

RESEARCH ARTICLE

Speech outcome evaluation in post-cleft palate closure patients with two flaps pushback technique

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ABSTRACT

Speech quality is an essential output in assessing the success of a palatoplasty. The goal of a palatoplasty is not merely to create a simple anatomical closure of the palate, but also to create an adequate velopharyngeal mechanism for a normal speech outcome and to prevent abnormal maxillofacial development after surgery. The aim of this study is to find out the difference in speech outcome between post-cleft palate closure patients and patients without cleft palate. An analytical retrospective study was conducted on 22 children (n = 22) with complete unilateral cleft palate, who had been treated using two flap push back technic of palatoplasty during 2014-2017 by purposive sampling method, and 22 children without cleft palate as the control group. The evaluation of speech outcome was done using an assessment of perception by doing a speech pathologist and instrumental examination by taking a lateral cephalometry radiograph. The perception was assessed by the articulation pattern, hypernasality, and speech intelligibility. The lateral cephalometry radiograph was taken at /i/ phonation to measure the distance velum to the posterior pharynx wall. Data were analyzed using Mann Whitney test. The velopharyngeal competence in post-palatoplasty group consisted of 22.8% adequate result, 0.1% marginal result, and 68.1% inadequate result. Meanwhile, in the control group, there were 72.7% adequate and 27.3% inadequate competence. According to the result of the statistical test, this study concluded that there was a significant difference in speech outcome based on articulation pattern, hypernasality, speech intelligibility, and velopharyngeal distance between post-cleft palate closure patients and patients without cleft palate ($p < 0.05$). Majority of patients after cleft palate closure with two flaps pushback technique had inadequate velopharyngeal competence with moderate-severe hypernasality, severe nasal emission, abnormal speech intelligibility, and velopharyngeal distance ≥ 5.0 mm, whereas the majority of control group had an adequate velopharyngeal competence.

Keywords: cleft palate; cleft palate closure; palatoplasty; speech outcome; two flap push back

INTRODUCTION

Cleft lip and palate are commonly found congenital disorders. Individuals with cleft lip and palate are commonly showing several problems, such as swallowing disorders, nutrition problems, delays of speech development and/or abnormal resonance, dentofacial and orthodontic disorders, hearing disorders, and probably psychosocial problems.¹ Several factors were reported to influence the speech outcome post palatoplasty treatment, such as the type of cleft palate, time of surgery, surgeon's skills, speech therapy, and surgery techniques.² The goal of a palatoplasty is not merely to create a simple anatomical closure of the palate, but also to create an adequate velopharyngeal mechanism for a normal speech outcome and to prevent

abnormal maxillofacial development after surgery. One of the most commonly used techniques of palatoplasty is two flap push back, because the technique could extend the soft palate so that the soft palate is expected to reach the posterior pharynx wall while the patient is speaking. In that case, a competent velopharyngeal closure could be achieved.

A retrospective study conducted by Kenneth, et al. who reviewed the two flap palatoplasty technique during a period of 20 years concludes that the two flap palatoplasty techniques were reliable in achieving the surgery outcome and were able to provide a very good result in speech outcome.³ Alla, et al. mentioned that two flap palatoplasty technique gave a significant

development in patient's speech outcome, and lowered the incidence of oronasal fistule.⁴ However, this result contradicted the research result conducted by Y. Dong, et al. on 88 patients with incomplete cleft palate, which revealed that palatoplasty using Furlow technique provided a better velopharyngeal closure result than palatoplasty using two-flap technique.⁵

Two flap push back technique of palatoplasty is commonly used as a protocol of complete unilateral cleft palate closure at the cleft centre unit of dental hospital of Padjadjaran University. On this basis, this research aims to analyze the speech outcome of patients after cleft palate closure with two flaps pushback technique in cleft centre unit of Dental Hospital of Padjadjaran University, with the hypothesis that there is a difference in speech outcome between post-cleft palate closure patients and patients without cleft palate.

MATERIALS AND METHODS

An analytical retrospective study was conducted on children of four to ten years old with complete unilateral cleft palate, who had been treated using two-flap push back palatoplasty technique during period of 2014-2017 at the cleft centre unit of Dental Hospital of University of Padjadjaran. The study by Sakran, et al stated that children's cooperation and validity of the perceptual assessment result can be achieved if the child is older than three years old, because during this age, a child is able to follow instructions better.⁶

Research sampling was done using purposive sampling method. Patients with oronasal fistula, post-recurrent palatoplasty, having a mental retardation history, hearing disorder or was having flu was excluded in this study. Each patient's family had previously signed the informed consent for approval as the research subjects.

The samples were divided into two groups. The first group was the post-palatoplasty and the second was the group without cleft palate as control. Each group consisted of 22 patients. The speech outcomes of both groups, including the articulation pattern, hypernasality,

speech intelligibility and lateral cephalometrical examination, were assessed using perceptual assessment by a speech pathologist.

Perceptual assessment was done by a speech pathologist at the cleft centre unit, with instructions given to the subject articulating words and sentences in Bahasa Indonesia, which consisted of nasal and oral consonants, as presented in Table 1 and 2. Voices were recorded using a high-quality digital recorder in a quiet room with a distance of 15 cm between the microphone and the patient's mouth. Each voice sample that had been recorded was analyzed by a speech pathologist. The assessment criteria were the articulation pattern, hypernasality, and the speech intelligibility as shown in Table 3.

Lateral cephalometry examination was done at the radiology installation of dental hospital of Padjadjaran University with instructions given to the patients pronouncing phonation /i/ during the picture taking process. The result of the lateral cephalometry was traced according to the landmarks measuring the distance of the velum to the posterior pharynx wall at phonation /i/ (Figure 1).

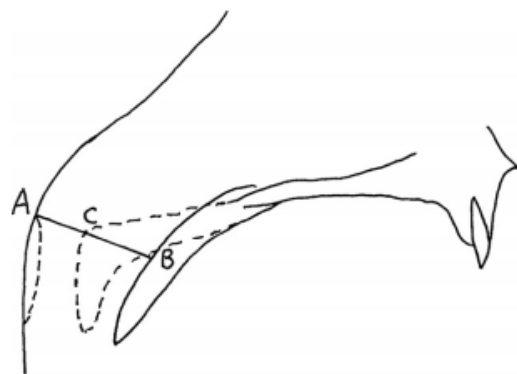


Figure 1. Cephalometry Landmark. (A) Midpoint contact of velopharyngeal; (B) Midpoint of pharyngeal side while velum at rest position; (C) Top of pharyngeal side while velum at /i/ phonation; AB. V-P distance while static; BC. V-P distance while /i/ phonation.⁷

The velopharyngeal competence assessment was done based on the perceptual assessment result and lateral cephalometry. Velopharyngeal competence was distinguished into three categories, adequate, marginal, and inadequate (see Table 4). The collected data were

Table 1. Articulation test in words

Consonant	At the beginning	In the middle	At the end
/p/	palu	api	Asap
/b/	bola	buku	Jilbab
/m/	mata	tomat	Jam
/w/	wayang	bawang	-
/h/	harimau	pohon	lidah
/t/	tas	satu	dompet
/d/	dasi	kuda	daud
/n/	nasi	nanas	daun
/ny/	nyamuk	monyet	-
/ng/	ngantuk	bunga	burung
/k/	kapal	ikan	badak
/g/	gelas	gigi	-
/y/	yoyo	ayam	-
/c/	cat	kucing	-
/r/	roti	jeruk	Air
/l/	lima	balon	mobil
/s/	sapu	pisang	bis
/j/	jagung	gajah	-

Source: Cleft Center Unit of Padjadjaran Univeristy Dental Hospital

Table 2. Articulation test in a sentence

Consonant	Sentence
/p/	Tidak pernah ada api tanpa asap
/b/	Buku Ibu baik
/m/	Dimana nomor enam
/w/	Wadah ini untuk bawang
/h/	Hani melihat pohon buah
/t/	Tuan Toto kuat
/d/	Didalam lumbung ada padi
/n/	Nasi panas dimakan Nani
/ny/	Nyamuk itu gigit banyak orang
/ng/	Ngarai itu ada singa tadi siang
/k/	Kami makan lauk pauk
/g/	Gelas itu bagus
/y/	Yatim piatu itu saya asuh
/c/	Saya cari kucing
/r/	Saya mau roti buah durian dan telur
/l/	Labu itu belinya mahal
/s/	Simpan pisang ini diatas
/j/	Jagung ada di meja

Source: Cleft Center Unit of Padjadjaran University Dental Hospital

subsequently analyzed statistically using Mann Whitney-test.

RESULTS

The average age of post-palatoplasty group was around 6.5 years old and the average age of the control group was 7.5 years old. In this research, there were three samples of subjects aged 4 years old who were quite cooperative to follow the assessment and whose voice were assessed with good result by the speech therapy professional. Post-palatoplasty group consisted of 63.6% girls and 36.4% boys. Control group consisted of 54.5% girls and 45.5% boys. In the sample group of post-palatoplasty, around 86% had followed speech therapy and only 13.6% had not followed the speech therapy. The study subject's characteristics were shown in Table 5.

The results of the perceptual assessment in post-palatoplasty group were nine patients (40.9%)

who had normal articulation, five patients (22.7%) who had omission, and eight patients (36.4%) who had combination of substitution, addition or distortion. Meanwhile, 22 patients of the control group had normal articulation. On parameters of hypernasality in post-palatoplasty group, there was one patient (4.5%) with normal hypernasality, seven patients (32.8%) with mild hypernasality, six patients (27.3%) with moderate hypernasality, and eight patients (36.4%) with severe hypernasality. Meanwhile, in the control group, there were 15 patients (68.2%) with normal hypernasality and seven patients (31.8%) with mild hypernasality. From the view of speech intelligibility in post-palatoplasty patients, eleven patients (50%) had a normal speech intelligibility and the rest of them had abnormal speech intelligibility. In the control group, all samples were shown to have a normal speech intelligibility (Table 6).

Based on the result of lateral cephalometry radiograph, the average distance of velopharyngeal

Table 3. Assessment criteria of perception

Parameter	Criteria	Information
Articulation	Normal	Normal production of phonation
	Substitution	Substitution of consonants
	Distortion	Rioting, changed the meaning
	Omission	Removal of consonants
	Addition	Addition of consonants
Hypernasality	Normal	No nasal emissions
	Mild	Audible nasal emissions
	Moderate	Audible nasal emissions, low weak consonants, probably existing compensation production
	Severe	Inaudible nasal emissions, weak consonants, short articulation range, compensation production
Speech intelligibility	Normal	Clear/understandable
	Abnormal	Not clear/need special audience's attention

Table 4. Assessment criteria of velopharyngeal competence

Velopharyngeal competence	Speech	Speech intelligibility	Lateral cephalometry
Adequate	Normal resonance, normal/mild nasal emission	Normal	VP distance at /i/ phonation < 5.0 mm
Marginal	Mild hypernasal, mild-moderate nasal emission	Abnormal	VP distance at /i/ phonation < 5.0 mm
Inadequate	Moderate-severe hypernasal, severe nasal emission, audible	Abnormal	VP distance at /i/ phonation ≥ 5.0 mm

(VP) in post-cleft palate closure patients was 7.1 mm with the maximum VP-distance of 13.3 mm and the minimum VP-distance of 1.40 mm. On the other hand, in the control group, the average distance of VP was 3 mm with the maximum VP-distance of 7.5 mm and the minimum VP-distance of 0 mm (Table 7).

The velopharyngeal competence in post-palatoplasty group in five patients (22.8%) was adequate; two patients (0.1%) were marginal and 15 patients (68.1%) were inadequate. In contrast, in the control group, there were 16 patients (72.7%) with adequate competence and six patients (27.3%) with inadequate velopharyngeal competence (Table 8).

DISCUSSION

The functional goal of cleft palate surgery is to facilitate normal speech and hearing function

without impairing facial growth.⁸ Various factors, such as surgical technique, time of surgery, operator skill, type and size of the cleft can affect the outcome of the operation.⁹ A retrospective study shows that doing a two flap mucoperiosteal palatoplasty in children with cleft palate before the age of two years old results in a better speech ability.¹⁰ From a clinical point of view, optimal speech function and facial growth can be achieved after repair of the hard palate before the age of 3 years.¹¹ The Cleft Centre Unit Dental Hospital of Padjadjaran University consistently applies the palatoplasty protocol to patients aged 18 to 24 month. In this study, both variables of surgical technique and time of surgery had been controlled.

The differences in age, sexes and speech therapy of each child can affect the assessment of speech function. In children under 6 years old, there were adenoid pads on the posterior wall of pharynx

Table 5. Characteristics of study subjects

Characteristic	Post palatoplasty (n = 22)	Control (n = 22)
Age		
Mean	6.55	7.27
SD	1.55	1.96
Sex		
Male	8 (36.4%)	10 (45.5%)
Female	14 (63.6%)	12 (54.5%)
Speech therapy		
Yes	19 (86.4%)	0 (0.0%)
No	3 (13.6%)	22 (100.0%)

Table 6. Results of perceptual assessment

Perceptual assessment	Post palatoplasty (n = 22)	Control (n = 22)	SD	p-value
Articulation			34.17	0.000
Normal	9 (40.9%)	22 (100.0%)		
Omission	5 (22.7%)	0 (0.0%)		
Distortion/Substitution/Addition	8 (36.4%)	0 (0.0%)		
Hypernasality			40.68	0.000
Normal	1 (4.5%)	15 (68.2%)		
Mild	7 (31.8%)	7 (31.8%)		
Moderate	6 (27.3%)	0 (0.0%)		
Severe	8 (36.4%)	0 (0.0%)		
Speech intelligibility			31.96	0.000
Normal	11 (50%)	22 (100.0%)		
Abnormal	11 (50%)	0 (0.0%)		

Table 7. Results of lateral cephalometrical measures

VP-distance (mm)	Post palatoplasty (n = 22)	Control (n = 22)	p-value
Mean	7.1	3.0	0.000
Median	7.35	3.75	
Minimum	1.4	0.0	
Maximum	13.3	7.5	
SD	2.8	2.7	

Table 8. Velofaringeal competence

Velofaringeal competence	Post palatoplasty (n = 22)	Control (n = 22)	SD	p-value
Adequate	5 (22.8%)	16 (72.7%)	36.87	0.0036
Marginal	2 (0.1%)	0 (0.0%)		
Inadequate	15 (68.1%)	6 (27.3%)		

in the natural contact area with velar. As a result, most children actually possessed a veloadenoidal closure until the natural thyroid atrophy because of aging. After the age approximately reach 6 years old, the thyroid begins naturally to shrink. Post-cleft palate closure or cleft submucosal patients could experience damage in the velopharyngeal closure, mainly when they reach the teenage period, because of this atrophy.⁷ The age difference of post palatoplasty sample group and control in this study was not significant because individual matching had been conducted to variable age between two sample groups.

Different sexes showed a systematic difference in the communication and verbal skills. According to Adhani, many epidemiological studies showed a significantly higher prevalence in communication, language, and speech disorders in boys than in girls. Girls mature more rapidly than boys in verbal communication process and language development skills. Those different abilities are commonly linked to their sexual hormones. Estrogen was found to be correlated with increased social and verbal skills, and to enhance the central speech growth and related area in the brain, while testosterone has an inverse effect.¹² The limitation of this study is on the ignorance of different sexes of each sample group.

Velopharynx is a complex structure that is responsible for the segregation of the oral cavity and nasal cavity during speech and swallowing production. Velopharyngeal closure refers to normal position of the soft palate or velum with the posterior and lateral wall of pharynx. This is a sphincteric mechanism that consists of velar and pharynx components. Movements of those velar components are mainly produced by levator veli palatini muscle action. Perceptual assessment

is considered as a gold standard for assessing the speech outcome and is commonly validated using the instrumental examination.⁸ Similarly, in this study the assessment of speech function with perceptual assessment included assessment of articulation, hypernasality and language intelligence by a speech pathologist, which was validated by instrumental examination using lateral cephalometric radiographs. One of the applicable instruments to measure the velopharyngeal gap is the lateral cephalometric radiograph. Radiographic examination with lateral cephalometry is an initial method to measure the shape of the soft palate and the depth of the pharynx. This is in line with the studies of Morris et al and Mazaheri et al, who reported the use of lateral cephalometry to analyze velopharyngeal function.⁴

In this study, the assessment of speech outcome based on the perceptual assessment through assessing the pattern of articulation, hypernasality and speech intelligibility in post palatoplasty group is significantly different from that of the control group ($p < 0.05$). This is in line with the research conducted by Shprintzen, who reported that 10 to 20% of post primary palatoplasty children experienced permanent hypernasality, while the research done by Sell on a high-scale measurement of cleft palate treatment results in the United Kingdom found out that 45% of the children had a nasal emission.¹³ According to Safaiean, et al., speech intelligibility in children with cleft lip and palate could be disturbed by the presence of articulation disturbance.¹⁴ McWilliams concluded that there is a direct relation between the speech intelligibility and the severity level of nasality and articulation errors.¹⁵ Nasal emission could cause additional secondary characteristics, including

weak or loss of consonants, the production of compensation of articulation, short pronunciation range, and even dysphonia. Overall, this condition influences the quality and intelligibility of speech.⁸

Based on the measurement of the velopharyngeal distance on the lateral cephalometry radiograph, we found seven post palatoplasty patients and eleven patients of the control group, who had a velopharyngeal gap of less than 5 mm. According to Y. Dong, et al., based on the research conducted by Chen, et al. who observed that a complete velopharynx closure was achieved in 16 post palatoplasty patients using Furlow technique and the majority of these patients had a velopharyngeal gap of less than 5 mm.⁵ Another research done by Rajesh, et al. mentioned that the ideal distance of velopharyngeal gap at rest position was approximately 6 mm to obtain a probable maximum velopharynx closure and to prevent velopharynx insufficiency.¹⁶

Velopharyngeal disfunction is a condition, where the velopharyngeal valve is not able to close perfectly, which causes an airway to the nasal cavity during speaking.¹⁷ Resonance disturbance that is mostly reported because of velopharyngeal disfunction is the presence of hypernasality. This event could happen because of inadequate anatomical structure or functional incompetence.⁷ In this research, we found a hypernasality of moderate to severe in patients with a velopharynx gap of more than 6 mm, while in the control group there were no samples with moderate-severe hypernasality. In post palatoplasty group, we found that the broader the velopharynx distance, the higher the severity of hypernasality and as a result, there was an articulation which was compensated but did not highly influence the speech intelligibility. These findings are in line with the study conducted by Lipira, et al. and Scarmagnani, et al. which showed that hypernasality and nasal emission were strong predictors of the velopharynx's gap size.⁸ This was also supported by Kummer's theory, which stated that the audibility of hypernasality and nasal emission depended on the opening size of velopharyngeal valve. However, the opening

size of this velopharyngeal valve was not well correlated with the severity level of speech disturbance that was felt and those affected the speech intelligibility.⁷

Velopharyngeal competence is divided into adequate, marginal, and inadequate. Adequate and marginal velopharyngeal competence refer to a successful velopharynx closure.⁸ In this study, 68.1% of patients of post palatoplasty using two flap push back technique possessed inadequate velopharyngeal competence and showed a significantly different competence than that of the control group ($p < 0.05$). The result of this study is in line with the retrospective study conducted by Y. Dong, et al., which showed that palatoplasty using Furlow technique provided better velopharyngeal function result than two flap palatoplasty, where there were only 10.5% of 88 patients of post palatoplasty using Furlow technique who had an inadequate velopharynx function.⁵

Sakran, et al. reported that the velopharyngeal speech outcome was significantly better in patients who were treated using palatoplasty modification with Z-plasty technique as compared to Von Langenbeck and two flap palatoplasty technique. According to Sakran, et al this event could be caused by a unique characteristic of Furlow Z-plasty palatoplasty. There was an extended soft palate, replacement of divergent palatal muscle to the right position, and avoidance of median scar on the velum that could reduce anteroposterior shrinkage post surgery.⁶ In this study, two flaps push back palatoplasty were chosen because it was a commonly used technique in Cleft Centre Unit of Dental Hospital of Padjadjaran University and this may be considered a limitations in our study. Thus, it is recommended to have further research with other palatoplasty technique such as Furlow Z-plasty.

The majority of post palatoplasty group who had followed speech therapy still had inadequate velopharyngeal competence. This is in line with Kummer, et al who stated that speech therapy was not able to correct velopharynx insufficiency as a condition that was caused by a structural disorder. In general, structural disorder is categorized as

a small velopharynx gap and needs of physical management. Otherwise, speech therapy is done to correct functional disorders (e.g. abnormal articulation placement). Speech therapy is effective in correcting compensatory articulation production that develops as a result of velopharyngeal insufficiency. Placement of such compensatory production occurs in the pharynx, where there is airflow. Due to this placement, the air is released through the nose causing nasal emission. By changing the voice placement from pharynx to oral by doing speech therapy, nasal emission can be eliminated.¹⁸

Postoperative speech therapy is essential to help individuals eliminate compensatory articulation production and learn to produce sounds with normal oral placement. In this study, 19 post-palatoplasty patient had a different history of speech therapy in terms of intensity and the chosen speech therapy procedure. Some patients had undergone speech therapy by following home program by the cleft centre unit, where patients were able to do the speech therapy together with their parents or other family members at home using a guidance by distance by a speech therapy professional from the cleft centre. Environmental or family factors have a major impact on a successful speech outcome in post palatoplasty patients. Therefore, a cooperation by the parents/family to take an active role in training the child's speech outcome at home is highly expected.

CONCLUSION

This study concludes that there is a significant difference in the speech outcome between post-cleft palate closure patients and patients without cleft palate. Most of patients after cleft palate closure with two flaps pushback technique had inadequate velopharyngeal competence with moderate-severe hypernasality, severe nasal emission, abnormal speech intelligibility, and velopharyngeal distance of ≥ 5.0 mm, whereas the majority of control group had an adequate velopharyngeal competence. The speech quality in post palatoplasty patients could be influenced by several factors, such as surgery

techniques, operator skills, time of surgery, age and sex of patients, environmental factors and speech therapy intensity. Further studies should be conducted by doing a long-period follow up to evaluate the speech outcome in post palatoplasty using two flap push back technique.

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