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Maxillary Growth and Speech Outcome in Patient with Cleft Lip and Palate after Two-Stage Palate Repair: A Systematic Review

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Background: The management of patients with cleft lip and palate is complex, where the treatment outcome is judged on the balance between aesthetics, speech, and maxillary growth. Up to now, there is no generally accepted treatment protocol. Every center must find the best-suited protocol treatment for their population.

Methods: A systematic review through literature search was conducted for English-language studies in PubMed. This search was conducted in September 2011 using EndNote X3 with keywords: Two-stage Palate Repair and Maxillary Growth and Two-stage Palate Repair and Speech Outcome. Both retrospective and prospective studies on maxillary growth and speech outcome in patient with cleft lip and palate after two-stage palate repair published from 2001 to 2012 were included.

Result: From the reviewed of 37 articles, only 14 articles fit the inclusions criteria, three articles discussed the outcome of maxillary growth and speech outcome, eight articles only discussed the maxillary growth and the rest of articles only discussed the speech outcome.

Conclusion: From this review we found that most of the two-stage palate repair results in better maxillary growth, but only few of them results in good speech outcome. We will perform further study based on this review to discover a new protocol for the management of palate repair in our center.

Keywords: *two-stage palate repair, maxillary growth, speech outcome competency*

Latar Belakang: Penanganan terhadap pasien dengan sumbing bibir dan langit-langit merupakan masalah yang kompleks dengan penilaian hasil terapi berdasarkan keseimbangan antara nilai estetik, suara, dan pertumbuhan maksila. Sampai saat ini, tidak ada protokol terapi yang dapat diterima secara luas. Setiap *center* diharuskan memiliki protokol terapi yang paling sesuai dengan populasi mereka.

Metodologi: Sebuah sistematik review dilakukan dengan mencari literature berbahasa inggris yang terdapat di PubMed. Pencarian dilaksanakan pada September 2011 menggunakan EndNote X3 dengan kata kunci Rekonstruksi Palatum Dua tahap dan Pertumbuhan Maksilla, serta Rekonstruksi Palatum Dua tahap dan Kemampuan Berbicara. Termasuk artikel baik berupa studi retrospektif maupun prospektif terhadap pertumbuhan maksila dan kemampuan berbicara pasien dengan sumbing bibir dan langit setelah menjalani rekonstruksi menggunakan dua tahap prosedur yang di publikasikan dari tahun 2001 sampai 2011.

Hasil: Berdasarkan review terhadap 37 makalah, hanya 14 yang memenuhi kriteria, tiga makalah membahas tentang pertumbuhan maksila dan kemampuan berbicara, delapan artikel membicarakan tentang pertumbuhan maksila dan sisanya merupakan makalah yang membahas tentang kemampuan berbicara.

Kesimpulan: Berdasarkan *review*, kami mendapatkan rekonstruksi menggunakan dua tahap prosedur memberikan hasil yang baik terhadap pertumbuhan maksila, dan hanya sebagian yang memberikan hasil yang baik terhadap kemampuan berbicara. Kami akan melakukan penelitian lebih lanjut berdasarkan penelitian ini untuk menemukan protocol terapi yang tepat untuk rekonstruksi palatum di *center* kami

Kata Kunci: *two-stage palate repair, maxillary growth, speech outcome competency*

The management of patients with cleft is complex, where the treatment outcome is judged on the balance between esthetics, speech, and facial growth.¹ The best treatment

should ensure good aesthetic and functional outcomes when these patients are adults. The patient should be able to speak and eat without problems and have an invisible scar and no

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growth disturbances. Improvement in the treatment of the cleft lip and palate required the determination of the optimal treatment protocols.²

Palate repair can be performed as a one- or two-stage procedure. There are different opinion about the technique and the ideal timing of surgery. The aim of palate repair is to create a complete closure, having an intact hard and soft palate with normal functioning velopharyngeal mechanism. If not, there are resonance disorder, with an altered voice, compensatory articulation, and chronic middle ear infections.³

In our center, currently we performed one-stage palate repair protocol using two-flap palatoplasty with repositioning of the muscle to treat complete cleft palate whether unilateral or bilateral. Our retrospective study showed the quality of facial growth is fair to poor (mean GOSLON index score = 3.53),⁴ and the speech outcome is good (72.7%).⁵ Many of our patients came from remote areas and low socio-economic background, most of them do not feel the need for maxillary hypoplasia correction (orthognatic surgery). Therefore, we are trying to find the best palate repair protocol which would result in good maxillary growth without sacrificing the speech outcome, so that our patients would not need to come back for further interventions in the future and yet their aesthetic appearance is satisfying and their speech outcome is good.

Up to now, there is no generally accepted treatment protocol. Every centers must find the treatment protocol best-suited for their own population. Further clinical study based on this review is needed to establish a new and better protocol for the management of cleft palate in our center.

PATIENT AND METHODS

This study is designed to answer the question of whether two-stage palate repair results in better maxillary growth and speech outcome or not. A systematic review through literature search was conducted for English-language studies in PubMed. This search was conducted in March 2012 using EndNote X3

with keywords: 'Two-stage Palate Repair and Maxillary Growth' and 'Two-stage Palate Repair and Speech Outcome'. Both retrospective and prospective studies on maxillary growth and speech outcome in patient with cleft lip and palate after two-stage palate repair published from 2001 to 2011 were included. Exclusion criteria are letter to editor, article, comment, discussion, literature research, publication review, prevalence study, animal study and lecture.

Good result refers to good maxillary growth and/or speech outcome based on conclusions made by author. Poor result refers to poor maxillary growth and/or speech outcome based on conclusions made by authors.

RESULT

The search strategy resulted in 37 articles, only 14 articles fit the inclusions criteria, three articles discussed the outcome of maxillary growth and speech outcome, seven articles only discussed the maxillary growth and the rest of the articles only discussed the speech outcome. Among 11 articles which discussed maxillary growth, five articles (45.4%) revealed better result in maxillary growth, two articles (18.2%) revealed no difference in maxillary growth, and 4 articles (36.4%) revealed poor maxillary growth. Seven articles which discussed speech outcome, only 2 articles (28.6%) revealed good speech outcome (Table 1).

There are two centers reported good result for both maxillary growth and speech outcome, namely Johannesburg and Göterburg. Chait et al in Johannesburg used a modified intravelar veloplasty technique for soft palate closure. The average age of patients at first-stage soft palate repair was 10.7 months (range 6 – 17 months). The residual cleft (second-stage repair) was closed at 32.7 months on average (range 26 – 34 months). Repair of residual cleft is undertaken using two hinge flaps for the nasal lining and two narrow posterior based mucoperiosteal flaps for cover (Fig.1).¹⁵

The Göterburg center used two-stage technique in which soft palate repair was performed at age 7.5 ± 2.6 months while the

Table 1. Studies from Literature Maxillary Growth and Speech Outcome Two-stage Palate Repair

Study Number/ Author(s)	Growth studies		Speech studies	
	Study Methods	Outcome	Study Methods	Outcome
1 Gaggl, et al ⁶	Cephalometric	Poor	N/A	N/A
2. Corbo et al ⁷	Cephalometric	no difference	N/A	N/A
3. Gaggl et al ⁸	Dental Model and Cephalometric	Poor	N/A	N/A
4. Nishio et al ⁹	Dental model	Good	N/A	N/A
5. Zemann et al ¹⁰	Cephalometric	no difference	N/A	N/A
6. Liao et al ¹¹	Cephalometric	Good	speech sample	Poor
7. Friede et al ¹²	Cephalometric and Dental Cast	Good	N/A	N/A
8. Holland et al ¹³	Cephalometric	Poor	Audio recording	Poor
9. Friede et al ¹⁴	Cephalometric	Good	N/A	N/A
10 Chait et al ¹⁵	Cephalometric	Good	N/A	good
11 Pradel W et al ³	Dental Model	Poor	N/A	poor
12 Lierde et al ¹⁶	N/A	N/A	Audio/video recording	poor
13 Lohmander A, Friede H, Lijla J ¹⁷	N/A	N/A	Audio Recording	Good
14 Lohmander A, Persson C ¹⁸	N/A	N/A	Audio recording	Poor

delayed hard palate repair was done in the early mixed dentition age of 8.1 ± 0.7 months. Closure of the soft palate was performed using a technique in which incisions were made close to the border between hard and soft palate. At repair of the residual cleft in the hard palate, incisions were made close to the teeth and flaps of the whole mucoperiosteum within the dental arch were raised. At the end of surgery the flaps were joined in the midline and the united palatal mucoperiosteum was sutured back in place, leaving no denuded bone in the palate (Figure 2).¹⁴

DISCUSSION

The disturbance of maxillary growth can be caused primarily by congenital anomaly and secondarily by surgery. The cleft surgery is among the surgeries which can affect the maxillary growth. Interference with maxillary growth caused by surgical scarring results in maxillary-deficient class III patterns.¹⁵ The retardation of maxillary growth can be an effect of early palatoplasty before 1 year of age which was caused by insufficiency of the soft tissue to cover the defect. This lacking of the soft tissue will cause scarring and thus will also have a

bigger risk to develop a velopharyngeal insufficiency beside maxillary growth retardation. To counter these speech development problems, the best time to do the surgery is in the 10-20 months of patient's age.¹⁹

The two-stage operation concept for palatal closure has long been advocate in many cleft centers to minimize maxillary growth inhibition induced by palatal surgery.¹⁹ These protocol basically consist of soft palate closure carried out at a relatively early period and delayed hard palate closure at 3, 5, 7 to 8, or 15 years of age. Moll et al (1998) showed that the width of the cleft in UCLP subjects decreased during the first year after veloplasty. After that the average size of the residual clefts remained stable.²⁰ Although patients treated with delayed hard palate closure have eventually shown severe articulation problems, the results maxillary growth have been encouraging.⁶ Thus delayed surgical invasion in the hard palate appears to be effective in protecting the palate from iatrogenic growth impairment, although, some controversies remain. Rohrich et al (2000) recommended 3 to 5 months as the ideal age for soft palate repair and 15 to 18 months as the age when the hard palate preferably should be closed.²¹

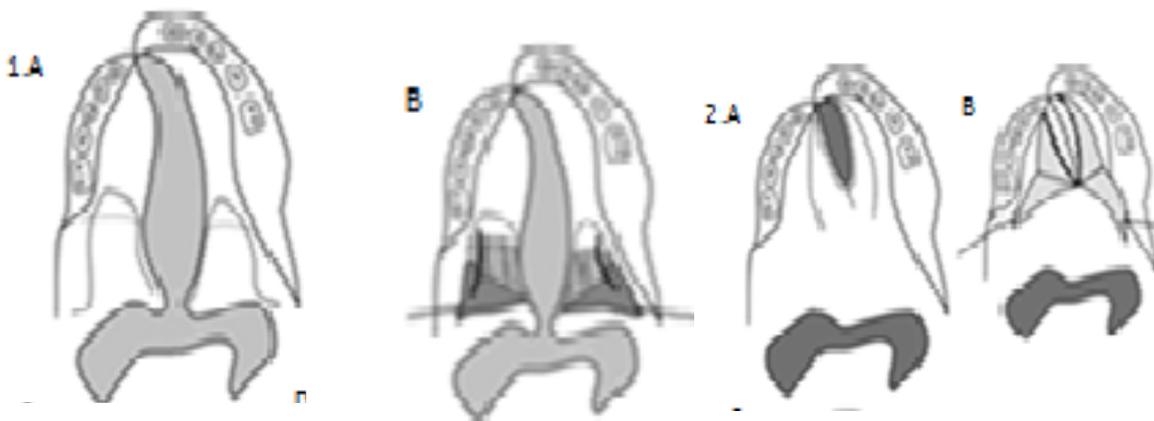


Figure 1 Primary palatoplasty. A. Two short posteriorly based mucoperiosteal flaps are raised. B. Levator muscles are freed from the posterior edge of the hard palate. 2. Repair residual cleft. A. Design of flaps for closure of the residual cleft. B. Two hinge flaps are used for nasal lining.

From our review, the Göterburg and Johannesburg studies revealed good maxillary growth and speech outcome. In Johannerburg, modified intravelar veloplasty technique (Figure 1) was used to close the soft palate. Intravelar veloplasty was performed with extension of the soft palate repair up to 1 cm beyond the border of hard palate. Good speech outcome in this study most probably due to the long soft palate created with this modified intravelar veloplasty technique. The residual cleft narrows in time, and the second-stage repair was undertaken using narrow flaps, thus reducing dissection and scarring, and in the long run resulted in good maxillary growth.¹⁵

In Göterburg, during the soft palate repair, incisions were made close to the border between hard and soft palate (Figure 2). Medially subperiosteal dissection was performed, while laterally, the cuts mostly involved the mucosa behind the maxilla. Also a subperiosteal flap was raised from the posterior vomer and turned backwards to be sutured to the anterior nasal layer of velum. Thus only small areas of palatal and vomer bone were left denuded close to the midline. At repair of the residual cleft in the hard palate, incisions were made close to the teeth and flaps of the whole mucoperiosteum within the dental arch were raised. At the end of surgery the flaps were joined in the midline and the united palatal

mucoperiosteum was sutured back in place, leaving no denuded bone in the palate. Soft palate repair involving anchoring of the velum to the posterior vomer seems to be one of the keys for good speech results in two stage regimens with delayed hard palate closure. As described by Friede (2009) it is obvious that 'the initial velar closure will result in a long soft palate, if its muscles are dissected from the posterior palatal shelves and reoriented to a transverse course.¹⁴ In addition, the inclusion of a posterior vomer flap to help repair the anterior nasal layer of the velum is another important step in the creation of a long soft palate. The Göterburg technique also resulted in good maxillary growth. By using this technique, only small areas of palatal and vomer bone were left denuded close to the midline in soft palate repair, while in the hard palate repair no denuded bone were left in the palate. A widely accepted view is that denuded bone will lead to development of scar tissue, which depending on the size and position, might risk subsequent maxillary growth.¹⁴

The speech outcome revealed from other studies found in our review was poor.^{3,11,13,16,18} Even in 1 study which used Furlow technique for the soft palate repair, the speech outcome is poor despite the rationale that this procedure lengthens the velum so that reliably provide adequate speech development. In this study, the

Table 2. Detailed of included studies in patient with cleft lip and palate

Study number/author(s)	Design	Sample size	Cleft type and subtype	Population Studied	Sequence (mean age at surgery in months)	Technique of palate repair	No. of Sur-geon	Age at assess-ment (yrs)
1 Gaggl, et al ⁶	Prospective	30	UCLP	Austria	L(3-6) → SP(1824) → HP(72)	Tennison → Widmaier → Veau	?	4
2 Corbo et al ⁷	Retrospective	11	UCLP	Austria Brussel	L(3-6) → SP&HP(11-14) L,SP&HP(3) → Quad Helix (84)	Tennison → Veau Malek Technique	?	4 7 & 12
3 Gaggl et al ⁸	Retrospective	30	UCLP	Brussel	Device → ABG(120) SP(3) → L&HP(6) → Quad Helix Device (84) → ABC(120) SP(6-10) → HP(48-72)	?	?	7 & 12
4 Nishio et al ⁹	Retrospective	29	UCLP	Austria	SP&HP(25-35)	widmaier → veau	2	18,9
5 Zemann et al ¹⁰	Retrospective	30	UCLP	Austria	PSO(03) → L(3) → SP(12) → HP(18)	Veau	?	17,8
6 Liao et al ¹¹	Prospective	41	UCLP	Taipei	PSO(0-3) → L(3) → PB(12) PSO(0-3) → L(3) → SP&HP(12) PSO(0-6) → L(6) → SP(12) → HP(30)	? → modified furrow → bridge flap technique Wardill Kliner Modification Milliard → Veau Randall's → intravelar veloplasty → simultaneous mucoperios closure of the alveolus Modified rotation advancement → widmaier → vomerine mucoperiosteal flap	1 1 1	48,6 6-8,3 6-8,3
7 Friede et al ¹²	Longitudinal	20	UCLP	Taipei	L(14,4) → SP(19,2) → HP(67,2)	Modified rotation advancement → two flap palatoplasty	9	10,7 ± 4,8
8 Holland et al ¹³	Retrospective	31	UCLP	Göteborg	L(4,8) → SP&HP(12)	Modified rotation advancement → two flap palatoplasty	9	9,6 ± 3,4
9 Friede et al ¹⁴	Study cohort	41	UCLP	Riga	LA(2,1) → SP(8,1) → L&N(17,2) → HP(102,5) → ABG(102,5) L&N(7,8) → SP(20,2) → HP(61,9) → ABG(114,8)	? → ? → ? → mucoperiosteal flap	?	7,10,13,16
10 Chait L et al ¹⁵	Study cohort	42	UCLP	Pittsburgh	SP(12) → a pinned-retained speech prosthesis → HP(84)	? → Frolowa/ schweckendiek → mucoperiosteal palatal flap intravelar veloplasty → vonlangenbeck	?	7,10,13,16
11 Pradel W et al ³	Study cohort	22	UCLP	Pittsburgh Göteborg Johannesburg	LA(3) → SP(7,5) → L&N(17,3) → HP(97,2) SP(10,7) → HP(32,7)	intravelar veloplasty, Vonlangenbeck Göteborg Modified intravelar veloplasty → mucoperiosteal flap	?	5,7,16,19
12 Lierde et al ¹⁶	Retrospective	20	UCLP/BCLP	Dresden	L(4,6) → SP(9-12) → HP(24-36)	intravelar veloplasty, Vonlangenbeck	?	5,7,16,19
13 Lohmander A, Friede H, Lijla J ¹⁷	Study cohort	55	UCLP/BCLP	Göteborg	L(3-8) → SP(13-28) → HP(96-138) L(3-9) → SP and HP(12-24) LA(3) → SP(7,5) → L&N(17,3) → HP(97,2)	intravelar veloplasty, vomer flap and bipedicle flap Wardill -Kliner Göteborg	1 1 ?	5,7,16,19
14 Lohmander A, Persson C ¹⁸	Longitudinal	18	UCLP	Göteborg	SP(6-8) → HP(36-46)	?	?	3,5,7

subject who received a two-stage Furlow palatoplasty statistically showed more hypernasality and have a higher nasalance scores in comparison with the one stage Wardill-Kilner palatoplasty.¹⁶

While in the maxillary growth evaluation, four other studies found in our review revealed poor results.^{3,6,8,13} Gaggl et al concluded that the technique which they use produce extensive scarring in the border region between the hard and soft palate, resulted in poorer maxillary growth compared to the one-stage palate repair group.^{6,8} Holland et al concluded that the poor maxillary growth they obtained is likely due to the scar produced by a cleft obturator prosthesis that can inhibit the growth of the maxilla.¹³ An early orthopaedic appliance might have facilitated speech development, however, current opinion is that the effect from early palatal appliances on speech production usually is insignificant.¹⁸

In our review, we found heterogeneity of the studies included i.e. a wide range of populations, hard palate repair timings, hard palate repair techniques, number of surgeons who performed the palate repair, duration of the study period (Table 2), tools used to evaluate maxillary growth and speech outcome, sample size, and sampling technique. This resulted in biases in drawing a conclusion whether two-stage palate repair resulted in better maxillary growth and speech outcome.

CONCLUSION

Despite many biases from the studies included in our review, we found that most of the two-stage palate repair studies resulted in better maxillary growth (45.4%), but only a few resulted in good speech outcome (28.6%). Among the studies reviewed, only 2 studies resulted in good outcome both in maxillary growth and speech. It is worthwhile to explore the possibilities of implementing the techniques used in these 2 studies to improve the maxillary growth and speech outcome in our center. Further clinical study based on this review is needed to establish a new and better protocol for the management of cleft palate in our center.

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