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Case report

Long-term follow-up after arthroplasty for pediatric temporomandibular joint ankylosis performed before the critical period of mandibular growth: A case report

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ABSTRACT

Introduction and importance: Pediatric temporomandibular joint (TMJ) ankylosis can lead to significant difficulties in opening the mouth, as well as stunted mandibular growth resulting in a small mandible and facial asymmetry. For pediatric TMJ ankylosis, the ideal time to perform TMJ mobilization in order to achieve standard mandibular growth is unclear.

Case presentation: An 11-year-old boy with limited mouth opening was referred to our hospital. The patient had previously sustained a fracture of the left articular process of the mandible. Clinical examination revealed bony ankylosis of the left TMJ, and the condylar head was dislocated anteromedially. The bony ankylosis was removed at the age of 12 years. Mouth-opening exercises were started postoperatively. His mouth opening recovered without the development of severe facial asymmetry, and the dislocated condylar process served as a new joint and provided good jaw function until the most recent follow-up at the age of 21 years.

Clinical discussion: It is practical to perform surgical intervention after the child has achieved some growth and at a time when the surgery would not interfere with jaw development because the mouth opening can be improved by postoperative physical therapy. No interpositional implant was used because strict postoperative mouth opening exercises and the displaced condylar process, which was maintained to almost normal TMJ structure, were expected to preserve the gap between the ramus and the zygomatic arch.

Conclusion: Surgical intervention in the appropriate growth period is important to prevent the sequelae of mandibular growth suppression due to pediatric TMJ ankylosis.

1. Introduction

Temporomandibular joint (TMJ) ankylosis in children can lead to significant difficulties in opening the mouth, mastication, and pronunciation, as well as stunted mandibular growth resulting in a small mandible and facial asymmetry [1,2]. Post-traumatic TMJ ankylosis often develops in childhood. Kaban et al. stated that the release of TMJ ankylosis should be performed as soon as possible [3]. However, if the procedure is performed when the patient is too young, postoperative opening exercise may be inadequate because of non-compliance, and there may be a risk of re-ankylosis. The ideal time to perform TMJ mobilization in order to achieve standard mandibular growth is unclear.

Sawhney's classification of TMJ ankylosis defines type III ankylosis

as the type in which the bone fragment containing the condylar head is dislocated anteromedially, and the lateral portion of the ramus and the zygomatic arch are attached by bone [4]. In these cases, it is unclear how the articular structure of the dislocated bone fragment will change over time

In this study, we found that performing arthroplasty before the critical period of mandibular growth prevented adverse effects on jaw growth, and the dislocated condylar process and articular disc recovered to almost normal TMJ structure and achieved normal jaw movement.

This case report has been presented in accordance with the SCARE 2020 criteria [5].

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Abbreviations: CT, computed tomography; MMO, maximal mouth opening; MRI, magnetic resonance imaging; TMJ, temporomandibular joint.

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Fig. 1. Facial and intraoral photographs at the first visit.

- (A) Slight deviation of the chin to the left.
- (B) Mild poor growth of the mandible.
- (C) Leftward deviation of the mandibular dental midline.
- (D) Leftward deviation of the opening path.

2. Case presentation

An 11-year-old boy with limited mouth opening was referred to our hospital. At the age of 6 years, the patient had sustained a fracture to the left articular process of the mandible from a fall. As a result of observation without any active treatment or surgery, his maximal mouth opening (MMO) had eventually decreased.

The MMO at the first visit was 18 mm with deviation to the left with slight deviation of the chin in the same direction. On lateral view, mild poor growth of the mandible was observed (Fig. 1). Computed tomography (CT) showed fibrous-bony ankylosis of the left TMJ between the lateral portion of the ramus and the zygomatic arch, and the condylar head was dislocated anteromedially and connected to the medial surface of the ramus of the bone. Further, it was observed that the articular disc was located on the condylar head, and the disc position with respect to the condyle appeared to be correct on magnetic resonance imaging (MRI) (Fig. 2).

The persistence of the opening disorder during the growth period and its adverse effects on jaw development were concerning; therefore, resection of the bony ankylosis was planned when the patient was 12 years old. The operation was performed via a temporo-preauricular

incision under general anesthesia. After confirmation of the bony adhesion, the augmented bone on the lateral portion of the ramus was resected and reshaped for gap formation. Intraoperative findings confirmed that the joint structure, including the upper and lower joint spaces and the articular disc, was well-maintained on the medially dislocated condylar process, and the outer surface of the articular disc was connected to the scar tissue of the fusion area (Fig. 3). Because of these conditions, only the bony bridge between the lateral aspect of the mandibular ramus and the surface of the zygomatic arch was resected because it was expected that the preserved medially deviated condylar process and the articular disc would function as a new joint and prevent re-ankylosis.

Mouth-opening exercises using hands were started by the patient on the second day postoperatively for TMJ rehabilitation, and an MMO of 40 mm was regained 7 months postoperatively. The MMO was maintained at 45 mm at the age of 21 years, i.e., 9 years after the surgery. In addition, there was no facial asymmetry or limitation of jaw movement (Fig. 4). The MRI findings showed that the positional relationship between the medially displaced condylar process and the articular disc was adequate. In addition, the articular disc was positioned on the condyle during opening and closing mouth movements, with a normal anterior

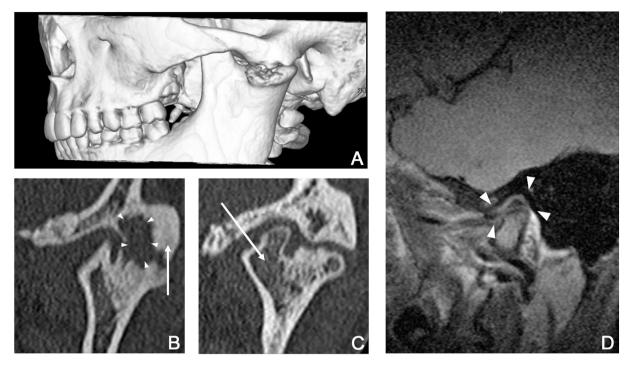


Fig. 2. Computed tomography findings and magnetic resonance imaging findings at the first visit.

- (A) Three-dimensional image of the left temporomandibular joint (TMJ).
- Fibrous-bony ankylosis of the left TMJ between the lateral portion of the ramus and the zygomatic arch was found.
- (B) The arrow indicates osteogenesis and bony deformity on the lateral aspect of the left TMJ; the arrowheads show soft tissue shadow.
- (C) The arrow indicates the condylar head displaced anteromedially and fused with the medial aspect of the mandibular ramus.
- (D) The arrowheads show the articular disc, and it is located on the condylar head, and the disc position appears to be correct.

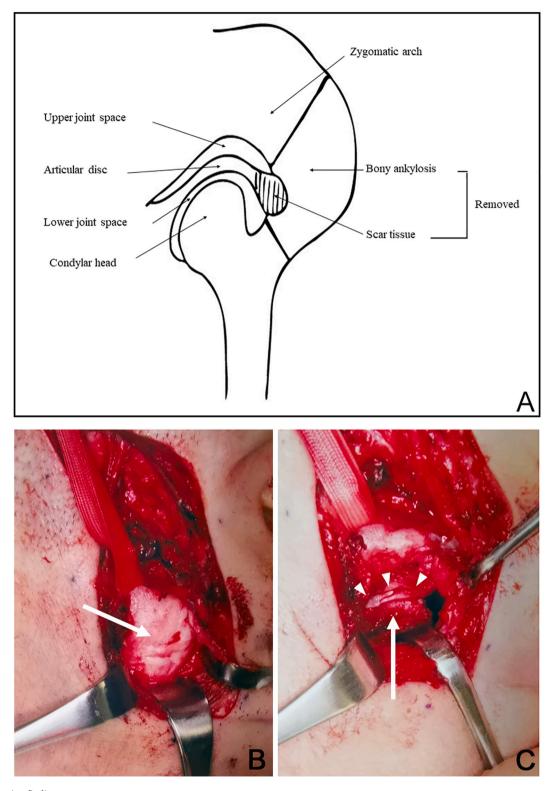
sliding movement (Fig. 5). Fortunately, the condylar protrusive sliding movement induced during mouth opening indicated that this medially displaced condylar component had a function similar to that of a normal TMJ.

3. Discussion

In patients with pediatric unilateral TMJ ankylosis, restriction of mandibular movement can adversely affect mandibular growth, resulting in micrognathia and facial asymmetry [6,7]. Therefore, mobilization of the mandible by releasing the bony ankylosis of the TMJ is recommended at an early stage. Kaban et al. stated that TMJ ankylosis should be released in children as soon as possible if it is feasible to expect patient cooperation post-surgery [3]. Erol et al. reported in their review of 59 patients with post-traumatic TMJ ankylosis that the most common age at which surgery was performed was ≥18 years, despite the most common age of injury being 0-10 years [8]. Roychoudhury et al. reported that there was no significant difference in the postoperative MMO between patients with TMJ ankylosis who underwent surgery before the critical period of jaw development and those who underwent surgery after this period [9]. For this reason, it is practical to perform the surgery after the child has achieved some growth, and it is important to perform it at a time when it will not interfere with jaw development because MMO can be improved by postoperative physical therapy. In the

present case, on performing the surgery at the age of 12 years, he was able to gain mobility of the TMJ before the critical period for mandibular growth, and thus, could achieve a normal MMO and avoid micrognathia and facial asymmetry.

Generally, it is thought that an interpositional implant is required to prevent re-ankylosis after gap arthroplasty [10,11]. In case of pediatric TMJ ankylosis, reconstruction of the mandibular head by rib cartilage grafting might be performed to take into consideration the growth of the mandible [12]. Furthermore, in case of re-ankylosis, reconstruction of the mandibular head using TMJ replacement prostheses might be necessary [13]. In the present case, no interpositional implant was used because strict postoperative mouth opening exercises and the displaced condylar process, which was maintained to almost normal TMJ structure, were expected to preserve the gap between the ramus and the zygomatic arch. Interestingly, MRI performed 9 years postoperatively showed that the condylar process and the articular disc, which were displaced medially due to the fracture, healed with almost normal articular structures, and normal jaw movement was observed. These elements were thought to have resulted in good postoperative recovery of the mandibular movements, and the suppression of maxillofacial growth during the growth period was avoided.



 $\textbf{Fig. 3.} \ \ \textbf{Intraoperative findings.}$

- (A) Schema of the intraoperative findings.
- (B) The arrow indicates the bony ankylosis of the lateral aspect of the left temporomandibular joint (TMJ).
- (C) Bony adhesions between the zygomatic arch and the ramus of the mandible have been removed with adequate gap formation. The arrowheads indicate the articular disc, and the arrow indicates the condylar head.

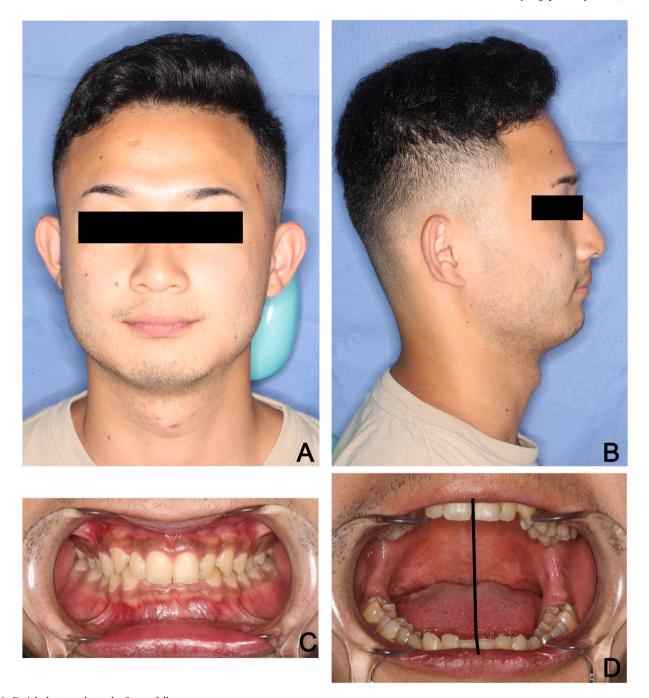


Fig. 4. Facial photographs at the 9-year follow-up.

(A, B) There is no facial asymmetry or obvious micrognathia.

(C, D) Deep bite is observed, but no deviation of the opening path is observed. The maximal mouth opening is maintained in the normal range.

4. Conclusion

In the present case, normal growth in the maxillofacial region was ensured by carrying out surgical intervention before the growth period. In addition, the positional relationship between the articular disc and the mandibular condyle, which was displaced anteromedially, was maintained correctly. Thus, a joint structure similar to the normal structure was constructed, and adequate jaw movement was achieved; these were essential elements that prevented micrognathia and facial asymmetry.

$CRediT\ authorship\ contribution\ statement$

Yasuhiro Kurasawa: Data collection, Writing original draft preparation.

Hiroyuki Yoshitake: Surgical management and care of patient, Writing - review & editing, Supervision.

Nobuyoshi Tomomatsu: Supervision.

Tetsuya Yoda: Supervision.

Declaration of competing interest

All authors report no conflicts of interests.

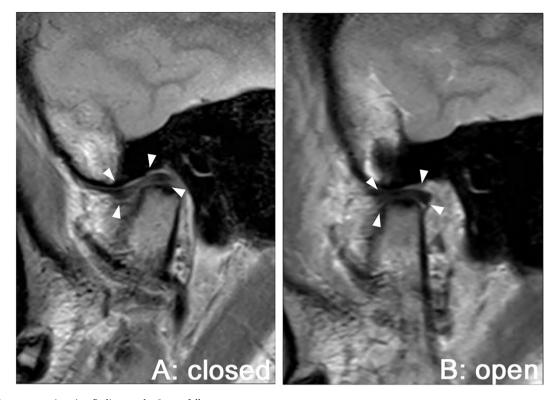


Fig. 5. Magnetic resonance imaging findings at the 9-year follow-up.

(A) In the closed-mouth condition, the displaced mandibular head and the articular disc are maintained normally; the arrowheads indicate the articular disc.

(B) In the open-mouth condition, the articular disc is located above the mandibular head, and anterior sliding movement of the mandibular head can be observed; the arrowheads point to the articular disc.

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Ethical approval

Single case reports are exempt from ethical approval in our institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Registration of research studies

None.

Provenance and peer review

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Guarantor

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