Comparison of Postoperative Stability and Complications Following Orthogenetic Surgery between Patients with Skeletal Class III Deformity with/without Cleft Lip and Palate

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Abstract

Objective: To assess the postoperative stability and complications following orthognathic surgeries for patients with skeletal class III deformity with/without cleft lip and palate (CLP). **Subjects and methods**: The subjects were 34 patients with CLP who underwent orthognathic surgeries, including sagittal split ramus osteotomy in 11 patients, Le Fort I osteotomy in 9 patients, and two-jaw surgery in 14 patients. As a control, 7 patients treated with two-jaw surgery and 18 patients treated with SSRO without clefts were used. Retrospectively, the amount of jaw movement and intra- and postoperative complications were analyzed, and then compared among the five groups. Furthermore, the pre- and postoperative facial landmarks and relapse distances were measured based on lateral cephalograms. **Results**: There were no significant differences in the amount of jaw movement, operation time, or intraoperative bleeding. Cephalometric analysis demonstrated significantly greater maxillary hypo-growth in both the anteroposterior and the vertical directions in patients with CLP. No significant differences were observed in the relapse distance in the same operation between with and without CLP by ANOVA or velopharyngeal closure among the five groups by χ square test. **Conclusions**: Our orthognathic surgeries provided the same levels of stability and intraor postoperative complications for patients with skeletal class III with or without CLP.

Key Words: Cleft lip and palate, Skeletal class III deformity, Orthognathic surgery, Relapse

Introduction

In patients with CLP exhibiting Class III illegal occlusion, jaw corrective surgery is often performed [1,2]. In these patients, significant hypo-growth of the maxilla often occurs during patient growth, and many of them require orthognathic surgery for maxillary deformity. However, the following accompanying difficulties due to soft tissue scars following palatal repair can occur during orthognathic surgery in these patients: 1) unstable circulation of maxillary bone fragments after osteotomy, 2) limitation of the amount of maxillary advancement during surgery, 3) difficult fixation of the bone fragments, and 4) postoperative relapse. Therefore, careful attention is recommended during orthognathic surgery with maxillary advancement for patients with CLP [3-10].

Postoperative relapse has been one of the most serious issues in the field of orthognathic surgery for patients with CLP. The causes and countermeasures have been reported over the last 30 years. Previous reports described that the rate of relapse following maxillary advancement was much higher in patients with CLP, ranging from 25% up to 50%, while the corresponding rate for non-cleft patients with maxillary hypoplasia has been reported to be approximately 10% [7-9,10]. Cheung et al. [9] reported in their meta-analysis of 72 reports involving 1,418 cleft palate patients treated with maxillary osteotomy between 1966 and 2003 that surgeons who performed maxillary advancement took the following countermeasures: 1) modification of the osteotomy design to minimize the tension of palatal scars, 2) protection of the mucosa to preserve the blood flow to bone fragments, 3) combination with mandibular osteotomy compensating for the extent of maxillary advancement, and 4) consideration of miniplate fixation and overcorrection. However, there remain many difficulties in the treatment of jaw deformity in patients with CLP compared with those without clefts. Moreover, although the complication of velopharyngeal insufficiency (VPI) after surgery has also been reported [11], preoperative prediction of the development of velopharyngeal insufficiency after maxillary advancement is not yet possible [12-16]. There are few reports of studies comparing cleft subjects and non-cleft subjects.

Since its establishment, our department has been engaged in the comprehensive treatment of cleft lip patients for more than 35 years. To prevent postoperative relapse, we have performed orthognathic surgery considering the following points: 1) completing preoperative orthodontic treatment, when possible, 2) sufficient dissection around the posterior edge of the palatal bone from the fibrous scar tissues, 3) secondary bone graft in the clefts to unionize the maxillary segments, and 4) bone graft into the gap produced by maxillary advancement. In this study, we analyzed the current state of orthognathic surgery for patients with CLP who were treated at our department, focusing on the changes in relapse after surgery using lateral cephalometric radiographs and velopharyngeal (VP) closure function, in comparison with patients without CLP.

Subjects and Methods

The data were collected from the records of the Oral Maxillofacial Department and the CLP Clinic at Kagoshima University Hospital. This study was approved by the Clinical Research Ethical Review Boards of Kagoshima University Medical and Dental Hospital (#20-150).

In this study, in the 35 years from April 1981 to March 2015, of the 40 patients who underwent primary cleft lip and

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cleft palate treatment in our department, we performed back mandibular posterior movement and mainly forward movement of the maxilla. There were 34 cases that were treated. Side cases of maxilla bone expansion, mandibular extension for Robin syndrome, and 6 cases of upper and lower jaw movement performed in 2 stages were excluded.

The subjects were 34 consecutive patients who had received primary lip and palate repairs followed by orthognathic surgery at our department between April 1981 and March 2015. The patients' ages ranged from 15 to 18 years old and they included 16 males and 18 females. All patients received primary lip repair using a modified Randal technique at 3~4 months after birth, and palatoplasty using modified pushback surgery, in which the mucoperiosteal tissue was preserved at the anterior part of the hard palate, with veloplasty consisting of lavatory muscle repositioning. The surgical procedure of orthognathic surgery was mandibular setback by sagittal split ramus osteotomy (SSRO) (CLP-S group) in 11 patients; bilateral cleft lip and palate (BCLP): 4, unilateral cleft lip and palate (UCLP): 6, isolated cleft palate (OCP): 1, two-jaw surgery (CLP-T group) in 14 patients (BCLP: 7, UCLP: 7), and maxillary advancement by Le Fort I osteotomy (CLP-L I group) in 9 patients (BCLP: 1, UCLP: 8) (*Figure 1*).

group	Total no.	Gender (f/m)	Mean age (y)		Cleft type		
					BCLP	UCLP	CP
CLP-S (CLP-SSRO)	11	6/5	17.6 ± 2.1	#	4	6	1
CLP-T (CLP-Two jaw)	14	7/7	16.7 ± 1.0	##	7	7	0
CLP-L (CLP-Le Fort I)	9	5/4	16.0 ± 0.9	##	1	8	0
Cont-S (Control-SSRO)	18	16/2	19.3 ± 4.0		—	—	—
Cont-T (Control-Two jaw)	7	2/5	19.2 ± 9.2		—	—	—

#, ##; significant difference between each group (ANOVA, #: $p<0.05$, ##: $p<0.01$)

Figure 1. *Tabular representation of subjects.*

In addition, as a control, 18 patients who underwent SSRO (Cont-S group) and 7 patients who underwent two-jaw surgery (Cont-T group) without clefts during the same period were used (*Figure 1*). The mean age of the Cont-T group was slightly older than that of the others.

For choosing the operation type, maxillary anterior movement was within 5 mm as a standard, and for cases with overjet of 5 mm or more, mandibular retraction surgery was used in combination. For cases in which velopharyngeal incompetence was expected, only the mandibular osteotomy was performed.

As SSRO had been performed after 2008 in the CLP-S group, we selected patients from 17 to 29 years of age who received SSRO after 2008 for the Cont-S group. We selected patients who had within a 4-mm difference between the left and light side set-back distance at the first molar in SSRO. For two-jaw surgery, patients with anterior, with/without inferior, movement of maxilla were selected. Additionally, patients without documents were eliminated in this study.

Procedures of orthognathic surgery

A summary of our procedures for orthognathic surgeries is provided below;

1) Preoperative orthodontic treatment was carried out ensuring an adequate occlusal relationship at orthognathic surgery, when possible.

2) In the CLP-S group, mandibular setback was carried out by sagittal split ramus osteotomy in accordance with the Obwegeser-Dal Pont method. Bone segments were fixed using metal screws (2 screws in each side) or titanium miniplates after movement of the mandible.

3) In the CLP-L group, down-fracture of the maxilla was performed employing Le Fort I osteotomy, followed by forward movement of the maxilla. Upon this maxillary advancement, sufficient dissection around the posterior edge of the palatal bone from the fibrous scar tissues was achieved, and bone fragments, which had been donated from the iliac

crest, were inserted bilaterally into the gaps produced by maxillary advancement.

4) In the CLP-T group, mandibular setback by SSRO was performed following Le Fort I maxillary advancement. The early stages of the fixation used wire, and titanium miniplates were applied to fix maxillary segments. An iliac bone was inserted into the gap produced by maxillary advancement before miniplate fixation.

5) In the Cont-S and Cont-T groups, surgical procedures for mandibular setback, maxillary advancement, and fixation of bone segments were the same as those in cleft lip patient groups.

6) Intermaxillary fixation was performed for 3 weeks in all groups at an early stage, but the fixation period has recently been decreased to 1 week.

The data on the amount of jaw movement, operation time, and blood loss during surgery were obtained from the operation records and compared between the CLP groups (CLP-S, CLP-T and CLP-L) and control groups (Cont-S and Cont-T). The amount of jaw movement was measured from the changes in the relationship between the first upper and lower molars.

Cephalometric evaluation of pre- and postoperative skeletal configuration and relapse distance

A total of 34 patients were analyzable until six months: 11 patients in the CLP-S group, 14 patients in the CLP-T group, 9 patients in the CLP-L group, 18 patients in the Cont-S group, and 7 patients in the Cont-T group. Lateral cephalograms acquired before surgery, and 1 and 6 months after surgery were scanned into a personal computer, and measurement and analysis were performed by one oral and maxillofacial surgeon using 3D-Rugle® (Medic Engineering, Kyoto, Japan).

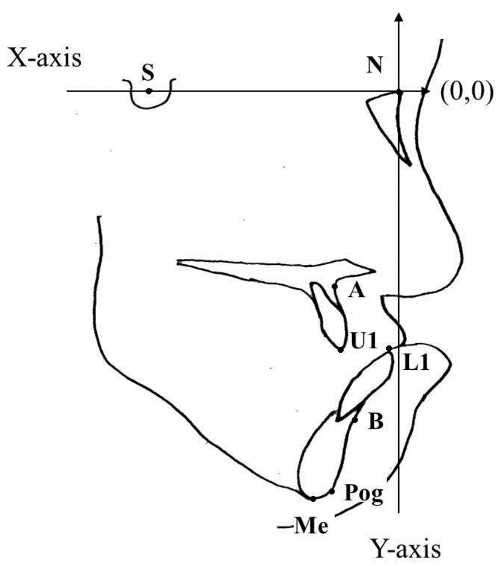

Figure 2. Evaluation of lateral cephalogram.

We used the same cephalostat for each patient and took cephalometric films at the standard head position for cephalometry. To avoid inter-examiner error, cephalometric measurement was performed by one oral surgeon who was not the operator.

On the lateral cephalometric radiographs, the X-axis was set as a straight line crossing the S-N line, and the Y-axis was set as a line crossing S and perpendicular to the X-axis (*Figure 2*). Point N was set as the origin (0,0).

In this study, changes in the X- and Y-coordinates of A, U1, L1, B, Pog, and Me, and changes in SNA, SNB, and ANB were measured. Changes in the X- and Y-axis directions from the coordinates before surgery (T1) to those at 1 month after surgery (T2) were regarded as the amount of cephalometric movement, and those from 1 month to 6 months after surgery (T3) were regarded as the relapse distance in the lateral cephalograms.

Intra and postoperative complications

Complications excluding intraoperative hemorrhage were investigated in all patients. The velophaligeal (VP) closure functions before and after orthognathic surgery was also analyzed. The VP closure function was evaluated based on perceptual judgment of the presence of hyper-nasality and nasal emission by oral surgeons and speech therapists during speech therapy before surgery, and 1 month and 6 months after surgery. The VP closure function was classified into two categories: good: no or mild hyper-nasality and slight nasal emission; or poor: moderate or severe hyper-nasality and moderate or severe nasal emission.

Statistical analysis

Of the measured values, the mean and standard deviation of the amount of movement, operation time, and blood loss were calculated in each group and compared using ANOVA. On lateral cephalometric analysis, the positions of the skeletal landmarks, SNA, andSNB between before and after surgery were compared among the groups using ANOVA. Movement and relapse distances in each group were compared using the paired t-test. The changes in velopharyngeal function were evaluated by χ The significance level was set at 5% or lower.

Results

Amount of jaw movement, operation time, and blood loss (*Figure 3*)

The measured amounts of jaw movement during the operation were 7.3 ± 2.1 (4.5~9.0) mm in CLP-S, 11.7 ± 2.4 (7.5~16.0) mm in CLP-T, and 6.0 ± 1.4 (3.5~8.5) mm in CLP-L. In CLP-T, the amount of jaw movement was divided into 4.5 ± 0.9 (3.0~6.0) mm in the maxilla and 7.2 ± 1.8 (4.0~10.0) mm in the mandible. The movement distance in the Cont-S group was 6.3 ± 1.8 (4.5~13.5) mm. The total amount of jaw movement in the Cont-T group was 12.5 ± 5.0 (5.3~19.0) mm, which was divided into 3.9 ± 1.3 (2.3~5.0) mm in the maxilla and 7.4 ± 4.0 (2.3~14.0) mm in the mandible. There were no significant differences in the amount of jaw movement between CLP-S and Cont-S, as well as CLP-T and Cont-T.

Group	Movement distance (mm) total (Wits' appraisal)	Maxilla	Mandible	Operation time	Amount of bleeding (ml)
CLP-S (n=11)	7.3 ± 2.1 (4.5 ~ 9.0)	–	7.3 ± 2.1 (4.5 ~ 12.5)	4:11 ± 0:41 (3:20 ~ 5:28)	207 ± 89 (50 ~ 360)
CLP-T (n=14)	11.7 ± 2.4 (7.5 ~ 16.0)	4.5 ± 0.9 (3.0 ~ 6.0)	7.2 ± 1.8 (4.5 ~ 10.0)	6:59 ± 1:13 (5:37 ~ 10:43)	609 ± 314 (205 ~ 1171)
CLP-L (n=9)	6.0 ± 1.4 (3.5 ~ 8.5)	6.0 ± 1.4 (3.5 ~ 8.5)	–	3:42 ± 0:27 (3:14 ~ 4:07)	613 ± 332 (200 ~ 1334)
Cont-S (n=18)	6.3 ± 1.8 (4.5 ~ 13.5)	–	6.3 ± 1.8 (4.5 ~ 13.5)	3:57 ± 0:52 (2:58 ~ 6:28)	152 ± 94 (30 ~ 400)
Cont-T (n=7)	12.5 ± 5.0 (5.3 ~ 19.0)	3.9 ± 1.3 (2.3 ~ 5.0)	7.4 ± 4.0 (2.3 ~ 14.0)	6:36 ± 1:58 (6:04 ~ 9:05)	756 ± 320 (389 ~ 1100)

#, ##; significant difference between each group (ANOVA, #: p<0.05, ##: p<0.01)

mean ± S.D. (min ~ max)

Figure 3. *Information of four operation group.*

The operation times were 3:57~4:11 in the SSRO groups (CLP-S and Cont-S) and 6:36~6:59 in the two-jaw surgery groups (CLP-T and Cont-T). Naturally, the operation times for the two-jaw groups tended to be longer than for the SSRO groups. On the other hand, there were no significant differences in the operation times between CLP-S and Cont-S, as well as CLP-T and Cont-T.

The levels of blood loss were 152~207 mL in the SSRO groups, and 609~756 mL in the two-jaw groups that underwent maxillary advancement operations (CLP-T and Cont-T). The blood loss in the SSRO groups was significantly less than that in the groups that underwent maxillary advancement operations (p<0.01).

Cephalometric analysis of skeletal landmarks in each stage

Changes in the positions of the skeletal landmarks on lateral cephalograms were measured in T1, T2, and T3. The measurement values and statistical results for CLP-S, CLP-T, CLP-L, Cont-S, and Cont-T are shown in *Figures 4-6*.

CLP-S		T1			T2			T3			2)	3)			movement			relapse			4)
		mean	SD	1)	mean	SD	1)	mean	SD	1)		T1-2	T1-3	T2-3	mean	SD	1)	mean	SD	1)	
X	A	-16.4	± 4.7	**	-16.5	± 4.6	**	-16.6	± 4.8	**	-	-	-	-	0.1	± 0.5	-	-0.1	± 0.5	-	-
	U1	-17.5	± 7.7	**	-16.6	± 7.7	**	-16.0	± 7.8	**	#	-	-	#	-0.9	± 1.5	-	0.5	± 1.2	-	*
	L1	-12.6	± 7.6	**	-20.1	± 6.7	**	-18.9	± 7.1	**	##	##	##	-	7.5	± 2.2	-	1.1	± 1.6	-	**
	B	-22.1	± 7.8	**	-30.1	± 6.7	**	-28.6	± 7.5	**	##	##	##	-	8.0	± 2.8	-	1.5	± 1.8	-	**
	Pog	-23.8	± 9.6	*	-32.5	± 7.7	**	-30.4	± 8.9	**	##	##	##	-	8.7	± 4.0	-	2.1	± 2.3	-	**
	Me	-31.6	± 9.5	*	-40.1	± 7.5	**	-38.1	± 8.7	*	##	##	##	-	8.6	± 4.4	-	2.0	± 2.5	-	**
Y	A	-60.5	± 5.1	-	-60.4	± 5.0	-	-60.5	± 5.1	-	-	-	-	-	-0.1	± 0.2	-	-0.1	± 0.3	-	-
	U1	-80.3	± 5.6	**	-80.6	± 5.5	**	-80.3	± 5.4	**	-	-	-	-	0.4	± 0.7	-	0.3	± 0.7	-	-
	L1	-80.1	± 5.2	**	-79.2	± 5.4	*	-78.9	± 5.3	*	-	-	-	-	-0.9	± 2.6	*	0.3	± 1.4	-	*
	B	-100.6	± 6.0	**	-100.0	± 6.3	-	-99.4	± 6.1	-	-	-	-	-	-0.6	± 1.6	**	0.6	± 1.4	-	*
	Pog	-117.8	± 8.1	*	-117.3	± 8.2	-	-117.1	± 8.2	-	-	-	-	-	-0.5	± 1.8	**	0.2	± 1.3	-	-
	Me	-123.3	± 8.5	-	-122.8	± 8.5	-	-122.2	± 8.3	-	-	-	-	-	-0.5	± 1.4	**	0.5	± 1.0	-	-

Cont-S		T1		T2		T3		2)	3)			movement		relapse		4)
		mean	SD	mean	SD	mean	SD		T1-2	T1-3	T2-3	mean	SD	mean	SD	
X	A	-10.3	± 2.9	-10.4	± 2.8	-10.4	± 2.8	-	-	-	-	0.1	± 0.3	0.1	± 0.2	
	U1	-6.8	± 3.9	-6.6	± 3.8	-6.5	± 3.3	-	-	-	-	-0.2	± 1.0	0.0	± 1.2	-
	L1	-2.1	± 3.8	-10.6	± 3.3	-9.8	± 3.4	##	##	##	-	8.5	± 2.8	0.8	± 0.9	**
	B	-13.1	± 5.5	-21.6	± 5.6	-20.3	± 5.3	##	##	##	-	8.5	± 3.3	1.3	± 0.8	**
	Pog	-14.8	± 7.8	-23.3	± 8.0	-21.5	± 7.6	##	##	#	-	8.5	± 4.0	1.8	± 0.9	**
	Me	-23.0	± 8.4	-31.4	± 8.3	-29.5	± 8.0	##	##	-	-	8.4	± 4.1	1.9	± 1.1	**
Y	A	-62.7	± 3.1	-62.7	± 3.1	-62.8	± 3.1	-	-	-	-	0.0	± 0.2	0.0	± 0.2	-
	U1	-86.2	± 4.5	-86.6	± 4.5	-86.1	± 4.8	-	-	-	-	0.4	± 0.6	0.5	± 0.8	-
	L1	-87.2	± 4.8	-84.0	± 4.3	-83.5	± 4.3	#	-	#	-	-3.2	± 1.9	0.6	± 1.3	**
	B	-107.2	± 6.1	-104.8	± 6.5	-104.3	± 6.9	-	-	-	-	-2.5	± 1.7	0.4	± 1.0	**
	Pog	-123.6	± 6.4	-121.2	± 6.6	-120.6	± 7.2	-	-	-	-	-2.4	± 1.6	0.6	± 1.1	**
	Me	-127.9	± 6.7	-125.7	± 6.6	-125.3	± 7.2	-	-	-	-	-2.3	± 1.3	0.3	± 0.8	**

1) Comparison between CLP-S and Cont-S by t-test (*; P,0.05, **; P<0.01)

2) Comparison of intragroup by ANOVA (#; P,0.05, ##; P<0.01)

3) Multiple comparison test among T1,T2 and T3 (#; P,0.05, ##; P<0.01)

4) Comparison between movement and relapse distance by t-test (*; P,0.05, **; P<0.01)

Figure 4. *Change of measurement points on cephalogram (CLP-S & Cont-S).*

CLP-T		T1			T2			T3			2)	3)			movement			relapse			4)
		mean	SD	1)	mean	SD	1)	mean	SD	1)		T1-2	T1-3	T2-3	mean	SD	1)	mean	SD	1)	
X	A	-19.6	± 4.8	-	-15.5	± 4.9	-	-15.7	± 5.0	-	-	-	-	-	-4.0	± 1.3	-	-0.2	± 0.6	-	**
	U1	-20.8	± 5.7	**	-17.3	± 6.2	*	-16.5	± 6.4	*	-	-	-	-	-3.6	± 2.0	-	0.7	± 1.0	-	**
	L1	-11.8	± 6.4	-	-20.9	± 6.0	*	-19.6	± 6.3	*	##	##	##	-	9.1	± 2.0	-	1.3	± 1.7	-	**
	B	-20.9	± 7.4	-	-31.0	± 6.7	*	-28.7	± 7.0	-	##	##	#	-	10.1	± 1.9	-	2.2	± 1.3	*	**
	Pog	-22.3	± 8.7	-	-33.2	± 8.0	-	-30.3	± 8.6	-	##	##	#	-	10.9	± 2.2	-	2.9	± 2.0	-	**
	Me	-31.2	± 8.7	-	-42.5	± 8.1	*	-39.3	± 8.8	-	##	##	#	-	11.3	± 2.4	-	3.3	± 2.3	-	**
Y	A	-63.3	± 4.6	**	-65.9	± 4.9	-	-65.5	± 4.9	-	-	-	-	-	2.6	± 1.5	**	0.4	± 0.7	-	**
	U1	-80.7	± 5.8	**	-83.3	± 5.1	**	-82.6	± 5.5	**	-	-	-	-	2.6	± 1.4	**	0.7	± 1.0	-	**
	L1	-81.4	± 6.0	**	-81.8	± 5.2	**	-81.4	± 5.4	**	-	-	-	-	0.3	± 2.8	**	0.3	± 0.7	-	-
	B	-102.2	± 6.8	**	-102.5	± 6.0	**	-101.5	± 6.3	**	-	-	-	-	0.3	± 2.0	**	0.9	± 1.2	-	-
	Pog	-121.1	± 8.1	**	-121.2	± 6.6	*	-120.3	± 7.0	*	-	-	-	-	0.0	± 2.4	**	0.9	± 1.1	-	-
	Me	-126.0	± 7.7	**	-125.8	± 6.9	**	-125.3	± 7.0	**	-	-	-	-	-0.2	± 1.9	**	0.5	± 0.9	-	-

Cont-T		T1		T2		T3		2)	3)			movement		relapse		4)
		mean	SD	mean	SD	mean	SD		T1-2	T1-3	T2-3	mean	SD	mean	SD	
X	A	-15.3	± 6.0	-11.0	± 6.0	-11.4	± 6.3	##	##	##	-	-5.5	± 2.5	-0.7	± 0.7	**
	U1	-12.1	± 8.0	-7.5	± 10.0	-7.7	± 10.3	##	##	##	-	-5.0	± 3.1	-0.1	± 1.3	*
	L1	-4.9	± 11.1	-12.4	± 9.2	-11.8	± 9.1	##	##	##	-	8.4	± 5.3	1.0	± 1.2	**
	B	-14.5	± 13.7	-22.1	± 12.9	-21.2	± 13.0	##	##	##	-	8.6	± 6.2	1.0	± 0.7	**
	Pog	-15.2	± 15.9	-22.9	± 15.2	-21.4	± 15.4	##	##	#	-	8.8	± 7.2	1.6	± 0.9	*
	Me	-23.8	± 16.5	-31.3	± 15.7	-29.8	± 16.3	#	#	-	-	8.4	± 7.8	1.6	± 1.3	*
Y	A	-70.4	± 5.9	-69.5	± 8.1	-69.0	± 8.1	-	-	-	-	-0.8	± 3.3	0.5	± 0.8	-
	U1	-95.5	± 6.4	-94.7	± 8.8	-94.1	± 8.8	-	-	-	-	-0.8	± 3.7	0.5	± 0.7	-
	L1	-95.4	± 8.7	-90.9	± 9.3	-90.8	± 9.7	##	##	##	-	-4.4	± 3.2	0.1	± 0.8	**
	B	-116.9	± 9.2	-113.4	± 10.4	-112.9	± 10.8	##	##	##	-	-3.5	± 2.3	0.5	± 0.9	**
	Pog	-136.3	± 12.0	-132.7	± 12.9	-132.3	± 13.4	##	##	##	-	-3.6	± 2.1	0.4	± 0.8	**
	Me	-141.9	± 11.4	-138.2	± 12.4	-138.0	± 12.9	##	##	##	-	-3.6	± 1.7	0.2	± 0.9	**

1) Comparison between CLP-T and Cont-T by t-test (*; P,0.05, **; P<0.01)

2) Comparison of intragroup by ANOVA (#; P,0.05, ##; P<0.01)

3) Multiple comparison test among T1,T2 and T3 (#; P,0.05, ##; P<0.01)

4) Comparison between movement and relapse distance by t-test (*; P,0.05, **; P<0.01)

Figure 5. *Change of measurement points on cephalogram (CLP-T & Cont-T).*

CLP-LF		T1		T2		T3		2)	3)			movement			relapse		4)
		mean	SD	mean	SD	mean	SD		T1-2	T1-3	T2-3	mean	SD		mean	SD	
X	A	-19.5	± 3.0	-14.8	± 3.7	-15.5	± 3.4	##	##	##	-	-4.7	± 1.7	-	-0.7	± 0.8	**
	U1	-20.3	± 4.9	-15.9	± 5.7	-16.0	± 5.9	##	##	##	-	-4.4	± 2.6	-	-0.2	± 0.8	**
	L1	-15.5	± 6.1	-18.8	± 6.0	-18.7	± 6.0	##	##	##	-	3.3	± 1.2	-	0.0	± 1.0	**
	B	-26.8	± 5.2	-30.0	± 5.3	-28.3	± 4.3	##	##	#	#	3.1	± 1.3	-	1.6	± 1.6	*
	Pog	-29.9	± 5.2	-33.5	± 6.0	-31.3	± 4.6	##	##	-	#	3.7	± 1.6	-	2.3	± 2.4	-
	Me	-38.5	± 6.2	-42.2	± 6.3	-40.2	± 5.6	##	##	-	-	3.7	± 1.5	-	2.0	± 2.7	-
Y	A	-63.8	± 4.4	-66.7	± 4.5	-66.3	± 4.2	##	##	-	-	2.8	± 1.5	-	0.4	± 0.9	**
	U1	-80.3	± 4.8	-83.0	± 5.0	-82.6	± 4.3	##	##	##	-	2.8	± 1.9	-	0.5	± 1.1	**
	L1	-80.5	± 5.0	-81.4	± 4.2	-80.6	± 4.0	-	-	-	-	0.9	± 1.4	-	0.8	± 1.2	-
	B	-99.4	± 6.0	-100.5	± 5.9	-99.8	± 5.5	-	-	-	-	1.1	± 0.9	-	0.6	± 1.4	-
	Pog	-116.8	± 6.7	-117.8	± 6.6	-117.2	± 6.3	-	-	-	-	0.9	± 0.8	-	0.5	± 1.4	-
	Me	-121.3	± 6.9	-122.5	± 6.8	-122.0	± 6.6	#	##	-	-	1.2	± 1.0	-	0.5	± 0.6	-

2) Comparison of intragroup by ANOVA (#; P,0.05, ##; P<0.01)

3) Multiple comparison test among T1,T2 and T3 (#; P,0.05, ##; P<0.01)

4) Comparison between movement and relapse distance by t-test (*; P,0.05, **; P<0.01)

Figure 6. *Change of measurement points on cephalogram (CLP-L).*

In CLP-S, all measurement points positioned posteriorly significantly matched with Cont-S, as a result of the shortness of x values. Postoperatively, x values of movement and relapse distances were not different between CLP-S and Cont-S; y values of movement distances in CLP-S were smaller than those in Cont-S.

On the x-axis, the movement distance of point B was 8.0 mm, and the relapse distance was 1.5 mm. The rate of relapse was 18.8%.

On the x-axis of CLP-T and Cont-T, which underwent two-jaw surgery, all measurement points of CLP-T were located more posterior than those of Cont-T. Point A was located significantly more posteriorly at the time of T1, T2, and T3 in CLP-T. On the x-axis, the movement distance of point A was 4.0 mm and the relapse distance was 0.2 mm. The movement distance of Point B was 10.1 mm and the relapse distance was 2.2 mm. On the y-axis, point A of CLP-T was located more superior than that of Cont-T before operation and moved inferiorly after the operation. On the y-axis, point A of CLP-T was located more superiorly than that of Cont-T before the operation. Postoperatively, point A of CLP-T moved downward and was not significantly different from Cont-T. Point A of CLP-L moved 4.7 mm forward and 2.8 mm downward at operation, and relapse distances were 0.7 mm and 0.4 mm, respectively.

Before surgery, the y-coordinates of points A, U1, L1, B, Pog, and Me were greater in the control group than in the other groups, demonstrating that the height of the maxilla of CLP subjects was shorter than in non-cleft subjects.

Changes of two angles (SNA and SNB) in the five groups are shown in *Figure 7*. Postoperatively, SNA was greater in CLP-T, CLP-L, and Cont-T, and SNB tended to be smaller in all groups.

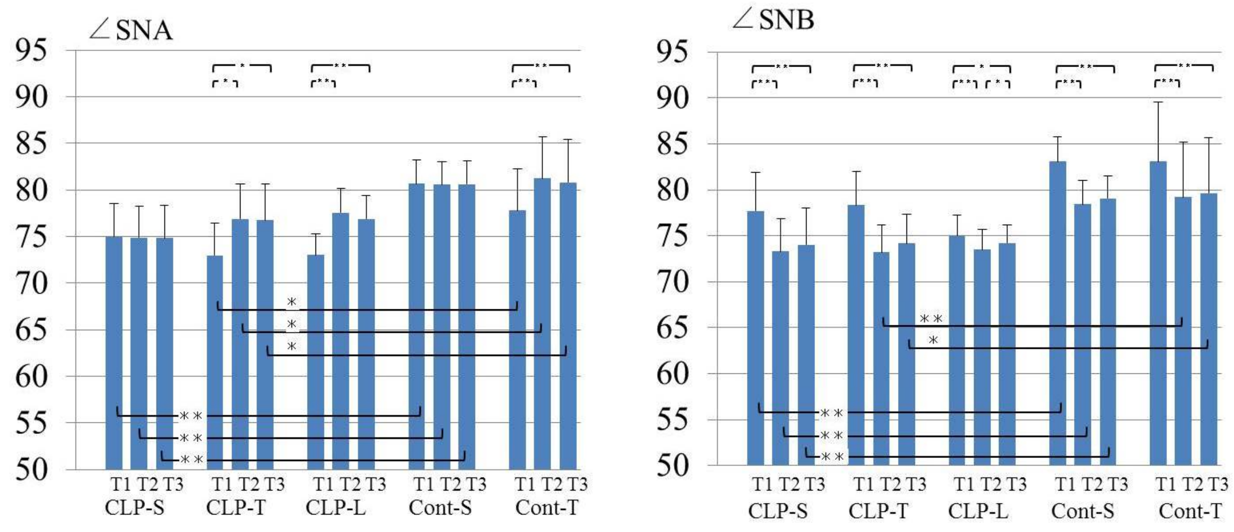

Figure 7. *Change of ∠SNA and ∠SNB.*

Pre- and postoperative changes of ∠ANB are shown in *Figure 8*. The findings demonstrate that the total movement of the maxilla and mandible tended to be greater in the two-jaw surgery groups. The changes between T2 and T3 postoperatively were used to represent the amount of relapse. Pre- and postoperative changes of ∠ANB (T2-T1) were significantly greater in the CLP-T group than that in the CLP-S, CLP-L, and Cont-S groups (P<0.01). The mean relapses of

∠ANB (T2-T3) were 0.7 ± 0.8 degrees in CLP-S, 1.1 ± 0.6 degrees in CLP-T, 1.3 ± 0.7 degrees in CLP-L, 0.5 ± 0.5 degrees in Cont-S, and 0.8 ± 0.9 degrees in Cont-T.

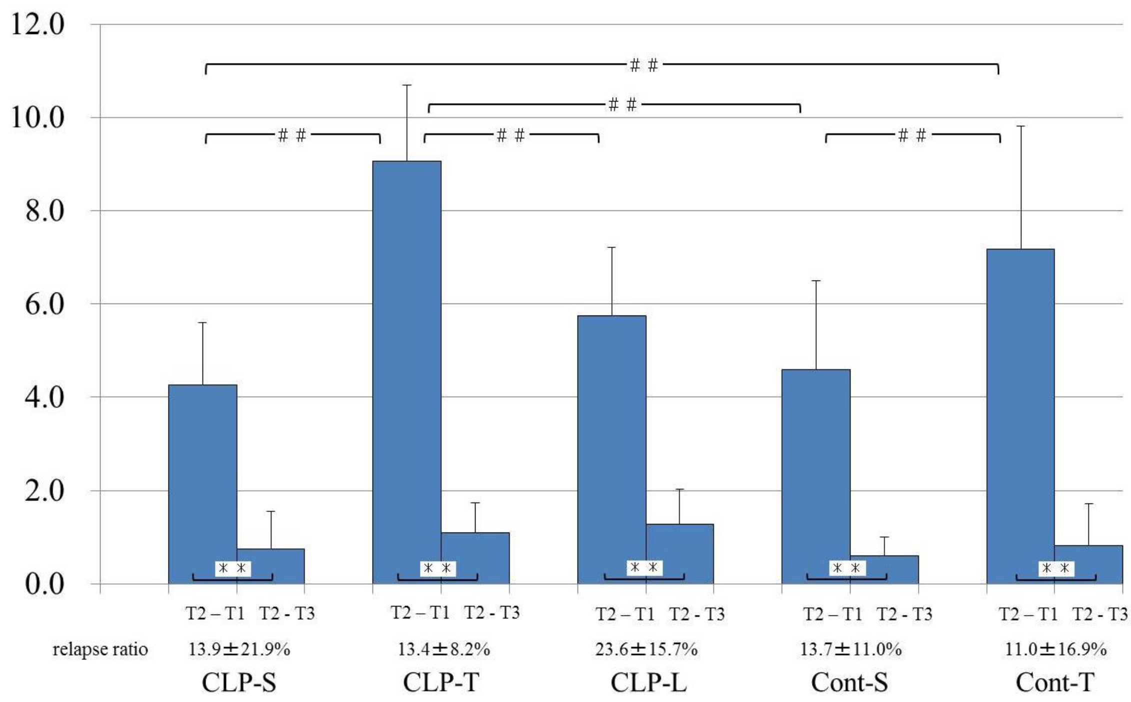

Figure 8. *Movement and relapse distances of ANB.*

The relapse amounts of ANB in CLP-T and CLP-L, which included maxillary advancement, tended to be greater than that in other groups. However, there were no significant differences in the relapse amount of ANB among the 5 groups. Analysis by t-test revealed that T2-T1 was significantly greater in T2-T3. The relapse ratios of ANB were 16.3% in CLP-S, 12.1% in CLP-T, 22.8% in CLP-L, 10.6% in Cont-S and 11.1% in Cont-T. There were no differences in the relapse ratio between cleft and non-cleft subjects.

Intra and postoperative complications and changes in velopharyngeal function

VP closure functions at the preoperative stage were assessed as poor in three cleft lip patients. One month postoperatively, poor VP closure was observed in 4 of the cleft lip patients. Transient VPI was noted after surgery in one patient, but it was improved by speech therapy after 6 months. Two patients with poor VP closure function before surgery were treated with pharyngeal flap operation at over 6 months after orthognathic surgery. VP closure functions of the patients in non-cleft groups were assessed as good throughout all stages of the treatment.

In terms of complications, postoperative gingival necrosis in the upper anterior tooth region was noted in 1 patient in CLP-T; no other complications, such as bone necrosis or bone loss, were noted in any group.

Discussion

In patients with CLP, malalignment of the upper dentition frequently occurs due to the surgical stress of palatoplasty, narrowing of the upper jaw by postoperative scar tissue present in the palatal mucosa, as well as the presence of alveolar cleft [4,14]. For these patients, it is difficult to acquire favorable occlusion. Moreover, latent hypo-growth of the maxilla is present in many patients [8], thus orthognathic surgery is needed when the acquisition of a favorable overlap is difficult by ordinal orthodontic treatment. Previous reports state that more than 25% of patients with CLP require surgical intervention and maxillary osteotomy [10]. Cheung et al. [9] reported in a meta-analysis of 72 reports involving 1,418 cleft palate patients that Le Fort I osteotomy was performed in 83.6% and mandibular movement was concomitantly performed in 24.4% of patients with cleft palate. This suggests that, in the great majority of cases, the surgeons approach the maxillary bone. However, there are serious problems relating to orthognathic surgery in patients with CLP that differ from those in patients with general skeletal reverse occlusion [6,10,13,17]. Palatal scar tissue may limit maxillary advancement, cause relapse, change the external nose morphology, and cause velopharyngeal insufficiency after surgery [12]. Thus, it is necessary to select the timing of treatment and a surgical procedure for each patient in consideration of several morphological and functional problems [9,18].

Our department has performed primary cleft palate surgery in 410 cases without cleft lip, and cleft lip and alveolus during the same period (35 years) of this series; orthognathic surgery was performed in 46 patients (40/410, 9.8%). This rate seems to be relatively low when compared with the rates of other previous reports [8]. In Kagoshima University Hospital, we have an integrated team for CLP, consisting of oral and maxillofacial surgeons, orthodontists, pedodontists, speech therapists, and nurses. Generally, palate repair is performed employing the modified pushback method preserving the periosteal tissue in the anterior part of the palate at approximately 1 and a half years old, and management of occlusion by orthodontists starts at 4 years of age. In the deciduous tooth period, a maxillary protraction appliance (MPA) is utilized for patients with marked maxillary growth disturbance, and after expansion of the upper alveolar arch, autologous iliac cancellous bone and marrow transplantation in the alveolar cleft is performed at 8-11 years old before eruption of the canines. In this series, bone transplantation to the alveolar cleft region was performed in patients treated with maxillary movement in order to move the upper jaw as one segment, except in three patients who underwent orthognathic surgery before bone graft treatment on the alveolar cleft. Orthognathic surgery is then performed after the completion of facial growth, if necessary. The low rate of orthognathic surgery in our department may be due to active interventions, such as the long use of MPA in the period of growth, by the orthodontists for the occlusal management of patients with CLP.

The results of the present study on orthognathic surgery comparing the patients with/without CLP suggest the following points. First, regarding the amount of jaw movement, operation time, or blood loss in orthognathic surgery, no significant differences were observed when compared with the patients with same operation method without clefts. According to reports by Cheung et al. the mean amount of maxillary movement in previous studies varied from 3.9 mm to 7.2 mm. These results are not significantly different from this series, with the mean amount of maxillary movement varying from 3.0 mm to 8.5 mm. Operation time was slightly longer than in previous reports in all groups; however, there were no differences in the same operation method between the cleft and non-cleft patients. The reasons why the operation time of orthognathic surgery in our department was long were thought to be related to the surgical procedures for mandibular setback by SSRO, with repositioning of the medial segment, and to the bone harvesting and graft in the gap at the maxilla and pterygoid process upon maxillary advancement. The blood loss associated with orthognathic surgery, including maxillary advancement, was greater than that in mandibular retropositioning alone. However, there was no significant difference in terms of the blood loss between the patients with/without CLP. Blood loss over 1,000 mL was recognized in one case each in CLP-T, CLP-L, and Cont-T. It was thought that such blood loss was caused by injury of the maxillary artery or its branches, but these cases occurred only at an early stage. Recently, a special team for orthognathic surgery was organized in our department, thereby shortening the operation time and reducing intraoperative bleeding.

The second point is that the relapse of patients with clefts was not significantly different and the level of stability was the same as in patients without clefts, and pre- and postoperative cephalometric analyses revealed that two-jaw surgery provided significant movement of the maxilla and mandible in both groups with/without CLP. Before surgery, the Y-coordinates of points A, U1, L1, B, Pog, and Me were significantly greater in the Cont-T group than those in the other groups ($p<0.05$ each), demonstrating that the height of the maxilla in CLP subjects was less than in non-cleft subjects.

In this study, the changes of ∠ANB in CLP-T were significantly greater than those in the other groups. However, relapse of ∠ANB was not significant and an adequate lateral profile was obtained in all groups. Furthermore, the present results of the relapse ratios of ∠ANB varying from 13.4% to 23.6% in CLP-T and CLP-L, respectively, were thought to be favorable, when compared with previous reports describing relapse rates varying from 16.7% to 50% [9,10,12,14]. It was stated in previous reports that the desirable distance of maxillary advancement is approximately 10 mm, but Loren et al. reported in 1992 that the risk of relapse increased when the distance was 4 mm or greater. Our department has developed a protocol to minimize postoperative relapse, as mentioned previously, in which orthodontic treatment is ensured before surgery, aimed at stabilizing occlusion after surgery. The maxillary mucosa is sufficiently dissected to the posterior region to separate scar and bone, and a bone graft prepared from excessive bone produced by mandibular setback by SSRO or another region is inserted into the bone gap to prevent relapse. Preoperative cephalometric analysis revealed that the maxillary height between N and the point of the upper central incisor (U1) was significantly shorter, and point A was retropositioned in the groups with CLP (CLP-S, CLP-T, and CLP-L) compared with those in the control groups. These findings may have been caused by inherent hypo-growth of the maxilla in not only the anteroposterior but also the vertical direction in patients with CLP, and/or the counterclockwise rotation of the mandible by orthodontic treatment, including MPA. It can be considered that the favorable stability following the orthodontic surgeries in the present study was provided by the combination of presurgical orthodontic treatment and surgical procedures.

For the treatment of patients with relapse, adequate occlusion was obtained by orthodontic treatment in all patients except one in the CLP-T group. In this patient, re-operation by SSRO was performed and occlusion improved uneventfully. In terms of complications, deterioration of VP closure function was observed after surgery in only one patient with BCLP, who received two-jaw surgery. The maxillary movement of this patient was 4.0 mm. VP closure function of this patient was improved by postoperative speech therapy (*Figure 9*). When we decide on the surgical procedure, we hold conferences with speech therapists, orthodontists, and oral and maxillofacial surgeons preoperatively, and discuss whether the patient is likely to develop VPI. In all patients with CLP, we perform velopharyngeal fiberscopy and lateral cephalometric analysis preoperatively at rest, and during the pronunciation of /i/ and blowing. Based on the results of these preoperative assessments, we explained the possibility of postoperative VPI to the patient and his/her family. In previous studies, pharyngeal flap closure was performed after maxillary advancement in CLP patients [10,15,17].

	Velopharyngeal closure					
	T1		T2		T3	
Group	good	poor	good	poor	good	poor
CLP-S (n=11)	10	1	10	1	10	1
CLP-T (n=14)	13	1	12	2	13	1
CLP-L (n=9)	8	1	8	1	8	1
Cont-S (n=18)	18	0	18	0	18	0
Cont-T (n=7)	7	0	7	0	7	0

$\chi 2$ test; P=0.35

Figure 9. *Table of velopharyngeal closure.*

In our study, pharyngeal flap closure was performed in 2 patients with persistent VPI after orthognathic surgery, and the recovery of good VP closure function was achieved. After Le Fort I osteotomy, the palatal mucosa was detached during surgery in one patient, and mucosal necrosis of the upper anterior tooth region was observed after surgery in one patient. Both complications were improved by postoperative cleaning and hyperbaric oxygen therapy.

There are several limitations of this study, as follows: 1) the number of subjects in each group was not large because this study was performed at a single institution, 2) there was more than one operator, and 3) there was a relatively short postoperative follow-up period of up to 6 months. Nonetheless, postoperative occlusion of cleft lip and palate patients was improved, achieving normal overbite and overjet; patients were satisfied with their occlusion and facial appearance. However, shortening and retrogression of the midface and counterclockwise rotation in the lower part of the face remained in the patients with CLP when compared with those without clefts. Further study on the causes and functional effects of these facial characteristics of patients with CLP in our department will therefore be necessary.

Conclusion

Our orthognathic surgeries provided the same levels of stability and intra- or postoperative complications for patients with skeletal class III with/without cleft lip and palate.

References

1. Harrington C, Gallagher JR, Borzabadi-Farahani A. A retrospective analysis of dentofacial deformities and orthognathic surgeries using the index of orthognathic functional treatment need (IOFTN). *International Journal of Pediatric Otorhinolaryngology.* 2015; **79**: 1063-1066.

2. Borzabadi-Farahani A, Eslamipour F, Shahmoradi M. Functional needs of subjects with dentofacial deformities: A study using the index of orthognathic functional treatment need (IOFTN). *Journal of Plastic, Reconstructive & Aesthetic Surgery.* 2016; **69**: 796-801.

3. Kufner J. Four-year experience with major maxillary osteotomy for retrusion. *Journal of Oral Surgery.* 1971; **29**: 549-533.

4. Epker BN, Wolford LM. Middle-third facial osteotomies:their use in the correction of congenital dentofacial and craniofacial deformities. *Journal of Oral Surgery.* 1976; **34**: 324-342.

5. Drommer R, Luhr HG. The stabilization of osteomized maxillary segments with Luhr miniplates in secondary cleft surgery. *Journal of Oral and Maxillofacial Surgery.* 1981; **9**: 166-169.

6. Loren BE, Schendel SA. An analysis of Le Fort 1 Maxillary advancement in cleft lip and palate patients. *Plastic and Reconstructive Surgery.* 1992; **90**: 779-786.

7. Hochban W, Ganss C, Austermann KH. Long-term results after maxillary advancement in patients with clefts. *The Cleft Palate-Craniofacial Journal.* 1993; **30**: 237-243.

8. Hirano T, Suzuki H. Factors related to relapse after Le Fort I Maxillary advancement osteotomy in patients with cleft lip and palate. *The Cleft Palate-Craniofacial Journal.* 2001; **38**: 1-10.

9. Chung LK, Chua HDP. A meta-analysis of cleft maxillary osteotomy and distraction osteogenesis. *International Journal of Oral and Maxillofacial Surgery.* 2006; **35**: 14-24.

10. Saltaji H, Major M, Alfakir H, Al-Saleh M, Flores-Mir C. Maxillary advancement with conventional orthographic surgery in patients with cleft lip and palate: Is it a stable technique? *Journal of Oral and Maxillofacial Surgery.* 2012; **70**: 2859-2866.

11. Janulewicz J, Buckley M, Ford M, Gassner R. The effects of Le Fort osteotomies on velopharyngeal and speech functions in cleft patients. *Journal of Oral and Maxillofacial Surgery.* 2004; **62**: 308-314.

12. Smedberg E, Neovius E, Lohmander A. Impact of maxillary advancement on speech and velopharyngeal function in patients with cleft lip and palate. *The Cleft Palate-Craniofacial Journal.* 2014; **51**: 334-343.

13. Pereira V, Shell D, Tuomainen J. The impact of osteotomy on speech outcomes in cleft lip and palate: An evidence-based approach to evaluating the literature. *The Cleft Palate-Craniofacial Journal.* 2013; **50**: 25-39.

14. Figueroa AA, Polly JW, Friede H, Ko EW. Long-term skeletal stability after maxillary advancement with distraction osteogenesis using a rigid external distraction device in cleft maxillary deformities. *Plastic and Reconstructive Surgery.* 2004; **114**: 1382-1392.

15. Posnick JC, Ewing MP. Skeletal stability after Le Fort maxillary advancement in patients with unilateral cleft lip and palate. *Plastic and Reconstructive Surgery.* 1990; **85**: 706.

16. Posnick JC, Dagys AP. Skeltal stability and relapse patterns after Le Fort maxillary osteotomy fixed with miniplates: The unilateral cleft lip and palate deformity. *Plastic and Reconstructive Surgery.* 1994; **94**: 924-932.

17. Kumar A, Gabbay JS, Nikjoo R. Improved outcomes in cleft patients with severe maxillary deficiency after Le Fort I internal distraction. *Plastic and Reconstructive Surgery.* 2006; **117**: 1499-1508.

18. Okawachi T, Nozoe E, Nishihara K, Nakamura N. 3-Dimensional analysis of outcomes following secondary trestment of unilateral cleft lip nose deformity. *Journal of Oral and Maxillofacial Surgery.* 2010; **69**: 322-332.