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Late maxillary distraction osteogenesis in a middle-aged cleft lip and palate patient: A multidisciplinary approach

Takuma Watanabe, Kazuo Yamashita, Motoki Katsube, Akihiko Yamaguchi, Makoto Hirota

ABSTRACT

Introduction: Patients with cleft lip and palate often present maxillary hypoplasia owing to early surgical scarring, necessitating surgical advancement to restore facial balance and functional occlusion. Maxillary distraction osteogenesis, particularly with internal devices, is an effective technique for advancing the maxilla and surrounding soft tissues in patients with cleft lip and palate. These patients often present with complex jaw deformities requiring a multidisciplinary approach.

Case Report: A 43-year-old male with unilateral cleft lip and palate presented with facial asymmetry and malocclusion. A multidisciplinary team performed maxillary distraction osteogenesis using an internal device, sagittal split ramus osteotomy, and plastic surgery. Maxillary distraction osteogenesis achieved maxillary advancement, clockwise rotation, and rolling. Subsequently, genioplasty and structural rhinoplasty were performed. The patient was satisfied with skeletal improvements and stable occlusion.

Conclusion: A middle-aged patient with unilateral cleft lip and palate, who required maxillary distraction osteogenesis was successfully treated through close collaboration among multiple professionals. Maxillary distraction osteogenesis effectively corrects maxillary malposition by enabling three-dimensional movement of the maxilla. A multidisciplinary team approach is crucial for addressing jaw deformity.

Keywords: Genioplasty, Oral and maxillofacial surgeon, Plastic surgeon, Rhinoplasty

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INTRODUCTION

Patients with cleft lip and palate (CLP) commonly present with a constricted maxillary dental arch and restricted anteroposterior growth of the maxilla, primarily owing to scar tissue formation during early surgical repair of the cleft [1]. As a result of maxillary hypoplasia, patients with CLP often require surgical advancement of the maxilla to normalize facial morphology and occlusion, as well as to improve psychosocial well-being [2].

Maxillary advancement via conventional Le Fort I (LFI) osteotomy or maxillary distraction osteogenesis (DO) is the most widely used surgical approach for correcting maxillary hypoplasia in patients with CLP [3]. Maxillary DO using internal devices has been shown to be an effective technique for achieving the

maxillary advancement in these patients [4]. Distraction osteogenesis is a surgical procedure that gradually elongates both the facial bones and associated soft tissues [5–7].

Adult patients with CLP frequently exhibit severe maxillary hypoplasia, maxillary arch constriction, missing teeth, and periodontal disease [3, 8]. As these complex problems affect multiple functions and aesthetic aspects, a multidisciplinary approach is essential to restore the patient's functional and esthetic integrity [9]. Actually, with regard to the comprehensive management of such cases, plastic surgeons, oral and maxillofacial surgeons, orthodontists, nurses, psychologists, and speech therapists can be required [9, 10].

Here, we report a case of a middle-aged patient with unilateral CLP who underwent maxillary DO and plastic surgery to address jaw deformity through a multidisciplinary approach.

CASE REPORT

A 43-year-old male patient with left-sided CLP was referred to our department with the chief complaint of dissatisfaction with the appearance of his face and occlusion. The patient had undergone cheiloplasty and palatoplasty during childhood. However, no alveolar bone grafting was performed. The patient maintained a stable social life and had no speech difficulties or psychosocial problems. Extraoral examination revealed a maxillo-mandibular retrusion with a midline shift to the left. Notable facial asymmetry and mental deviation toward the left side were also observed (Figure 1A and B). Intraoral examination revealed a crossbite in the left molar region due to narrow maxillary arch (Figure 1C). Dense scar tissue in the palatal area without evidence of oronasal fistulae was observed (Figure 1D). Panoramic radiography revealed the absence of multiple teeth, as well as caries and periodontitis of several teeth (Figure 2A). The frontal cephalogram showed an occlusal cant and left-sided deviation of the mandible (Figure 2B). Lateral cephalometric analysis revealed the following skeletal parameters: ANB angle, 2.7°; SNA angle, 79.5°; SNB angle, 76.9°; and FMA angle, 24.7°. Dental measurements included overjet (OJ) of 2.1 mm, overbite (OB) of 3.1 mm, upper incisor to Frankfort horizontal (U1 to FH) angle of 95.6°, and lower incisor to mandibular plane (L1 to MP) angle of 79.8° (Figure 2C). Three-dimensional computed tomography (CT) confirmed the absence of alveolar bone in the left maxillary anterior region (Figure 3A and B).

The clinical diagnosis was facial asymmetry and maxillo-mandibular retrusion accompanied by malocclusion. The patient had a large bone defect in the cleft region of the maxilla, and reconstruction of the continuity between the bilateral bone segments through bone grafting appeared to be highly challenging. After consultation with an orthodontist, we recommended surgical orthodontic treatment, including maxillary DO of both segments, to address maxillary retrusion and occlusal cant in a middle-aged patient with CLP. The patient ultimately consented to the proposed treatment. Preoperative orthodontic treatment was initiated, and the patient successfully completed all scheduled procedures over a 3-year period following the initial visit. At this stage, model surgeries were performed using plaster casts to achieve proper occlusion and improve facial aesthetics. The first and second splints as well as a palatal splint designed to retain bilateral bone segments during the surgery and distraction phases were fabricated in advance by a dental technician. Maxillary DO using the Maxillary Distractor System (Synthes, Paoli, PA) was planned to achieve 5 mm advancement at the anterior nasal spine, clockwise rotation of the maxilla, and 5 mm impaction at the right maxillary first molar through maxillary rolling. The maxillary distractor consists of a distractor body, an anterior footplate, and a posterior footplate, all made of stainless steel. Intraoperatively, alignment rods are used to facilitate the parallel placement of the device and to indicate the vectors of advancement. During the distraction period, the activation screwdriver is attached to the distractor body and turned counterclockwise, resulting in distraction at a rate of 0.5 mm per turn. For mandibular repositioning, a 4 mm setback at the right molar and a 7 mm advancement at the left molar were planned using sagittal split ramus osteotomy (SSRO). The patient was also evaluated for articulation and velopharyngeal function by a plastic surgeon and speech therapist using cephalometry and nasoendoscopy, both of which revealed no significant abnormalities. A 3D model was fabricated from preoperative CT data. The distractor footplates were bent and adapted to fit the contours of the maxillary alveoli and zygomatic buttress on the 3D model, and the vector of the distractor rods was determined (Figure 4A and B).

After hospitalization, an Le Fort I osteotomy was performed under general anesthesia. During the procedure, a palatal splint was secured to the maxillary teeth using a wire, and the distractor footplates were bilaterally fixed to the zygomatic buttresses. Intraoperatively, the distractors were activated using an activation screwdriver to confirm their function and mobility in the maxilla and to assess the distraction vector using alignment rods (Figure 5). Postoperatively, distractor settings were evaluated using a lateral cephalogram (Figure 6A). After a latency period of six days, distraction was initiated at a rate of 1 mm per day (two turns). During the distraction phase, re-evaluation by the plastic surgeon and speech therapist revealed no deterioration in velopharyngeal function. Distraction was completed over 10 days, achieving advancement of 9 mm on the right side and 10 mm on the left side. The amount of distraction was set to be greater on the left than on the right side to facilitate smooth maxillary rolling. Maxillary advancement was evaluated using lateral cephalography (Figure 6B). Twenty-one days after the initial surgery, the palatal splint and distractors were surgically removed,

and SSRO was performed. The maxilla was positioned using a prefabricated first splint and fixed bilaterally with plates. Similarly, the mandible was positioned using a prefabricated second splint and secured with screws. During the distraction and perioperative phases, appropriate oral care was provided by a dental hygienist, and the patient's postoperative course was uneventful. After the patient was discharged on the 42nd hospital day, the occlusion was stabilized through postoperative orthodontic treatment.

The patient sought further improvement in facial appearance and underwent genioplasty performed by a plastic surgeon 10 months after the second surgery. Intraoperatively, a horizontal osteotomy was performed after marking both the deviated and desired midlines (Figure 7A). Subsequently, the chin segment was shifted approximately 5 mm to the right and fixed with a plate in a position where the right part of the segment was yawed anteriorly by approximately 3 mm (Figure 7B). Furthermore, seven months later, a plastic surgeon performed a structural rhinoplasty. After harvesting the costal cartilage from the rib cage, the cartilage was shaped, prepared, and placed in the nose to augment and reshape the nasal structure (Figure 8).

Currently, eight years after the initial visit, facial asymmetry has improved (Figure 9A and B), and the patient's occlusion remains stable without velopharyngeal incompetence (Figure 9C). A panoramic radiograph revealed the continued absence of several teeth (Figure 10A), while frontal and lateral cephalograms showed improvements in the maxillary occlusal cant, mandibular deviation, and maxillo-mandibular retrusion (Figure 10B and C). The patient was satisfied with the postoperative outcome, and prosthetic treatment is planned for the future.

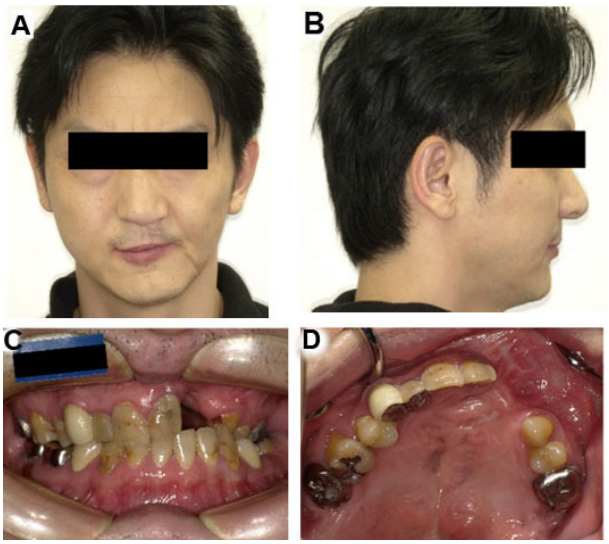

Figure 1: Frontal (A) and lateral (B) facial photographs showing facial asymmetry and maxillo-mandibular retrusion, respectively. Intraoral frontal (C) and palatal (D) photographs showing a crossbite in the left molar region and alveolar defects in the left anterior region.

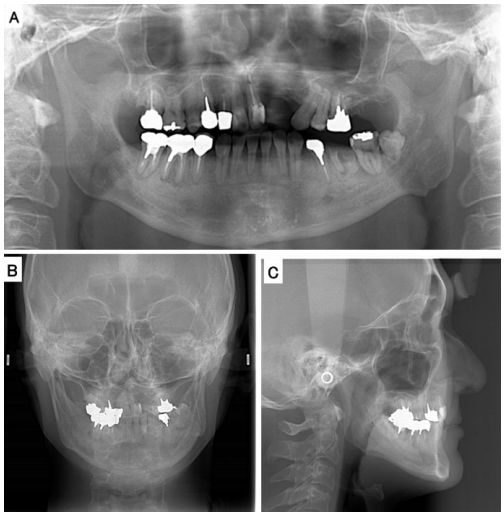

Figure 2: Panoramic radiograph showing the absence of multiple teeth, as well as several teeth with severe caries and periodontitis (A). Frontal (B) and lateral (C) cephalograms showing a maxillary occlusal cant, mandibular deviation to the left side, and maxillo-mandibular retrusion.

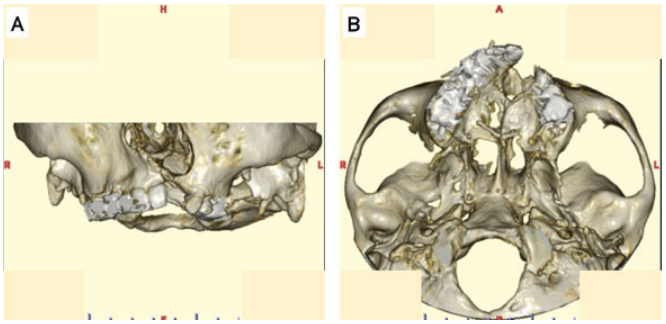

Figure 3: Three-dimensional computed tomography (3DCT) images showing the absence of alveolar bone in the maxillary left anterior region (A, B).

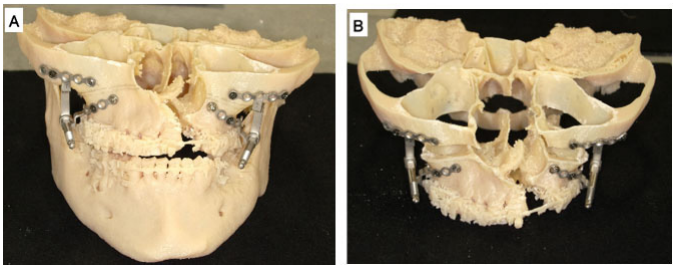

Figure 4: Photograph of a 3D model from preoperative CT images showing positioned distractor foot plates and rods (A, B).

Figure 5: Intraoperative photograph showing assessment of the distraction vector using paralleling pins.

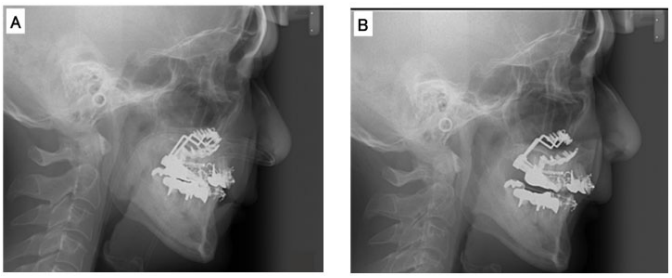

Figure 6: Lateral cephalograms obtained before (A) and after (B) distraction, demonstrating maxillary advancement.

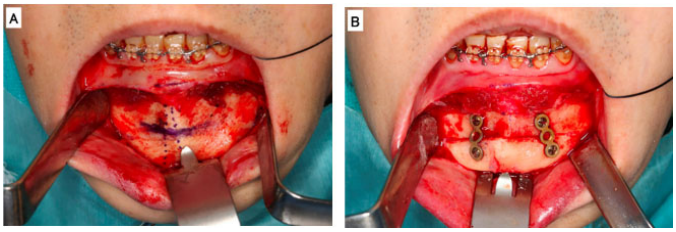

Figure 7: Intraoperative photograph showing horizontal osteotomy and plate fixation after marking both the deviated and ideal midlines (A, B).

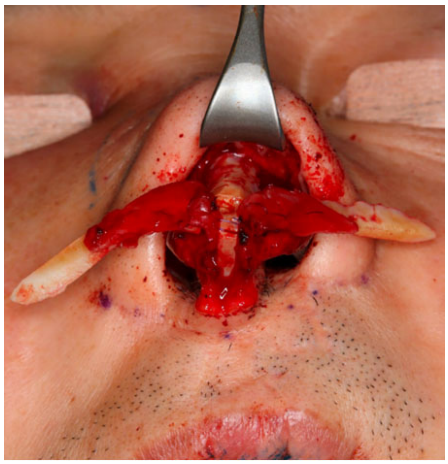

Figure 8: Intraoperative photograph showing placement of harvested costal cartilage in the nose to augment and reshape the nasal structure.

Figure 9: Frontal (A) and lateral (B) facial photographs showing improvements in facial asymmetry and maxillo-mandibular retrusion, respectively. Intraoral frontal photograph showing stable occlusion and improvement in the crossbite in the left molar region (C).

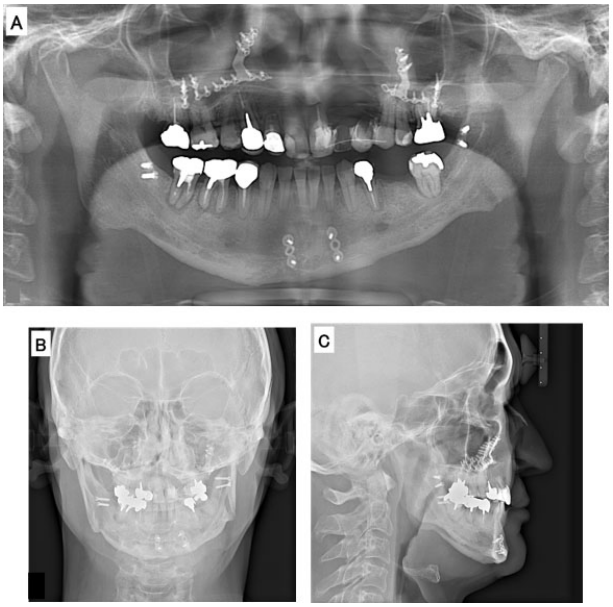

Figure 10: Panoramic radiograph showing the continued absence of several teeth, consistent with previous findings (A). Frontal (B) and lateral (C) cephalograms showing improvements in maxillary occlusal cant, mandibular deviation, and maxillo-mandibular retrusion.

DISCUSSION

The management of this case highlights important clinical considerations. Maxillary DO in patients with unilateral CLP allows sufficient mobilization of the maxilla and can be effective for correcting occlusal cant. Facial asymmetry, maxillo-mandibular retrusion, and malocclusion associated with unilateral CLP can be successfully treated through a multidisciplinary approach.

Patients with CLP usually present with a depressed midface and a narrow maxillary dental arch because of dentofacial anomalies compounded by scar tissue from early surgical repair of CLP [6]. Scarring from previous surgeries involving the lip, alveolus, palate, and pharynx can hinder mobilization of the maxilla during surgery and increase the risk of maxilla contraction postoperatively [6]. For many oral and maxillofacial surgeons, a 10 mm advancement is considered the upper limit for a single-stage LFI procedure because exceeding this threshold increases the risk of postoperative instability and relapse [11]. In patients with maxillary hypoplasia secondary to CLP repair, the safe advancement range may be further reduced to 6 mm or less owing to local scarring and preexisting velopharyngeal incompetence [11, 12]. In the present case, the maxillary advancement was limited to 5 mm, and no postoperative deterioration in velopharyngeal function or significant relapse was observed.

Maxillary advancement with DO has become a viable option for patients with CLP experiencing medium to severe midfacial or maxillary deficiencies [1, 11, 13]. Distraction osteogenesis gradually lengthens both

the bone and soft tissue, greatly reducing soft tissue restrictions around the distracted segment and lowering relapse rates [11]. The LFI-DO technique using an internal device is considered a predictable, stable, and convenient option for correcting a severe, unfavorable intermaxillary relationship [11]. Adequate skeletal stability during the active distraction and consolidation phases, as well as a sufficiently long consolidation period, are required for proper mineralization of the distraction zone [14]. Previous studies have suggested that consolidation periods range from eight weeks to eight months [2–4, 7, 11]. Internal devices may be removed earlier if clinically indicated, depending on the distraction amount and occlusal loading conditions [7, 11]. However, the active DO and consolidation phases can be socially, physically, and psychologically challenging for patients [2]. It is an intensive treatment that can cause significant anxiety and short-term distress, requiring frequent follow-up visits during the distraction phase [7, 15, 16]. In the present case, the consolidation period was limited to three weeks, and all procedures were performed during hospitalization. This decision was based on several factors: favorable bone union owing to impaction contact on the right side of the maxilla; the primary objective of increasing soft tissue mobility rather than promoting bone regeneration; concerns over long-term physical and psychological burden; and the need for frequent care during the distraction phase.

Regarding the amount of distraction, a previous study on Zurich maxillary distractors reported that activation ranged from 4.3 to 14.8 mm, while actual advancement at the anterior nasal spine ranged from 2.3 to 11 mm [17]. The resulting distraction ratios ranged from 51.1% to 95.0%, with a mean of 69.5% [17]. In the present case, owing to strong soft tissue tension caused by severe scar contracture in this middle-aged patient, distraction efficiency was expected to be relatively low. Therefore, distraction of 9–10 mm was performed to achieve and maintain a 5 mm maxillary advancement, based on an estimated distraction ratio of approximately 50%. This strategy enabled smooth 3D repositioning of the maxilla to the predetermined position location.

Multidisciplinary treatment involving professionals from various specialties who collaborate to evaluate and manage patients with CLP should be considered essential [18]. Intervention for patients with CLP is based on a team approach that includes pediatric dentists, restorative dentists, oral and maxillofacial surgeons, orthodontists, plastic surgeons, and speech therapists [13]. Oral and maxillofacial surgeons perform orthognathic surgeries—including maxillary expansion, DO, and mandibular setback—to normalize the occlusion between the maxillary and mandibular arches [18, 19]. Orthodontists manage dental and skeletal malocclusions and promote proper jaw relationships to improve dental and facial aesthetics as well as the functional efficiency of the dentition [18, 20]. Plastic surgeons not only repair the lip, palate, and facial anomalies but also improve overall facial aesthetics, feeding, and speech through surgery [18, 21]. Speech therapists evaluate feeding and swallowing, general development, speech, language, resonance, and velopharyngeal function; they provide treatment recommendations and therapy for communication and feeding-related issues [18]. Notably, overlap exists between the roles of plastic surgeons and oral and maxillofacial surgeons, particularly in procedures such as lip and palate repair, orthognathic surgery, and secondary aesthetic enhancements. Effective teamwork helps avoid conflicts and ensures cohesive care [18]. Clear communication among all providers is critical to achieving successful functional and aesthetic outcomes [22]. In the present case, the oral and maxillofacial surgeon performed the orthognathic surgery, the orthodontist managed the pre- and postoperative orthodontic treatment, the plastic surgeon conducted the genioplasty and rhinoplasty, and the speech therapist carried out speech evaluations. As the patient had no psychosocial issues, compliance with our multidisciplinary treatment was favorable. A successful outcome was achieved that accommodated his preferences for facial appearance and occlusion.

CONCLUSION

In this case, a severe jaw deformity associated with CLP was successfully treated through close collaboration among oral and maxillofacial surgeons, orthodontists, plastic surgeons, and other healthcare professionals. Maxillary DO allows for sufficient mobilization of the maxilla, smoothly enabling three-dimensional repositioning to the predetermined location by alleviating soft tissue tension caused by scarring. A multidisciplinary approach is essential for the definitive treatment of patients with CLP, addressing both esthetic and functional issues.

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Author Contributions

Takuma Watanabe – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Kazuo Yamashita – Acquisition of data, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Motoki Katsube – Acquisition of data, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Akihiko Yamaguchi – Conception of the work, Design of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Makoto Hirota – Conception of the work, Design of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission
The corresponding author is the guarantor of submission.

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None.

Consent Statement
Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest
Authors declare no conflict of interest.

Data Availability
All relevant data are within the paper and its Supporting Information files.