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Case Report

Orthognathic surgery with iliac bone grafting for an interpositional gap in a patient with type III hemifacial microsomia: A case report

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ABSTRACT

Hemifacial microsomia (HFM) is characterized by uni- or bilateral microtia and hypoplasia of the mandible, orbits, facial nerve, and adjacent soft tissues. Patients with Pruzansky–Kaban type III HFM show the most severe facial deformities and often encounter difficulty obtaining treatment. In recent years, orthognathic surgery for HFM-related deformities has often been performed after the patient has stopped growing. However, few detailed reports have described the difficulties of orthognathic surgery for patients with type III HFM. This report describes the case of a patient with type III HFM who underwent three unilateral mandibular reconstructions while still growing, including autogenous reconstructions and secondary distraction osteogenesis, followed by orthognathic surgery with iliac bone grafting for an interpositional gap between

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the proximal and distal segments after she had stopped growing to improve facial asymmetry and malocclusion.

Hemifacial microsomia (HFM) is the second most common congenital craniofacial anomaly and is characterized by microtia and hypoplasia of the mandible, orbits, facial nerve, and adjacent soft tissues.[1,2] Patients with type III HFM show the most severe facial deformities, due to the absence of a ramus, temporomandibular joint (TMJ), and glenoid fossa, and often encounter difficulty receiving treatment for facial asymmetry, even after multiple surgeries since childhood. Such early surgeries make corrections after the patient has stopped growing even more technically difficult to achieve.

We report the case of a girl with type III HFM who had undergone three unilateral mandibular reconstructions while still growing, including autogenous reconstruction and distraction osteogenesis. These were followed by orthognathic surgery and iliac bone grafting for an interpositional gap between the proximal and distal segments after she had stopped growing, to improve facial asymmetry and malocclusion.

Case presentation

A 15-year-old girl with a chief complaint of facial asymmetry was referred to our hospital for correction of facial deformity. She had been diagnosed with bilateral HFM at birth, but did not have any difficulty breathing. The left side of her face was more severely affected, showing type III deformity, while the right side was affected by type I deformity.

She had already undergone three mandibular reconstructions, comprising reconstruction of the left ramus with calvarial bone graft at 8 years old and two rounds of distraction osteogenesis at 10 and 11 years at another hospital.

Clinical examination showed clear asymmetry of the facial skeleton, including chin deviation toward the left and severe soft tissue deficiency on the left. Severe lip canting in the transverse dimension to the upper left and lip incompetency were noted. Mild facial nerve palsy was also present on the left. Orbital size and positions were bilaterally normal. The point of maximum mouth opening was deviated to the left, but the right condyle showed appropriate translatory movement. A mild maxillary cant was seen during occlusion, inconsistent with the soft tissue findings. Unilateral posterior scissor bite on the right side and crossbite on the left side were also observed (Figure 1). Radiography revealed markedly reduced height of the reconstructed left ramus.

Facial asymmetry was diagnosed as mainly due to hypoplasia of the left mandible and associated soft tissues. The ideal treatment for facial asymmetry and malocclusion would have been distraction osteogenesis, which could have been expected to lengthen both the left mandibular ramus and associated soft tissue. However, the left mandibular ramus was so thin due to three mandibular reconstructions that this would have been difficult. We therefore decided to perform orthognathic surgery to correct facial asymmetry and malocclusion.

At 19 years old, after waiting for the patient to reach skeletal maturity, a left mandibular impacted wisdom tooth was extracted under general anesthesia.

After pre-surgical orthodontics, the final amount of movement was examined. The surgical objectives were to move the body of the mandible 3.5 mm to the undeviated side to correct the dental midline and achieve 9 mm of advancement on the left side and 4 mm of setback on the right side, to adjust the maxillary occlusal cant. A 3-dimensional simulation showed a large gap between the proximal and distal segments of the left mandible (Figure 2), so iliac bone graft was planned to fill the interpositional gap.

Bilateral sagittal split and Le Fort I osteotomies were performed simultaneously at the age of 20 years and 3 months. LeFort 1 osteotomy was performed with downward rotation of the anterior max-

Figure 1. Initial findings of facial and oral photograph.

illa, parallel shift to the left to align the midline and slight downward shift on the left side to correct the cant. The maxillary position was corrected with an intermediate splint, and fixation with four miniplates was conducted. Sagittal split osteotomy on the normal right side was performed using Dal Pont's technique via an intraoral approach. Next, osteotomy was carefully carried out on the left reconstructed mandible to avoid pathological fractures, using an extraoral approach at the junction between the regenerated bone and original bone. Vertical cuts were made with a Lindeman bar, then a sagittal cut was made along the inferior border of the regenerated bone straight inward. The split was completed with chisels in combination with separators. After manually mobilizing the proximal and distal segments of the mandible, intermaxillary wire fixation was performed using the final occlusal splint. The left proximal segment was unstable and was easily moved backward due to the absence of the condyle and TMJ, and transferring the distal segment to the ideal position indicated by the 3D simulation was extremely difficult due to tension from surrounding soft tissues. Correcting the mandibular yaw proved very difficult. A block of cortico-cancellous iliac bone was harvested for grafting and tightly positioned between the proximal and distal segments using bicortical screws to stabilize the bony segments. The planned occlusion was achieved (Figure 3). Antibiotics were administered within 72 h after surgery. Intermaxillary wire fixation was applied for 7 days, then changed to elastic traction. Two weeks after surgery, the patient was discharged with good wound healing. Postoperative orthodontic treatment was started 5 weeks after surgery.

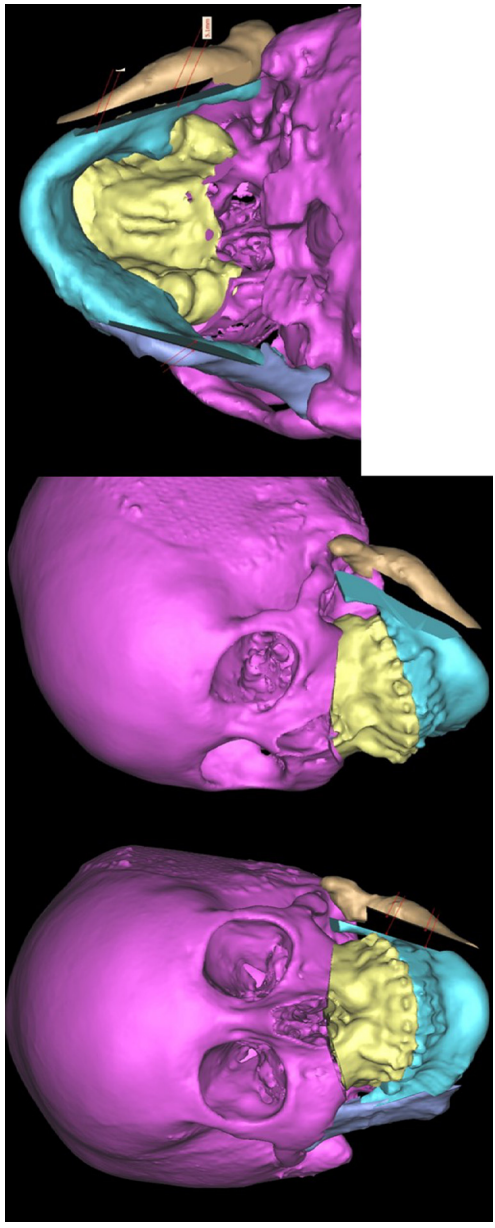

Figure 2. Three-dimensional computed tomography-based virtual surgical simulation.

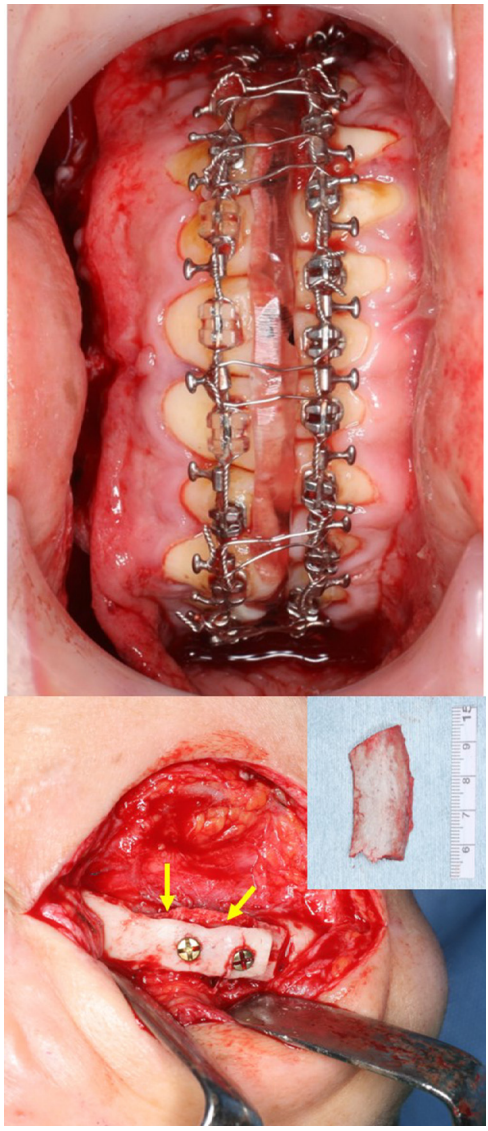

Figure 3. Intraoperative photograph displaying iliac bone grafting to fill the interpositional gap between the proximal and distal segments with bicortical fixation (arrow), and final occlusion with a splint.

Figure 4. Facial and oral photographs obtained 4 months after orthognathic surgery.

The patient was very satisfied with the improvements in facial symmetry and with being able to close her lips following the correction of lip cant (Figure 4). Postoperative changes in cephalometry are shown in Supplementary Table 1. Postoperatively, the left-sided occlusal cant improved from 4° to 2° from oblique to horizontal. However, skeletal discrepancies and mild deformities in facial features due to facial nerve paralysis remained, and gradual occlusal relapse has been observed in the 4 months since the surgery.

The patient lives in another prefecture and has had difficulty attending the hospital as she has started college and due to the restrictions caused by the COVID-19 pandemic across the country. We will continue to perform careful follow-up with orthodontists in contact with her.

Discussion

Type III HFM deformities are the most challenging to reconstruct, requiring an individualized approach to the diverse clinical presentations. Eighteen percent of patients with severe HFM deformities require secondary distraction osteogenesis following conventional autogenous reconstruc-

tion.[3] However, early mandibular distraction osteogenesis does not appear to significantly influence whether orthognathic surgery is needed at skeletal maturity.[4] Even after undergoing a series of such surgeries, our patient continued to exhibit severe facial asymmetry due to postoperative relapse and an "occlusal disaster".[5]

In patients with type III HFM, preoperative surgical planning and simulations are particularly important to facilitate complex maxillary and mandibular movements during orthognathic surgery.[6] Three-dimensional planning for orthognathic surgery facilitates superior improvements in the symmetry of facial contours for congenital anomalies compared to 2-dimensional planning.[7,8] With the help of simulations, we were able to determine in advance that the reconstructed mandible of our patient would be so thin that iliac bone grafting would need to be performed to fill the interpositional gap between proximal and distal segments and allow sufficient bony contact and rigid fixation. The intersegmental space can cause postoperative instability. In cases of severe deformity, such as Pruzansky type 3, where the mandibular is thinned after repeated mandibular distraction, bone grafting should be considered to increase bone volume and obtain better bony contact. However, some cases are encountered in which moving the mandible to the ideal position identified during simulation proves difficult intraoperatively, and postoperative occlusal relapse may occur due to the absence of the TMJ and glenoid fossa, thinness of the reconstructed mandible, or soft tissue tension.[9] In such cases, where replicating the idealized procedure indicated in simulations ends up being intraoperatively impractical, the application of custom plates is becoming more widespread. The use of this technology in our country is anticipated.[10] Use of bicortical positional screws with maintenance of the intersegmental gap has also been reported as a stable fixation approach.[11]

In recent years, alloplastic reconstruction of the missing TMJ with total joint replacement has been considered the optimal solution for salvaging patients with type III HFM in whom autogenous reconstruction has not gone well. This harmonizes both hard and soft tissues. Several other reports have described custom alloplastic TMJ replacement combined with orthognathic surgery, demonstrating favorable results in terms of both facial deformities and malocclusion.[12,13] Orthognathic surgery is of limited use for reconstruction with autologous tissue, and the application of prosthetics may become the main approach to the reconstruction of HFM-related facial deformities in the future.

Conclusions

We have described a case in which orthognathic surgery with bone graft was used to treat type III HFM in a patient who exhibited severe deformities after previous mandibular reconstructions. Despite the use of 3D simulation, postoperative relapse of occlusion occurred. Our case report provides some warnings regarding the treatment of type III HFM deformity after such early reconstructions.

Patient consent

Informed consent was provided by patient to publish the case details and associated images.

Ethical approval

Not required.

Declaration of Competing Interest

The authors have no potential conflicts of interest to declare with respect to the research, authorship, and/or publication of this article.

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Supplementary materials

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