will be achieved. This research can be used as a reference for speech outcome evaluation in cleft palate patients in Indonesia.

Keywords: *speech outcome, speech assessment, speech evaluation, Indonesian language*

Latar Belakang: Fungsi bicara adalah tujuan utama palatoplasty, namun tidak terdapat evaluasi jangka panjang untuk fungsi bicara pada rumah sakit kami yang merupakan rumah sakit rujukan nasional yang memiliki pusat sumbing dan kraniofasial satu-satunya di Indonesia. Studi ini bertujuan untuk mendapatkan data awal dengan evaluasi fungsi bicara pasien pasca palatoplasty dengan kata asesmen persepsual teradaptasi berbahasa indonesia, dan menjabarkan faktor yang mempengaruhi fungsi bicara.

Metodologi: Penelitian ini merupakan penelitian *cross-sectional* untuk mengevaluasi hasil bicara pasien yang menjalani palatoplasti di Rumah Sakit Cipto Mangunkusumo dari Oktober 2010-Desember 2012 yang dilakukan pada Desember 2017 - Juli 2018.

Hasil: Dua puluh tiga total sampel diukur untuk tingkat artikulasi di mana 17 (74%) pasien memiliki produksi normal dari mayoritas fonem, dan 6 (26%) pasien memilki distorsi predominan dari fonem. Penilaian hipernasalitas normal pada 12 (52%) pasien, ringan pada 5 (22%) pasien dan sedang pada 6 (26%) pasien. Penilaian inteligibilitas suara secara dominan normal di mana semua kata dapat dimengerti pada 17 (74%) patients dan sisanya yaitu 6 (26%) pasien membutuhkan perhatian pendengar. Kemampuan velofaringeal baik pada 16 (70%) pasien, sedang pada 1 (4%) pasien dan buruk pada 6 (26%) pasien.

Kesimpulan: Manajemen pasien sumbing langit-langit akan dicapai dengan layanan yang terintegrasi dengan baik termasuk ahli patologi wicara dan ortodontis. Dengan memberikan tindak lanjut jangka panjang kepada pasien, hasil optimal akan tercapai. Penelitian ini dapat digunakan sebagai referensi untuk evaluasi hasil bicara pada pasien sumbing langit-langit dan mulut di Indonesia.

***Keywords**: speech outcome, speech assessment, speech evaluation, Indonesian language*

Received: 15 November 2018, Revised: 17 Desember 2018, Accepted: 21 Desember 2018

ISSN 2089-6492 ; E-ISSN 2089-9734
This Article can be viewed at www.jprjournal.com

INTRODUCTION

Cleft lip and/or palate are the most common congenital craniofacial anomalies faced by plastic surgeon. Among the cleft lip and palate population, the most common diagnosis is cleft lip and palate at 46%, followed by isolated cleft palate at 33%, then isolated cleft lip at 21%. The majority of bilateral cleft lip (86%) and unilateral cleft lip (68%) are associated with cleft palate. In our center, the most common type of cleft is unilateral complete cleft lip and palate which we know as veau 3 classification of cleft palate.[1]

The treatment objective in palatoplasty is not only simple closure of the soft and hard palate but also create an adequate function of velopharyngeal mechanism for normal speech production. Speech quality remains the most important standard to assess clinical outcomes and success of surgical procedures. That is why the primary goal of palatoplasty is normal speech. [1,2] The cleft palate patients have speech problem due to velopharyngeal insufficiency, hypernasal speech, and hoarse quality. This speech problem occurred because the airflow is difficult to be directed through the mouth. The cleft repair must be performed to obtain both anatomic and functional outcome to avoid compensatory mechanism for sound to be learned and resulting abnormal sound. [1]

There are many factors that influence speech outcome for the children with cleft palate, for example the timing of primary palatal surgery and also the palatoplasty technique. Speech itself is also affected by Intelligence Quotient (IQ), hearing impairment, environment and nutritional status. However, there is no current data available about the long term speech evaluation after cleft palate repair in our center. The initial data of speech outcome is required for further research which assessment should be standardized and applicable to Indonesian children that mostly speak Indonesian language.

Treatment of Cleft Palate

The main goal of palate repair is to achieve a normal speech. The two most important aspects to achieve the goals are: (1) surgical technique and (2) timing of palate repair. The first observation of a correlation between age at repair and speech outcome was made 1931. The children who had undergone repair at about 12 months of age were much more likely to have normal speech than those with repair between two and four years of age. Children who underwent repair after nine years of age had the worst speech outcome. Holland et al in 2007 proved that delayed closure of the hard palate leads to speech problems, due to palatal scarring. The optimal time of cleft repair still remains scientifically unproven. Confounding variables of technique, surgeon's skill also play roles.[1,5]

Complication Cleft Palate Repair

Fistula formation is one of the complication of cleft palate repair. It may be a source of persistent air nasal loss even in the face of functioning soft palate, they are also a source of nasal regurgitation of fluids. The fistula with diameter more than 5 mm can affect speech outcome do to air escape causing hypernasality.[1,7]

Speech Therapy

The main goal of speech therapy in cleft palate patients is to let the patients produce optimal speech based on their structural anatomy by stimulating speech production throughh vocal play activity in varied sounds, sound imitating training, and stimulating oral stop consonants production. Oromotor therapy such as blowing, sucking, whistling, horn therapy, palatal massage and electrical stimulation do not give benefit to facilitate the speech accuracy due to significant difference of velpharyngeal closure between speech activity and non-speech activity. The orormotor therapy should be avoided in cleft palate patients, because the patients have structural deformity despite of muscle weakness.[7]

Speech Production

Speech is produced through three mechanism: 1) vibration mechanism from vocal cord; 2) stimuli mechanism from breathing pressure; and 3) vocal tract as resonance mechanism to produce voice energy. In order to produce speech, good supporting structure and function is required, involving upper and lower respiratory tract, and orofacial organ, in this term normal palate plays important role.[8] The function of muscle palate is to

Disclosure: *The authors have no financial interest to disclose.*

control the air passage between oral and pharyngeal cavity, while the contribution of palatal vault to the resonance of the oral cavity is varied due to many variation of hard palate curvature.[8]

Speech is a coordination of physiological subsystems, which are respiration, phonation, resonance and articulation, It is also including other components such as intonation, precision and fluently of speech. Speech production is a complex interaction between respiratory and masticatory structure involving nasal, labial, maxilla, mandibular, glottis, abdomen, diaphragm, thoracic wall, laryngeal, oropharyngeal, and both hard and soft palate. (Figure 1), while di air passage control is occurred at five regions that are laryngeal, velopharyngeal valve, nasal valve, between glottis and both hard and soft palate, glottis and dental, and also lip and dental. (Figure 2)

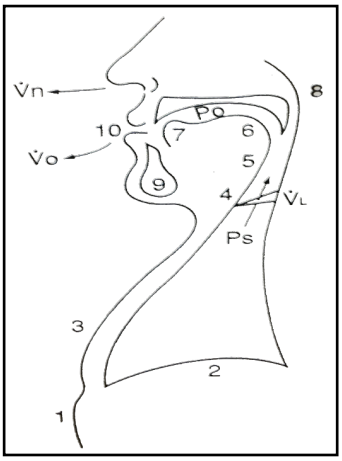

Figure 1. Vocal tract – Ten functional components that produce air valve during speech. 1-abdominal muscle; 2-diaphragma; 3-costae; 4-laryngeal; 5-pharyngeal; 6-posterior glottis; 7-tip uf glottis; 8-velopharyngeal; 9-mandible; 10-labial; Ps-subglottal air pressure; Po-intraoral air pressure; Vn-nasal air passage; Vo-oral air passage; Vl-

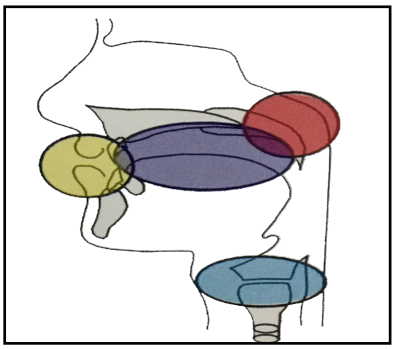

Figure 2. Vocal tract and location of air passage

Velopharyngeal Dysfunction

Normal velopharyngeal function depends on 3 basic components: normal structure (anatomy), normal movement (neurophysiology), and normal articulation learning. When the velopharyngeal valve does not close consistently or completely during the production of oral sounds, this is often called velopharyngeal dysfunction (VPD). VPD is used as a general term that encompasses disorders of any of the 3 basic components of velopharyngeal function (structure, function, and learning). Other terms are used for more specificity as to the type and causation of VPD. For example, velopharyngeal insufficiency (VPI) is most often used to describe a structural defect that prevents complete velopharyngeal closure (Fig. 3). Velopharyngeal insufficiency is the most common type of VPD because it can be caused by a history of cleft palate or submucous cleft. In contrast, velopharyngeal incompetence (also abbreviated as VPI) is used to refer to a neurophysiologic disorder in which poor movement of the velopharyngeal structures results in incomplete velopharyngeal closure. (Fig. 4) Finally, velopharyngeal mislearning refers to an articulation disorder in which speech sounds are inappropriately produced in the pharynx. As a result of this placement, the velopharyngeal valve is open, thus mimicking VPI during attempted production of certain speech. [10.12,13]

Hypernasality implies that too much sound energy emerges through the nose. Thus, there is an oral/nasal resonance imbalance. This lingering speech disorder typically is attributed to VPI. With VPI, incomplete velopharyngeal closure may result in hypernasality during vowel segments and nasal emission of air during non-nasal consonant segments. For example, in the word *baby*, there may be abnormal nasal emission of air during production of the non-nasal /b/ consonant segments and hypernasality during production of the /i/ vowel segments.[14]

In contrast to hypernasality, if there is blockage of the velopharyngeal port or the nasal passages, this may result in hyponasality (also called denasality) during production of nasal speech sounds. Hyponasality implies that there is too little sound energy emerging through the nose. For example, in the word *mama*, there would be too little sound energy emerging through the nose during the production of the nasal

consonants /m/. Thus, hyponasality is also an oral/nasal resonance imbalance, but in the opposite direction from that of hypernasality.[7]

Speech development

Infants and young children with cleft palate should be given normal language stimulation. Parents should talk to their children frequently and listen to them. Parents should avoid using nonsense words and should speak clearly, using correctly formed words and short phrases. Normally, speech articulation to produce vocal and consonants starts at one to three years old, and majority of seven year-old children are using most of consonants..[15] Infants with cleft palate should be allowed to babble freely and naturally. Children should be encouraged to communicate using speech. Speech development itself is affected by Intelligence Quotient, hearing impairment, environmental

Speech Evaluation in Cleft Palate

Perceptual speech assessment is considered the gold standard in the diagnosis of speech disorders of persons with cleft palate and VPD. Additional instrumental assessment and imaging are considered adjunct to the perceptual speech findings, which are the ultimate arbiter of a patient's need for treatment.[7]

Perceptual assessment is evaluated by Medical Rehabilitation Doctor. The patients' should speak consonants words which are familiar or using their own language. In Indonesia, where Indonesian language is the most common language used, speech assessment tool should be adjusted with Indonesian language including the consonant words that isi commonly used in Indonesian languange. (Table 1)[8] Various speech parameters in cleft palate patients is shown in Table 2.[17]

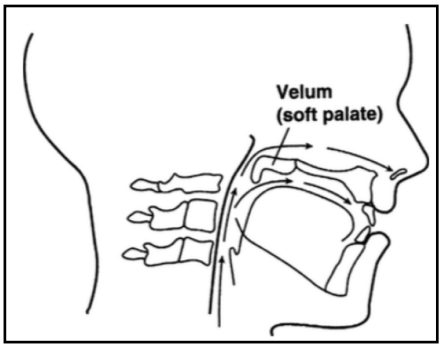

Figure 3. Velopharyngeal insufficiency

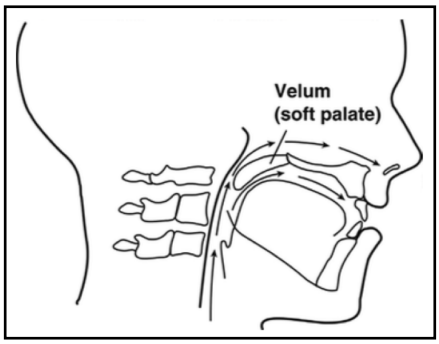

Figure 4. Velopharyngeal incompetence

T**able 1.** Perceptual Assessment Words in Indonesian Languange

	A	B	C
B	balon	bibir	buku
C	cacing	cicak	cuci
D	daun	mandi	duduk
G	gajah	gigi	dagu
H	paha	hijau	hujan
J	jambu	jinjit	keju
K	kaca	kaki	kuda
L	lalat	tali	palu
M	mandi	minum	mulut
N	nanas	anisa	banu
R	kerang	lari	rumah
S	sapi	dari	susu
P	papa	api	sapu

Table 2. Evaluation Criteria for Various Speech Parameters Scoring

Characteristic	
Articulation rating	1-Normal production of majority of phenomes
	2-Predominantly distortion of phenomes
	3-Distortion and substitution of phenomes
	4-Phenomes are substituted and ommited
Hypernasality rating	1-Normal
	2-Mild hypernasality
	3-Moderate hypernasality
	4-Severe hypernasality
Speech intelligibility rating	1-Normal
	2-Listeners attention needed
	3-Occassional repetition of words required
	4-Repetition and rephrasing necessary
	5-Isolated words understood
	6-Ocassionally understood by others
	7-Unintelligible speech
Velopharyngeal competence	1-Good
	2-Fair
	3-Poor

METHOD

This research is a cross-sectional study to evaluate speech outcome of patients underwent palatoplasty in Cipto Mangunkusumo Hospital from October 2010 – December 2012. Conducted from December 2017 – July 2018. The exclution criteria are patient under seven years old, non contactable patients, and patient's parent refuse to participate.

This research will be submitted to the Ethical Committee of the Faculty of Medicine Universitas Indonesia for ethical approval and informed consent will be obtained from the parents.

RESULT

We had total of 23 samples of patients to be evaluated. Most of the samples were 17 males where the rest were females. The mean of the age was 8.3 (7.3-10.1) years old. (Table 3).

Table 3. Patients' Demographic .

No.	Variable	
1.	Age (years)	
	Mean	8,3
	Range	7.3 - 10.1
2.	Sex	
	Male	17
	Female	6

Patients' Clinical Characteristics

According to the data, the most cleft palate type were veau 3 classification which was 19 patients (82%), while the others were 2 patients (9%) each for veau 2 and veau 4 classification. None of the patient had veau 1 cleft palate classification. Almost all the palatoplasty technique performed was two-flap palatoplasty which was done in 21 (91%) patients and the rest 2 (9%) others underwent VY pushback palatoplasty..

The timing of surgery was done before the patient's age before two years old in 17 (74%) patients, and after or at two years old in 6 (26%) patients. None of the patients underwent speech therapy after palatoplasty procedure. There were 2 (91%) presented with fistulae more than five mm, while the rest 21 9 (%) of the patients were not. (Table 4).

T**able 4.** Patients' Clinical Characteristics

No.	Variable	n	Percentage
1.	Cleft type before palatoplasty		
	Veau 1	0	0
	Veau 2	2	9
	Veau 3	19	82
	Veau 4	2	9
2.	Technique performed		
	Furlow Palatoplasty	0	0
	VY Pushback Palatoplasty	2	9
	Von Langenback Palatoplasty	0	0
	Two-flap Palatoplasty	21	91
3.	Patients' age when palatoplasty performed		
	< 2 years old	17	74
	>/= 2 years old	6	26
4.	History of Speech therapy		
	Yes	0	0
	No	23	100
5.	Presentation of Fistulae more than 5 mm		
	Yes	2	9
	No	21	91

The outcomes were measured in four parameters by the speech pathologist. For articulation rating parameter, there were 17 (74%) patients had normal production of majority of phonemes, while there were 6 (26%) patients had predominantly distortion of phonemes. There were no patients with distortion an substitution of phonemes and whose phonemes are substituted and ommited. The hypernasality rating were normal in 12 (52%) patients, mild in 5 (22%) patients and moderate in 6 (26%) patients, while there no patients with severe hypernasality The speech intelligibility rating were dominantly normal which all speech is understood in 17 (74%) patients and the rest of 6 (26%) patients were listeners attention needed. There were no patients were measured to other speech intelligibility rating. The velopharyngeal competence were good in 16 (70%) patients, fair in 1 (4%) patients and poor in 6 (26%) patients. (Table 5)

Table 5. Speech Outcomes of Cleft Palate Patients Underwent Palatoplasty

No.	Variable	n	Percentage (%)
1.	Articulation Rating		
	1-Normal production of majority of phonemes	17	74
	2-Predominantly distortion of phonemes	6	26
	3-Distortion and substitution of phonemes	0	0
	4-Phonomes are substituted and ommited	0	0
2.	Hypernasality Rating		
	1-Normal	12	52
	2-Mild hypernasality	5	22
	3-Moderate hypernasality	6	26
	4-Severe hypernasality	0	0
3.	Speech Intelligibility Rating		
	1-Normal, all speech is understood	17	74
	2-Listeners attention needed	6	26
	3-Occasional repetition of words required	0	0
	4-Repetition and rephrasing necessary	0	0
	5-Isolated words understood	0	0
	6-Ocassionally understood by others	0	0
	7-Unintelligible speech	0	0
4.	Velopharyngeal Competence		
	1-Good	16	70
	2-Fair	1	4
	3-Poor	6	26

Other Factors Affecting Speech Outcome

From 23 patients, there were 3 (13%) patients whose IQ scores were under 100, while there were 2 (9%) patients had IQ more than or equal to 100. The IQ of 18 (79%) patients were not evaluated due to several reasons.

All of the 23 (100%) patients had normal hearing function and had playmates at home for environmental role parameter. Only 1 (4%) patient categorized as underweigt while the other 22 (96%) were in normal weight category. (Table 6).

Table 6. Other Factors Affecting Speech Outcome

No.	Variable	n	Percentage
1.	IQ Score		
	<100	3	13
	>/=100	2	9
	N/A	18	79
2.	Hearing Function		
	Normal	23	100
	Impaired	0	0
3.	Environmental Role		
	Have playmate	23	100
	Have no playmate	0	0
4.	Nutritional Status (Weight/Height)		
	Underweight	1	4
	Normal weight	22	96
	Over weight	0	0

DISCUSSION

There are established data about speech outcome in cleft palate palate patients, but there are no data available in Indonesia specifically in our hospital, where as Cipto Mangunkusumo hospital is the national referreal hospital that has the only cleft craniofacial center in Indonesia. We had 23 samples of patients underwent palatoplasty whose age above seven years old, since children normally speaks all consonants at the seventh year of age.[8] Cleft Craniofacial Center Ciptomangunkusumo Hospital was founded in 2012, the patients data before that time were not well filed, but in this study, the data was taken from October 2010 in consideration of the data availability from the previous studies conducted in our hospital.[4,22] In 2012 Cleft Craniofacial Center Cipto Mangunkusumo hospital has conducted 68 cleft palate surgery with complete data filing, but the contactable patients whose age were 7 years old by June 2018 were only 12 patients, while the rest of 11 patients were collected from contactable patients in the previous studies. Male samples are more than female, it is consistent to the previous literature study that stated the incedence of cleft were predominantly in male.[2]

The patients clinical characteristic obtained the type of cleft where unilateral cases showed much more than unilateral which is accordant to the theory that unilateral cleft was nine times higher than bilateral.[2] The technique performed were actually depend on the type of cleft palate.[1] The most common technique was two-flap palaoplasty. However this technique will result in lateral palatal defect. Some of the plastic surgeon in our

center will leave the periosteum at the lateral side by beveling the incision which is usually known as non-denuded two-flap palatoplasty. From the previous conducted study, this technique was proven to fasten the epithelization rate of the lateral defects. Faster epithelization is expected to decrease wound contraction thus reducing scar formation, and in the long run will affect the speech outcome.[1,2,3,4] This field of study can be conducted in the future to compare the speech outcome in standard two-flap palatoplasty technique with non-denuded two-flap palatoplasty.

Published studies has stated that palatoplasty on patients below two years old will improve speech outcome compared to patients above two years old,[1,5] but in our hospital there were still 6 patients who underwent palatoplasty above two years old. This may be happened due to lack of education of the patients where they could not gather much information about cleft. It gives lesson to the institution to socialize more about cleft in order to achieve better outcomes. None of the patients in this study received speech therapy regardless the indication. By establishing cleft center, this problem could be diminished since the management of patients are integrated. There are two patients presented with fistulae. Fistulae rate post cleft palate repair was the indicator in our center, but since 2014 there were no more fistula recorded in our center.

Since perceptual assessment is the gold standard for speech evaluating,[9] we need to have the standardized words in Indonesian language which applicable for speech perceptual assesment. According the the articulation manner with their responsible consonants, the author formulating specific words in Indonesian language.[7] This words can be used for further speech assesment in Indonesia to produce many further researches. Previous study conducted in India in 2010 showed the articulation rating mean was 1.63 (+/-0.42), while the hypernasality rate 1.96 (SD 0.48). For the intelligibility rating it was 2.42 (0.76).[17] Although the intelligibility rating in our hospital were no more than 2.0, the data were not comparable due to lack of sample size. The patients with poor velopharyngeal competence had further endoscopy examination, and one of them received repalatoplasty using furlow palatoplasty technique. Seven patients needs alveolar bone grafts and were refered to orthodontist in our center for further treatment, where two of them also received lip scar revision.

Only five patients had IQ assessment, this is due to lack of facility, since some of the samples were taken by visiting the small village, where the patients live. There are no significant hearing impairment detected by the examiner, concluded by all the samples were cooperatively following the words spoken by the examiner. Although some of the patients have no sibling, they mostly have playmates that live near to their house. One of the underweight patient was found at the patient whose family was in low economic status.

CONCLUSION

In summary, This study provided initial data that can be used for further research. The good data filing is very important for the sake of education and patients management. By giving the long term follow up to the patients, the optimal outcomes will be achieved. Management of cleft palate patients will be achieved by well integrated services including speech pathologist and orthodontist. This research can be used as reference for speech outcome evaluation in cleft palate patients in Indonesia.

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