Orthodontic-surgical approach for treating skeletal Class III malocclusion with severe maxillary deficiency with isolated cleft palate

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ABSTRACT

Orthodontic treatment in patients with orofacial cleft such as cleft lip and palate or isolated cleft palate is challenging, especially when the patients exhibit severe maxillary growth retardation. To correct this deficiency, maxillary expansion and protraction can be performed in the first phase of orthodontic treatment. However, in some cases the malocclusion cannot be corrected by these procedures, and thus skeletal discrepancy remains when the patients are adolescents. These remaining problems occasionally require various orthognathic treatments according to the degree of the discrepancy. Here we describe one case of a female with isolated cleft palate and hand malformation who exhibited severe maxillary deficiency until her adolescence and was treated with multiple orthognathic surgeries, including surgically assisted maxillary expansion (SARPE), maxillary LeFort I and mandibular bilateral sagittal split osteotomy (BSSO) in order to correct severe skeletal discrepancy and malocclusion. The treatment resulted in balanced facial appearance and mutually protected occlusion with good stability. The purpose of this case report is to show the orthodontic treatment outcome of one patient who exhibited isolated cleft palate and subsequent

severe skeletal deformities and malocclusion which was treated by an orthodontic-surgical approach.

INTRODUCTION

Isolated cleft palate (CP) is one of the common forms of orofacial cleft, occurring in about 0.3 in 1000 live births, with some variation depending on race (Dixon et al., 2011; Mossey and Modell, 2012). In order to achieve normal orofacial functions such as feeding and speech, surgical palate closure is essential at an age of around one or two. It is well known that the scar produced by this surgical intervention can cause maxillary growth retardation that results in vertical as well as horizontal upper jaw deficiency (Ishikawa et al., 1998; Iwasaki et al., 2014; Smahel et al., 2003). For these reasons, CP patients frequently require orthodontic treatment in order to correct various malocclusions caused by maxillary deficiency (Baek et al., 2002; Ishikawa et al., 1998). The treatment often requires long-term, complex, comprehensive treatment, sometimes including orthognathic surgery which requires an interdisciplinary team approach by orthodontists and craniofacial surgeons (Susami et al., 1996). Because of the wide diversity of malocclusions resulting

from various types of cleft, numerous reports and discussions of CP treatments are essential in order to establish efficient and stable treatment protocols in many institutes and clinics (Iwasaki et al., 2014). This case report describes one example of successful treatment of a CP patient who exhibited severe skeletal discrepancy that resulted in midfacial deficiency, skeletal Class III malocclusion, total cross bite and severe crowding. Additionally, this patient exhibited general symptoms such as a malformed right hand, short stature, coxarthrosis, cardiac murmur, and strabismus, which indicate the possibility of a genetic cause of the CP.

CASE HISTORY

A 13-year-old girl first came to the Department of Orthodontics at Osaka University Dental Hospital in Suita City with complaints of mandibular protrusion and occlusal disturbance. She was referred for orthodontic treatment by her hospital's pediatrics department because of midface deficiency and anterior crossbite. She had a history of pushback palatoplasty for cleft palate repair at the age of one. She also exhibited several general symptoms, such as a malformed right hand, short

stature, coxarthrosis, cardiac murmur, and strabismus.

At the age of 13 years 2 months, maxillary protraction was initiated with a reverse headgear and lingual arch. At the same time, growth hormone treatment was carried out for 6 months in order to promote an increase of the patient's stature. Even after 2 years of using the reverse headgear, skeletal class 3 and partial crossbite remained because of greater mandibular growth than the promoted maxillary growth during the first phase of orthodontic treatment. After the maxillary and mandibular growth spurt confirmed by serial lateral cephalogram superimposition, all records were taken for use in the diagnosis for planning second-phase treatment at the age of 18 years 4 months.

The extraoral examination showed severe midfacial deficiency and concave type profile with mild lip incompetency. A large buccal corridor space could be seen upon posed smile (Fig 1). The occlusion showed right-side lateral and anterior cross bite. The upper dental arch showed lateral constriction and severe crowding with blocked-out right lateral incisor and left canine. The lower dental arch exhibited moderate crowding (Fig 1). The maxillary dental midline was deviated 1.5 mm to the left relative to the facial midline (Fig 1). The mandibular midline was deviated 3.0 mm to the

right relative to the facial midline (Fig. 1). No symptoms or signs of any temporomandibular joint disorder were observed.

The panoramic radiograph showed congenitally missing maxillary left lateral incisor as well as right upper wisdom tooth (Fig. 1).

The cephalometric analysis showed a skeletal Class III relationship (ANB, -6.6°) with a retrusive maxilla (SNA, 68.7°). The mandibular plane angle was larger than the norm (FMA, 39.0°). The maxillary incisors were palatally inclined (U1-FH, 98.6°), and the mandibular incisors showed lingual inclination (L1-MP, 62.5°) (Table).

There was no evident speech problem such as hypernasality or velopharyngeal insufficiency at the beginning of the second-phase treatment.

TREATMENT PLAN AND PROGRESS

The treatment objectives were to correct the constricted maxillary arch and lateral cross bite, improve the midfacial deficiency with concave type facial profile associated with the skeletal Class

III relationship and the anterior crossbite, and to improve the asymmetrical facial appearance caused by mandibular deviation and improve the crowding of the upper and lower jaws to obtain proper occlusion. The following treatment plan was proposed: (1) orthopedic maxillary expansion with surgically assisted rapid palatal expansion (SARPE) (2) placement of preadjusted edgewise appliances in both dental arches to align the dentition, (3) forward movement and rotation of maxilla with Lefort1 osteotomy (4) mandibular setback and rotation by bilateral sagittal split ramus osteotomy (BSSO) (5) obtain mutually protected occlusion (6) retention.

At 18 years 4 months, the second phase of orthodontic treatment was initiated by expansion of the maxilla using SARPE (Fig. 2). The osteotomy line was drawn at the middle of the maxillary incisor in order to obtain symmetrical expansion. A Hyrax-type appliance was cemented to the first premolar and the first molar after an osteotomy was performed. A total of approximately 12.0 mm of expansion was achieved (Fig. 3). All of the wisdom teeth that existed were extracted during this SARPE. After a stabilizing period of 6 months, the Hyrax-type appliance was replaced with a transpalatal arch. Then pre-adjusted fixed appliances were bonded on the maxillary and mandibular

dentition to align and level the teeth, and preoperative orthodontic treatment was started. The retroclined maxillary and mandibular incisors were corrected for preparation for the orthognathic surgery. The preoperative orthodontic treatment continued until the patient underwent orthognathic surgery at the age of 21 years 8 months (Fig. 3).

The patient underwent two jaw orthognathic surgeries after the orthodontic preparation. The maxilla was set forward 6 mm to improve the midfacial deficiency and rotated horizontally for midline correction with LeFort I osteotomy. The mandible was set back 1 mm and rotated horizontally for midline correction with BSSO.

After the postoperative orthodontic treatment was continued for 7 months to obtain mutually protected occlusion, all appliances were removed. Begg-type retainers were placed on both arches for retention, and posttreatment records were taken after debonding of all appliances (Fig. 4).

TREATMENT RESULTS

The first phase of orthodontic treatment prevented further progression of the skeletal Class III relationship and the anterior crossbite. However, lateral and anterior cross bite remained at the time of the second phase of orthodontic treatment. The SARPE corrected the laterally constricted maxillary arch and posterior crossbite with large buccal corridor space (Fig. 3). The surface alveolar bone of the upper first molar after SARPE still remained after the appliances were removed (Fig. 2, red arrowhead). The subsequent LeFort I osteotomy improved the midfacial deficiency, skeletal Class III relationship and asymmetrical facial profile together with setting back of the mandible by BSSO. The posttreatment facial photographs showed a preferable straight-type facial profile without lip incompetency (Fig. 4). Intraoral photographs showed mutually protected occlusion with Class I canine relationships and proper overjet and overbite. The molar relationship was Class I on the right side and Class II on the left side (Fig 4). The maxillary crowding was eliminated by the surgical lateral expansion and proclination of the incisors. The maxillary left canine-premolar position was transpositioned at the beginning of the treatment, and the maxillary left premolar was reshaped to resemble a lateral incisor (Fig 4). Mandibular crowding was eliminated by proclination of the

mandibular incisors. The posttreatment cephalometric analysis showed a skeletal Class I relationship with ANB angle of 1.2° (Figs. 4,5 and Table). The lingual inclinations of both maxillary and mandibular incisors were corrected with U1-FH from 98.6° to 110.2° and L1-FH from 78.6° to 52.3°, respectively. The IIA angle (122.1°) was almost an ideal value at the end of the treatment (Figs. 4,5 and Table).

DISCUSSION

Controlling maxillary deficiency is one of the main purposes of orthodontic treatment of CP patients. For correcting the maxillary transverse deficiency, many ways of expanding the maxillary arch for the first phase of treatment have been described, for example the fixed quad helix (Henry, 1993). For a severe maxillary transverse deficiency in post-adolescent patients, surgical maxillary expansion can in some cases be the first-choice treatment. In this case we used SARPE because the patient exhibited cleft palate with severe maxillary transverse deficiency (Suri and Taneja, 2008; Susami et al., 1996; Woods et al., 1997). Segmental osteotomy is another way of orthopedic

maxillary expansion. However, for present case we needed more than 8 mm expansion and segmental osteotomy could have resulted in an unstable result (Koudstaal et al., 2005; Suri and Taneja, 2008). To improve the midfacial deficiency, we employed Lefort1 osteotomy for maxillary forward movement. There are also other methods which have shown successful forward movement of the maxilla in facial cleft patients with severe maxillary deficiency such as distraction osteogenesis (DO) or maxillary anterior segmental distraction osteogenesis (MASDO) (Hirata et al., 2016; Kageyama-Iwata et al., 2017; Kloukos et al., 2016). Any of these methods could have been utilized in this case, but we chose LeFort I osteotomy because of its relatively simple and less invasive procedure. Several reports suggested possible side effects, such as worsened velopharyngeal insufficiency, after maxillary advancement (Guyette et al., 2001; Nohara et al., 2006; Satoh et al., 2004). However, we did not detect any speech problem in this case before or after orthodontic treatment. The treatment outcomes of different surgical methods for maxillary advancement still remain elusive and thus require further investigation. Precise and comprehensive diagnostic procedures are essential for choosing the appropriate surgical procedure for maxillary

advancement.

About 15% of orofacial cleft cases can occur as one of the phenotypes of syndromic disease (Mossey and Modell, 2012). Interestingly, isolated CP tends to have more syndromic features than cleft lip and/or palate (Mossey and Modell, 2012). Even among non-syndromic orofacial cleft patients, we encounter some with other less-defined general symptoms. Thus, it is also important to mention other general phenotypes of the patients in order to improve our understanding of orofacial cleft. In this case, the patient exhibited several general symptoms, such as a malformed right hand, short stature, coxarthrosis, cardiac murmur, and strabismus with isolated CP in the oral region (Fig. 6). Notably, some previous reports showed concurrent facial cleft and hand malformation in humans with specific gene mutations (Barrow et al., 2002; Buss et al., 1995; Wattanarat and Kantaputra, 2016). However, there are only a few reports about the progress of orthodontic treatment of CL/P patients with malformed hands (Agnieszka et al., 2012). For these reasons, this case report should become a useful reference for future orthodontic treatment in patients with similar symptoms. Since many genes are commonly used in embryonic craniofacial and limb development, it is possible that

there was a genetic contribution to this patient's condition.

CONCLUSIONS

For isolated CP patients, comprehensive orthodontic treatment should be performed in collaboration with treatments in other departments. Particularly in patients with severe maxillary growth retardation and crowding, as in this patient, orthodontic treatment becomes complicated, and accurate diagnosis is essential. In cases of severe skeletal Class III malocclusion due to severe maxillary deficiency, most patients require upper and lower jaw osteotomy in the second phase of orthodontic treatment. In the present case, we utilized SARPE to improve the constricted maxillary alveolar width and severe crowding. Further improvement of the anterior-posterior intermaxillary relationship was achieved by maxillary advancement and setting back of the lower jaw after preoperative orthodontic treatment. The occlusion and facial aesthetics of this patient were substantially improved and she showed high satisfaction at the end of the treatment.

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