CRANIOFACIAL

Speech Outcome in Cleft Palate Patients After Soft Palatoplasty (Stage 1) in Two-Stage Palatoplasty Technique: A Review of Two Cases

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Background: The challenge in palatoplasty is no longer a successful closure of the cleft palate but an optimal speech outcome without compromising maxillofacial growth. On the contrary, repairing cleft palate as soon as possible is recommended to improve speech. The surgeon proposed two-stage palatoplasty in early aged, range between 4 to 30 months. The delayed hard palate closure started approximately between the age of three years and one year after soft palatoplasty to avoid temporary retracted oral articulation of certain consonant.

Patient and Methods: 11-months old and 9-months old patients with non-syndromic unilateral cleft lip and palate had undergone soft palatoplasty. Speech was examined as a perceptual assessment, using protocol in Craniomaxillofacial Center Cipto Mangunkusumo Hospital, in the age of 29 months by an experienced speech pathologist.

Result: Patient 1 has mild hypernasality, adequate velopharyngeal competence, normal articulation pattern, phonation and speech intelligibility. Patient 2 has misarticulation pattern, which are omission or weak consonants, substitution of pharyngeal stop, mild to moderate hypernasality, mild impairment in speech intelligibility, and inadequate velopharyngeal competence.

Summary: Speech outcome is influenced by intense speech practicing done by their parents. The patient can work on the misarticulation. We need further study to know the result of speech outcome in two stage palatoplasty.

Keywords: Two-stage palatoplasty, soft palatoplasty, hard palate closure, speech outcome, velopharyngeal competency

Latar Belakang: Tantangan dalam palatoplasti tidak lagi penutupan celah yang sukses tetapi hasil bicara optimal tanpa mengganggu pertumbuhan maksillofasial. Ahli bedah mengusulkan palatoplasti dua tahap. Soft palatoplasty awal dilakukan dalam batas 4-30 bulan. Penundaan penutupan palatum durum dilakukan hingga kisaran usia 3 tahun dan minimum setahun setelah soft palatoplasty untuk mencegah artikulasi oral teretraksi temporer pada beberapa konsonan tertentu.

Pasien dan Metode: Dua pasien dengan bibir dan langit-langit sumbing unilateral non-syndromic telah menjalani soft palatoplasty pada usia 11 bulan dan usia 9 bulan. Keduanya dilakukan pemeriksaan bicara pada usia 29 bulan menggunakan penilaian perseptual sesuai protokol di RS. Dr. Cipto Mangunkusumo Pusat Craniomaksillofasial oleh seorang ahli patologis bicara yang berpengalaman.

Hasil: Pasien 1 memiliki hipernasalitas ringan, kompetensi velopharyngeal adekuat, pola artikulasi, fonasi dan pemahaman bicara normal. Pasien 2 memiliki pola artikulasi yang salah, berupa penghilangan atau pelemahan konsonan, substitusi dari letupan pharyngeal, hipernasalitas ringan hingga sedang, pemahaman bicara terganggu ringan, dan kompetensi velopharyngeal tidak adekuat.

Ringkasan: Hasil bicara dipengaruhi oleh intensitas latihan bicara oleh orang tua pasien. Pasien dapat berlatih memperbaiki artikulasi yang salah. Dibutuhkan penelitian lebih lanjut.

Kata kunci: Two-stage palatoplasty, soft palatoplasty, hard palate closure, speech outcome, velopharyngeal competency

left lip and palate are the most common congenital craniofacial anomalies treated by plastic surgeons. 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%. There might be many reasons why a certain treatment protocol, whether about timing or technique operation, is chosen. Many studies have been done to compare different protocol.

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Therefore, surgical protocols for cleft palate are continuously evolving to give better results in successful closure of the cleft palate, an optimal speech outcome without compromising maxillofacial growth.^{1,2}

Many studies have revealed that patients with unrepaired clefts have normal skeletal cephalometric relationships compared with those with repaired clefts. The effect of hypoplasia maxilla may be influenced by excessive scar tension in the antero-posterior vector which retards normal midface growth. As the opposite, there is recommendation to repair cleft palate as soon as possible to improved speech. In order to get the better results in these two opposites' outcomes, two stage palatoplasty has been proposed with different timing and procedure operation.

Two stage palatoplasty is two-stage approach to cleft palate repair with different protocols aimed at early repair of the soft palate, followed by delayed repair of the hard palate. By early soft palate closure, speech was possible from an early age as which we considered that the most important period for speech commences immediately after birth, and not as was previously believed- in the third year of life. Residual cleft palate is left to grow without being disturbed and expected to get narrower. As a result, delayed hard palate closure will have less lateral defects without any periosteal coverage which give a less scar. So, we expect to decrease maxillary growth disturbance caused by operation.^{1,3-6}

Delayed hard palate closure in the age of 12 to 14 years as on Schweckendiek protocol has been reported to have the most favorable maxillary growth (Bardach et al., 1984) but less satisfactory in speech outcome. Recently, many studies reported that delayed hard palate closure to 5 years old indicates difficulties in articulating consonants produced at or before the site of articulation (Cosman and Falk).^{5,7,8}

To avoid this speech impairment in this study, we delay the closure of residual cleft, approximately until in the age of three years. We expect to get characteristics of speech outcome in cleft patient with two-stage palatoplasty as good as another procedure but get a better characteristic result of maxillary growth.

Patients with non-syndromic unilateral cleft lip and palate underwent soft palatoplasty (stage 1) in Medan on August and October 2010 (Figures 1). Patients were operated by single

PATIENT AND METHODS

surgeon. They underwent soft palatoplasty, using straight line technique, with intravelar veloplasty (release vellum muscle from posterior edge of hard palate and approximate it into midline position). Patient 1 underwent soft palatoplasty and labioplasty at the same time in the age of 11 months, and patient 2 had undergone labioplasty in the age of 9 months. Both of the patients were examined for speech outcome in the age of 29 months and none of them underwent speech therapy before. The speech examination included alloanamnesis, physical examination of mouth cavity and speech recording. Speech examination is a perceptual assessment. Because there is no protocol for speech examination in Medan, speech had been assessed using universal parameters for reporting speech outcome in individuals with cleft palate as a first protocol (Table 1).12

Because this is a perceptual assessment, we consulted their speech recordings to Medical Rehabilitation Department, Faculty of Medicine University of Indonesia, Cipto Mangunkusumo Hospital Jakarta and changed speech protocol examination. Both patients after soft palatoplasty (stage 1) underwent speech examination in April 2012 using protocol in Craniomaxillofacial Center of Dr. Cipto Mangunkusumo Hospital (Table 2)

Speech was recorded using digital recorder in a noise free room. The microphone was placed 15-20 cm away from the mouth. The patients were asked to follow 39 single words in Bahasa Indonesia, including words predominantly consisted of nasal and oral consonant with phonation emphasis of the vowels /a/, /i/, and /u/. The words are listed in Table 2. Patients also count numbers from 1 to 10 in Bahasa Indonesia. Speech samples was analyzed by a speech pathologist who is experienced in the assessment of cleft palate speech. Perceptual analysis of intelligibility, articulation, and resonance are analyzed

Table 1. The rating scale value that applies for each parameter based on universal parameters for reporting speech outcome in individuals with cleft palate

Rating Scale	Words and Sentences			
Hypernasality –single words and 0 = Within normal limits 1 = Mild 2 = Moderate	Papa Dadu Tat, tat, tat, tat Bip, bip			
3 = Severe				
X = missing data		Pipa ada dua buah Ada dua puluh dua batu di atap		
Hyponasality – Sentences 0 = within normal limits/ none 1 = present X = missing data	Mama makan mangga Mama minum			
Audible Nasal Air Emission and Nasal Turbulence – single words sentences 0 = within normal limits/none 1 = present		Sisi Susu Bantal		
I = intermittent or variable F = frequent or pervasive X= missing data	Susi dan Teddi saling membantu Siska makan malam di tenda			
Consonant Production Errors – single words and/ or sentences 0 = Within normal limits/none 1 = Present	Abnormal backing of oral targets to post uvular place	Bapak Budi duduk di bus dekat supir		
	Abnormal backing of oral targets, but place remains oral	Tali, Duduk, Gagak, Susi, Zaza, Zaki, azan		
	Nasal fricative	Sip, Zip, Visa, Shinta, Vivi Bibi Shinta duduk dekat ibu		
	Nasal consonant for oral pressure consonant	Bapak,Tidur,Data, Gagak, Lupa, bulu, lalu lintas		
	Nasalized voiced pressure consonants	Badak, badai, Guru, Gugus		
	Weak oral pressure	Didi datang tidak ditandu		
	Other oral misarticulations	Susah, Tinta, Fanta, Visa Lupa lihat lalu lintas		

Speech Understandability – conversational speech and/ or alloanamnesis

- 0 = within normal limits : speech is always easy to understand
- 1 = mild : speech is occasionally hard to understand
- 2 = moderate : speech is often hard to understand
- 3 = severe : speech is hard to understand most or all of the time
- X = missing data

Speech Acceptability – conversational speech and/ or alloanamnesis

- 0 = wthin normal limits : speech is normal
- 1 = mild : speech deviates from normal to a mild degree
- 2 = moderate : speech deviates from normal to a moderate degree
- 3 = severe : speech deviates from normal to a severe degree
- X = missing data

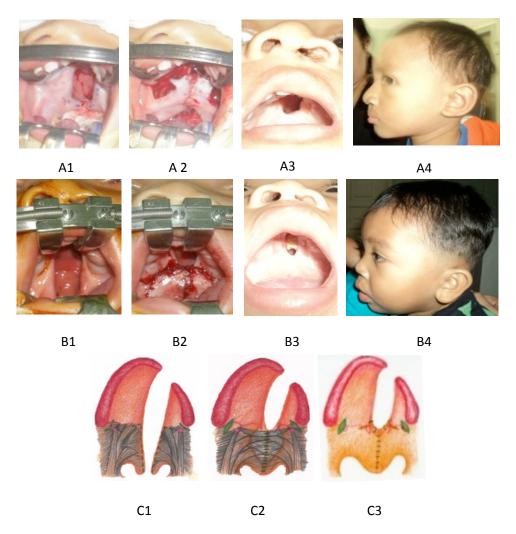


Figure 1. A1. Patient 1 with unilateral labiopalatoschizis in the age of 11 months; A2. Patient 1 Post Soft palatoplasty with intravelar veloplasty and labioplasty; A3, A4. Patient 1 in the age of 29 months; B1.Patient 2 with unilateral labiopalatoschizis post labioplasty in the age of 9 months; B2. Patient 2 post soft palatoplasty; B3, B4. Patient 2 in the age of 29 months; C1. Diagram illustrates abnormal muscle insertions and orientation in cleft palate⁴; C2. Diagram illustrates soft palatoplasty using straight line technique with Intravelar veloplasty; C3. Diagram illustrates suture in mucosal oral post soft palatoplasty.

following the Murthy rating criteria for speech parameters. Based on these parameters, the velopharyngeal competence level is divided in whether good, fair, or poor (Table 3). Good result was preferred to definite and probable adequate velopharyngeal competency. Fair result means marginal velopharyngeal competency, while poor score means a probable or definite inadequate velopharyngeal competency.

Delayed hard palate closure (stage 2) is designed to be done approximately in the age of three years old and minimum one year after soft palatoplasty using two flaps or one flap on non-cleft side technique. Speech outcomes is design to be examined before, three months after hard palate closure (stage 2), and followed every year after delayed hard palate closure until the patient in the age of 8 years.

Early soft palatoplasty was done by straight line incision along margin of soft palate cleft. Velar muscles were released from their attachment to the posterior edge of palatal shelves and then being reoriented across the midline. Small transversal incision on oral mucosa was done in hard palate-soft palate transition to facilitate intravelar veloplasty and closure of soft palate.

Table 2. Words in Bahasa Indonesia using protocol in Craniomaxillofacial Center of RSCM

	/a/	/i/	/u/	
/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	
/1/	lalat	tali	palu	
/m/	mandi	minum m	mulut	
/n/	nanas	anisa	banu	
/r/	kerang	lari	rumah	
/s/	sapi	dasi	susu	
/p/	papa	api	sapu	

Table 3. Rating scale values in Craniomaxillofacial Center Cipto Mangunkusumo Hospital

Rating Scale	Characteristics			
Articulation	0 = Normal production of			
Rating	majority of phonemes			
	1 = Predominantly distortion			
	of phonemes			
	2 = Distortion and substitution			
	of phonemes 3 = Phonemes are substituted			
	and omitted			
Hypernasality	0 = Normal			
Rating	1 = Mild hypernasality			
	2 = Moderate hypernasality			
	3 = Severe hypernasality			
Phonation	0 = Normal			
	1 = Mild			
	2 = Moderate			
	3 = Severe			
Speech	0 = Normal, all speech			
Intelligibility	is understood			
Rating	1 = Mild			
	2 = Moderate			
	3 = Severe, almost all speech			
is hard to be understood				
Velopharyngeal	0 = Good			
competence	1 = Fair			
	2 = Poor			

In post operative time of soft palatoplasty, patients were hospitalized for 1 day and would be discharged if the systemic or local complication had not occurred. Antibiotic and analgesic were given orally. Patients were instructed to come for follow up visits every three days. Post operative nutrition was liquid diet with normal temperature in one week. First to second week after operation, patients were given liquid diet. Second to third weeks after operation, patients were given soft diet. After three weeks, patients may consume normal diet.

Two patients have been examined for speech in the age of 29 months on April 2012 using protocol in Craniomaxillofacial Center of Cipto Mangunkusumo Hospital. Their speech was assessed by an experienced speech pathologist. The result is in Table 4. Patient 1 has normal articulation pattern, mild

hypernasality, normal phonation, normal speech intelligibility, and adequate

RESULT

velopharyngeal closure. Patient 2 has misarticulation pattern, mild to moderate hypernasality, phonation within normal limit, mild impairment in speech intelligibility, and inadequate velopharyngeal closure. Even though, both of them still have residual cleft of the hard palate. Misarticulating pattern in patient 2 is omission or weak consonants in oral pressure sounds, where there is a problem in manner of formation articulation for instance "/p,b,d,g,k,t,s,j/", and substitution of pharyngeal stop.

From alloanamnesis, both patients were in a good condition to be recorded. Parent of patient 1 gives an intensive speech practice, for instance teach to pronounce words well. We found that parent of patient 2 did not give an intense speech practice. On the physical examination, no broken suture were found in post-operative.

There is still controversy in speech outcome post two stage palatoplasty. Schweckendiek has opinion that in spite of the remaining cleft in the hard palate, intelligible speech can be learned.³ Soft palatoplasty during the first year of life enables normal growth of the upper jaw and the closure of the residual cleft of the hard palate is generally postponed until the age of 12 to 14 years. On average, the residual cleft becomes at least 60 to 70% narrower, in over 95% of all cases. Speech difficulties were found in the first few years.

DISCUSSION

Long term speech results after soft palatoplasty indicates that normal speech was found in 57.2% of cases, intelligible speech in 37.6% of case, moderate speech in 4.5% of case, poor speech in 0.7% of cases. There is no intravelar veloplasty in Schweckendiek's patients. Another experimental retrospective randomized study, done by Akamatsu et.al. in Japan (2004), compared between one stage done in the age of 18-24 months and two stage done in the age of 6 months (stage 1) then 18-24 months (stage 2). The final speech evaluations

in the age of 4-5 years from both groups were no difference between one stage and two-stage procedures.^{4,9}

Chait et al. did the first-stage repair of the soft palate defect involved by mobilizing two short posterior based flaps and freeing of the muscle followed by an intravelar veloplasty, they found 86% of the sample had good to excellent speech (41% excellent, 45% good, 14% poor). ⁴ Before that report, the result from surgery using straight-line repair without intravelar veloplasty, similar to those described by Cosman and Falk, was included poor speech development and difficulty in closing the residual cleft. ⁷

Speech development depends on anatomic of orofacial structure, timing of cleft closure, hearing, sensory perception, cognitive, language development, and social quotient skills. In this study, primary veloplasty was done with intravelar veloplasty to restore vellum muscle on anatomical position. After complete separation of the levator muscles from the posterior edge of the hard palate, they could be rotated medially and backwards, and sutured as a strong muscle sling with less tension, which improve healing and ruptures did not arise frequently. 10-12

Speech development is a very gradual process that begins in infancy and sometimes continues until the age of 7 or 8. Babbling, started in the age of 2-6 months, is the

Table 4. Speech Assessment in the Two Patients

No	Name	Sex	LAHSHAL	Operation Stage I Age Technique		Follow up Age Speech Outcome					
				(mo)		(mo)	A	Н	P	SI	VPC
1	S	M	SHAL	11	Soft palatoplasty + labioplasty	29	0	1	0	0	0
2	NG	M	SHAL	9	Soft Palatoplasty	29	3	2	0	1	3

A = Articulation pattern SI = Speech Intelligibility

H = Hypernasality VPC = Velopharyngeal competency

P = Phonation

repetition of syllables in sequence like "ba-ba-ba" and typically not attach any meaning to these playful sounds. Laling, started in the age of 7-9 months, is the repetition of their sound. Echolali, started in the age of 18-36 months, is repetition of sound which comes from outside. They are practicing these oral motor skills that they will later need for actual speech. Soft palatoplasty (stage 1) can be done in the age of 4 months so they can start earlier to use their vellum in speech. We expect the patient can catch up with their speech development close to normal child and gain normal speech in five year-old to eight year-old.^{4,11,13,14}

In this report, two patients were operated on the soft palate at the age of 9 months and 11 months. We expected that after soft palatoplasty, they could start developing their speech in the laling and echolalia period and start to learn articulation. Patient 1 who underwent soft palatoplasty in the age of 11 months has a better result than patient 2 who underwent soft palatoplasty in the age of 9 months. Patient 1 has mild hypernasality and within normal limit for phonation, articulation pattern, intelligibility, and velopharyngeal competence even though there is still residual cleft in the hard palate. Patient 2 has misarticulation pattern, omission (some consonants was disappeared and changed by another consonant), certain weak consonants in oral pressure sounds and substitution of pharyngeal stop. There is a disturbance in manner process of formation oral pressure sounds for instance /p,b,d,g,k,t,s,j/ because of suboptimal intraoral pressure to produce the consonants. Substitution of pharyngeal stop is an effort to compensate closure of vocal tractus to the distal because of disturbance closure in the proximal part of the vocal tractus.

From alloanamnesis with their parents, we found that parent of patient 1 who gave intense speech practice to their child have their child better speech outcome. Parent of patient 2 is a single parent and the intensity of practicing speech is not enough. Patient 1 is more obedient than patient 2.

Considering about maxillary growth, Graber stated that early traumatic palatoplasty can interfere with maxillary growth laterally, anteroposteriorly, and vertically, and recommended that palatoplasty should be postponed until the end of the fourth year, when fifty-sixths of the total maxillary width has been attained.¹⁰

Slaughter performed the soft palate closure at 11 months to 4 years of age and suggested that the primary suture of the velum should promote growth processes by creating a muscle balance around the defect (theory of the functional matrix). In another study on development of the residual cleft of the hard palate after primary veloplasty, it was found that the residual cleft narrowed markedly during the first year after surgery.⁶

However, Cosman and Falk emphasize the fact that children with an uncorrected cleft palate encounter difficulties in articulating consonants produced at, or before, the site of articulation. There is relative high prevalence oral retraction articulation which has found in the age of three to seven years. Spontaneous speech concern is about temporary retracted oral articulation of certain consonants.^{9,10,15}

In this study, we started to close residual cleft palate approximately in the age of three years old but minimum one year after soft palatoplasty. We expected to avoid retracted oral articulation and get residual cleft narrower. Furthermore, less scar tension will diminish disturbance of maxillary growth.

There is another fifty four patients with non-syndromic unilateral and bilateral cleft lip and palate were undergone soft palatoplasty (stage 1) in Medan since November 2009. Six of them have been operated delayed hard palate closure (stage 2). Patients were operated by single surgeon. They underwent soft palatoplasty in the age range of 4 to 30 months, using straight line technique, with intravelar veloplasty with or without posterior vomer flap. Delayed hard palate closure was done approximately from three years old and minimum one year post soft palatoplasty, using two flap or one flap on non-cleft side technique. Six of them, one BCLP and five UCLP, underwent hard palate closure in the range age from 31 months to 48 months (average 39.2)

months). Speech outcomes were examined before, three month after, and followed every year after delayed hard palate closure (stage 2) until the patient in the age of 8 years. We expected to make a preliminary study.

SUMMARY

Speech outcome is influenced by intense speech practicing done by their parents. The patient can work on the misarticulation. We need further study to know the result of speech outcome in two stage palatoplasty.

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