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Surgical and Prosthetic Treatment for Bilateral Temporomandibular Joint Ankylosis With Micrognathia: A Case Report

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Temporomandibular joint（TMJ）ankylosis is a joint disorder characterized by the fibrotic or bony adhesion of anatomical joint components, leading to severe limitation or loss of joint mobility. TMJ ankylosis affects mastication, digestion, speech, and oral hygiene. Onset during childhood results in secondary micrognathia, leading to a reduction in the oropharyngeal airway and psychological problems. We present the case of a 39-year-old female patient with bilateral TMJ ankylosis and micrognathia that developed during childhood. The patient had a history of a traffic accident at the age of 5 years and presented with trismus. Computed tomography revealed bony ankylosis of the right TMJ and fibrotic ankylosis of the left TMJ. Bilateral interpositional gap arthroplasty was performed using an intraoral approach, followed by mandibular distraction osteogenesis（DO）. Following DO, oral rehabilitation with dental implants was performed. The patient's facial profile and mastication significantly improved and were maintained for 10 years after treatment.

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INTRODUCTION

Temporomandibular joint（TMJ）ankylosis is a joint disorder characterized by the fibrotic or bony adhesion of anatomical joint components, leading to severely limited or lost joint mobility. TMJ ankylosis is primarily caused by trauma, infection, autoimmune diseases, and failed surgeries［1］. A limited mouth opening can lead to issues related to mastication, digestion, speech, and oral hygiene. Onset during childhood affects the growth of the facial skeleton, resulting in secondary maxillofacial deformities, commonly observed as facial asymmetry and micrognathia. A retrognathic mandible can affect the oropharyngeal dimensions, potentially compromising or obstructing the airways in severe cases. TMJ ankylosis and maxillofacial deformities can also lead to psychological problems［2］. The treatment of TMJ ankylosis with micrognathia is often performed in one or two phases, involving surgeries for TMJ ankylosis, including gap arthroplasty with or without interpositional materials and arthroplasty with condylar reconstruction using different materials. Additionally, surgeries for secondary skeletal deformities,

such as orthognathic surgery, distraction osteogenesis (DO), and esthetic surgery, have been conducted [2]. In this case report, we describe a patient with bilateral TMJ ankylosis and micrognathia who, with a history of a traffic accident during childhood, underwent interpositional gap arthroplasty, mandibular DO, and oral rehabilitation using dental implants.

CASE REPORT

A 39-year-old Japanese female patient was referred to the Department of Oral and Maxillofacial Surgery at Shimane University Hospital with the chief complaint of trismus. She was involved in a traffic accident at the age of 5 years and gradually developed trismus. The initial examination revealed micrognathia with a 2 mm mouth opening. Multiple dental caries were observed, which presented with treatment difficulties (Fig. 1). On radiography, the right TMJ appeared massive, and the fossa was not visible, with a narrow joint space in the left TMJ. The mandibular molars, especially on the right side, were decayed because of dental caries. Cephalography revealed mandibular micrognathia, with the midline shifting to the right side (Fig. 2). On computed tomography (CT), the right TMJ exhibited a bony mass, whereas the left TMJ showed no bony adhesions although the joint space was narrow (Fig. 3). The clinical diagnosis was bilateral TMJ ankylosis (bony ankylosis on the right side and fibrotic ankylosis on the left side) with micrognathia. The treatment plan included gap arthroplasty to enable mouth opening, followed by treatment for dental caries and DO of the mandible.

Bilateral interpositional gap arthroplasty was performed using an intraoral approach. The bilateral coronoid processes were removed to reach the site of ankylosis, and osteotomy was performed under the bony mass on the right side and the condylar neck on the left side. A gap of 10 mm or more was formed, and a buccal fat pad was inserted as an intermediate material (Fig. 4). Mouth opening exercises were initiated early in the postoperative period to maintain mouth opening, and procedures

Fig. 1. Extraoral and intraoral findings at the initial examination.
Frontal and lateral views showing micrognathia (a, b) and deep overbite (c).

Fig. 2. Radiographs at the initial examination.
Panoramic radiograph showing a mass in the right TMJ, narrow joint space in the left TMJ, and decayed mandibular molars due to dental caries (a). The cephalogram shows mandibular micrognathia, and the midline has shifted to the right (b, c).

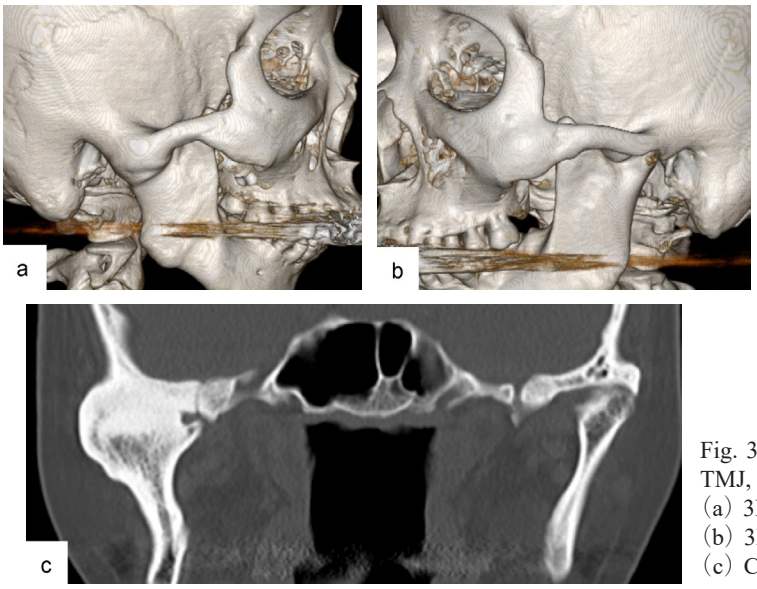

Fig. 3. Preoperative CT showing a mass in the right TMJ, narrow joint space in the left TMJ.
(a) 3D CT of the right TMJ.
(b) 3D CT of the left TMJ.
(c) Coronal view.

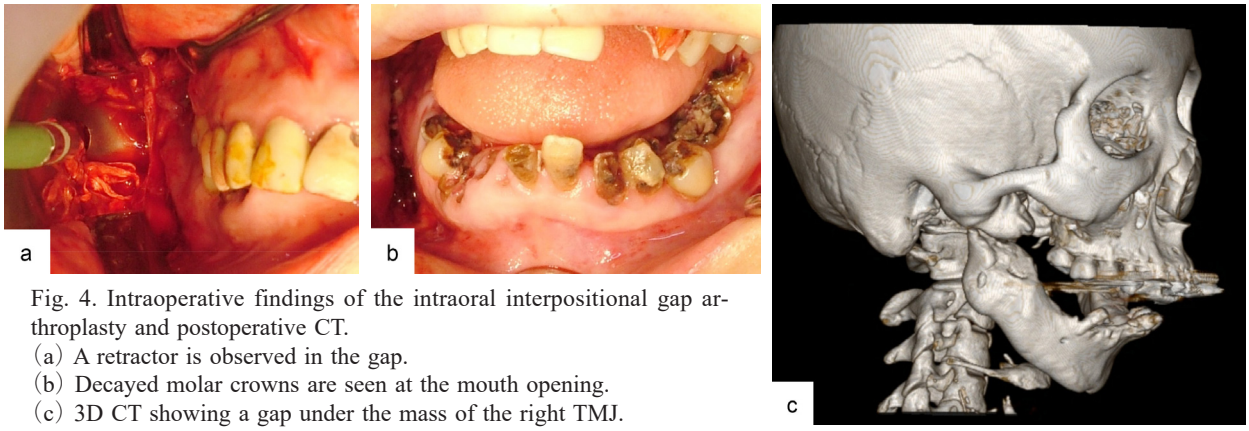

Fig. 4. Intraoperative findings of the intraoral interpositional gap arthroplasty and postoperative CT.
(a) A retractor is observed in the gap.
(b) Decayed molar crowns are seen at the mouth opening.
(c) 3D CT showing a gap under the mass of the right TMJ.

for tooth extraction, dental caries, and periodontal disease treatment were initiated. The adhesive left condyle was separated and luxated, leading to bone union between the condyle and the mandibular ramus.

After the interpositional gap arthroplasty, DO using an external distractor was planned because of the need for multidirectional movement. Two extra-oral distraction devices were placed bilaterally in the mandible. Subperiosteal dissection of the mandible was performed using the submandibular approach. Before osteotomy, a percutaneous anterior and posterior pair of pins was placed using a trocar. The devices were attached to the pins, and an osteotomy between the anterior and posterior pins was performed to preserve the inferior alveolar nerve. After a latency period of 7 days, distraction started at a rate of 0.5 mm twice a day. Lengthening of 22.5 mm on the right side and 11 mm on the left side was achieved. During distraction, orthognathic intermaxillary elastics were used to mold the regenerated bone and optimize occlusion. The devices were left in place to serve as external fixators for an 8-week consolidation period and then removed (Fig. 5).

After interpositional gap arthroplasty and DO, mouth opening was possible and both the occlusion and facial profile improved (Fig. 6). However, the patient's mandible was edentulous. The all-on-4 treatment concept using a fixed dental prosthesis was proposed as an appropriate treatment option. Two regular platform (RP) implants (Brånemark Mk III; Nobel Biocare, Göteborg, Sweden) with a diameter of 4.3 mm and length of 10 mm were placed in the bilateral lateral incisor region, and two RP implants with a length of 11.5 mm were placed

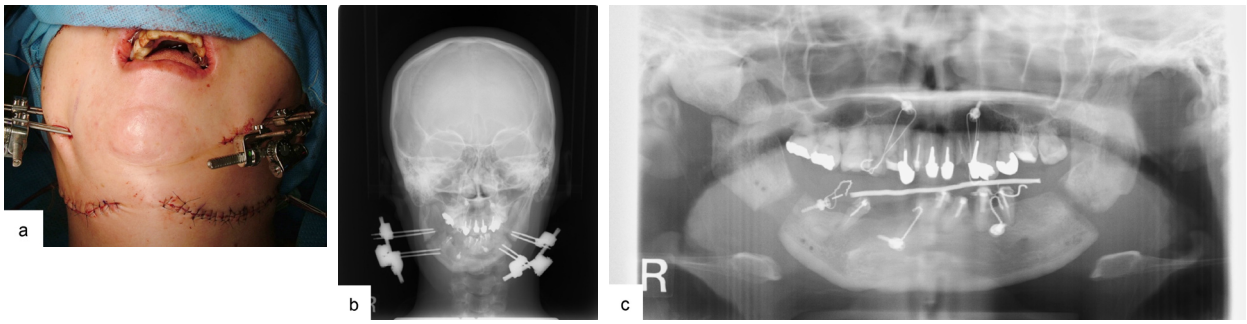

Fig. 5. Findings of DO.
(a) Postoperative findings of the placement of the two extraoral distraction devices.
(b) Frontal radiograph during DO.
(c) panoramic radiograph after DO showing the gap in the bilateral side of the mandible.

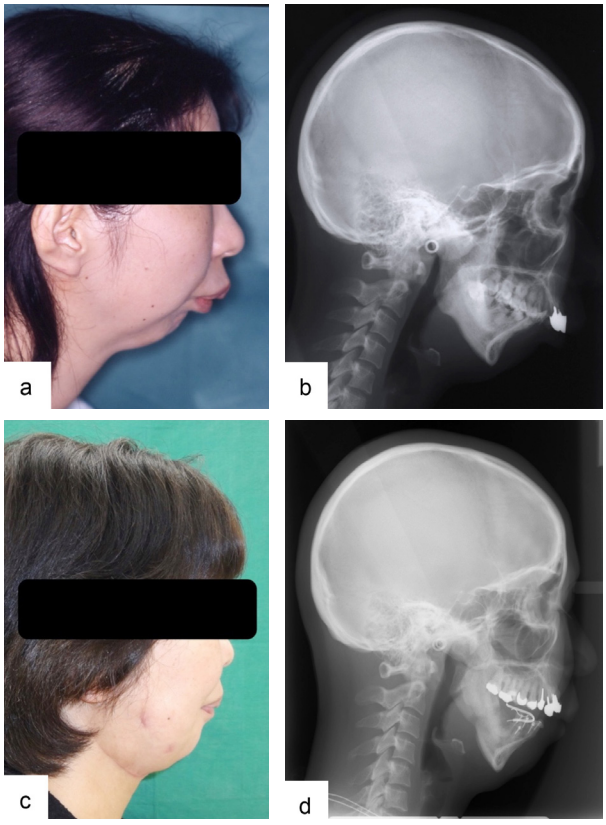

Fig. 6. Comparison between preoperative and postoperative lateral views.
(a) Preoperative lateral view.
(b) Preoperative cephalogram (lateral view).
(c) Postoperative lateral view.
(d) Postoperative cephalogram (lateral view).

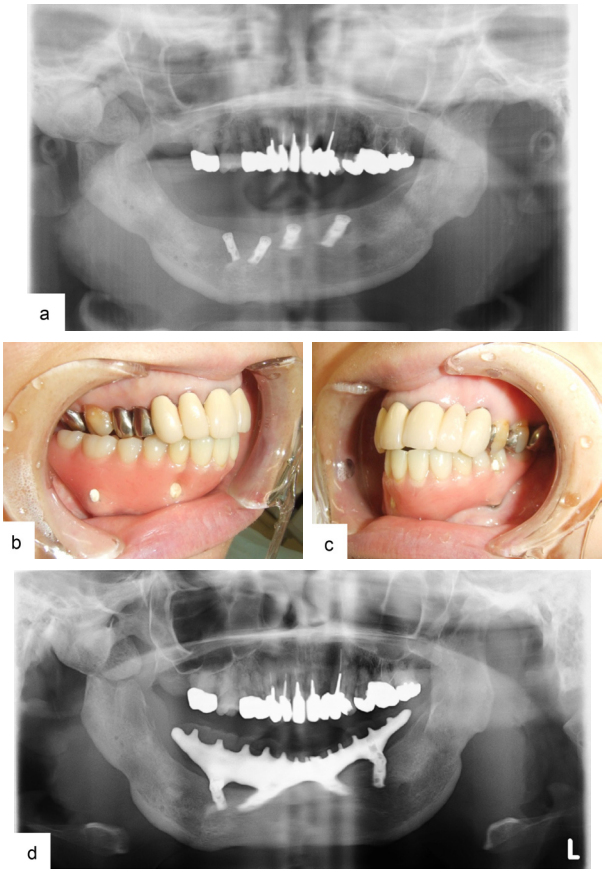

Fig. 7. The all-on-4 implant treatment.
(a) Postoperative panoramic radiograph of placement of the four dental implants.
(b, c) Intraoral findings of fixed prosthesis supporting 4 dental implants.
(d) Panoramic radiograph after the implant treatment.

in the second premolar region of the mandible. After the placement of the implant-supported provisional prosthesis, the final prosthesis was placed (Fig. 7). Following prosthetic treatment with dental implants, the patient has maintained an opening of 28 mm and maintained a stable occlusion 10 years after the treatment.

DISCUSSION

Controversies persist regarding the management of TMJ ankylosis with micrognathia using DO, including post-arthroplastic, simultaneous arthroplastic, and pre-arthroplastic DO. The potential drawbacks of post- and simultaneous arthroplastic DO include in-

stability of the proximal segment and noncompliance with active physiotherapy [3]. Ideally, during distraction, it is crucial to minimize unwarranted jaw mobility to prevent pseudarthrosis between segments. Although post-and simultaneous arthroplastic DO provide faster functional movement of the mandible than pre-arthroplastic DO, there is a risk of re-ankylosis [3]. In their systematic review, Chugh *et al*. suggested that pre-arthroplastic DO appears to be the most effective in correcting mandibular deformities. Pre-arthroplastic DO allows distraction against a stable fixed point, ensuring that true distraction of the mandible occurs in the desired direction with better vector control. Predicting and controlling the vector of the post-arthroplastic DO is challenging. In other protocols, some distance from the distraction is lost because of the upward or backward movement of the condyle [4]. However, Albert and Muthusekhar described in their systematic review that maximum mouth opening and mandibular length increased, and chin and mandibular position improved by the end of treatment in all three protocols [3]. In our case, post-arthroplastic DO was performed because mouth opening was severely restricted, and compliance with active physiotherapy was better with post-arthroplastic DO than with simultaneous arthroplastic DO. Post-arthroplastic and simultaneous arthroplastic DO do not limit the amount of distraction, unlike pre-arthroplastic DO, in which occlusion becomes the limiting factor [4]. As expected, the distraction vector control was challenging.

In this case, intraoral gap arthroplasty [5, 6] was carried out. Ko *et al*. reported that intraoral gap arthroplasty can mitigate the complications associated with the extraoral approach for TMJ arthroplasty. The advantages of intraoral arthroplasty include the absence of a facial scar, lower likelihood of injury to the facial and auriculotemporal nerves, no sialocele, less hemorrhage, and simultaneous coronoidectomy or coronoidotomy with improved direct access to the ankylosed condyle via the same incision. Its disadvantages include a limited surgical field and constraints on the selection of the interpositional material [5]. They also noted that cases with large ankylosed bony masses involving part of the skull base and/or occupying the sigmoid notch area were good indications for the intraoral approach. The intraoral technique, in which the narrowest part of the ankylosed bony mass is located usually under the original condylar neck region, can be performed easily after cutting off the coronoid process [7].

Using the intraoral approach, coronoidectomy facilitates improved access to the ankylosed site. Kumar *et al*. reported that ipsilateral and contralateral coronoidectomies enhanced mouth opening in patients with unilateral TMJ ankylosis, concluding that coronoidectomy plays a crucial role in improving mouth opening in TMJ ankylosis treatment [8]. In the present case, the right TMJ contained an ankylosed bony mass occupying the sigmoid notch. Intraoral gap arthroplasty was performed to avoid additional facial scarring using subsequent DO with external distractors. Bilateral coronoidectomy was performed to access the ankylosed sites, potentially influencing mouth opening.

In gap arthroplasty, various local or distant tissues are used as interpositional materials to prevent re-ankylosis. Heterotopic bone formation and fibrosis around the gap are the main causes of re-ankylosis. Different types of autogenous interpositional materials, including temporalis myofascial flap, temporalis muscle, temporalis superficial fascia flap, auricular cartilage, costochondral graft, skin, and fat, have been used over the years [9]. More recently, the buccal fat pad (BFP) has been introduced as an alternative to these options. Rattan was the first to describe a technique in two patients with TMJ ankylosis, in which pedicled BFP was used as the interpositional tissue [10]. The primary advantages of the BFP include its availability in the proximity of the surgical site and retrieval from the same preauricular incision. Owing to its pedicled nature, its independent vascular supply contributes to its long-term survival. In a radiological study using MRI of the long-term fate of pedicled BFP used for interpositional arthroplasty in TMJ ankylosis, the BFP remained viable after 1 year and prevented heterotopic bone formation following TMJ ankylosis release [11, 12]. In our case, BFP harvested from the same surgical site was used as an interpositional material, and re-ankylosis did not occur in the right condyle. The left condyle was luxated and united with the mandibular ramus. Ko *et al*. also used the BFP in two cases using the intraoral approach for gap ar-

throplasty. In these cases, re-ankylosis was not observed, and a maximum mouth opening range of 36 mm and 40 mm apertures was maintained [5].

Several prosthetic treatment options are available for edentulous jaws, including complete dentures, removable implant-supported prostheses, and fixed implant-supported prostheses. However, removable implant-supported prostheses or fixed implant-supported prostheses provide a higher degree of patient satisfaction than complete dentures [13, 14]. In 2003, the "All-on-4" treatment concept was introduced for prosthetic rehabilitation based on only four implants: two in the anterior region of the jaw, oriented straight, and two in the posterior region, tilted distally [15]. This treatment concept was developed to maximize the use of available remnant bone in atrophic jaws, allowing immediate functioning and avoiding regenerative procedures that increase treatment costs and patient morbidity, as well as complications inherent to these procedures. The results obtained indicate a survival rate of 99.8% for more than 24 months and 99.0 ± 1.0% at 36 months in the two systematic reviews. However, it is necessary to conduct long-term clinical and laboratory studies to determine the long-term success criteria in all-on-4 implant designs because the current evidence is limited by the quality of available studies and the paucity of data on long-term clinical outcomes [16, 17].

In our case, because the alveolar bone width in the posterior mandible was insufficient, fixed implant-supported prosthetic rehabilitation based on the all-on-4 treatment concept was used to avoid additional regenerative surgery. The prosthetic treatment was effective in this case, resulting in patient satisfaction.

Ethical approval

Not required

Funding

None

Conflict of Interests

None

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