

Evaluation of the Maxillary Morphological Changes Following Distraction Osteogenesis in Patients with Repaired Cleft Palate Using Three-dimensional CT Images

Junko Nagata^a, Koichi Satoh^b,
Sumio Sakoda^c, Ryosuke Shiba^d,

^aDepartment of Orthodontics, Kagoshima University Graduate School of Medical and Dental Science, Kagoshima,

^bDepartment of Oral and Maxillofacial Surgery, Kyushu Dental College, Kitakyushu,

^cDepartment of Oral and Maxillofacial Surgery, University of Miyazaki, Miyazaki,

^dProfessor Emeritus, University of Miyazaki, Miyazaki.

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Abstract

The purpose of this study was to document the morphological changes of maxilla following distraction osteogenesis (DOG) using three-dimensional CT images. The maxillary distraction using a face mask and/or an expansion screw was applied in nine cleft palate patients aged from 9 to 11 years. Cephalograms, photographs, and three-dimensional CT images before and three months after distraction were evaluated. Cephalometric analysis revealed that the estimated advancement and lateral expansion were achieved, and photographs showed the improvement of facial profile and occlusion. Superimpositions of pretreatment and posttreatment tracings of three-dimensional CT images facilitated to visualize the maxillary changes with distraction. It is confirmed that the whole maxilla moved forwards with a counterclockwise rotation, the maxillary lateral segments were moved forwards and outwards, and posterior elongation of the maxillary tuberosity, followed by the backward migration of the maxillary second molar.

From these results, it is suggested that advantages of the maxillary distraction osteogenesis during growing period include not only improvement in facial appearance and occlusion but provision of an osseous environment to permit spontaneous eruption of the second maxillary molar into the elongated area.

Key words; Distraction osteogenesis, Cleft lip and palate, Three-dimension, CT images, Maxilla

Introduction

Severe maxillary hypoplasia accompanied by midface retrusion and anterior and/or lateral crossbite were observed in a significant number of patients with repaired cleft palate^{1,3}. Orthodontic maxillary protraction, however, does little to restore the maxillofacial relations^{4,8}. Moreover, advancement of the maxilla by LeFort I osteotomy in those patients is often difficult and

has a greater tendency to relapse, because of the scarring from previous operations^{9,11}. Since the feasibility of distraction osteogenesis (DOG) in the membranous bone of the maxilla and midface has been demonstrated^{12,13}, DOG has rapidly developed for managing maxillofacial deformities as an alternative procedure to conventional orthodontic surgery.

Until now, several articles have been published presenting successful results of maxillary DOG using

Corresponding author: Dr. Koichi Satoh, D.D.S., Ph.D.

Address: Department of Oral and Maxillofacial Surgery,

Kyushu Dental College, 2-6-1 Manazuru,

Kokurakita-ku, Kitakyushu, 803-8580, Japan.

E-mail: satoh@ku-dent.ac.jp.

various types of distraction devices in patients with repaired cleft palate¹⁴⁻¹⁸⁾. Although those patients present with maxillary hypoplasia in vertical, horizontal, and transverse dimensions, there have been few articles on three-dimensional evaluation of maxillary changes following distraction. The purpose of this study was to evaluate the changes in the maxilla itself following distraction using three-dimensional CT images in addition to photographic and cephalometric evaluation. A key distinguishing feature of the present study is its approach towards evaluating them by superimpositions of pretreatment and posttreatment CT images with three-dimensional reconstruction.

Subjects and methods

Subjects

The subjects consisted of nine patients aged from 9 to 11 years, five with repaired unilateral and four with repaired bilateral cleft lip and palate, in whom the primary lip and palate repairs were performed during infancy or early childhood at several hospitals in Miyazaki Prefecture, Japan. Although attempts were made to treat their midfacial retrusions and anterior and/or lateral crossbites through the use of the maxillary protracting appliance and/or the lateral expansion appliance, their responses fell short of expectations. Therefore, after obtaining informed consent from all patients, the maxillary distraction osteogenesis was selected as an alternative method. They were classified into three groups according to the type of maxillary

Table 1. Number and Cleft Type of Subjects

Group	UCLP	BCLP	Total
A	1	1	2
B	4	0	4
C	0	3	3

UCLP: unilateral cleft lip and palate
BCLP: bilateral cleft lip and palate

hypoplasia: Group A (n=2); anteroposterior hypoplasia, Group B (n=4); anteroposterior and lateral hypoplasia, and Group C (n=3); anteroposterior and lateral hypoplasia with the premaxilla in a normal position (Table 1). All patients and their parents or conservators granted the distribution or publication of their photographs as a voluntary contribution in the interest of public education.

Treatment

The treatment procedure in the present study was varied depending on requirements of each Group (Fig.1). The lingual arch appliance with vestibular hooks was placed in the maxillary dental arch in Group A, while the expansion screw with vestibular hooks was placed in Groups B and C prior to surgery (Fig.2). The high LeFort I osteotomy across the whole maxilla in Groups A and B and on both maxillary lateral segments except the premaxilla in Group C was performed, and the maxilla was down-fractured softly to ensure its mobility. After a 5-day latency period, the advancement was performed with elastics linked from a face mask to vestibular hooks with force of 900 g all day, which proceeded over the following 3 to 7 weeks in all Groups. The direction of advancement

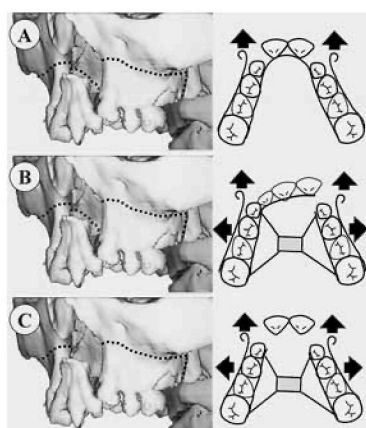


Fig. 1. Design of maxillary distraction in each Group. Broken lines indicate the site of osteotomy and arrows indicate the directions of advancement and lateral expansion.



Fig. 2. Intraoral appliances in place prior to surgery. A lingual arch in Group A (*top*) and an expansion screw in Groups B and C (*bottom*).



Fig. 3. Advancement of the hypoplastic maxilla with a face mask and elastics.

was parallel to the occlusal plane, and the elastics were renewed once per day (Fig. 3). Simultaneous lateral expansion was started at a rate of 0.67 mm per day and proceeded over the following 11 to 18 days in Groups B and C. When the estimated advancements were achieved, the amount of force was reduced to 300 g for an additional 3 months to maintain the maxilla in the new position, and when the estimated lateral expansions were achieved, the intraoral appliance was left in place as a retainer for a further 3 months and replaced by the palatal bar. Approximately at 3 to 6 months after distraction, alveolar bone grafting was performed in all subjects. The palatal bar was removed 6 months after the alveolar bone grafting.

Evaluation

Facial and intraoral photographs, lateral and posteroanterior (P-A) cephalograms, and standardized CT images taken before the operation and 3 months after distraction were utilized for analysis. Landmarks and

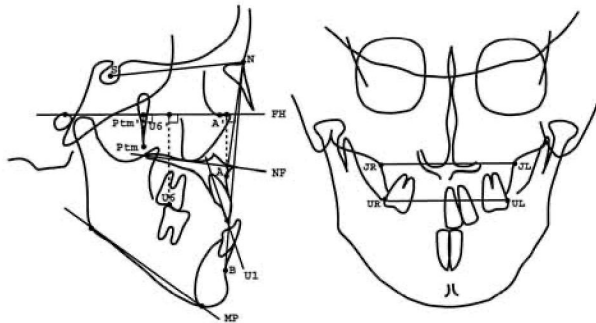


Fig. 4. Cephalometric landmarks and reference lines: N - nasion, S - sella turcica, Ptm - pterygomaxillary fissure, Ptm' - Ptm projected to FH, A - point A (subspinale), A' - A projected to FH, B - point B (supramentale), U6 - buccal groove of the maxillary first molar, U6' - U6 projected to FH, JR and JL - jugular points, UR and UL - the most lateral point on the buccal surface of the maxillary first molar, FH - Frankfort horizontal plane, NF - nasal floor, U1 - axis of upper incisor, MP - mandibular plane.

Table 2. Measurement Variables

Maxillary body	
NF to FH :	angle between lines NF and FH
U1 to NF :	angle between U1 axis and NF
U6'- Ptm' :	projected distance of U6 and Ptm to FH
A' - Ptm' :	projected distance of A and Ptm to FH
JR - JL :	distance between points JR and JL
UR - UL :	distance between points UR and UL
Facial skeletal pattern	
SNA :	angle between lines SN and NA
SNB :	angle between lines SN and NB
ANB :	angle between lines NA and NB
MP to FH :	angle between lines MP and FH

reference lines on lateral and P-A cephalograms are illustrated in Fig. 4, and measurement variables are shown in Table 2. In the present study, standardized CT images were reconstructed three-dimensionally and oriented using midsagittal and Frankfort-horizontal planes. Tracings of these three-dimensional CT images were superimposed on the zygomatic arch or the maxillary teeth to better describe the maxillary changes between pretreatment and posttreatment. Cephalograms and standardized CT images were traced on acetate paper and all tracings were carried out by a single experienced investigator (J.N.).

Results

Facial and intraoral findings

Pretreatment and posttreatment facial and intraoral photographs of one typical case in each Group are shown in Fig. 5 A-C.

Group A (Fig. 5A): Midface retrusion and overclosure of the mandible were improved with the backward rotation of the mandible. The anterior crossbite was improved and the molar relation changed from Class I to II.

Group B (Fig. 5B): Midface retrusion was improved with the advancement of the midface and the backward rotation of the mandible. The anterior and lateral crossbite was improved and the molar relation changed from Class I to II.

Group C (Fig. 5C): The flattened cheek line



Fig. 5A. Lateral facial photographs (*top*) and intraoral photographs (*bottom*) of one typical case in Group A (*left*-pretreatment, *right*-posttreatment). Midface retrusion and overclosure of the mandible in the pretreatment photographs were improved with the backward rotation of mandible as shown in the posttreatment photographs. The anterior crossbite before treatment was improved.

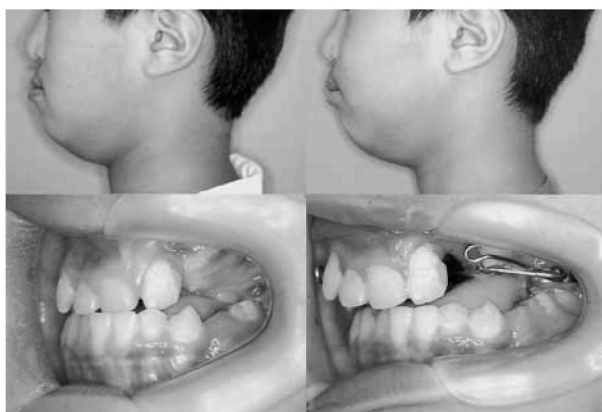


Fig. 5B. Lateral facial photographs (*top*) and intraoral photographs (*bottom*) of one typical case in Group B (*left*-pretreatment, *right*-posttreatment). Midface retrusion before treatment was improved with the advancement of midface and the backward rotation of the mandible after treatment. The anterior and lateral crossbite before treatment was improved

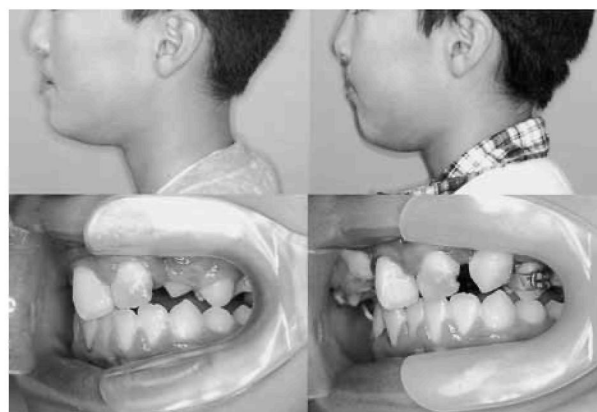


Fig. 5C. Lateral facial photographs (*top*) and intraoral photographs (*bottom*) of one typical case in Group C (*left*-pretreatment, *right*-posttreatment). The flattened cheek line before treatment changed to round and the mandible rotated backwards. The upper central incisors were in a normal position before and after treatment. Before treatment, the upper lateral incisors were congenitally missing and both lateral segments were occluded lingually to the mandibular dental arch.

changed to round and the mandible rotated backwards. The upper central incisors were in a normal position before and after treatment. Before treatment, the upper lateral incisors were congenitally missing and both lateral segments were occluded lingually to the mandibular dental arch. After treatment, the spaces of missing incisors were closed and the molar relation changed from Class I to II.

3D CT images

Pretreatment and posttreatment three-dimensional standardized CT images and their superimpositions of the lateral and basal tracings of one typical case in each group are shown in Fig. 6A-C. Their characteristic

findings in each Group were as follows.

Group A (Fig. 6A): From the superimpositions of the lateral and basal tracings on the zygomatic arch (e, f), the maxillary body was moved forwards with an upward rotation of the anterior part of the maxilla. However, the shape and the position of the pterygomaxillary fissure were unchanged. From the superimpositions of the lateral and basal tracings on the maxillary teeth from canine to first molar (g, h), the maxillary tuberosity was elongated posteriorly and the second molar migrated backwards.

Group B (Fig. 6B): The findings in the superimpositions on the zygomatic arch were similar to those in Group A except that the maxillary body and maxillary

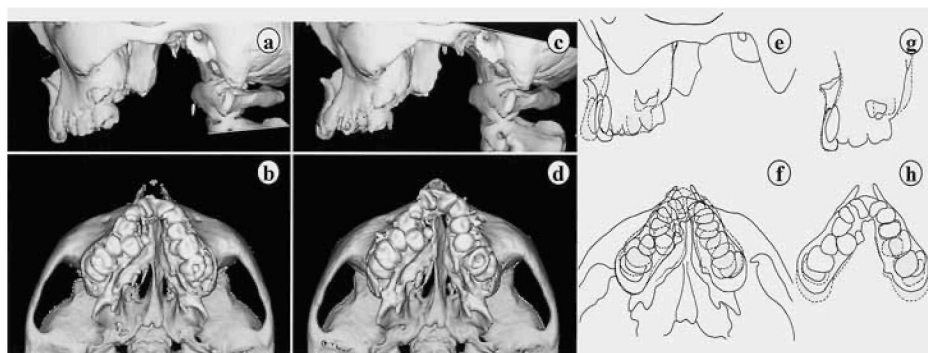


Fig. 6A. Three-dimensional CT images and superimpositions of tracings of one typical case in Group A. Pretreatment (a-lateral, b-basal) and posttreatment (c-lateral, d-basal) CT images, superimpositions on the zygomatic arch (e-lateral, f-basal) and on the maxillary teeth (g-lateral, h-basal). Gray areas and dotted lines in superimpositions indicate posttreatment.

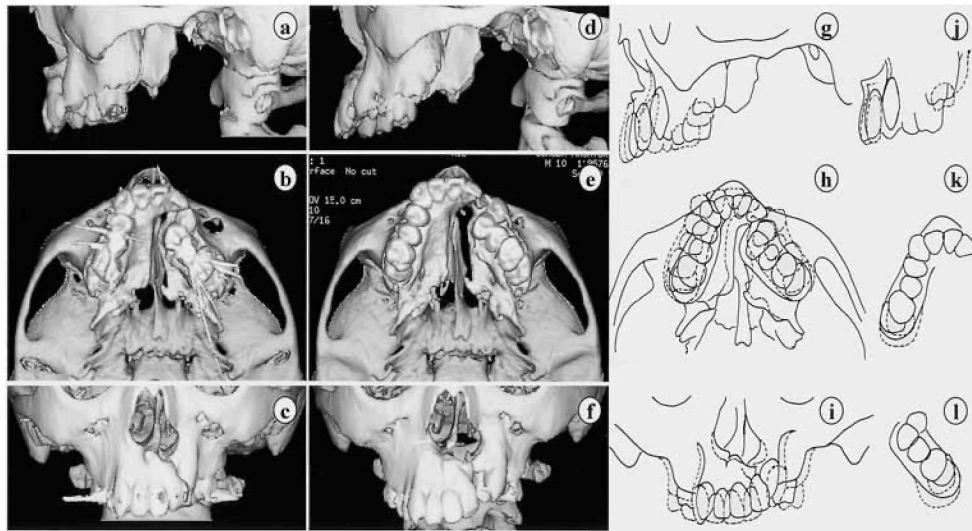


Fig. 6B. Three-dimensional CT images and superimpositions of tracings of one typical case in Group B. Pretreatment (a-lateral, b-basal, c-frontal) and posttreatment (d-lateral, e-basal, f-frontal) CT images, superimpositions on the zygomatic arch (g-lateral, h-basal, i-frontal) and on the maxillary teeth (j-lateral, k-basal of the right segment, l- basal of the left segment). Gray areas and dotted lines in superimpositions indicate posttreatment.

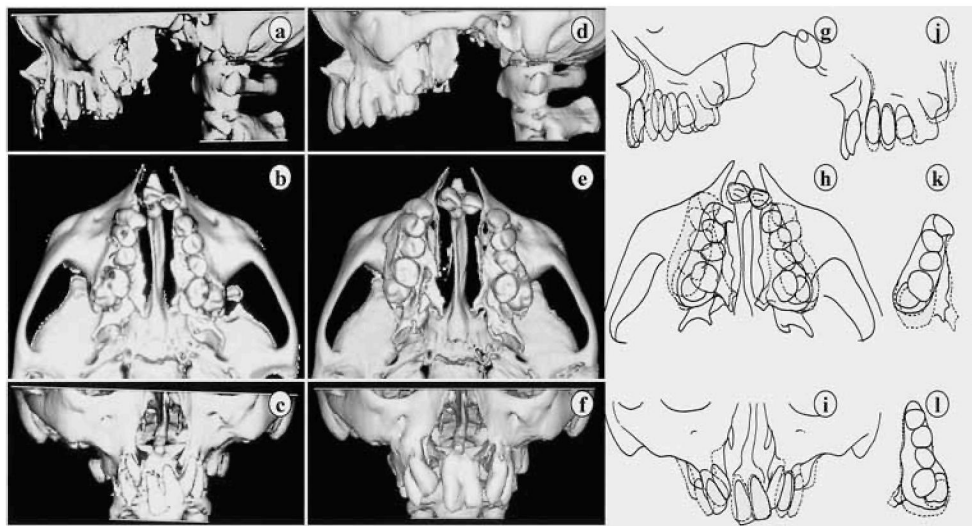


Fig. 6C. Three-dimensional CT images and superimpositions of tracings of one typical case in Group C. Pretreatment (a-lateral, b-basal, c-frontal) and posttreatment (d-lateral, e-basal, f-frontal) CT images, superimpositions on the zygomatic arch (g-lateral, h-basal, i-frontal) and on the maxillary teeth (j-lateral, k-basal of the right segment, l-basal of the left segment). Gray areas and dotted lines in superimpositions indicate posttreatment.

teeth were moved forwards and downwards in the lateral tracing (g). In the basal tracing (h), both the major and minor segments were moved forwards and outwards with widening of palatal and alveolar clefts. In the frontal tracing (i), both the major and minor segments were moved with slight outward inclination and an increase of the maxillary width. The bone step indicating the osteotomy site was recognized on the pyriform margin and the pyriform was widened below the bone step. From

the superimpositions of the lateral and basal tracings on the maxillary teeth of each segment (j, k, l), the maxillary tuberosity was elongated posteropalatally and the second molar migrated backwards.

Group C (Fig. 6C): From the superimpositions on the zygomatic arch, the lateral segment was moved forwards and downwards with an upward rotation of the anterior part of the lateral segment, however, both the position and shape of the pterygomaxillary fissure were

unchanged as in Groups A and B in the lateral tracings (g). In the basal and frontal tracings (h, i), both lateral segments were moved forwards and outwards without reduction of the palatal inclination. The palatal cleft was also widened and the premaxilla moved into that space with correction for the rotation of the incisors, and the maxillary width was increased. The findings of the pyriform were similar to those in Group B. From the superimpositions of the lateral and basal tracings on the maxillary teeth of each segment (j, k, l), the findings were similar to those in Groups A and B.

Cephalometric analysis

Cephalometric changes in the maxillary body and in the facial skeletal pattern in each Group are presented in Tables 3 and 4.

Group A: Counterclockwise rotation of the nasal

floor (NF to FH) decreased 3.9 degrees in Case 1 and 4.4 degrees in Case 2. Advancement of the maxilla was measured as the increase of 6.2 mm in Case 1 and 6.9 mm in Case 2 at the U6'-Ptm'. This caused increases in their maxillary body lengths (A'-Ptm') of 4.8 mm and 6.0 mm, respectively. SNA increased 3.8 and 4.6 degrees and SNB decreased 1.7 and 2.0 degrees for the increased ANB of 5.5 and 6.6 degrees in Case 1 and Case 2, respectively. Clockwise rotation of the mandibular plane (MP to FH) increased 2.1 degrees in Case 1 and 1.4 degrees in Case 2.

Group B: Counterclockwise rotation of the nasal floor (NF to FH) decreased 0.5 to 2.6 degrees. Advancement of the maxilla was measured as the increase of 4.2 to 6.4 mm at the U6'-Ptm'. This caused increases in their maxillary body length (A'-Ptm') of 3.8 to 5.0 mm. Uprighting of the U1 axis was indicated by the

Table 3. Cephalometric Changes of the Maxillary Body

Variables	Pre	Post	Diff.	Pre	Post	Diff.	Pre	Post	Diff.	Pre	Post	Diff.		
Group A			Case 1 (BCLP)			Case 2 (UCLP)								
NF to FH (deg)	4.1	0.2	-3.9	-1.4	-5.8	-4.4								
A'-Ptm'(FH) (mm)	44.8	49.6	4.8	43.8	49.8	6.0								
U6'-Ptm'(FH)(mm)	32.5	38.7	6.2	30.9	37.8	6.9								
U1 to NF (deg)	96.1	95.0	-1.1	128.1	127.5	-0.6								
Group B			Case 3 (UCLP)			Case 4 (UCLP)			Case 5 (UCLP)			Case 6 (UCLP)		
NF to FH (deg)	2.3	-0.3	-2.6	12.0	11.5	-0.5	5.1	4.5	-0.6	-2.9	-5.3	-2.4		
A'-Ptm' (mm)	47.5	52.5	5.0	41.4	45.2	3.8	48.3	52.3	4.0	33.9	33.9	0.0		
U6'-Ptm' (mm)	37.3	43.7	6.4	33.5	37.7	4.2	30.9	35.1	4.2	33.8	38.9	5.1		
U1 to NF (deg)	115.1	112.1	-3.0	98.0	97.0	-1.0	105.0	102.1	-2.9	96.2	97.0	0.8		
JR-JL (mm)	70.1	77.2	7.1	68.8	74.6	5.8	67.5	74.8	7.3	59.8	64.8	5.0		
UR-UL (mm)	59.2	65.7	6.5	60.6	66.0	5.4	54.3	62.3	8.0	52.2	58.0	5.8		
Group C			Case 7 (BCLP)			Case 8 (BCLP)			Case 9 (BCLP)					
NF to FH (deg)	-2.8	-3.4	-0.6	5.3	8.0	2.7	9.1	9.8	0.7					
A'-Ptm' (mm)	51.2	51.0	-0.2	52.8	52.4	-0.4	64.3	63.0	-1.3					
U6'-Ptm' (mm)	28.3	34.9	6.6	34.2	38.3	4.1	24.4	26.8	2.4					
U1 to NF (deg)	98.7	96.1	-2.6	90.3	84.2	-6.1	94.5	92.0	-2.5					
JR-JL (mm)	68.7	76.2	7.5	59.8	70.5	10.7	60.0	72.0	12.0					
UR-UL (mm)	51.4	60.1	8.7	58.5	70.2	11.7	55.9	69.5	13.6					

Pre: pre distraction, Post: posttreatment, Diff.: difference between Pre and Post.

Table 4. Cephalometric Changes of the Facial Skeletal Pattern

Variables	Pre	Post	Diff.	Pre	Post	Diff.	Pre	Post	Diff.	Pre	Post	Diff.	
Group A			Case 1 (BCLP)			Case 2 (UCLP)							
SNA (deg)	76.5	80.3	3.8	71.0	75.6	4.6							
SNB (deg)	76.7	75.0	-1.7	74.9	72.9	-2.0							
ANB (deg)	-0.2	5.3	5.5	-3.9	2.7	6.6							
MP to FH (deg)	31.9	34.0	2.1	23.1	24.5	1.4							
Group B			Case 3 (UCLP)			Case 4 (UCLP)			Case 5 (UCLP)			Case 6 (UCLP)	
SNA (deg)	79.4	84.0	4.6	80.5	84.2	3.7	76.2	79.8	3.6	71.3	75.5	4.2	
SNB (deg)	79.0	77.8	-1.2	83.0	81.9	-1.1	74.0	72.6	-1.4	75.0	72.7	-2.3	
ANB (deg)	0.4	6.2	5.8	-2.5	2.3	4.8	2.1	7.2	5.1	-3.7	2.8	6.5	
MP to FH (deg)	29.0	31.3	2.3	30.5	32.0	1.5	32.5	34.1	1.6	18.5	21.5	3.0	
Group C			Case 7 (BCLP)			Case 8 (BCLP)			Case 9 (BCLP)				
SNA (deg)	78.5	78.0	-0.5	79.4	78.6	-0.8	81.0	80.5	-0.5				
SNB (deg)	72.0	71.3	-0.7	68.5	67.3	-1.2	74.8	73.3	-1.5				
ANB (deg)	6.5	6.7	0.2	10.9	11.3	0.4	6.2	7.2	1.0				
MP to FH (deg)	27.4	28.5	1.1	32.7	35.4	2.7	28.0	31.1	3.1				

Pre: pre distraction, Post: posttreatment, Diff.: difference between Pre and Post.

decrease in the U1 to NF except for Case 6. The maxillary width (JR-JL) increased 5.0 to 7.3 mm, and the intermolar width (UR-UL) increased 5.4 to 8.0 mm. SNA increased 3.6 to 4.6 degrees and SNB angle decreased 1.1 to 2.3 degrees for the increased ANB of 4.8 to 6.5 degrees. Clockwise rotation of the mandibular plane (MP to FH) increased 1.5 to 3.0 degrees.

Group C: Counterclockwise rotation of the nasal floor (NF to FH) decreased 2.7 degrees in Case 8. Although an advancement of both lateral segments was measured as an increase of 2.4 to 6.6 mm at the U6'-Ptm', there was no increase in the maxillary body length (A'-Ptm'), which differed from the findings in Groups A and B. The palatal inclination of the U1 axis (U1 to NF) decreased 2.5 to 6.1 degrees. The maxillary width (JR-JL) increased 7.5 to 12.0 mm, and the intermolar width (UR-UL) increased 8.7 to 13.6 mm. SNB was similar to that in Groups A and B, but the increases of SNA and ANB shown in Groups A and B were not recognized. Clockwise rotation of the mandibular plane (MP to FH) increased 1.1 to 3.1 degrees.

Discussion

The maxillary hypoplasia and the anterior and/or lateral crossbites are commonly seen in patients with repaired cleft lip and palate. Ross⁹ showed that about 25% of patients with repaired cleft lip and palate developed maxillary hypoplasia that did not respond to orthodontic treatment alone. Conventional orthodontic treatment does little to restore dentofacial relations until later in adolescence when growth of the jaws is near completion, and orthognathic surgery is carried out subsequently^{7,8}. In the last decade, DOG has been rapidly developed for managing hypoplastic maxillae in patients with repaired cleft palate. With the description by Swennen et al¹⁷, and Polley and Figueroa¹⁸, compared with orthognathic surgery, DOG offers the advantage of not having to wait until the facial growth is completed, of enhancing further growth and development of jaws, and of no limitation to the age when patients can be treated. The early aesthetic and functional rehabilitation of these patients has been considered as a major goal¹⁹.

Most maxillary hypoplasia in patients with repaired cleft lip and palate varies in the vertical, horizontal, and transverse directions that are often difficult to mobilize because of scarring from previous surgeries^{9,11}. For

instance, the small maxillary advancement of 2.4 mm and the large lateral expansion of 13.6 mm were estimated in Case 9, then the normal facial appearance and occlusion could not be anticipated by the traditional orthodontic/surgical approaches. Although the use of the maxillary protracting appliance was applied in all patients in this study, their responses fell short of expectations. Consequently, the maxillary distraction osteogenesis combining maxillary advancement and lateral expansion was provided as an alternative method during the mixed dentition. As the results showed, the estimated advancement of 2.4 to 6.9 mm and lateral expansion of 5.4 to 13.6 mm were achieved, then the midface retrusion was improved and the anterior and/or lateral crossbite was corrected. No complications such as skin irritation on the chin, compliance of the devices or tooth loss were observed.

In most previous reports of DOG in patients with cleft maxillary hypoplasia, attention was only paid to the amount of maxillary advancement and associated changes in facial appearance. These changes were evaluated two-dimensionally using cephalograms and photographs, however, the morphological changes of maxilla itself was not shown precisely in those reports and description on the results in expanding the hypoplastic maxilla as shown in this report is rare. The results demonstrated that the hypoplastic maxilla can be successfully elongated and expanded in these patients, especially, those proved by three-dimensional CT images were of much interest. Molina et al.¹⁵ only used three-dimensional CT scan reconstruction to evaluate the maxillary changes in patients with repaired cleft lip and palate. In the present study, standardized CT images using midsagittal and Frankfort-horizontal planes were reconstructed three dimensionally. Tracings of three-dimensional CT images were superimposed on the zygomatic arch or the maxillary teeth to visualize the maxillary changes from pretreatment to posttreatment, and they facilitated to understand the cephalometric and photographic evaluations. Their findings confirmed that the whole maxilla moved forwards with a counterclockwise rotation, the maxillary lateral segments were moved forwards and outwards, and the second molars migrated to the posteriorly elongated maxillary tuberosity.

As described by Kusnoto et al.²⁰, the posterior elongation of the maxillary tuberosity is seemed to be the area where the new bone formation occurs as a result of

DOG. This is the region where stability of the maxilla after DOG is achieved. In addition, the new bone formation at the maxillary tuberosity seemed to be those in the normally growing maxilla. They also can be interpreted that the cleft maxillary hypoplasia in the mixed dentition responded well to the present DOG procedure, and that the impaired growth of the maxilla in patients with repaired cleft lip and palate presented a catch-up growth during distraction process for only three to seven weeks. It is suggested that the maxillary advancement by distraction osteogenesis during growing period could provide an osseous environment to permit spontaneous eruption of the second maxillary molar into the elongated maxillary tuberosity.

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口蓋裂術後患者における仮骨延長術による上顎骨の 形態変化の三次元CT画像を用いた評価

永田順子^a，佐藤耕一^b，迫田隅男^c，芝 良祐^d

^a鹿児島大学大学院医歯学総合研究科発生発達成育学講座顎顔面育成学分野，

^b九州歯科大学口腔顎顔面外科学講座病態制御学分野，

^c宮崎大学医学部歯科口腔外科学講座，

^d宮崎大学名誉教授

本研究の目的は、仮骨延長術による上顎骨の形態変化を特に三次元CT画像を用いて明らかにすることである。9歳から11歳の9名の唇顎口蓋裂患者を対象に、フェイスマスクと拡大装置を用いた上顎骨の仮骨延長を行った。術前と術後3ヵ月におけるX線規格写真、写真、および三次元CT画像を用いて上顎骨の形態変化を分析した。頭部規格写真分析から、予定した上顎骨の前方移動と側方拡大が達成されていることが示された。写真から顔のプロファイルと咬合の改善が示された。手術前後における3次元CT画像の重ね合わせから、仮骨延長による上顎骨の形態変化を容易に視覚化することができた。これらの分析から、上顎骨が反時計回りに回転しながら前方へ移動し、側方セグメントは前方および外方へ移動していることが確認された。また、上顎結節が後方へ延びたことにより、上顎第二大臼歯が後方移動していることが確認された。これらの結果は、成長期の上顎骨の仮骨延長が、顔貌と咬合を改善するだけでなく、上顎第二大臼歯の萌出空隙の確保にも有効であることを示した。