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Distraction osteogenesis promotes temporomandibular joint self-remodeling in the treatment of mandibular deviation caused by condylar ankylosis

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ABSTRACT

Craniofacial deformity and malocclusion are primary concerns following temporomandibular joint ankylosis (TMJa) in growing patients, and they pose even greater challenges in adult patients. The treatment objectives always involve restoring proper jawbone structure, achieving stable occlusion, and attaining satisfactory joint mobility. This report presents a 4-year follow-up of an adult patient with TMJa-induced mandibular deviation, who underwent a combined treatment approach involving distraction osteogenesis (DO) and orthodontic–orthognathic surgery. Orthodontic treatment resulted in favorable occlusion and improved facial esthetics. A new condyle with a reconstructed glenoid fossa in a forward position was established after mandibular DO and the damaged TMJ experienced self-remodeling owing to functional improvement. Thus, this case demonstrates the efficacy of DO in promoting adaptive TMJ self-remodeling with long-term stability when treating mandibular deviation caused by condylar ankylosis in adult patients.

1. Introduction

Craniofacial deformity and malocclusion commonly occur due to temporomandibular joint ankylosis (TMJa), which can have various causes, including trauma, tumors, and inflammation. TMJa often leads to restricted jaw movement, limited mouth opening, and irreversible facial deformities [1]. Facial asymmetry may result from unilateral ankylosis, causing psychosocial issues for patients, and bilateral ankylosis can lead to jaw deformity and airway obstruction. Moreover, growing patients may experience changes in the jawbone, leading to severe crowding of teeth and malocclusion.

Current surgical treatments for joint ankylosis and its associated deformities include arthroplasty or joint reconstruction,

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orthognathic surgery, bone transplantation, and distraction osteogenesis (DO). However, surgeons face technical challenges and a high recurrence rate. DO has emerged as a popular technique for restoring symmetry and function in TMJa treatment [2]. It allows for bone elongation and soft tissue changes, minimizing surgical trauma, donor area complications, and recurrence [3,4]. Auxiliary orthodontic treatment aids in achieving precise surgical movement as well as satisfactory occlusion. Combined operations are often necessary to attain desired outcomes, although the sequencing of TMJ release and DO remains debated [5]. Our treatment approach focuses on rebuilding joint function and achieving stable facial esthetics and occlusion based on previous clinical research [6,7].

This case report presents a 4-year follow-up of an adult patient with TMJa-induced facial asymmetry and malocclusion, showcasing our treatment outcomes.

Fig. 1. (A) Pretreatment extraoral views. (B) Pretreatment intraoral views.

2. Case report

2.1. Diagnosis and etiology

A 24-year-old woman sought orthodontic treatment for "crooked teeth" and facial asymmetry resulting from TMJa. The patient had a history of tympanitis at the age of eight, leading to restricted mouth opening. TMJa was diagnosed on the left side, and gap arthroplasty was performed to release the joint, significantly improving mouth opening. No other treatment was received during the intervening years.

Extraoral examination revealed severe facial asymmetry, with the chin shifted to the left, primarily due to TMJa. Closer examination showed uneven nostrils, mouth corners, and shoulders, along with a convex profile and slight retrusion of the mandible. The lower facial height was reduced, and the nasolabial angle was acute (Fig. 1A).

Intraoral assessment revealed proclined upper and lower anterior teeth, with normal overbite. There was a 2 mm crossbite in the upper lateral incisors. The lower midline was shifted 3.5 mm to the left, and molar relationships were Class II on the left and Class I on the right. Moderate crowding was observed in both arches (Fig. 1B).

Radiographic examination revealed a deformed left condyle. The left condyle showed irregular structure (Fig. 2A) in closing position compared with the right (Fig. 2B). It also appeared in abnormal location (Fig. 2C) in opening position compared with the right (Fig. 2D). The lateral cephalogram showed significant protrusion in profile (Fig. 3A) and the panoramic radiograph showed impacted lower third molars (Fig. 3B).

Bilateral TMJ examination revealed no clicking sounds and moderate limited mouth opening, with mandibular deviation to the right. Lateral movement to the left side was severely restricted, and movement to the right side was completely restricted due to the damaged left joint. Cephalometric analysis performed before treatment is presented in Table 1.

2.2. Treatment objectives

The overall treatment objectives were to improve profile esthetics, achieve facial symmetry, and establish a functional and stable occlusion. The specific goals were as follows.

1. Correct the left mandibular deviation (3.5 mm), achieve frontal facial symmetry, and improve the convex profile.
2. Align and level the dental arches, improve occlusal relationships, and correct the canted occlusal plane.
3. Improve masticatory efficiency, establish balanced and stable mandibular movement, and restore TMJ function.

2.3. Treatment alternatives

Initially, the patient declined TMJ surgery owing to uncertain outcomes. Given the severe mandibular asymmetry and the patient's

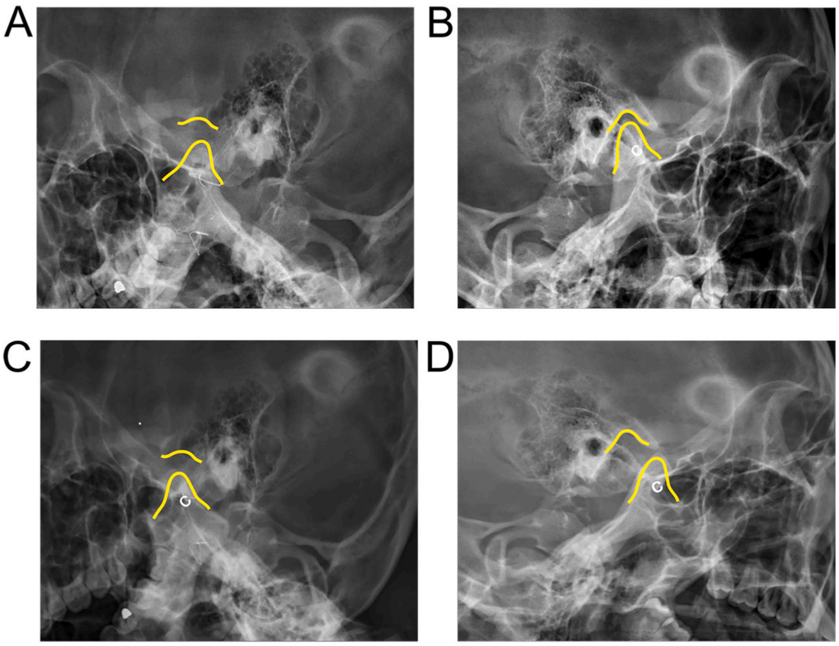

Fig. 2. Pretreatment oblique lateral transcranial radiographs of the TMJ. (A) Left condyle in closing position. (B) Right condyle in closing position. (C) Left condyle in opening position. (D) Right condyle in opening position.

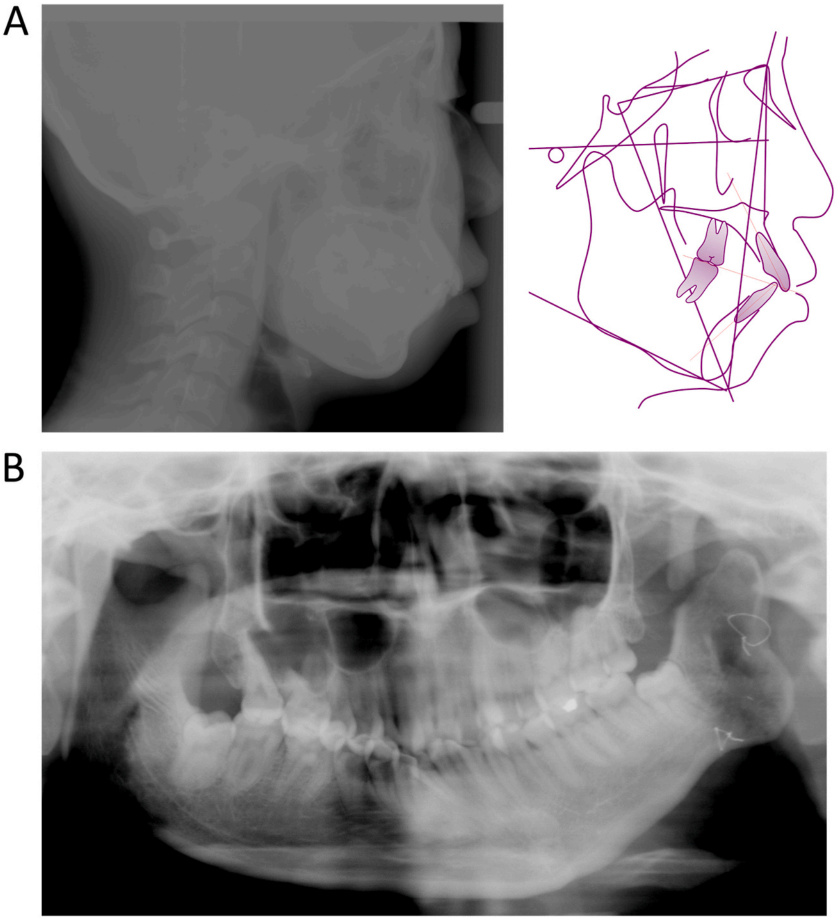

Fig. 3. (A) Pretreatment lateral cephalogram and cephalometric tracing. (B) Pretreatment panoramic radiograph.

Table 1
Cephalometric analysis before and after treatment.

Measurement	Norm± Std Dev	Pretreatment	Posttreatment
SNA (°)	82.8 ± 4.0	74.8	72.5
SNB (°)	80.1 ± 3.9	69.1	70.0
ANB (°)	2.7 ± 2.0	5.7	2.5
U1-NA (°)	22.8 ± 5.7	26.5	23.8
U1-NA (mm)	5.1 ± 2.4	8.8	6.3
L1-NB (°)	30.3 ± 5.8	38.5	29.4
L1-NB (mm)	6.7 ± 2.1	10.2	6.5
Po-NB (°)	1.0 ± 1.5	0.8	3.5
U1-L1 (°)	124.2 ± 8.2	107.2	119.0
OP-SN (°)	16.1 ± 5.0	30.5	24.0
GoGn-SN (°)	32.5 ± 5.2	40.5	38.2
FMA (°)	31.3 ± 5.0	27.5	31.0
FMIA (°)	54.9 ± 6.1	43.0	52.0
IMPA (°)	93.9 ± 6.2	109.5	97.0

SNA, sella-nasion-A point; SNB, sella-nasion-B point; ANB, A point-nasion-B point; U1, upper in-cisor; NA, nasion-A point; L1, lower incisor; NB, nasion-B point; Po, pogonion; OP, occlusal plane; SN, sella-nasion; Go, gonion; Gn, gnathion; FMA, mandibular plane angle; FMIA, Frankfort plane to mandibular incisor angle; IMPA, incisor mandibular plane angle.

desire for facial and dental improvement, the treatment plan combined surgery and orthodontics. The patient exhibited an inclined occlusal plane due to mandibular ramus hypoplasia caused by TMJa and excessive pressure from the masticatory muscles. Additionally, vertical development was insufficient. Considering the extensive bone filling required and the high risk of recurrence and unpredictable stability associated with a single orthodontic–orthognathic treatment, the final treatment plan involved unilateral DO and fixed orthodontic appliances before orthognathic surgery. Presurgical orthodontics was planned after DO to address severe crowding and proclined anterior teeth.

2.4. Treatment progress

2.4.1. Stage one: mandibular DO

According to the treatment plan, distraction was scheduled as the initial step after a thorough examination. After extracting all first premolars and third molars, an oblique osteotomy was performed on the left side of the ramus to place the distractor and facilitate distraction. The distraction rate was maintained at 1 mm/day and monitored every 7 days (Fig. 4A–C). After achieving a chin position close to the facial midline within 3 weeks, with a total shift of approximately 4 cm, distraction was ceased and maintained for 4 months to allow for consolidation and remodeling. Osteogenesis was observed in the gap. The left condyle appeared deformed compared with the right before treatment (Fig. 5A), while the left ramus had been elongated, and bone remodeling occurred on the top of the left condyle after DO (Fig. 5B). Two months after distraction cessation, the patient received treatment with self-ligating brackets (Damon

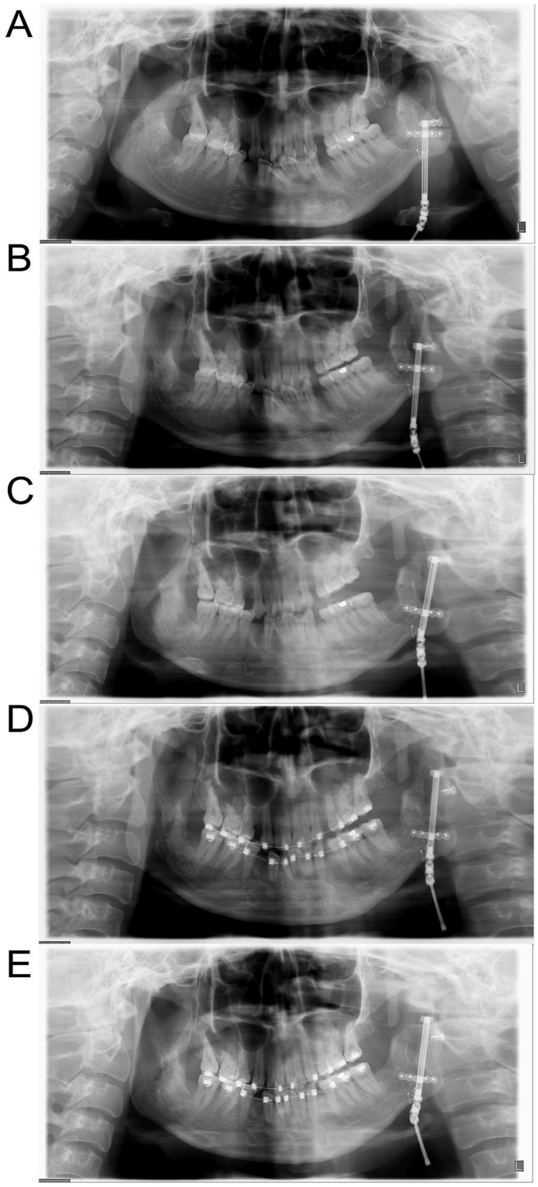

Fig. 4. (A, B, C) Distraction osteogenesis (DO) at 0 days, 7 days, and 1 month (1 week after ceasing distraction). (D) The patient received treatment with self-ligating brackets (Damon Q, 0.022-inch slot size) 2 months after ceasing distraction. (E) Before removal of the distractor, 4 months after ceasing distraction. New bone formation occurred at the left ramus.

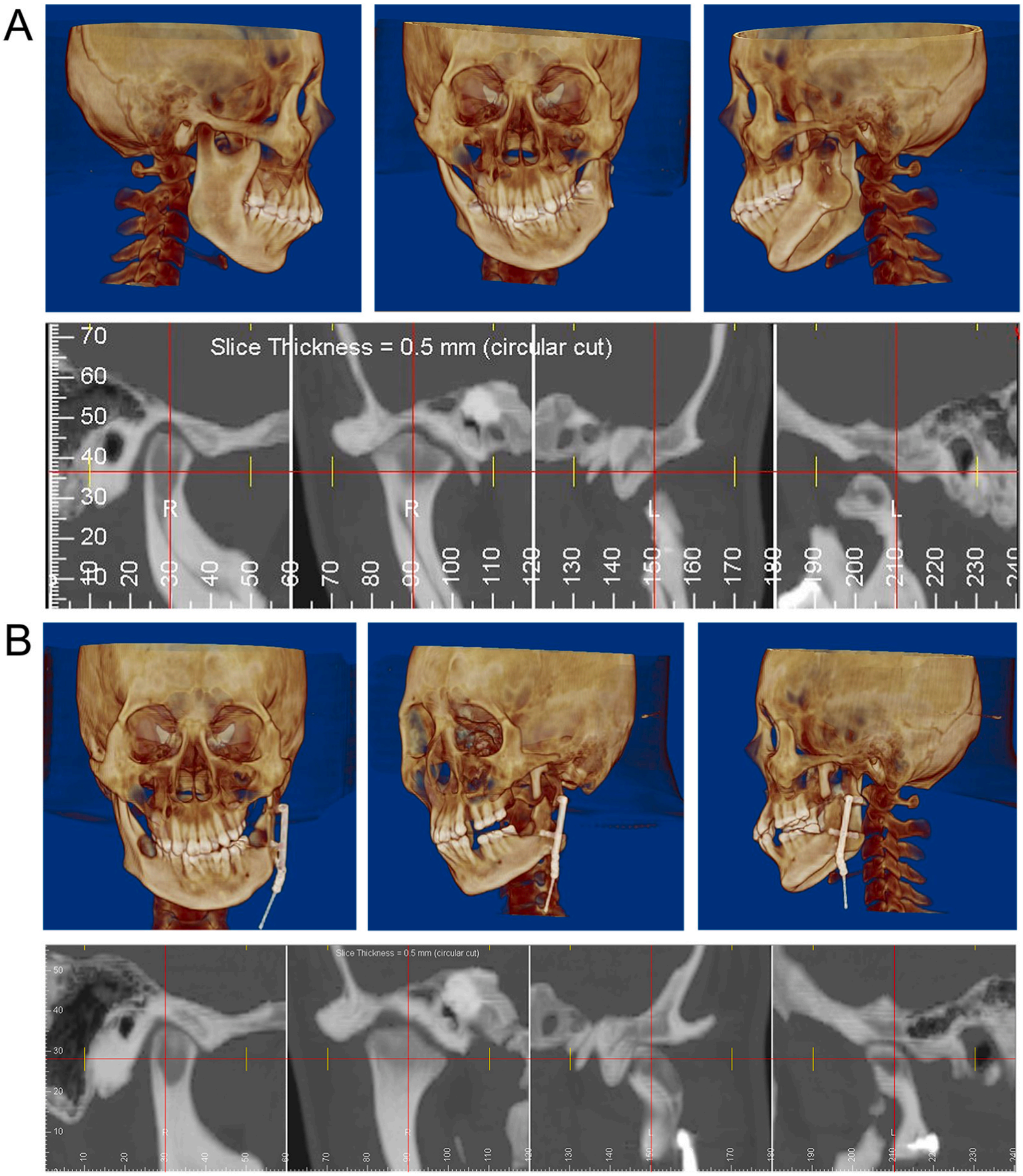

Fig. 5. TMJ morphology before and after DO. (A) TMJ morphology before treatment: the left condyle appeared deformed compared with the right. (B) TMJ morphology after DO: the left ramus had been elongated, and bone remodeling occurred on the top of the left condyle.

Q, 0.022-inch slot size; Fig. 4D and E).

2.4.2. Stage two: orthognathic surgery and orthodontic treatment

After removing the distractor, initial alignment and leveling of the dental arches were achieved using a 0.014-inch nickel–titanium thermal arch wire and occlusal splint. The profile and mouth opening views revealed significant chin movement (Fig. 6A). The occlusal splint opened the bite and eliminated occlusal interference, aiding in alignment (Fig. 6B). However, 7 months after distractor removal, relapse of distraction occurred, and the occlusal plane gradually became canted (Fig. 7A). Efforts were made to stabilize the occlusal relationship and correct the occlusal plane with an occlusal splint during the space-closing stage, but the results were unsatisfactory.

Considering the patient's high demands, it was decided to proceed with orthognathic surgery (Le Fort I + bilateral BSSO + genioplasty). Presurgical alignment, leveling, and closure of the extraction space with maximum anchorage were continued until a 0.019 × 25 stainless steel archwire was reached (Fig. 7B). Postsurgical orthodontics lasted 8 months to close the remaining space, adjust canine and molar relationships, and establish final occlusion (Fig. 7C). The patient was taught tongue muscle and masseter muscle training to practice throughout the entire recovery stage. The left condyle and ramus showed significant self-remodeling 27 months

Fig. 6. Two months after removal of the distractor. (A) The profile and mouth opening view showed significant movement of the chin. (B) Initial alignment and leveling of the dental arch were achieved using a 0.014-inch nickel–titanium thermal archwire and an occlusal splint. The occlusal splint opened the bite and eliminated occlusal interference, aiding in alignment.

after DO pre-surgery (Fig. 8A) and appeared more functional after orthognathic surgery (Fig. 8B).

2.5. Treatment results

2.5.1. Improvement of occlusion relationship, facial symmetry, and profile esthetics

Post-treatment extraoral (Fig. 9A) and intraoral views (Fig. 9B) revealed both upper and lower midlines consistent with the facial midline. Class I canine and molar relationships were achieved on both sides, and reasonable functional contacts were established. The profile appeared straight after extraction of the first premolars, as shown in the post-treatment lateral radiograph (Fig. 10A). The post-treatment panoramic radiograph showed the impacted lower third molars had been extracted (Fig. 10B). The total treatment time,

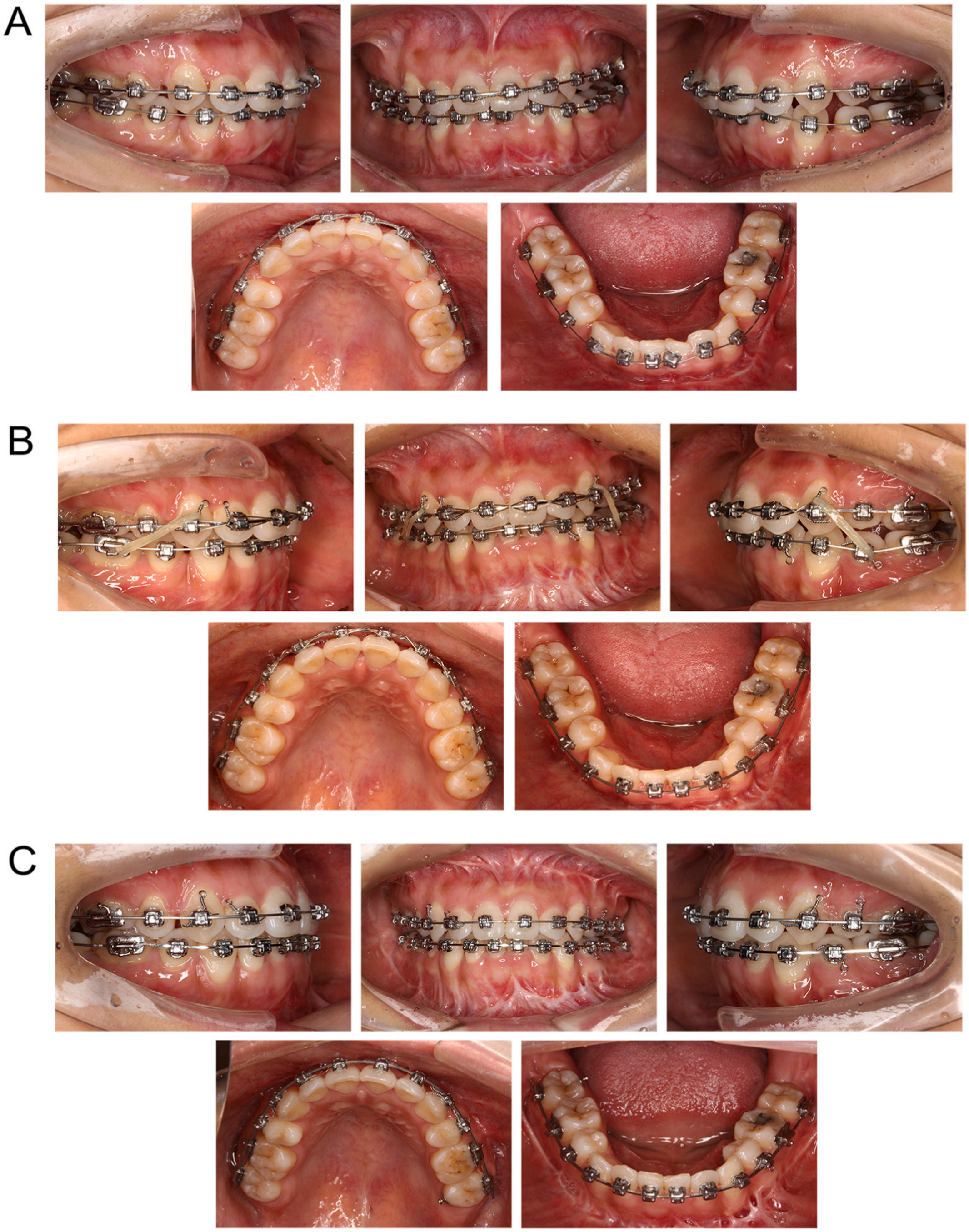

Fig. 7. (A) Seven months after removal of the distractor, the occlusal plane gradually canted (0.018 × 25 nickel–titanium wire in the upper arch; 0.018 nickel–titanium wire in the lower arch). (B) Presurgical alignment and leveling continued until a 0.019 × 25 stainless steel archwire was reached. (C) Two months after surgery. Class II elastics (3/16, 3.5 oz) helped adjust the canine and molar relationship (0.016 × 22 nickel–titanium wire in the upper arch; 0.017 × 25 nickel–titanium wire in the lower arch).

including orthognathic surgery, was approximately 26 months. Cephalometric evaluation (Table 1) and lateral superimpositions (Fig. 11) before and after treatment revealed prominent uprighting of incisors (reduced U1-L1 value). Six months postsurgery, when a reasonable occlusion was achieved, the appliances were debonded, and a vacuum-formed retainer was provided. The first-year extraoral (Fig. 12A) and intraoral views (Fig. 12B), the second-year intraoral views (Fig. 13) and the fourth-year extraoral (Fig. 14A) and intraoral views (Fig. 14B) confirmed long-term stability during follow ups.

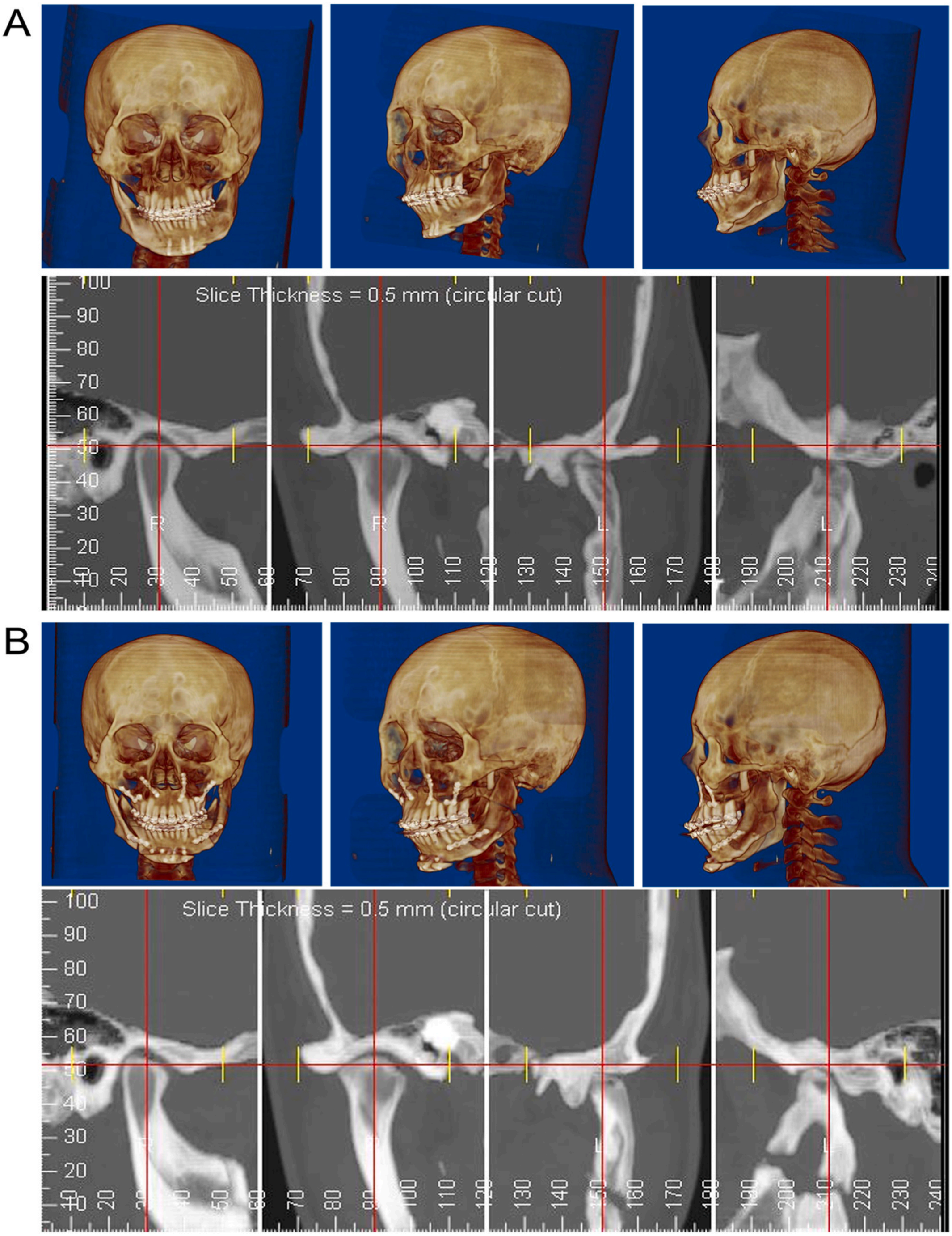

Fig. 8. TMJ morphology before and after surgery. (A) TMJ morphology before surgery: the left condyle and ramus showed significant self-remodeling 27 months after DO. (B) TMJ morphology after surgery.

2.5.2. *Improvement of mandibular movement, TMJ function and morphology*

Regular examinations were conducted to assess mandibular bilateral and protrusive movement after treatment. Typical canine guidance was established during left movement. Unfortunately, lateral movement to the right side remained completely restricted. Surprisingly, cone-beam computed tomography (CBCT) revealed a remarkable curve in the newly positioned fossa, along with increased bone mass (Fig. 15A). Additionally, the second-year (Fig. 15B) and the fourth-year (Fig. 15C) follow-ups confirmed the long-term stability of TMJ self-remodeling. 3D reconstruction of mandible at the time of pre (Fig. 16A) and post (Fig. 16B) treatment and the fourth-year follow-up (Fig. 16C) by 3D Slicer software (Version 5.2.1) indicated that the morphological structure of left condyle

Fig. 9. (A) Post-treatment extraoral views. (B) Post-treatment intraoral views.

appeared clearer along with TMJ self-remodeling after treatment as new bone formation was significant on the top of the condyle.

3. Discussion

3.1. The effect of DO in the treatment

An adult patient with facial asymmetry and condylar joint deformity caused by TMJ arthritis underwent surgery and fixed orthodontic treatment. DO was used to address bone insufficiency by gradually separating bony segments and promoting the formation

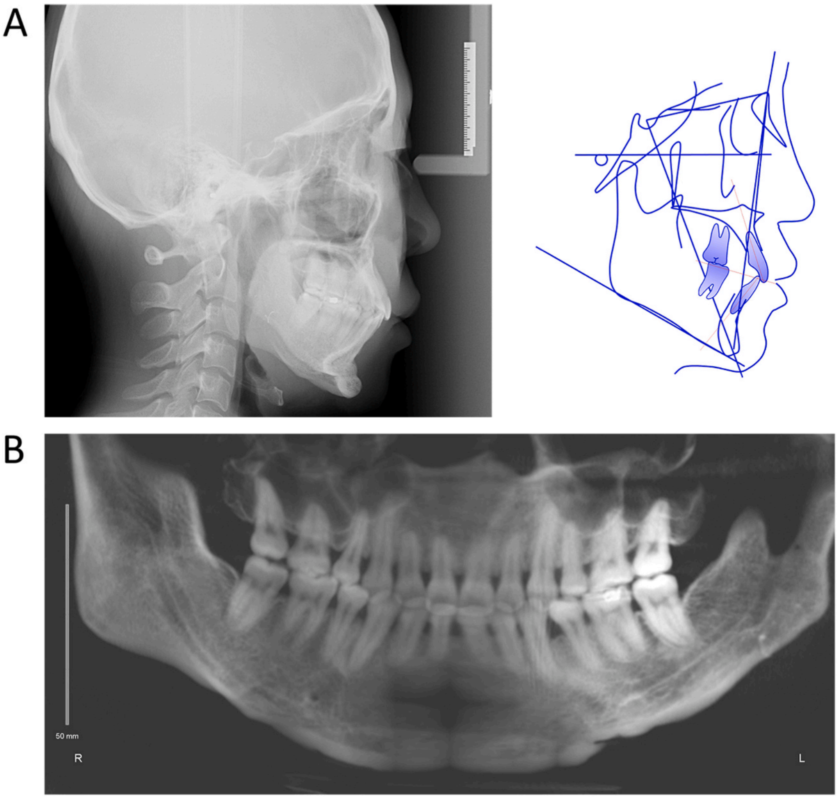

Fig. 10. (A) Post-treatment lateral cephalogram and cephalometric tracing. (B) Post-treatment panoramic radiograph.

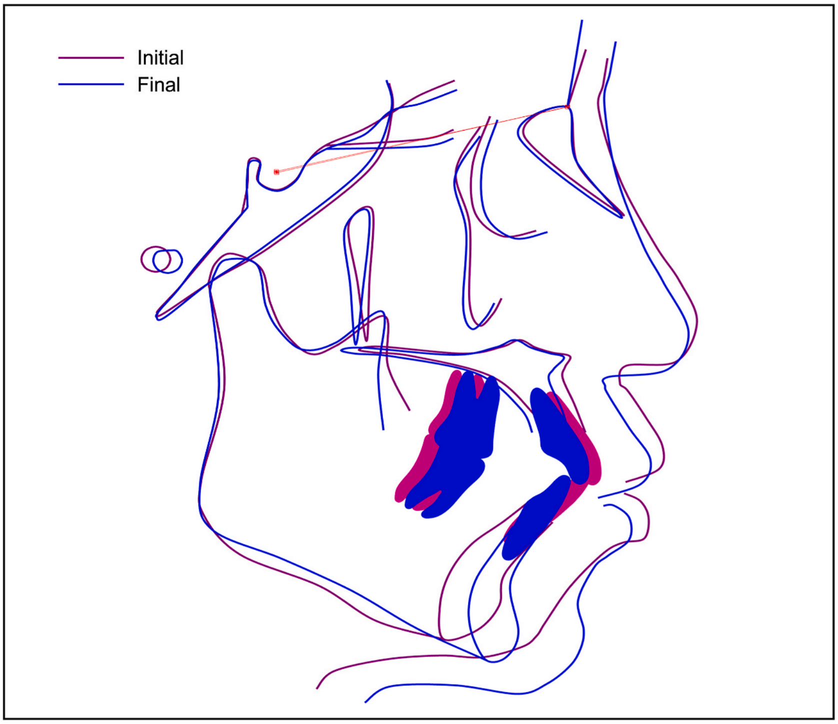

Fig. 11. Pretreatment and post-treatment lateral cephalometric tracing superimpositions based on the SN plane.

Fig. 12. (A) Extraoral views 11 months after treatment. (B) Intraoral views 11 months after treatment.

of new bone in the distraction gap. Simultaneously, the soft tissues, including the muscles, skin, blood vessels, and nerves, were elongated [8]. This approach eliminates the need for a second surgical procedure, such as bone grafting, and offers advantages over conventional orthognathic surgery, including reduced trauma, postoperative complications, and improved stability, although the same degree of precision might not be possible [9].

Mandibular DO has been widely used to lengthen the bone and improve facial asymmetry. However, its complications cannot be ignored, including chance of relapse, injury to the teeth and nerve, infection, device failure, inappropriate distraction vector, and fusion error [10]. Van strijen et al. reviewed 70 bilateral MDO cases and categorized all the complications into three categories: intra-operative, intra-distraction, and post-distraction [11]. A previous study also reported that TMJ injury accounts for 0.2 % of all the complications caused by MDO [12]. Xia et al. aimed to evaluate the remodeling of condyles reconstructed by MDO in patients with TMJa and found that MDO combined with gap arthroplasty was an effective method for the treatment of TMJa to improve maximum

Fig. 13. Intraoral view 2 years after treatment (unfortunately, the extraoral view is missing).

mouth opening (MMO), but the reconstructed condyle also exhibited a high frequency of resorption in height [13]. In this specific case, MDO not only promoted bone formation but also facilitated the reconstruction of the condyle. A new condyle with a reconstructed glenoid fossa in a forward position was established after MDO (Figs. 15 and 16), which was the key to gain new structure of a functional TMJ. However, due to stability issues, orthognathic surgery and genioplasty were performed to achieve facial esthetics. The post-treatment results showed excellent esthetics and facial symmetry.

3.2. *The relapse of DO in the treatment*

Despite the efficiency of DO in new bone formation, however, in the present case, the patient experienced distraction relapse after 3 months of maintenance. Several factors could have contributed to this relapse, including the speed and distance of distraction, the duration of the maintenance period, and the patient's age. A previous study concluded that high-angle patients are at a higher risk of relapse compared with low-angle patients [14]. Additionally, the patient's previous arthrolysis procedure at a young age may have influenced the instability and abnormal structure of the TMJ, potentially impacting the relapse of DO. In a study combining transport disk distraction osteogenesis with arthroplasty, the use of temporalis fascia and a pedicled buccal fat pad helped fill the gap and prevent recurrence [15].

After distraction, the maxillary fixation period lasted 4–6 months, whereas the mandibular fixation period lasted 3–4 months. Fixation is crucial to prevent the collapse of new bone tissue and the recurrence of deformity. However, stability depends on various factors, including new bone collapse, scar contracture, unilateral mastication, unbalanced occlusion, unstable joint position, and long-term muscle imbalance [8,9]. In addition to skeletal changes, DO also impacts the facial soft tissue profile [16]. Despite the limited volume of new bone, the myofiber extension resulting from osteogenesis and modification of surrounding soft tissues contribute to harmonic muscle strength for interfragmentary stability after orthognathic surgery, reducing the likelihood of long-term recurrence. Although the long-term stability of mandibular DO remains a topic of debate, satisfactory facial esthetics could be maintained for up to 7–12 years postoperatively [17].

3.3. *The treatment timing of DO*

Regarding the overall treatment plan for TMJa, various opinions are held on the sequencing of TMJ release and distraction osteogenesis. The three main approaches are release of TMJa followed by DO, DO followed by release of TMJa, and simultaneous release of TMJa and DO. The single-stage treatment saves secondary surgical procedures and costs but presents challenges in controlling the amount and vector of distraction owing to an unstable condyle. Performing DO first helps increase the airway and achieve stable results [18,19], making it suitable for patients with obstructive sleep apnea–hypopnea syndrome [20]. However, releasing TMJa first provides patients with a functional TMJ and mastication system [21], thereby restoring oral function [22] and allowing for a more accurate preoperative evaluation [23].

Regardless of the treatment sequence, DO remains an effective approach for correcting mandibular deviation caused by TMJa. However, many patients may require additional adjunctive surgeries, such as genioplasty or grafting, to enhance facial esthetics [23]. Notably, iatrogenic ankylosis can occur after autologous reconstruction of the ramus–condyle unit for severe mandibular hypoplasia [24].

Fig. 14. (A) Extraoral views 4 years after treatment. (B) Intraoral views 4 years after treatment.

3.4. *The potential causes of TMJ self-remodeling achieved by DO*

Jaw remodeling inevitably affects the occlusion relationship. In the present case, combining DO with a fixed appliance effectively eliminated occlusal interference, facilitating maximum mandibular shift. Postsurgical orthodontic treatment achieved a coordinated intercuspal relationship. Notably, there were observable changes in disk displacement and TMJ morphology. After treatment, left lateral movement of the jaw and normal canine guidance were established. Follow-up records demonstrated successful establishment of functional occlusion and balance in the oral and maxillofacial muscle system, leading to more effective mastication. CBCT images revealed improved morphology of the glenoid fossa and articular head 4 years after surgery, particularly on the left side, suggesting that function contributes to TMJ remodeling. Further studies are required to confirm these findings.

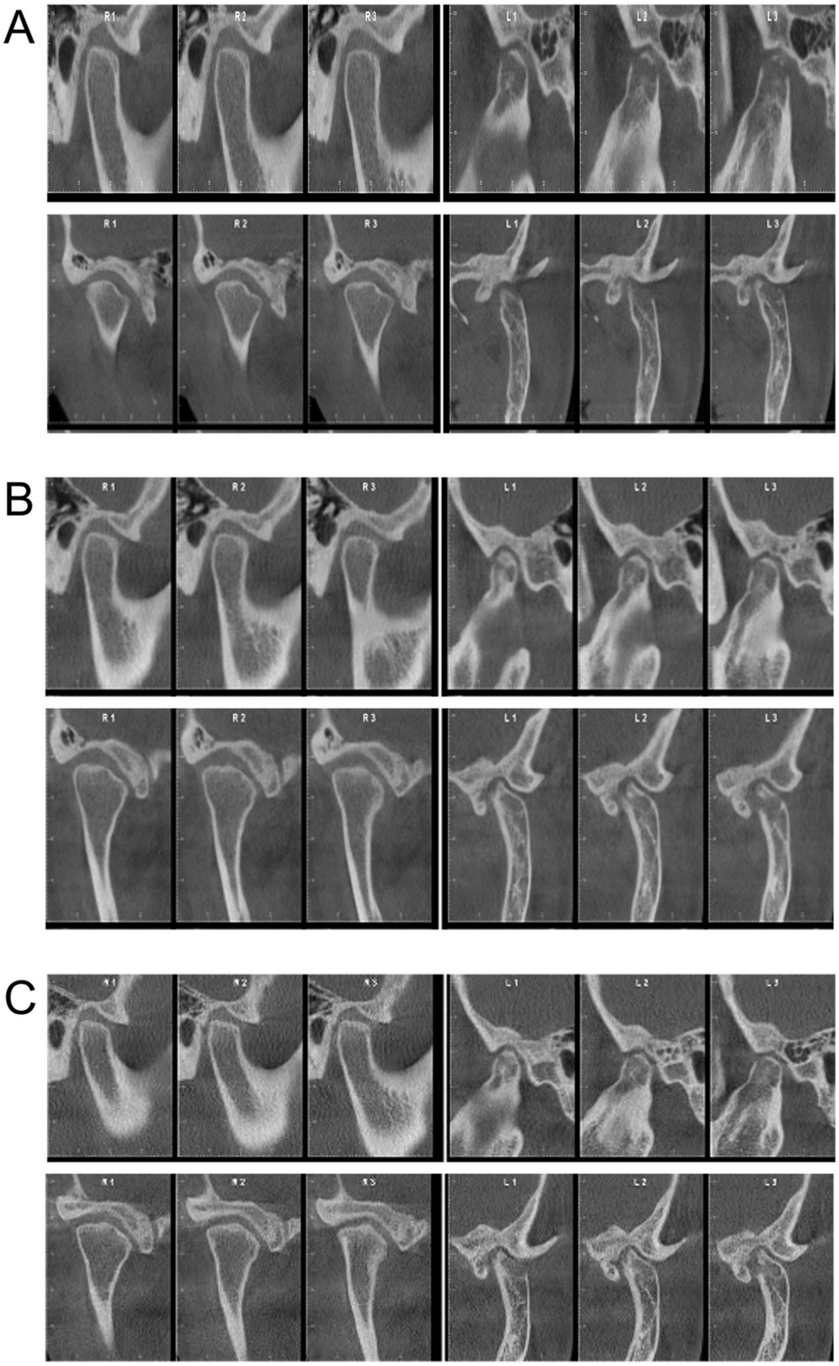

Fig. 15. (A) Bilateral CBCT image of the TMJ immediately after treatment. The newly positioned fossa formed a remarkable curve, and there was an increase in bone mass. (B) Bilateral CBCT image of the TMJ 2 years after treatment. (C) Bilateral CBCT image of the TMJ 4 years after treatment. Long-term stability of TMJ self-remodeling was confirmed.

McCormick et al. observed increased volume and optimized space orientation of the TMJ early after distraction [25]. A systematic review of animal experimental studies indicated that mandibular distraction osteogenesis (MDO) can induce adaptive changes in the TMJ, including thickening of the articular cartilage, increased bone formation, and remodeling of the condyle, depending on the daily distraction rates and total length of DO [26]. The physiological mechanism involves changes in condyle loading and the vector applied during DO. The biological principles of bone formation reported by these animal studies have confirmed that a neocondyle with functional shape can be created by MDO. However, given that these adaptations are reversible, achieving balanced occlusion and stable TMJ function is crucial for maintaining such reconstruction. Mihmanli et al. suggested that increased cartilage thickness is a

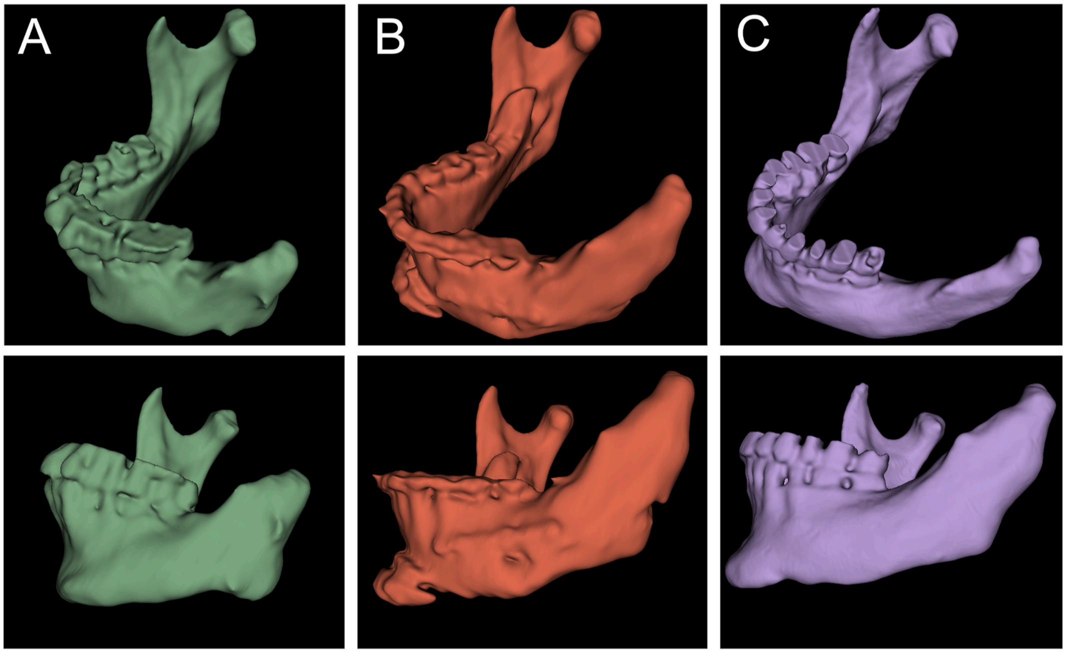

Fig. 16. (A) 3D reconstruction of mandible before treatment. The bone defect and absence of normal condyle structure was significant in the left ramus compared with the right side. (B) 3D reconstruction of mandible after left mandibular DO and orthognathic surgery (Le Fort I + bilateral BSSO + genioplasty). The height of left ramus was significantly improved, and the basic structure of condyle was preliminary obtained. (C) 3D reconstruction of mandible at the fourth year of follow-up. The morphological structure of left condyle appeared clearer with self-remodeling after treatment.

reactive response to maintain functional balance and resistance to external wear in the TMJ [27]. In the present case, CBCT images did not readily confirm the regeneration of condylar cartilage, and the lack of histological confirmation is one of the limitations of this case. Furthermore, longer-term follow-up is necessary, as MDO can lead to TMJa in cases with congenital TMJ deformities, necessitating careful monitoring of TMJ health during and after MDO [28].

Although DO can be a valuable tool for mandibular reconstruction in cases of TMJa-induced deviation, TMJ reconstruction and remodeling are most effective in growing children. In cases of ankylosis caused by injury or inflammation, the timing and type of reconstruction should be carefully evaluated, particularly in skeletally immature patients, considering the potential for future growth after reconstruction [29]. A study by Yu et al. indicated that simultaneous gap arthroplasty and DO in young patients with unilateral TMJa and micrognathia can restore normal mandibular growth by re-establishing correct skeletal and soft tissue function, as the forces produced by the distractor are similar to physiological forces during mandibular development [30]. Therefore, an early treatment plan combining these procedures may lead to better outcomes in mandibular growth and TMJ remodeling.

4. Conclusion

A successful treatment approach involved DO and orthognathic surgery combined with fixed orthodontics was employed for an adult patient presenting with mandibular deviation and proclined, crooked teeth due to unilateral TMJ ankylosis. The treatment results achieved improvement of occlusion relationship, facial symmetry, profile esthetics, mandibular movement, TMJ function and morphology. The patient's 4-year follow-up indicated sustained facial and occlusal stability, as well as significant adaptive self-remodeling of the TMJ. A new condyle with a reconstructed glenoid fossa in a forward position was established in the ankylosis side. Therefore, this case proves that DO enables to promote adaptive TMJ self-remodeling with long-term stability in the treatment of mandibular deviation caused by condylar ankylosis in adult patients, which highlighted the superior benefits of the combined approach compared with simple orthognathic surgery for the treatment plan.

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Ethics statement

This study was reviewed and approved by the Institutional Review Board of the School of Stomatology, the Fourth Military Medical University, Xi'an city, China, with the approval number: [IRB-REV-2022116]. The patient provided informed consent to participate in the study and for the publication of her anonymized case details and images.

Additional information

No additional information is available for this paper.

Data availability statement

Data included in article.

CRediT authorship contribution statement

Axian Wang: Writing – original draft, Methodology, Investigation, Funding acquisition, Data curation. **Sijie Wang:** Writing – review & editing, Writing – original draft, Visualization, Methodology, Investigation, Formal analysis, Data curation. **Yu Zhou:** Visualization, Software, Formal analysis, Data curation. **Yi Wen:** Visualization, Software, Investigation, Formal analysis. **Zuolin Jin:** Validation, Supervision, Conceptualization. **Xiaoyan Chen:** Validation, Supervision, Funding acquisition, Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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None.

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