Patient With Cleft Maxillary Hypoplasia Who Underwent Distraction Osteogenesis and Conservative Therapies to Postoperative Velopharyngeal Insufficiency

Katsu Takahashi, DDS, PHD, Khaled Gamal Dahy, MD,*† Kazuyuki Saito, DDS,* Hiromu Masuoka, MD,‡ Shigehiko Suzuki, MD, PHD,§ and Kazuhisa Bessho, MD, PHD**

Abstract: The authors performed distraction osteogenesis using The Maxillary Distractor System (SYNTHES) to maxillary hypoplasia patient with cleft lip palate, and consequently improved the aesthetic complexion of the patient. Velopharyngeal insufficiency developed after bone elongation; the authors improved the insufficiency with conservative therapies such as articulatory training using the bulb attached palatal lift prosthesis. And then the authors got successful and accepted postoperative speech outcome.

Key Words: Cleft lip and palate, maxillary distraction osteogenesis, orthognathic surgery, velopharyngeal insufficiency

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Maxillary hypoplasia is a common presentation of patients with repaired cleft lip palate. It is either a result of cleft its self, the genotype of the patient or maxillary growth affected by scarring associated with various procedures that the patients undergo during rehabilitation.[1] Management of this condition has been a challenge to reconstructive surgeons because velopharyngeal insufficiency (VPI) will develop after bone elongation and so speech outcome will be affected.[2]

In this clinical report, we clinically applied a new internal device for maxillary distraction osteogenesis, The Maxillary Distractor System to cleft maxillary hypoplasia patient.

PATIENT PRESENTATION

A 19-year-old male had suffered from malocclusion. The patient had a history of left cleft lip and palate that repaired at the age of 1-years-old in a certain hospital; then palatoplasty was done when he was 3 years old. After that, the patient missed follow-up and became aware of malalignment of teeth and malocclusion of the maxilla when he was 14-year-old, and he visited our department.

Clinical examination revealed that he had maxillary retrusion. We did speech and voice assessment, after that hearing test, snort mirror inspection, nasopharyngeal fiberscope, and lateral cephalometric radiograph to assess the preoperative velopharyngeal function (VPF).

We performed Le Fort I osteotomy and applied maxillary distraction osteogenesis apparatus under general anesthesia. Maxillary Distractor System used in this case composed of distractor body, anterior plate, and posterior plate. We set the planned osteotomy line to three-dimensional maxilla model created from the captured computed tomography images before surgery, we previously bent the device to fit well into this model (Fig. 1). After 6 days latency period, we started the maxillary distraction osteogenesis. Maxilla moved forward 13 mm (0.5–1.0 mm per day). The period was 14 days. It is difficult to control direction by the distraction osteogenesis alone technically and we need to correct the mandibular asymmetry of this case in addition to severe maxillary hypoplasia. We removed the osteogenesis distraction apparatus and then did plate fixation for the maxilla and bilateral sagittal split ramus osteotomy (SSRO) under general anesthesia. The mandible moved backward 1 mm and 2 mm shift to the right side because interincisal line shifts 2 mm to left side without use of bone graft. After that, intermaxillary fixation was performed and maintained for 3 days. We gained good occlusal relationship in both the anterior and posterior teeth (Fig. 2).We applied Bulb-PLP (Fig. 3), and then started articulatory training. Evaluation of the velopharyngeal function was performed after the end of bone elongation and after applying Bulb-PLP.

Articulation dynamics in lateral cephalometric radiograph showed that in preoperative status, no morphological abnormalities in the soft palate and pharynx during rest. But when the patient pronounced consonant (s), the soft palate elevated, and the gap in the posterior wall of the pharynx closed and when the patient pronounced the vowel (a), the degree of soft palate elevation was slightly weak; there is a small gap in the posterior pharyngeal wall. After the end of DOG, when the patient pronounces both vowel and consonant, the gap is larger than before the operation, and the soft palate elevation is seldom observed when the patient pronounces consonant (Fig. 4).

Nasofiber-optic examination revealed that in preoperative status, bubbling was seen in velopharyngeal closure, it was diagnosed as very mild VPI. Immediately after the end of bone distraction, compared with preoperative status, the distance between the soft palate and the posterior pharyngeal wall becomes far, so it is diagnosed as severe VPI, but after the articulatory training start (after applying Bulb-PLP) the nasopharynx was almost closed (Fig. 5).

Articulatory training was performed in the following 3 ways, voiced blowing method, tube vocalization method, and articulation training under Bulb-PLP fitting. Because we experienced small improvement of dysarthria after simple PLP usage, by changing it

From the *Department of Oral and Maxillofacial Surgery, Graduate School of Medicine, Kyoto University, Kyoto, Japan; †Department of Otolaryngology, Sohag University, Sohag, Egypt; ‡Department of Plastic and Reconstructive Surgery, Kansai Electric Power Hospital, Osaka; and §Department of Plastic and Reconstructive Surgery, Graduate School of Medicine, Kyoto University, Kyoto, Japan.

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Address correspondence and reprint requests to Khaled Gamal Dahy, MD, Department of Oral and Maxillofacial Surgery, Graduate School of Medicine, Kyoto University, Syogoin Kawaramachi, Sakyo-Ku, Kyoto 606-8507, Japan; E-mail: khgamal@kuhp.kyoto-u.ac.jp

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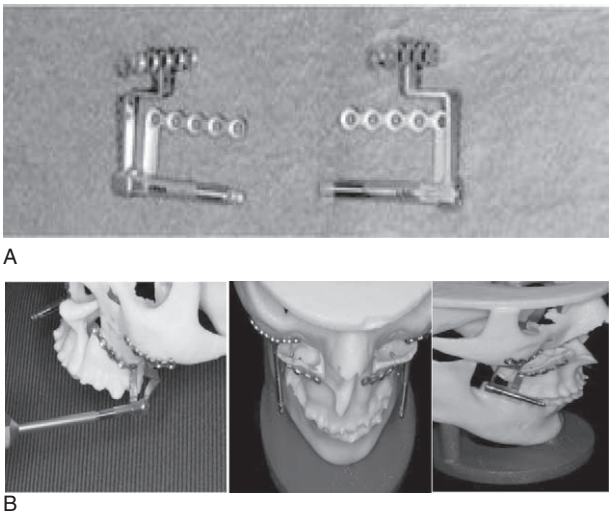

FIGURE 1. Maxillary distractor system of SYNTHES. (A) It is composed of distractor body, anterior plate, and posterior plate. (B) After bending the device and fitting it well into surgery model.

into the Bulb-PLP, dysarthria improved. After that Pharyngeal veloplasty was performed with the successful speech outcome.

DISCUSSION

Maxillary distraction osteogenesis devices are broadly divided into those: of the rigid external fixation type (RED system) and the rigid internal fixation type (Zurich distractor system).[3]

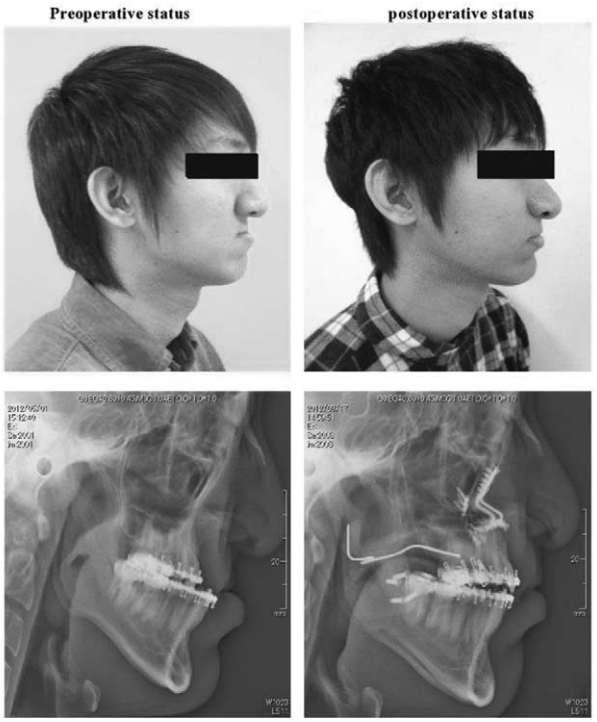

FIGURE 2. Face photos and lateral cephalometric radiograph before and after surgery (after the end of bone elongation, maxillary plate fixation, and bilateral SSRO).

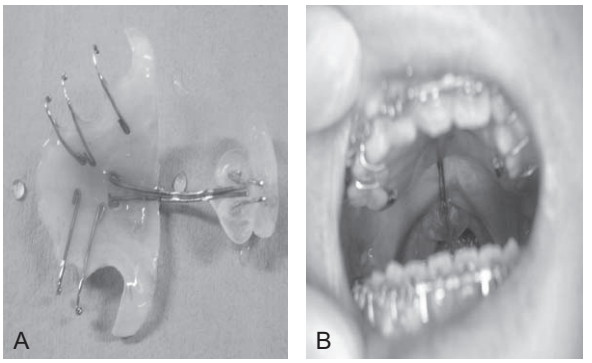

FIGURE 3. Bulb attached palatal lift prosthesis. (A) Before fitting. (B) After fitting.

The RED system device is adjustable, offering the ability to change the vertical and horizontal vectors of distraction, so the devices can perform three-dimensional corrections at any time, without discomfort to the patient during the distraction process. It has no limits on distraction distance, so its extension distance is high.[1] While it can correct direction during extension, the structure of the device is complicated, the force of the extension is hard to reach straight to the bone, the perforating wound, and local fractures of the skull due to the penetration of stabilizing pins intracranially and there is a problem in a social life with its appearance.[4]

The Maxillary Distractor System used in this patient is the internal fixation elongation device that became available in 2008 in Japan. It can settle up to a maximum of 25 mm distraction distance by the combination of its parts, so it can compensate for the disadvantages of the conventional internal fixation distraction devices by being with a limited distraction distance so that this device can increase the needs for internal fixation distraction device selection.

The maxillary elongation cases were 6 cases for the past 5 years in our hospital. The RED system cases were 5 cases of them. The Maxillary Distracter System case was only this case. Comparing the cases using the RED system with the case using Maxillary Distracter System, although there was no much difference in the average maxillary movement amount, there was a large difference in the extension efficiency. In our case, the extension efficiency of The Maxillary Distracter System is about 85%.

Also, in 4 cases of 5 cases using the RED system, the entire upper jaw bone rotated upward during bone elongation, measures to

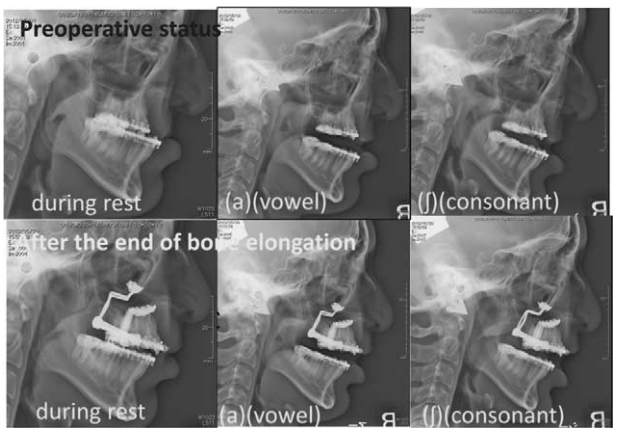

FIGURE 4. Articulation dynamics in lateral cephalometric radiograph before surgery and after the end of bone elongation.

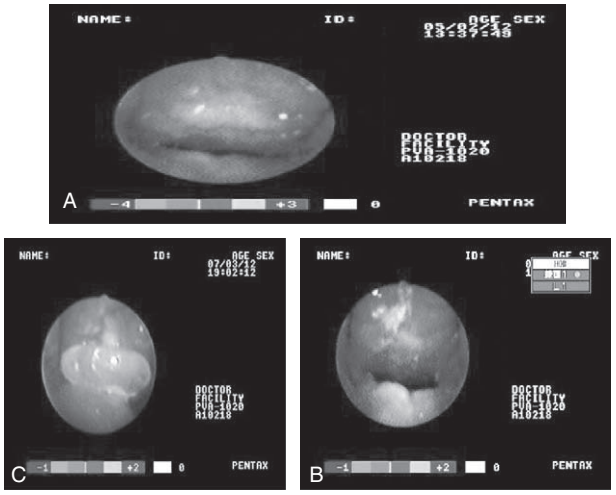

FIGURE 5. Nasofiberoptic finding. (A) Preoperative. (B) After the end of bone distraction. (C) After Bulb-palatal lift prosthesis fitting.

modify the vector of bone distraction downward had been taken. Harada et al[5] reported that the number of activations required is about 4 times as much as the amount of planned maxillary distraction. This difference related to various factors, one of them is the flexibility of the external traction hooks attached to the maxillary dentition, and another may relate to the amount of scar tissue present. The mechanical efficiency of the system decreases; therefore, more force is required to mobilize the cleft maxilla.

Maxillary osteotomy impacts negatively on VPF with increases in nasality and nasal turbulence in patients with cleft lip palate.[6] The cause of the VPI in repaired cleft palate patients may be due to insufficient muscle reconstruction when we performed push back method (due to excessive palatal scar tissue). Because separation of the oral mucosa and nasal mucosa of the Levator palati muscle is insufficient, it interferes with the extensibility of the Levator palati muscle, reduces the pushback, and Levator palati muscle does not move backward and goes close to the front. And also the VPI will increase after maxillary advancement due to a short, soft palate, and deep pharynx.[7]

By strong bone DOG, such as the present case, the soft palate pulled forward at the same time, deterioration of VPI occurred. Currently, an Intravelar Veloplasty operation in which oral mucosa, the Levator palati muscle, and nasal mucosa respectively divided into 3-layer recommended as the treatment of choice for VPI correction. By this method, when the maxillary bone moves forward, the Levator palati muscle does not move forward together, so we can prevent deterioration of the VPF.[8] Like this case, nasopharyngeal fiberscope evaluation of the soft palate revealed a high tension which is a strong example of severe scar tissue; it suggested caring about worsening of postoperative VPI.

In general, we take measures to improve the articulation by performing articulatory training using the soft palate elevation device and a PLP as a conservative therapy or by performing the pharyngeal valvoplasty operation as a surgical treatment. Satoh et al[9] reported that by using a lateral cephalometric radiograph in repaired cleft palate patients, they measured morphological ratio (soft palate length S/depth P of the pharyngeal cavity) and evaluated the velopharyngeal closure before and after surgical treatment; there is deterioration in their velopharyngeal closure and speech outcome postoperatively. Hoopes et al[10] defined the velar length-to-pharyngeal depth ratio (adequate ratio) as a relative length of the soft palate, and reported the ratio to be 1.35 in normal adults and 1.05 in patients with repaired cleft palate showing velopharyngeal incompetence.

Satoh et al[11] reported that adequate ratio indicated that how adequately the velum closes the pharyngeal depth. He suggested that the normal adequate ratio was stable, ranging from 1.2 to 1.4 which was consistent with the findings reported by Subtelny[12] and Hoopes et al.[10]

Morphological ratio after bone extension, in this case, was S/P = 1.11, which indicates good VP closure. It has passed 2 months after the start of the articulatory training; dysarthria improved. After pharyngeal Veloplasty, dysarthria improved more and nowadays the patient is on articulatory training, and we are going to continue follow-up.

We performed DOG for the cleft maxillary hypoplasia patient, after that, worsening of the VPF developed. By the articulatory training after surgery, dysarthria improved and after pharyngeal Veloplasty, we got an excellent speech outcome. We reported the summary, according to bibliographic consideration.

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