

Speech and Hearing Outcomes of the Patients with Cleft Lip and Palate: Comparison between One-Stage Simultaneous and Two-Stage Surgical Repairs

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Background: Conventional treatment for cleft lip and palate patients is lip repair at three to four months and then palatal repair at nine to 12 months of age. However, for the patients who delay seeing a doctor especially in a developing area such as Northern Thailand, simultaneous lip and palate repair is performed at 12 to 18 months of age or later, depending on the age at the first visit. It is a common belief that patients with cleft lip and palate will be behind non-cleft patients in early development phonemes because of the open palate. This delay persists until the palate is repaired and on into the postoperative period. This proposition has not been proven with long-term clinical outcomes in one-stage repairs.

Objective: To investigate the effects of one-stage repair on speech assessment, hearing, and incidence of palatal fistula. The results were compared with conventional two-stage surgical repairs.

Materials and Methods: The present study was designed two groups. Group 1 consisted of 25 children (mean age 11.28±1.93 years) treated with a one-stage repair. Cleft lip, palate, and alveolus were repaired at a single surgical session in the first 18 months of life (mean age at the time of surgery 13.52±4.51 months). Group 2 consisted of 17 children (mean age 11.02±2.23 years) treated in two-stage surgical repairs. Lip repair was performed at a median age of 4.01 months (IQR 3.62 to 5.46), and palate repair was performed at a mean age of 13.54±4.14 months. Both groups underwent cleft lip and palate repairs at the Division of Plastic Surgery, Department of Surgery, Faculty of Medicine, Chiang Mai University between January 1, 2004 and December 31, 2010. Speech and hearing for all patients were evaluated by experienced ENT doctors. The palatal fistula was evaluated by the same plastic surgeons.

Results: One-stage repair showed significant normal articulation and less articulation disorder when compared with two-stage surgical repairs. However, no significant difference was determined for other speech assessments, hearing, and incidence of palatal fistula.

Conclusion: Because one-stage repair seems to have a more positive influence on articulation, and both surgical treatment protocols give similar results on speech assessments, hearing, and incidence of palatal fistula, regardless of the timing of the surgery, the one-stage repair is not inferior to conventional two-stage surgical repairs for patients in developing areas. This is due to several important advantages, such as less hospitalization, lower cost, and less chance of nosocomial infection.

Keywords: One-stage repair, Speech, Hearing, Palatal fistula, Cleft lip, Palate

Received 24 April 2020 | Revised 19 June 2020 | Accepted 23 June 2020

J Med Assoc Thai 2020;103(11):1171-7

Website: <http://www.jmatonline.com>

An orofacial cleft is the interruption of the fusion of midline frontonasal, maxillary, lateral nasal, and

the mandibular portion that leads to cleft lip and cleft palate. The incidence of cleft lip and palate in white new-borns is approximately 1:1,000 live births⁽¹⁾. In Thailand, the incidence is between 1.10 and 2.49 per 1,000 live births; 59% of cleft patients lived in the North-East⁽²⁾, with a male to female ratio of 1.7:1⁽³⁾. The main objective of cleft lip and cleft palate surgery is to reconstruct the maxillary segments so that normal speech and hearing with reliable maxillofacial growth can be achieved. Although there is concurrence in some surgical practices (e.g., lip repair at three to six months of age, palate repair before 18 months of age)^(1,4), the best timing of treatment, the most reliable treatment regimen, and the efficacy of currently used surgical techniques remain controversial. The concept

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How to cite this article:

Chanpeng S, Khwanngern K, Sonsuwan N, Sirimaharaj W. Speech and Hearing Outcomes of the Patients with Cleft Lip and Palate: Comparison between One-Stage Simultaneous and Two-Stage Surgical Repairs. J Med Assoc Thai 2020;103:1171-7.

doi.org/10.35755/jmedassocthai.2020.11.10887

Table 1. Baseline characteristics of patients

Characteristics	One-stage (n=25) n (%)	Two-stage (n=17) n (%)	p-value
Sex			0.530
Male	13 (52.00)	11 (64.71)	
Female	12 (48.00)	6 (35.29)	
Diagnosis: lip			0.810
Unilateral incomplete	2 (8.00)	3 (17.65)	
Unilateral complete	12 (48.00)	7 (41.18)	
Bilateral incomplete	2 (8.00)	2 (11.76)	
Bilateral complete	9 (36.00)	5 (29.41)	
Diagnosis: palate			1.000
Unilateral complete	14 (56.00)	10 (58.82)	
Bilateral complete	11 (44.00)	7 (41.18)	
Incomplete	0 (0.00)	0 (0.00)	
Weight at CLP/CP surgery (kg); mean±SD	9.92±1.73	9.14±0.85	0.093
Age at cheiloplasty (months), median (IQR)	-	4.01 (3.62 to 5.46)	
Age at palatoplasty (months); mean±SD	13.52±4.51	13.54±4.14	0.988
Age at the time of study (years); mean±SD	11.28±1.93	11.01±2.23	0.685
Alveolar bone grafting			0.353
Yes	13 (52.00)	6 (35.29)	
No	12 (48.00)	11 (64.71)	

CLP=cleft lip and palate; CP=cleft palate; SD=standard deviation; IQR=interquartile range

of “one-stage repair” is based on early repair of the entire clefts within the first 12 months. According to this concept, cleft lip, palate, and alveolus are simultaneously repaired in one surgical session to obtain the best functional and developmental results. This procedure helps to form a full vestibule of the mouth and to close the oronasal fistula, which allows normal development of the dentoalveolar arch, and therefore, speech. This approach was introduced in 1966 by Davies⁽⁵⁾. Subsequently, many experimental and clinical studies have been published regarding this concept. Many of these⁽⁶⁻⁹⁾ demonstrated that simultaneous repair of cleft lip and cleft palate before 12 months of age provided better speech and hearing results and reliable maxillofacial growth compared to conventional two-stage repairs. However, there was no long-term follow-up for these issues.

Because the present study patients came to see doctors late, the authors used this concept to perform the operations. Therefore, a “one-stage repair” had more benefits for the patients. Additionally, the authors were interested in long-term clinical outcomes compared to conventional two-stage surgical repairs.

In the present article, the authors investigated the speech assessment, hearing, and incidence of palatal fistula results of the cleft lip and palate cases, compared between the two groups.

Materials and Methods

Populations

From the authors’ retrospective data, 75 new patients with cleft lip and palate were treated with either a one-stage simultaneous repair or two-stage surgical repair between January 1, 2004, and December 31, 2010, at the Division of Plastic Surgery, Department of Surgery, Faculty of Medicine, Chiang Mai University. Ten cases with a syndrome or mental retardation and twenty-three lost to follow-up cases were excluded. The remaining 42 patients were included in the present study.

Twenty-five patients treated with one-stage simultaneous repairs were gathered as Group 1. The remaining seventeen patients, treated with two-stage surgical repairs, constituted Group 2. Detailed information about sample sizes and the baseline characteristics in each group is listed in Table 1.

Surgical technique

The surgical techniques performed in both one-stage and two-stage surgical repairs were identical. The only differences between the two procedures in the present study were the timing of the operations and alveolar closures. In the one-stage, whole cleft defects were repaired at the one surgical session simultaneously in the first visit (between six and 30 months) of life (Table 1). The two-stage surgical repairs were the cheiloplasty and palatoplasty. They were performed separately at three to six months and 12 to 18 months of age, respectively. This approach is commonly used worldwide.

All the cleft patients were treated with the same surgical methods and by the same team of four plastic surgeons, in the Division of Plastic Surgery, Department of Surgery, Faculty of Medicine, Chiang Mai University. In the present protocol, cheiloplasty was performed according to Millard's rotation advancement⁽¹⁰⁾ and Tennison's technique⁽¹¹⁾ for unilateral cleft lip. The authors repaired bilateral cleft lip according to the Mulliken's technique⁽¹¹⁾. To achieve good functional results, the orbicularis oris muscles were dissected and joined carefully. Hard palate closure was achieved with two-flap palatoplasty according to the Bardach's technique⁽¹²⁾. In the patients who were suitable for a vomerine flap, a two-layer palatoplasty was performed. Soft palate closure was performed with intra-velar veloplasty in three layers according to Kriens⁽¹³⁾. Finally, the alveolar repair was completed with periosteoplasty according to Skoog⁽¹⁴⁾. All patients were given first-generation cephalosporins prophylactically to minimize the chance of infection.

Speech and hearing assessment

Speech and language: Speech is human vocal communication using language.

Articulation: The sound that results from phonation and resonance is further altered for individual speech sounds by articulators, lips, jaws, teeth, tongue, and velum. Functional articulation disorder means production in response to abnormal structure, compensatory articulation disorder means articulation is primarily produced in the pharynx or larynx.

Resonation: This is the transmission of sound energy through the oral, nasal, or pharyngeal cavities of the vocal tract during speech production. Hyponasality means too little sound energy is in the nasal cavity. Hypernasality means too much sound energy is in the nasal cavity. A nasal emission is

the presence of audible nasal airflow due to leakage intraoral air pressure from mouth to nose.

Voice: The vibration of the vocal cords (due to rapid cycles of opening and closing) causes the oscillation of the air stream, thus producing the sound of voices. Voice disorders are abnormal conditions involving abnormal pitch, loudness, or quality of the sound.

Intelligibility: A measure of how comprehensible speech is in given conditions. The present study divided intelligibility into intelligibility if the topic was known and unintelligible if the topic was unknown.

All the parameters were evaluated by an experienced ENT doctor using single sounds, syllable repetition, sentence repetition, counting, and connected speech.

The hearing was measured via air conduction testing, testing with earphones, and evaluation of the entire auditory system per audiogram. If the air conduction test indicated a hearing loss, bone conduction testing was used. The results were interpreted by the same experienced ENT doctor from the Otolaryngology Department, Faculty of Medicine, Chiang Mai University.

Palatal fistula

Palatal fistula defined as patency between the oral and nasal cavities were evaluated by the same four plastic surgeons at the Division of Plastic Surgery, Department of Surgery, Faculty of Medicine, Chiang Mai University.

Statistical analysis

Independent t-test analysis was used to compare the continuous data of baseline characteristics, speech, and hearing results in both groups, and Fisher's exact test or chi-square test was performed for categorical data, e.g., gender and diagnosis. All calculations were performed using Stata, version 14.0 (StataCorp LP, College Station, TX, USA) for Windows. A p-value of less than 0.05 determined statistical significance.

Ethical approval

The present study was approved by the Research Ethics Committee of the Faculty of Medicine, Chiang Mai University on April 30, 2018 (no.169/2561).

Results

Evaluations of patients were performed mainly in two sessions, speech and hearing assessment, and incidence of palatal fistula.

Table 2. Speech and hearing assessment

Speech and hearing results	One-stage (n=25) n (%)	Two-stage (n=17) n (%)	p-value
Speech and language			0.506
Normal	23 (92.00)	17 (100)	
Speech and language delay	2 (8.00)	0 (0.00)	
Articulation			0.037
Normal	10 (40.00)	3 (17.65)	
Functional articulation disorder	0 (0.00)	3 (17.65)	
Compensatory articulation disorder	15 (60.00)	11 (64.71)	
Resonance			1.000
Normal	5 (20.00)	3 (17.65)	
Hyponasality	0 (0.00)	0 (0.00)	
Hypernasality	20 (80.00)	14 (82.35)	
Nasal emission	0 (0.00)	0 (0.00)	
Voice			1.000
Normal	23 (92.00)	16 (94.12)	
Voice disorder	2 (8.00)	1 (5.88)	
Intelligibility			1.000
Intelligibility	24 (96.00)	17 (100)	
Intelligibility if the topic is known	0 (0.00)	0 (0.00)	
Unintelligibility	1 (4.00)	0 (0.00)	
Hearing			1.000
Normal	24 (96.00)	17 (100)	
Sensorineural hearing loss	1 (4.00)	0 (0.00)	
Conductive hearing loss	0 (0.00)	0 (0.00)	
Mixed hearing loss	0 (0.00)	0 (0.00)	

Speech and hearing assessments

Speech and language: Two patients in Group 1 had delayed speech and language. The overall speech and language for both cleft groups were normal and not significantly different (Table 2).

Articulation: One-stage repairs had more normal articulation and significant functional articulation disorders when compared to two-stage surgical repairs. There was no significant difference between the groups regarding compensatory articulation disorder (Table 2).

Resonance: Almost 80% of the cases per group had hypernasality and no significant differences between the groups (Table 2).

Voice: Almost 90% of the cases per group had a normal voice. Therefore, there were no significant differences between the groups (Table 2).

Intelligibility: One patient in Group 1 was unintelligible. The overall intelligibility of both cleft groups was normal and not significantly different

(Table 2).

Hearing: One patient in Group 1 had sensorineural hearing loss. The overall hearing of both cleft groups was normal and not significantly different (Table 2).

Discussion

Simultaneous repair of cleft lip and palate is an old protocol that has been the subject of debate during the past four decades. Most of these studies did not reveal comprehensive results or sufficient follow-up time. However, enough knowledge has been accrued to compare the procedures with other approaches to cleft lip and palate repair.

The repair of the cleft lip between three and six months of age is a concept that has been agreed on by most authors.

For lip repair, Manchester reported that he eliminated the muscle repair stage involved in lip closure to avoid undue tension⁽¹⁵⁾. Conversely, many authors have stated that the orbicularis muscle must

be repaired independently to allow each to fulfill its separate function and to increase speech and maxillary development due to the reduction of premaxilla anterior projection, repositioning of the premaxilla, especially in bilateral cleft lip and palate, and lingual tipping of the upper incisors^(16,17). The authors repaired the orbicularis oris muscle in both study groups. In the authors' opinion, muscle repair must be performed to obtain the best functional and aesthetic results. Currently, it is a common practice performed by many surgeons.

There are primarily two intensively used procedures for alveolar repair, which are bone grafting and periosteoplasty. Millard et al. found that the use of primary gingivoperiosteoplasty yields the advantages of moving the palate into a normal position, stabilizing dental arch, providing not only a potential conduit for the eruption of teeth, but also the asymmetrical platform for the early repair of the nose, and avoided anterior palatal fistula⁽¹⁸⁾. Sameshima et al revealed in their study that primary periosteoplasty appeared to have an additional advantage over bone grafting regarding increased vertical relations of maxillofacial morphology⁽¹⁹⁾. The periosteoplasty procedure was routinely used for alveolar repair in all the authors one-stage repair cases.

The main point of debate has been a palatal repair. There are still no standard protocols for ideal timing for cleft palate repair to obtain optimum speech and to avoid maxillofacial growth disturbance. In the literature, there were primarily two early simultaneous repair methods with some variations for cleft lip and palate patients. The first is a one-stage simultaneous palatal repair, which is the authors procedure in the present study. The timing of the procedure was described above. The second method is the Malek protocol⁽²⁰⁾, in which the soft palate was closed at a mean age of three months, followed by simultaneous repair of the lip and hard palate at a mean age of six months.

One-stage palatal repair between 9 to 18 months is the most common worldwide practice. As for the present study, the one-stage palatal repair was used in all the present cases.

Regarding speech and hearing, it has been demonstrated that the number of operations is an insignificant factor affecting the incidence of otitis media, tympanic membrane assessments, and audiometric evaluations⁽²¹⁾.

The present study revealed that patients with the one-stage repair had normal articulation and less significant compensatory articulation disorder

when compared to the two-stage surgical repairs, and almost 80% of the cases per group had hypernasality. This was due to no regular follow up to improve and correct their speech, So, they had mislearning that was compensated by compensatory articulation disorder and hypernasality.

As for palatal fistula, Landheer et al⁽²²⁾, in 2010, revealed a relatively high incidence of fistula formation after cleft palate repair. Their study suggested that a two-stage surgical repair had a higher rate of fistula formation when compared with a one-stage repair. The present study demonstrated that cleft width at the time of cleft palate repair played a crucial role in the development of fistulas.

The present study revealed that patients with the one-stage repair had normal articulation and less significantly articulation disorder when compared to the two-stage surgical repairs. Other speech and hearing assessments, and incidence of palatal fistula revealed similar deviations in both study groups. Regardless of the timing of surgery, an important question needs to be answered: which criteria must be considered as decisive factors in preference of the surgical method?

It is very difficult to state that the results and scope of the present study could be considered satisfactory, even though the articulation was significant because the sample size was too small, and the operations were performed by multiple surgeons. Multiple factors influenced speech development other than the cleft itself and the surgical method provided such as the brain structure and functions, environmental stimulation, hearing and vision, motivation, attention, and vocal tract anatomy and physiology. The authors believe that a multidisciplinary approach with follow-up of the cleft patients is necessary.

Conclusion

The authors' comparative study revealed that both the one-stage repair and the conventional two-stage surgical repairs affected to the same degrees of other speech assessments (except articulation), hearing, and incidence of palatal fistula. However, the one-stage repair offers several important advantages. First, the one-stage repair is less expensive than repeated operative procedures. A short hospitalization period saves the children from the high risk of nosocomial infections. Furthermore, the psychological stress connected with a hospitalization can be reduced if it occurred before the age when the baby is aware of himself or herself. An additional aspect of cleft surgery that is often seen in the developing countries

is a lack of continuity of follow-up because of socio-cultural and economic insufficiency. The one-stage repair may be a good alternative for solving these problems. The authors believe that the one-stage repair procedure in cleft lip and palate repair should not be undervalued.

What is already known on this topic?

1. Patients with cleft lip and palate will be behind non-cleft patients in early development phonemes due to the open palate. This delay persists until the palate is repaired and on into the postoperative period.

2. "One-stage repair" is based on early repair of the entire clefts of the child within the first 12 months. According to this concept, cleft lip, palate, and alveolus are simultaneously repaired in one surgical session to obtain the best functional and developmental results. This procedure helps to form a full vestibule of mouth and to close the oronasal fistula, which allows normal development of the dentoalveolar arch and provides better speech and hearing results compared to conventional two-stage repairs.

What this study adds?

Because no study provided long-term clinical outcomes in speech assessment, hearing, and the incidence of palatal fistula in patients who were performed the one-stage repair compared to conventional two-stage surgical repairs, the parameters were added in this study.

Acknowledgment

The authors would like to thank the Faculty of Medicine Research Fund for providing research funding. Special thanks are also given to the staff in the Department of Orthodontics, Faculty of Dentistry and Department of Otolaryngology, Faculty of Medicine, Chiang Mai University, Chiang Mai, Thailand for their cooperation.

Conflicts of interest

The authors declare no conflict of interest.

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