A Case of Unilateral Cleft Lip and Palate with Postoperative Skeletal Stability after Maxillary Distraction Osteogenesis

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Abstract

This case report describes postoperative skeletal stability in a unilateral cleft lip and palate (UCLP) patient who received maxillary distraction osteogenesis (DOG) using a rigid extraoral distractor (RED) for maxillary advancement.

The patient was a Japanese female with right UCLP who had presented at our orthodontic clinic at 5 years and 10 months of age. She received autologous secondary bone grafting (SBG) using cancellous bone from the iliac crest at 7 years and 6 months of age. Since the phase-1 orthodontic treatment to facilitate maxillary growth using a face mask did not work effectively, the objective of the phase-2 treatment was to correct midface hypoplasia.

The maxilla was advanced by DOG using RED with a significant improvement of facial esthetics and skeletal relationship. By long-term face mask therapy for 9 months, there was little postoperative relapse of maxilla in this case.

SBG and prolonged postsurgical face mask therapy are likely to improve postoperative skeletal stability in the sagittal dimension after maxillary DOG in CLP patients.

Key words: cleft lip and palate, postoperative skeletal stability, maxillary distraction osteogenesis

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Introduction

The treatment objectives of cleft lip and palate (CLP), besides achieving integrity of the primary and secondary palates, normal speech function and airway patency, are to obtain functional occlusion and improve the aesthetic appearance of the face1, 2). Many efforts have been made to improve these points for over eight decades3). However, it is well known that as a result of surgical stress and rigid scar formation, the cleft repair affects the maxillary growth with lesser or greater extent of midfacial deficiency with Class III malocclusion4-7). Therefore, further surgical interventions are often required to correct maxillary hypoplasia. For more than four decades conventional orthognathic surgical methods (including Le Fort I osteotomy) have been used for maxillary advancement in CLP patients, even though these methods frequently fail to fulfill expectations8, 9).

Since Ilizarov developed a technique for distraction osteogenesis of long bones10), this surgical procedure has also become an important alternative to correct midface hypoplasia11, 12). Nevertheless, some amount of relapse is reported in many studies13-15), and the average postoperative relapse rate after maxillary advancement has been reported to be as high as 25%16).

In this study, we demonstrate a unilateral cleft lip and palate (UCLP) case with severe maxillary retrusion who received maxillary distraction osteogenesis (DOG) using a rigid extraoral distractor (RED) (KLS Martin, Tuttlingen, Germany). The patient underwent secondary bone grafting (SBG) at 7 years and 6 months of age. For postoperative stability, postsurgical long-term face mask therapy was applied. The present case, together with previous studies12, 17), suggests that SBG and long-term face mask therapy might contribute to the postoperative skeletal stability in the sagittal dimension.

Case

Patient: 12 years and 9 months female
Chief complaint: total cross bite and midface hypoplasia
Medical history:
A Japanese female patient with right UCLP presented at

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our orthodontic clinic at 5 years and 10 months of age (T0: pretreatment). Her body height and body weight were comparable with the Japanese female norms (110cm and 19.0kg). She had received cheiloplasty using the Millard method and palatoplasty using the pushback method at 3 months and 15 months of age, respectively.

She presented a concave profile (Fig. 1A through C) with maxillary hypoplasia, an Angle Class III malocclusion with narrow upper arch, total crossbite, a reverse overjet of −4.0mm and an overbite of +3.0mm (Fig. 1D through H). The postero-anterior cephalogram showed a deviated nasal septum (Fig. 2A). Furthermore, analysis of the lateral cephalogram revealed a skeletal Class III intermaxillary relationship (ANB: −0.3°) due to the maxillary retrusion (SNA: 78.9°) and mandibular protrusion (SNB: 79.2°) compared to the age-matched Japanese norm (Fig. 2B and Table 1). Panoramic radiographs showed that the permanent maxillary left second premolar was congenitally missing (Fig. 2C) and the alveolar cleft was located distal to the upper right lateral incisor (Fig. 2C, D).

Our phase-1 treatment objectives were to integrate the alveolar cleft for normal tooth eruption and for promoting maxillary growth. She received autologous SBG using cancellous bone from the iliac crest at 7 years and 6 months of age. Six months after SBG, when compared to the preoperative conditions, the postsurgical radiographs showed an elevated height of the interdental septum, successful closure of the cleft (Fig. 3F, G), and eruption of the upper right lateral incisor (Fig. 3A through G). However, the upper right lateral incisor erupted with distal tipping and curved root (Fig. 3A, G). After the eruption of the maxillary right lateral incisor, she received a sectional arch appliance to level and align the maxillary bilateral central and lateral incisors. A face mask was placed to protract the maxilla at 8 years and 10 months of age. However, due to her poor compliance, the face mask treatment was unsuccessful. Also, the leveled maxillary central and lateral incisors were relapsed by her poor compliance of using retainer (Fig. 4D through H). After the phase-1 treatment, neither the soft tissue profile nor the occlusion was satisfactory.

1) Diagnosis and treatment plan

According to cephalometric measurements at 12 years and 9 months of age (T1: before the orthognathic treatment), severe maxillary hypoplasia was present (SNA:
Additionally, during the peak growth period, a large amount of mandibular growth was seen (SNB: 83.5°) (Table 2) (T1). The severe maxillary hypoplasia significantly deteriorated her profile (Fig. 4A through C) and her skeletal Class III disharmony became worse. The ANB angle changed to −10.0° (Table 2). Furthermore, her overjet decreased to −8.3mm (Fig. 4D through H). Arch length discrepancies were −6.3mm and +0.5 mm for the upper and lower arches, respectively.

Our phase-2 treatment objectives were to correct the midface hypoplasia. We planned to overtreat the maxilla by moving 20% more anterior to the position of Japanese norm. This was because postoperative maxillary relapse is frequently seen after DOG. DOG was chosen to move the maxilla by 14.0mm. Since the upper right lateral incisor was a microdont and deformed with insufficient space for proper alignment of the maxillary arch, we decided to extract this tooth. Since her bone age was comparable to those of adult female and there was no increment in her height, we decided to initiate the phase-2 treatment. We planned a surgical correction after DOG if there was a postoperative hypernasality.

As a treatment alternative, after the growing period "one step", Le Fort I osteotomy for the maxillary displacement combined with sagittal splint ramus osteotomy for mandibular setback was also considered. The patient showed severe maxillary deficiencies with rigid scar formation and needed

Fig. 2 Pretreatment cephalograms (A, B), panoramic radiography (C) and panagram (D).
arrow: congenitally missing maxillary left second premolar tooth in C, asterisk: alveolar cleft in C and D.
Table 1  Pretreatment cephalometric analysis

| Measurement                  | T0 (5Y10M) | Japanese norm ± SD |
|------------------------------|------------|--------------------|
| SNA (°)                      | 78.9       | 81.4 ± 2.8         |
| SNB (°)                      | 79.2       | 76.4 ± 2.1         |
| ANB (°)                      | −0.3       | 5.0 ± 2.4          |
| Mandibular plane (°)         | 30.2       | 31.1 ± 5.2         |
| U1 to FH (°)                 | 87.2       | 96.4 ± 4.7         |
| U1 to SN (°)                 | 79.1       | 88.8 ± 4.4         |
| L1 to Mandibular (°)         | 87.2       | 84.4 ± 6.2         |
| A’-Ptm (mm)                  | 40.6       | 41.9 ± 2.1         |
| Ptm’-Ms (mm)                 | 16.1       | 17.6 ± 2.3         |
| Ar-Go (mm)                   | 40.8       | 36.5 ± 2.8         |
| Go-Pog (mm)                  | 73.5       | 65.4 ± 3.1         |
| Upper lip to E-line (mm)     | −2.0       |                    |
| Lower lip to E-line (mm)     | +3.0       |                    |

T0: Pretreatment (5Y10M) ; S indicates sella tuitica; N, nasion; A, point A; B, point B; Ptm, pterygomaxillary point; Ar, articulare; Go, gonion; Pog, pogonion; A’, point on Ans-Pns projected from point A; Ptm’, point on Ans-Pns projected from pterygomaxillary point; Ms, point on Ans-Pns projected from center point of the upper first molar; FH, Frankfort horizontal plane; Mandibular plane (°), Angle between Frankfort horizontal plane (FH) and mandibular plane; U1, long axis of the maxillary central incisor; U1 to FH (°), Angle between upper incisor axis and FH plane; U1 to SN (°), Angle between upper incisor axis and SN plane; L1, long axis of mandibular central incisor; L1 to Mandibular (°), Angle between L1 and mandibular plane; A’-Ptm’ (mm), horizontal distance between point A’ and Ptm’; Ptm’-Ms (mm), horizontal distance between Ptm’ and Ms; Ar-Go (mm), distance between Ar and Go; Go-Pog (mm), distance between Go and Pog; E-line, line between tip of nose and chin; Upper lip to E-line (mm), distance between E-line and top of upper lip; Lower lip to E-line (mm), distance between E-line and top of lower lip. The mean ± S.D. represent the age-matched Japanese norm (Iizuka, 1958).18

Fig. 3 Oral photos (A-E) and dental X-rays (F, G) after secondary bone grafting (SBG) (8 years and 1 month). Arrow: the maxillary right lateral incisor was erupted, asterisk: alveolar cleft after SBG in F and G.
Table 2 Cephalometric analysis from T1 to T4

| Measurement                        | T1 (12Y9M) | T2 (13Y10M) | T3 (14Y2M) | T4 (15Y10M) | Japanese norm ± SD |
|------------------------------------|------------|-------------|------------|-------------|-------------------|
| SNA (°)                            | 73.6       | 73.4        | 87.0       | 87.0        | 81.5 ± 4.2        |
| SNB (°)                            | 83.5       | 84.4        | 83.7       | 84.0        | 77.1 ± 3.8        |
| ANB (°)                            | -10.0      | -11.0       | +3.3       | +3.0        | 4.4 ± 4.0         |
| Mandibular plane (°)               | 27.6       | 29.8        | 31.3       | 30.3        | 34.0 ± 3.8        |
| U1 to FH (°)                       | 116.6      | 112.6       | 114.5      | 114.8       | 111.5 ± 5.0       |
| U1 to SN (°)                       | 108.2      | 104.1       | 106.0      | 106.3       | 105.4 ± 5.2       |
| L1 to Mandibular (°)               | 82.2       | 77.9        | 80.9       | 78.6        | 95.4 ± 6.3        |
| A'-Ptm' (mm)                       | 40.7       | 40.6        | 53.4       | 53.5        | 46.3 ± 1.8        |
| Ptm'-Ms (mm)                       | 16.9       | 17.3        | 31.3       | 31.6        | 17.6 ± 2.7        |
| Ar-Go (mm)                         | 45.5       | 46          | 45.3       | 46.5        | 45.2 ± 4.8        |
| Go-Pog (mm)                        | 84.5       | 88.1        | 89         | 89.7        | 78.1 ± 4.2        |
| Upper lip to E-line (mm)           | -8.0       | -9.0        | ±0.5       | +0.0        |                   |
| Lower lip to E-line (mm)           | +4.0       | +4.0        | +0.5       | +0.5        |                   |

T1: before orthognathic treatment (12Y9M); T2: before maxillary distraction (13Y10M); T3: after maxillary distraction and before consolidation (14Y2M); T4: after active treatment (15Y10M); S indicates sella tucica; N, nasion; A, point A; B, point B; Ptm, pterygomaxillary point; Ar, articulare; Go, gonion; Pog, pogonion; A', point on Ans-Pns projected from point A; Ptm', point on Ans-Pns projected from pterygomaxillary point; Ms, point on Ans-Pns projected from center point of the upper first molar; FH, Frankfort horizontal plane; Mandibular plane (°), Angle between Frankfort horizontal plane (FH) and mandibular plane; U-1, long axis of the maxillary central incisor; U1 to FH (°), Angle between upper incisor axis and FH plane; U1 to SN (°), Angle between upper incisor axis and SN plane; L1, long axis of mandibular central incisor; L1 to Mandibular (°), Angle between L1 and mandibular plane; A'-Ptm' (mm), horizontal distance between point A' and Ptm'; Ptm'-Ms (mm), horizontal distance between Ptm' and Ms; Ar-Go (mm), distance between Ar and Go; Go-Pog (mm), distance between Go and Pog; E-line, line between tip of nose and chin; Upper lip to E-line (mm), distance between E-line and top of upper lip; Lower lip to E-line (mm), distance between E-line and top of lower lip. The mean ± S.D. represent the age-matched Japanese norm (Iizuka, 1958).
a large amount of maxillary advancement. In cases with moderate or severe maxillary deficiencies, DOG is recommended\(^6\)\(^,\)\(^{19}\) to obtain a large amount of movement. Baek stated that the choice of DOG should strongly depend on

Fig. 5 Facial photos (A-C), oral photos (D-H), cephalograms (I, J) and panoramic radiography (K) before the maxillary distraction (T2: 13 years and 10 months)
the amount of advancement. Patients with scars resulting from previous lip and palate operations are further indicated for distraction surgery. Consequently, as distraction methods have shown advantages over one-stage orthognathic surgery, we proposed the superiority of maxillary DOG over the conventional orthognathic surgery. If there is an unexpectedly large amount of mandibular growth and maxillary relapse after DOG, further surgery on the mandible might be needed.

2) Treatment progress
The upper and lower arches were aligned by a .022 slot edgewise appliance from 12 years and 9 months of age (T1). Prior to DOG (T2: 13 years and 10 months of age, before the maxillary distraction) (Fig. 5A through H), the lateral cephalogram and measurements of cephalometric analysis showed that the SNA angle remained stable, nevertheless slight mandibular growth occurred (SNB: 84.4°) and the ANB angle further decreased to −11.0° during the preoperative orthodontic treatment (Fig. 5J and Table 2). From the postero-
anterior cephalogram, the mandible was shifted slightly to the right (Fig. 5I). Panoramic radiography showed distal tipping of the maxillary right central incisor (Fig. 5K).

A high Le Fort I osteotomy was performed at 14 years of age. The entire procedure was carried out under general anesthesia with naso-endotracheal intubation. After osteotomy, the RED device was inserted to the skull with three scalp screws on each side. After 5 days of latency period, the

Fig. 7  Facial photos (A−C), oral photos (D−H), cephalograms (I, J) and panoramic radiography (K) after active treatment (T4: 15 years and 10 months)
The distractor was activated by half a turn two times daily. This resulted in a predicted maxillary advancement of 1.0 mm per day. The direction of maxillary traction was 30° downward to the FH plane. Distraction was continued to obtain facial harmony and achieve Angle Class I occlusion with 20% of overcorrection. The entire activation process (38 days) was performed on an inpatient basis (Fig. 6A through F). The postero-anterior cephalogram showed that the maxilla was protracted without significant asymmetry (Fig. 6G) at 14 years and 2 months of age (T3: after maxillary distraction and before consolidation). The lateral cephalogram showed that an appropriate overjet was obtained (Fig. 6H). After receiving full advancement of the maxilla, the RED was left in situ for an additional 15 days. Following this consolidation period, the distractors were removed (Fig. 6I) and the patient was advised to wear an orthodontic face mask for 12 hours a day for 9 months to prevent postoperative relapse. There was good compliance in using the face mask. The reason for this was that she had been satisfied with posttreatment facial appearance and had strongly desired to avoid relapse as well.

3) Treatment result

Favorable occlusion with an overjet of +2.0 mm and an overbite of +2.0 mm was achieved (Fig. 7D through H, J) at 15 years and 10 months of age (T4: after active treatment). Successful correction of the skeletal Class III relationship was obtained due to the significant advancement of the maxilla. The soft tissue profile of the patient became from concave to convex with a slight protrude midface. Also, she showed a somewhat short face, due to the insufficient vertical movement of the maxilla and mandible (Fig. 7C and 8). Additionally, it was found that there was no significant change in external nose form (Fig. 7A through C). The distance between E-line changed from −9.0 mm to +0.0 mm by the upper lip and changed from +4.0 mm to +0.5 mm by the lower lip. The postero-anterior cephalogram showed that the midlines of maxillary and mandibular dentition matched each other (Fig. 7D). The results of cephalometric measurements at T1, T2, T3 and T4 are shown in Table 2. The SNA angle changed significantly from 73.4° (T2) to 87.0° (T4) and the ANB angle from −11.0° (T2) to +3.0° (T4). The inclination of the mandibular plane slightly increased (Table 2). The panoramic radiograph showed that proper root paralleling was achieved (Fig. 7K). The superimposition before and after maxillary DOG and after active treatment showed that the maxilla was advanced by 14.0 mm parallel to the palatal plane, and the mandible showed slight clockwise rotation as planned. The occlusion and facial profile remained stable without showing any relapse (Fig. 8). Examined by a speech therapist, the condition of her hypernasality was comparable to her preoperative condition.

**Discussion**

Maxillary hypoplasia is a common deformity in CLP patients. To correct this deformity, either conventional orthognathic surgery or maxillary DOG is often planned, as they improve facial esthetics and advancement of the maxilla. However, it is well documented that some amount of postsurgical relapse is inevitable. Factors contributing to the relapse include scar formation following previous surgeries, muscle resistance, tension of soft tissues, delayed adaptation and stability of bony fragments. To date, no firm protocols and guidelines have been established to prevent the relapse.

In the present case, a considerable amount of maxillary advancement was obtained by maxillary DOG with RED device. The skeletal relationship improved significantly and remained stable. Table 3 compares the treatment outcome and relapse rates in cases undergone maxillary DOG. Most cases with only SBG or face mask therapy or neither of them reported higher relapse rates. Singh et al. evaluated the stability following maxillary distraction osteogenesis of 12 patients, and found a relapse rate after DOG of approximately 30.0%. Aksu et al. reported 7 cases who underwent maxillary DOG with an average advancement of 9.0 mm, and found that the effective maxillary length decreased significantly by 22.0%. Cho et al. and Baek reported similar results, with relapse rates of 23% and 21%, respectively. Kanno et al. reported slightly less relapse (11%) after maxillary DOG. However, Okada et al. reported no relapse in 3 patients by performing SBG and long-term
(6 months) postsurgical face mask therapy as in the present case.

One of the important factors related to the postoperative stability after the Le Fort I osteotomy is a stable occlusion\(^2\). SBG is a surgical procedure to integrate maxillary segment and maintain a stable occlusion\(^2\). Considering these points, it is likely that SBG could help prevent postoperative relapse after the maxillary DOG by maintaining her stable occlusion. There was no direct evidence on how much contribution was obtained by the postoperative face mask therapy to her treatment outcome. However, performing the same protocol, we reported three other CLP cases undergone maxillary DOG without any postoperative relapse in the sagittal dimension\(^1\), suggesting the effect of postoperative face mask therapy on the stability (Table 3).

In conclusion, the treatment result without postoperative relapse was achieved in our patient. It is likely that SBG and prolonged postsurgical face mask therapy might contribute to the treatment outcome. However, the amount of formed scar and surgical stress after lip and palate formation differ among CLP cases, and these factors are related to the amount of postoperative relapse in each case.

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### Table 3  Reported treatment protocol and outcome of maxillary distraction osteogenesis (DOG)

| Articles          | Gender | Cleft type | Type of distraction | SBG | Age at DOG (year) | Horizontal maxillary advancement (mm) | Latency period (days) | Consolidation period | Face-mask therapy | Fixation plates | Mean horizontal relapse rate (%) |
|-------------------|--------|------------|---------------------|-----|------------------|---------------------------------------|-----------------------|---------------------|-------------------|-----------------|-----------------------------|
| Present case      | Female | UCLP       | Extra               | +   | 14.0             | 14.0                                  | 5                     | 15 days             | 9 months          | -               | -                           |
| Tanaka et al., 2001 | Male   | UCLP       | Intra               | NA  | 21.5             | 11.4                                  | 7                     | 6 months            | -                | +               | 27.9%                       |
| Baba et al., 2003 | 4 Males | 1 CL       | Extra               | -   | mean             | mean                                  | 2.6                   | 22-36 days          | -                | 21.7%                        |
|                   | 4 Female| UCLP       | 1 CP                |     |                  |                                       |                       |                     |                  |                 |                             |
| Shirota et al., 2006 | Male   | BCLP       | Extra               | +   | 21.6             | 15                                    | 3                     | 28 days             | -                | +               | 13.3%                       |
| Watanabe et al., 2007 | Male   | UCLP       | Extra               | +   | 16.3             | 9.1                                   | NA                    | 8 days              | -                | +               | 31.8%                       |
| Kanno et al., 2008 | 8 Males | 9 UCLP     | 9 Extra             | +   | 20.7             | mean                                  | 5-7                   | 3-4 weeks           | +                |                | 6.0%                        |
|                   | 4 Female| 1 BCLP     | 3 Intra             |     |                  |                                       |                       |                     |                  |                 |                             |
| Aksu et al., 2010 | 4 Males | 3 UCLP     | Extra               | -   | mean             | mean                                  | 5                     | 12 weeks            | -                | -               | 22.0%                       |
|                   | 3 Female| 4 BCLP     |                     |     |                  |                                       |                       |                     |                  |                 |                             |
| Menon et al., 2010 | 3 Females| 5 ULCP     | Intra               | +   | mean             | mean                                  | 3                     | 8 weeks             | -                | +               | 7.1%                        |
|                   | 3 Males | 1 BCLP     |                     |     |                  |                                       |                       |                     |                  |                 |                             |
| Akarsu et al., 2012 | Male   | UCLP       | Extra               | NA  | 31.0             | 12.0                                  | 3                     | 4 weeks             | 8 weeks           | -               | 8.3%                        |
| Okada et al., 2012 | 2 Males | 2 UCLP     | Extra               | +   | mean             | mean                                  | NA                    | 27-30 days          | 6 months          | -               | 0.0%                        |
|                   | 1 Female| 1 BCLP     |                     |     |                  |                                       |                       |                     |                  |                 |                             |
| Singh et al., 2012 | 7 Males | 8 ULCP     | Extra               | -   | 17-34            | mean                                  | 4-6                   | 6-8 weeks           | -                | -               | 30.0%                       |
|                   | 5 Female| 4 BCLP     |                     |     |                  |                                       |                       |                     |                  |                 |                             |

NA, Not available; Extra, Extraoral distraction device; Intra, Intraoral distraction device; SBG, Secondary bone grafting; Fixation plates, Fixation plates between maxillary segments after DOG
上顎骨延長法後に術後安定性が得られた片側性唇顎口蓋裂の1例

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この症例報告は、Rigid extraoral distractor (RED) Systemを用いて上顎骨延長法（DOG）を施行した片側性唇顎口蓋裂症例の術後安定性を報告する。

患者は右側唇顎口蓋裂の日本人女性。当科初診時年齢5歳10か月であった。7歳6か月時に、自家腸骨海綿骨片を用いた顎裂部二次骨移植術（SBG）が施行された。矯正歯科におけるⅠ期治療は、上顎骨の成長促進を目的として上顎前方牽引装置を用いたが、患者の協力が十分でなく良好な効果が得られなかった。Ⅱ期治療の目標は外科的矯正治療による中顔面の劣成長の改善であった。

RED Systemを用いたDOGにより上顎骨の前方移動が行われ、顔面の審美性と顎間関係の著しい改善が達成された。術後の上顎骨後戻り防止のために上顎骨前方牽引装置を9か月の長期間用いた。本症例では、術後の上顎骨の後戻りはほとんど見られなかった。

骨延長法前のSBGによる歯槽堤の連続性の獲得と、延長術後の長期の上顎骨前方牽引装置の使用は、唇顎口蓋裂症例における上顎骨延長法の前後の術後安定性に寄与する可能性がある。

キーワード：唇顎口蓋裂、術後安定性、上顎骨延長法

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