Comprehensive treatment approach for bilateral idiopathic condylar resorption and anterior open bite with customized lingual braces and total joint prostheses

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This case report describes the successful treatment of a 14-year-old girl with severe bilateral idiopathic condylar resorption and resultant mandibular retrusion, increased overjet, and anterior open bite. The nonextraction treatment plan included (1) aligning and leveling the teeth in both arches, (2) performing Le Fort I maxillary osteotomy, bilateral condylectomy, and mandibular joint replacement, and (3) postsurgical correction of the malocclusion. The orthodontic treatment was initiated with the use of custom lingual appliances followed by orthognathic surgery planned with virtual surgical planning. Patient-fitted and customized temporomandibular joint implants were designed and manufactured based on the patient's stereolithic bone anatomic model. Treatment was concluded with detailed orthodontic finishing. Optimum esthetic and functional results were achieved with the cooperation of 2 specialties and the use of state-of-the-art technology. (Am J Orthod Dentofacial Orthop 2019;156:125-36)

Idiopathic condylar resorption (ICR), also known as idiopathic condylysis, is a progressive degenerative disease of the temporomandibular joint (TMJ). It is most commonly seen in female patients (approximate female:male ratio 9:1)[1] at the ages of 10-40 years (predominant in teenagers during pubertal growth phase).[2] Studies have shown that 25% patients with ICR have no TMJ dysfunction symptoms.[3] ICR can often cause skeletal and occlusal instability (mostly Class II malocclusion), dentofacial deformities, and TMJ dysfunction and pain.[4] Bilateral involvement of condyles creates progressively retrusion of the mandible, high occlusal and mandibular plane angle, and Class II occlusion with or without anterior open bite.[3] In unilateral cases, mandibular midline and chin shifts to the affected side, developing crossbite and posterior occlusal prematurities. Condylar degeneration is associated with several local and systemic factors, including osteoarthritis, traumatic injuries, rheumatoid arthritis, ankylosing spondylitis, and others.[5,6] However, condylar changes without specific local or systemic etiologies are described as "idiopathic." Thus, ICR has a different cause and pathosis compared with other condylar resorption conditions and therefore requires special consideration in treatment. Because ICR is often aggressive and fast-moving, precise diagnosis and proper treatment planning are crucial. The diagnosis of ICR is generally based on the patient's history, clinical evaluation, and radiographic examination. Previously recommended treatment options for ICR include splint therapy, anthroscopic lysis and lavage, condylar replacement with the use of a costochondral graft, and articular disk repositioning and stabilization.[3,7-9] In the present case report, the comprehensive treatment of girl with bilateral ICR and anterior open bite is discussed and an effective treatment protocol, including presurgical orthodontics for arch alignment and leveling, orthognathic surgery with condylar prosthesis, and postsurgical orthodontics, is presented with predictable and stable outcomes achieved.

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DIAGNOSIS AND ETIOLOGY

In January 2014, a 14-year-old girl presented to the Department of Orthodontics, University of Alabama at

Fig 1. Initial extraoral and intraoral photographs.

Birmingham, with the chief complaint of "I want jaw surgery to bring my lower jaw forward to get rid of my overbite and to get rid of my double chin." The patient said that she had difficulty eating food because her front teeth could not touch. Clinical examination revealed convex facial profile, retrognathic and hyperdivergent mandible, and retrusive chin in a skeletal Class II base. Her malocclusion was complicated by 3 mm of open bite (incisor), 6 mm of overjet, and mild crowding (2 mm) in the lower arch. The maxillary dental midline was coincident with the facial midline but the mandibular midline was deviated 2 mm to the left. There was no popping, clicking, or crepitation of the TMJ. During the next visit, extraoral and intraoral photographs and upper and lower impressions were taken along with the lateral cephalometric and panoramic radiographs and cone-beam computed tomographic (CBCT) scanning (Figs 1 and 2).

Dental casts showed an open bite from premolars to premolars with 3 mm open bite on the incisors. On the left side, the first molars were Angle Class I and on the right side, they were in an end-on Class II relationship. Mandibular incisors were severely proclined with upright maxillary incisors. The panoramic radiograph showed that all teeth were present except the third molars. Right and left condyles had undergone complete resorption. Lateral cephalometric analysis revealed bilateral ICR, skeletal and occlusal Class II deformity with ANB 8.9°, anterior open bite, hyperdivergent mandibles with increase mandibular plane angle (SN-MP 46.3°), decreased vertical height of ramus, and proclined mandibular incisors (L1-NB 34°). CBCT showed the resorption of the condyles to the level of the sigmoid notch. SICAT jaw motion tracking was performed and the data provided visualization of true mandibular movements and predicted the condylar position without further radiation exposure.

TREATMENT PLAN

The patient was diagnosed with bilateral ICR, mandibular retrognathism, Angle Class II malocclusion,

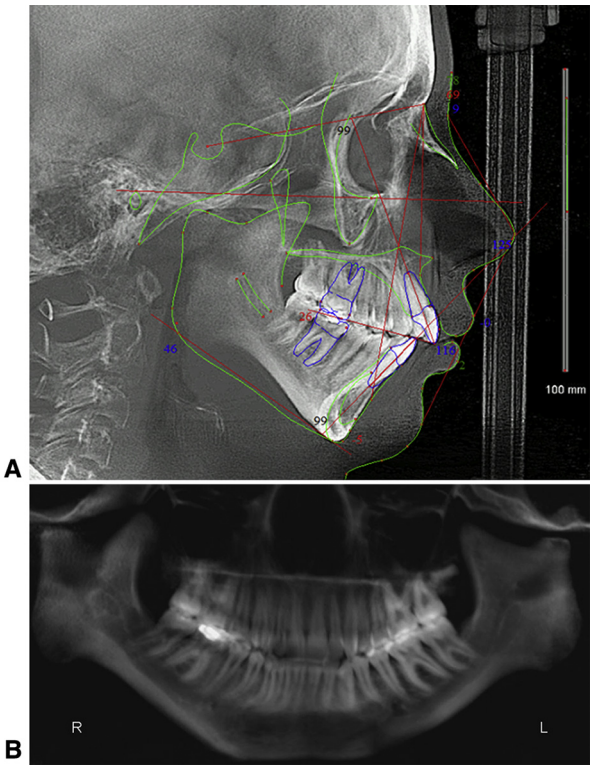

Fig 2. A, Pretreatment lateral cephalometric radiograph with tracing. **B,** Pretreatment panoramic radiograph.

anterior open bite, increased overjet, mild mandibular crowding, and lower midline deviation. The dental, skeletal, and soft-tissue treatment objectives for this patient were: (1) eliminate crowding in the lower anterior segment and correct anterior open bite and overjet; (2) decrease the high angle, reduce ANB with mandibular advancement, eradicate TMJ resorptive phenomenon by reconstruction of condyles bilaterally, and correct chin retrusion with genioplasty; and (3) correct the convex facial profile and retrusive chin.

After careful consideration of all treatment alternatives together with the patient, the final treatment plan was decided as nonextraction maxillary and mandibular arch fixed appliance orthodontic treatment to align and level the teeth, followed by Le Fort I maxillary osteotomy to advance the maxilla, and bilateral condylectomy and mandibular joint replacement with patient-fitted TMJ implants. Postsurgical orthodontic treatment was also planned to correct minor discrepancies. Furthermore, the patient was given the esthetic option of brackets despite the complexity of the case. Thus, a self-ligating fully customized lingual appliance (Harmony System, Paris, France) was used for the presurgical correction of dental alignment and leveling. The SICAT jaw motion tracking system was used to analyze mandibular movements. Preoperative virtual surgical planning (VSP) was performed before orthognathic surgery, and postsurgical interarch settling was done with the use of clear self-ligating brackets.

TREATMENT PROGRESS

Maxillary and mandibular high-accuracy polyvinyl siloxane impressions were obtained and sent to Harmony Systems Lab (American Orthodontics). Customized self-ligating lingual appliances were fabricated and careful consideration was made as to how the final occlusion would be created. Initial leveling was started with the use of 0.014″ NiTi (Fig 3). Wires were sequenced in 0.016 × 0.022 NiTi. In most cases with a skeletal open bite, the maxillary arch shows a skeletal compensation of the skeleton. Thus, in most cases, an accentuated curve of Spee wire in the finishing sequence is used to maintain the open bite. Alternatively, the maxillary arch is aligned in 3 segments. These 2 procedures prevents dental relapse of the dentition back to an open bite situation. However, in this clinical case, the maxillary dentition did not show any compensation and therefore it was leveled normally. The dental open bite was maintained as the mandibular dentition leveled and aligned. Once the leveling and alignment was completed, CBCT and dental casts were taken for medical modeling by virtual surgical planning (VSP Orthognathics; 3D Systems, www.3dsystems.com). Twelve months after the initial bonding, orthognathic surgery was discussed with an oral surgeon to determine a key point to decide the kind of maxillary movements during surgery and to fabricate the mandibular joint prosthesis.

The VSP workflow enabled the practitioners to visualize the expected final outcome with the intermediate and final position of the maxilla and mandible. A soft-tissue simulation was acquired as well. VSP was carried out to correct retrognathic mandible and anterior open bite (Fig 4; Table I). With the use of Digital Imaging and Communications in Medicine (DICOM) data, the stereolithic model was produced with the maxilla and mandible in the final position. The stereolithic model was then sent to TMJ Concepts (Ventura, Calif) for the design, blueprint, and wax-up of the TMJ prostheses. After the design was approved by the surgeon, the custom-fitted total joint prostheses were manufactured. It took ~8 weeks to manufacture the total joint patient-fitted implants. Figure 5 shows the design of the TMJ Concepts patient-fitted prosthesis. The mesh framework on the underside of the custom-fitted titanium shell

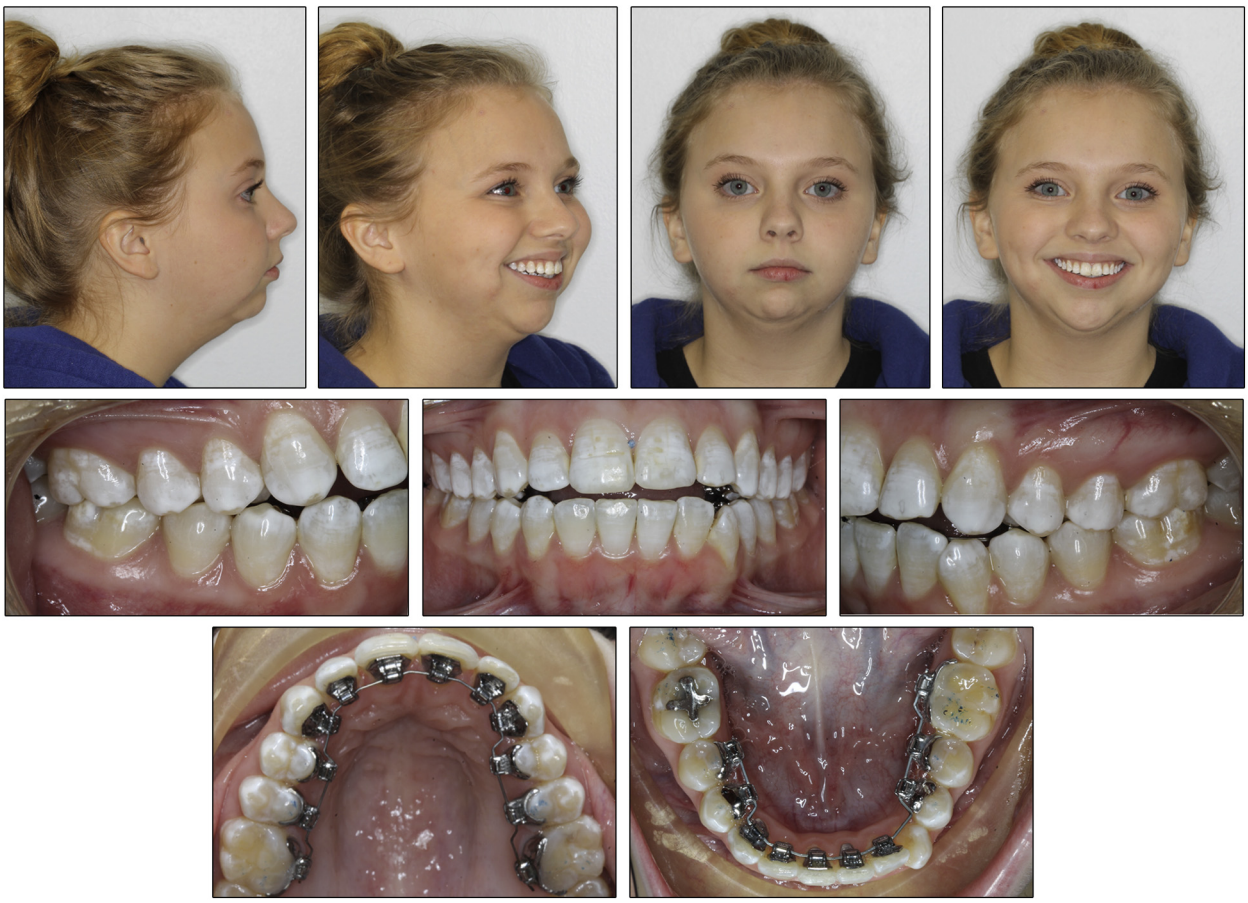

Fig 3. Initial appliance placement.

secures the polyethylene articulating portion of the fossa component and the mesh on the superior surface of the fossa component allows osseointegration with the fossa bone. Screw holes were placed to avoid the alveolar nerve and screw lengths were decided based on measurements taken from the bone model and implants.

Twenty months after the initiation of the orthodontic treatment, orthognathic surgery was performed (PDW; Fig 6). Clear orthodontic brackets were placed on the facial axis point on the labial and buccal surfaces of upper and lower arch to aid fixation of the occlusal splint and interarch rubber band setting. Subperiosteal dissection was achieved to remove the residual condylar stump, which was sent for pathologic examination. Coronoid process was resected below the level of the sigmoid notch. The intermediate splint fabricated by Medical Modeling was placed between the dentition and intermaxillary fixation was done with 26-gauge stainless steel wire. First, the custom condylar fossa was inserted on the right side and secured with screws. Then the condylar component was placed. Eventually, the same procedure was repeated on the left side and the condylar replacement along with the TMJ Concept custom fossa were inserted in the same manner. Le Fort 1 osteotomy was performed from the posterior maxilla through the piriform rim. Thus the pterygoid planes were separated and the maxilla down-fractured. The final splint was placed and maxillary-mandibular fixation was done. Four L-shaped plates were incorporated in the maxilla, one on each side on the nasomaxillary buttress and one on each side along the pterygomaxillary buttress. Advancement genioplasty was also performed to correct the retrusive chin and improve the cervicomental angle. Final occlusion was verified and found to be satisfactory. Light interarch elastics were placed and the patient was extubated without any complication. The patient was then transferred to postoperative care unit and subsequently discharged after 2 days of postoperative observation and care. She was given Norco elixir, Zofran, Robaxin,

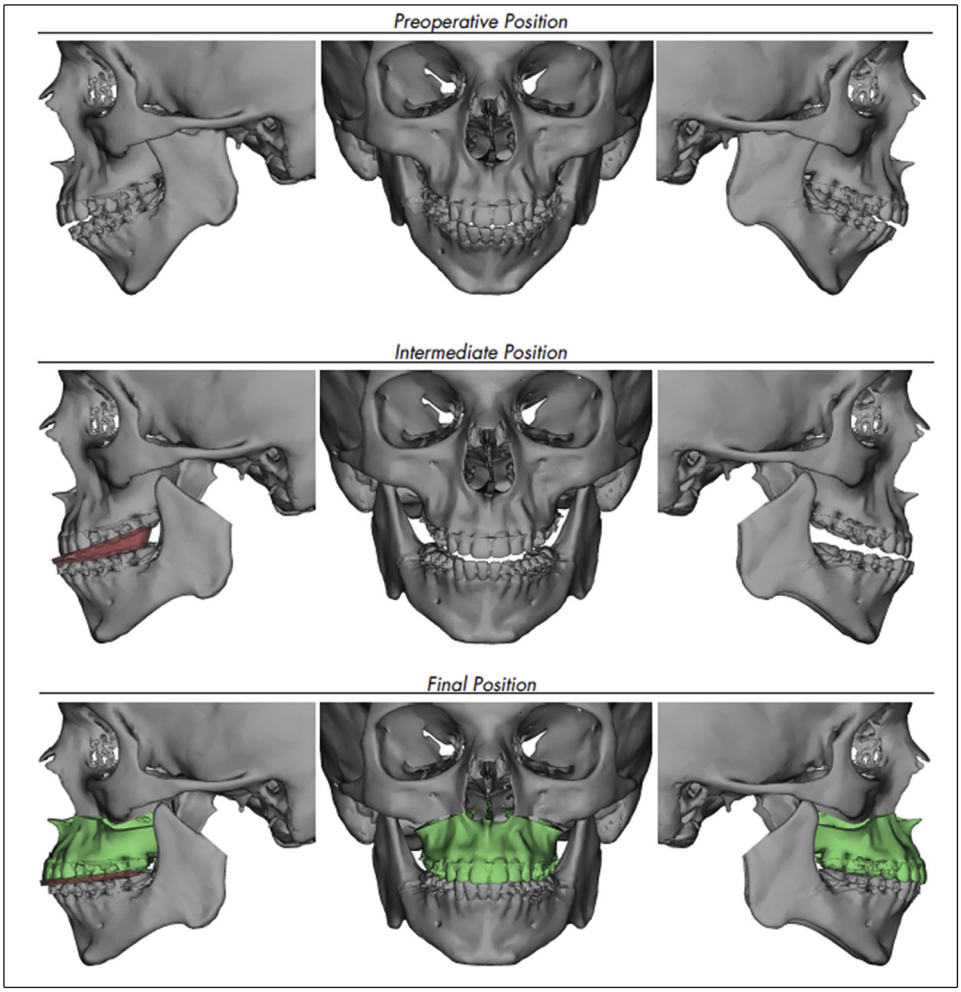

Fig 4. Virtual surgical planning workflow for Le Fort I maxillary osteotomies.

Table I. Occlusal and bony anatomic landmarks and their summarized movements from preoperative position (with mandible autorotated close) to simulated postoperative position

Point	*Name*	*Anterior/posterior*	*Left/right*	*Up/down*
ANS	Anterior nasal spine	1.33 mm anterior	0.99 mm right	2.96 mm up
A	A point	3.07 mm anterior	0.92 mm right	1.92 mm up
ISU1	Midline of upper incisor	7.00 mm anterior	1.00 mm right	2.50 mm up
U3L	Upper left canine	7.00 mm anterior	0.86 mm right	0.80 mm up
U6L	Upper left anterior molar (mesiobuccal cusp)	6.31 mm anterior	0.54 mm right	2.98 mm down
U3R	Upper right canine	6.41 mm anterior	0.88 mm right	1.09 mm up
U6R	Upper right anterior molar (mesiobuccal cusp)	5.11 mm anterior	0.55 mm right	2.71 mm down
ISL1	Midline of lower incisor	8.60 mm anterior	0.69 mm right	4.83 mm up
L6L	Lower left anterior molar (mesiobuccal cusp)	6.75 mm anterior	0.70 mm right	2.99 mm down
L6R	Lower right anterior molar (mesiobuccal cusp)	6.40 mm anterior	0.69 mm right	2.56 mm down
B	B point	14.56 mm anterior	0.76 mm right	0.74 mm up
Pog	Pogonion	18.47 mm anterior	0.81 mm right	1.12 mm down

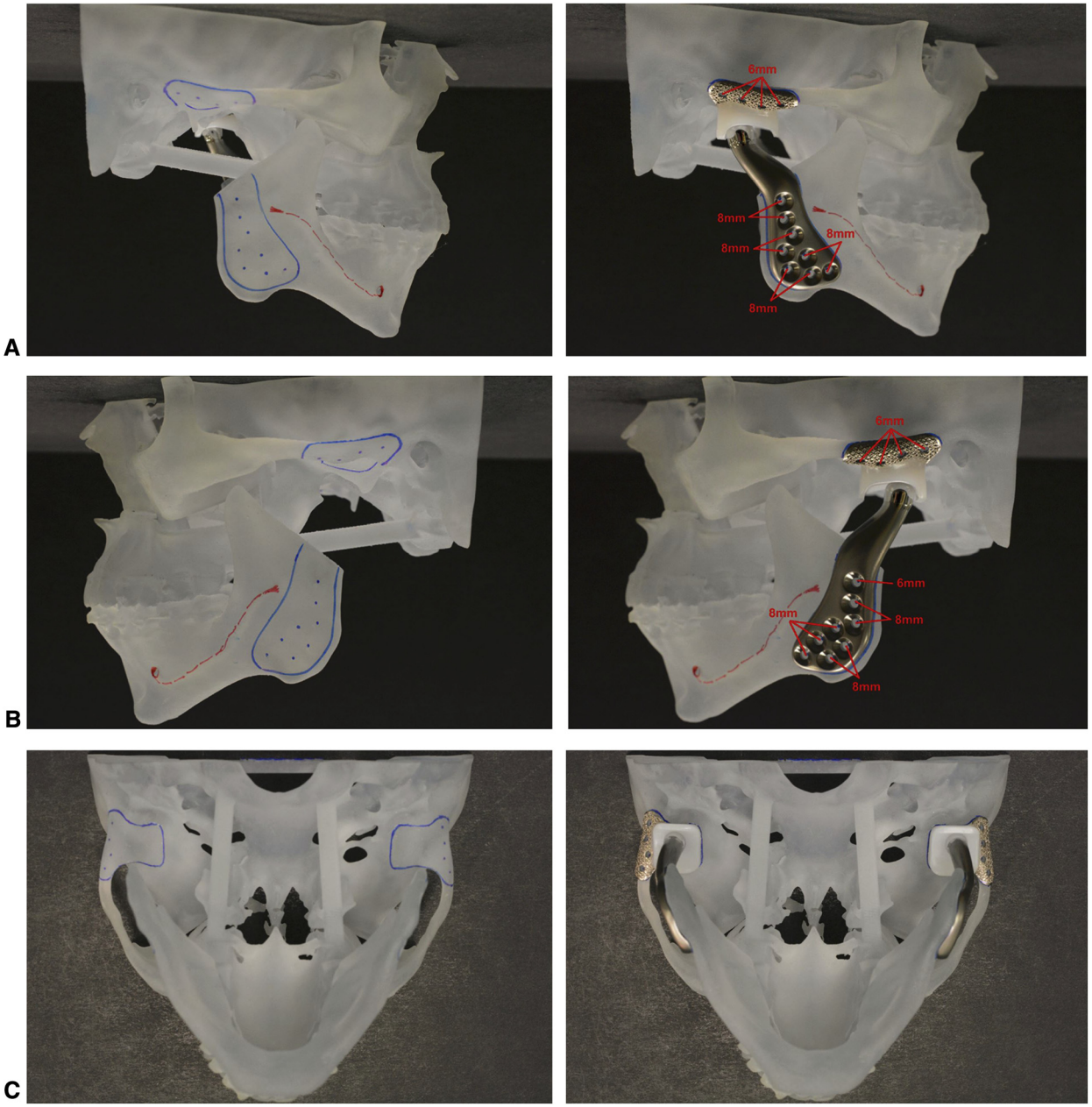

Fig 5. Patient-fitted TMJ implants for the orthognathic surgery of joint reconstruction. **A,** Right side TMJ implant on the anatomical bone model. **B,** Left side TMJ implant on the anatomical bone model. **C,** Bilateral TMJ implants on the anatomical bone model.

and Guaifenesin as required for standard orthognathic surgery.

Five weeks after the successful completion of the surgery, the splint was removed (C.H.K.). Maxillary and mandibular arch alignment along with the minor occlusal adjustments were started with the use of 0.014″ NiTi (Fig 7). Postsurgical setting was performed with elastic bands. Two months after the surgery, lingual brackets were removed and occlusal settlements were done with the use of labial orthodontics.

TREATMENT RESULTS

The posttreatment records show a balanced facial profile and occlusion. ICR was treated and bilateral condylar reconstruction was accomplished exactly as

Fig 6. Maxillary and mandibular teeth with clear brackets, just before surgery.

simulated in VSP. Intraorally, the anterior open bite was corrected, normal overjet and overbite were achieved, teeth were properly leveled and aligned, upper and lower midlines were coincided with the facial midline, and incisor and canine guidance were obtained. The patient's soft tissue profile was straight, neck throat angle significantly was reduced, and esthetically pleasant smile arc was achieved with lip competency and adequate gingival display. Skeletally, ANB angle was reduced to 2.6°, SN-MP was reduced to 31.4°. In addition, L1 to MP was increased by 107.1°. Furthermore, soft tissue convexity was reduced to normal (Fig 8; Table II).

Twenty-five months after the initiation of orthodontic treatment, debonding was carried out. Facial, intraoral, and extraoral photographs, CBCT, and dental impressions were taken for final records. Middle-postsurgical and final cephalometric radiographs were superimposed on the initial lateral cephalometric radiograph (Fig 9). The final outcome of our comprehensive treatment approach satisfied both functional and esthetic demands (Fig 10). Retrognathic mandible and anterior open bite were corrected successfully.

DISCUSSION

Condylar resorption is a rare event seen in normal orthodontic practice. However, it can result in malocclusion, TMJ dysfunction, facial disfigurement, and pain when it occurs. Therefore, careful differential diagnosis and treatment planning of condylar resorption is of utmost importance. Although the specific etiology of condylar degeneration has not been clearly understood, Sarver et al[5] summarized that condylar changes might be due to several localized factors, systemic factors, postoperative factors, or idiopathic origin. Possible localized factors are osteoarthritis, reactive arthritis, traumatic injuries, and infection. Systemic etiologic factors include rheumatoid arthritis, juvenile idiopathic arthritis, systemic lupus erythromatosus, Sjögren syndrome, and psoriatic arthritis.[5] In addition, parafunctional habits,

Fig 7. Postsurgical orthodontic correction.

third molar extractions, and previous orthodontic and orthognathic surgery might also increase the mechanical load in the TMJ, facilitating the resorption phenomenon.[1] In fact, several studies have suggested that incidence of condylar resorption is 2%-5% for all orthognathic surgery patients but 20%-30% for those with Class II high angle.[10-15] Notably, Scheerlink et al[16] showed that orthognathic surgery of >10 mm of mandibular advancement can lead to condylar resorption in 67% of cases owing to increased mechanical stress to the TMJ. In addition, studies have also reported the potential linkage of hormonal changes to condylar resorption.[17,18] It is found that hormonal changes within the TMJ may give rise to hyperplasia of the synovial tissue, which breaks down the ligamentous structure and stimulates active resorptive phenomenon around the head of the condyles.[3]

Precise analysis of the TMJ function with the use of condylar path tracing is always a challenge in dentistry. Our previous study showed that the SICAT jaw motion tracking function software can combine and merge the 3-dimensional CBCT and electronic jaw motion tracking data to provide visualization of true mandibular movement.[19] Thus it provides a way to predict the condylar position without further radioactive exposure of patients. Therefore, in this case, we integrated the SICAT jaw motion tracking system to evaluate the mandibular movement for our patient with ICR.

Maxillofacial morphology alteration is a common consequence of ICR. Studies showed that the application of 2D imaging for the treatment planning of 3D structures often leads to distortion of image and errors in magnification and landmark identification.[20,21] However, cast mounting on a semiadjustable articulator is not only difficult, but also time consuming and requiring of experienced skills. Therefore, VSP has been incorporated with CBCT to facilitate the implementation of the orthognathic surgery which overcomes the complexity and inconvenience of cast mounting.[22,23] Thus, medical modeling was adopted in this case to provide a more predictable preoperative orthognathic surgery plan to achieve excellent esthetic and functional outcome.

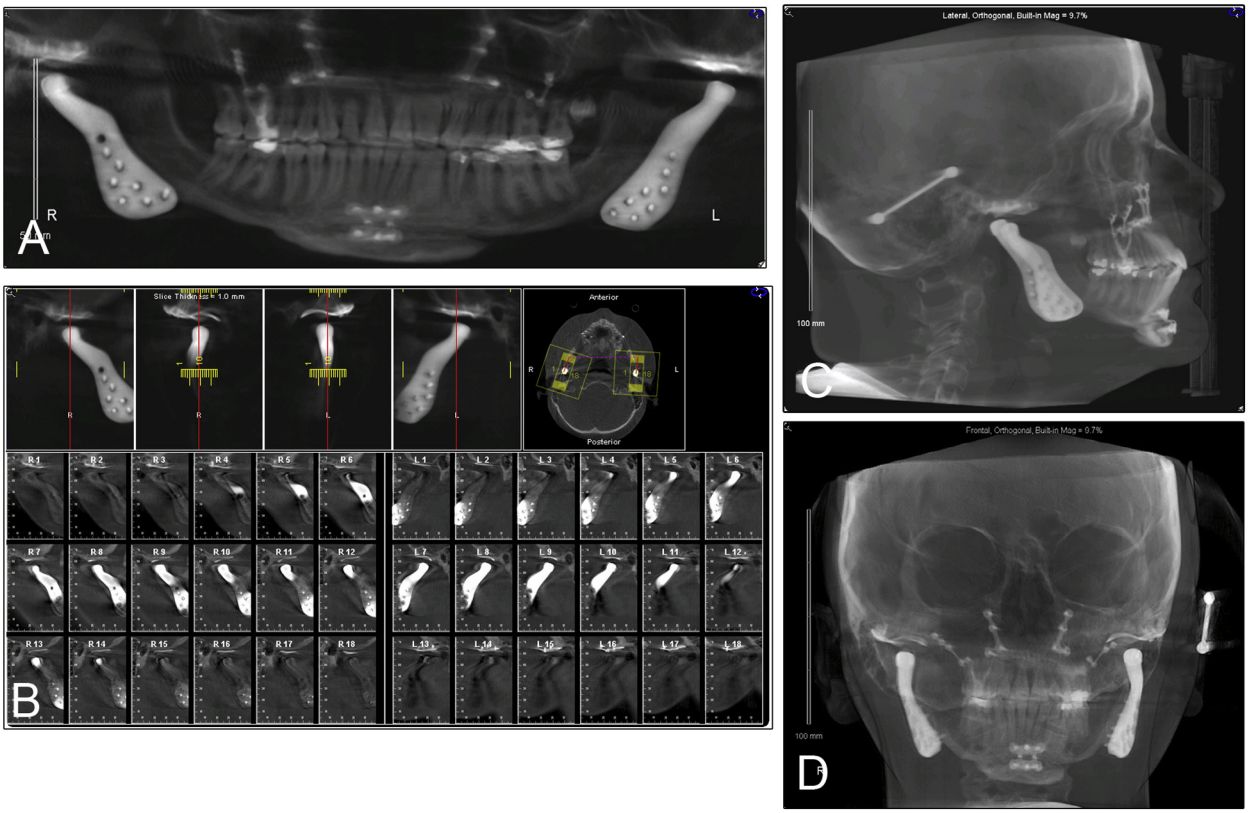

Fig 8. Postsurgical radiographs. **A,** Panoramic radiograph. **B,** Bilateral TMJ radiograph. **C,** Lateral cephalometric radiograph. **D,** Frontal radiograph.

Table II. Cephalometric analysis

Table	*Initial*	*Middle-postsurgical*	*Final*
SNA (°)	77.6	78.2	79.2
SNB (°)	68.7	74.4	76.5
SN-MP (°)	8.9	37.1	31.4
FMA (MP-FH) (°)	33.5	24.4	22.7
ANB (°)	8.9	3.8	2.6
U1-NA (mm)	5.1	5.8	6.8
U1-SN (°)	98.9	98.9	104.3
L1-NB (mm)	11.0	9.4	8.9
L1-MP (°)	99.0	106.8	107.1
Lower lip to E-plane (mm)	1.6	−4.1	−4.2
Upper lip to E-plane (mm)	−0.4	−5.0	−5.3

Determination of treatment options for ICR are indicated on the pathologic conditions of the articular disk and condyle. According to Wolford and Cardenas,[2,3] the treatment protocol for the ICR patients with salvageable articular disks and condyles includes pre- and postsurgical orthodontic treatment to reduce the mechanical load for the TMJ, orthognathic surgery consisting of disk repositioning and ligament repair, and bilateral mandibular ramus osteotomies together with maxillary osteotomies. Furthermore, Mercuri[24] reported that the replacement of TMJ with an autologous costochondral graft or total alloplastic TMJ prosthesis provides postoperative stability. Notably, in this study, we introduced a novel surgical approach to manage ICR and achieved excellent facial profile and dentoalveolar correction through bilateral TMJ replacement with patient-fitted condyle and fossa and Le Fort I osteotomies with maxillary advancement.

In addition, a lingual appliance was implemented in this complicated case as an esthetic option. Importantly, the drawbacks of working with lingual braces, such as limited visibility during treatment, short interbracket distance, and complicated attaching and detaching process were overcome with self-ligating customized brackets.[25-27] In a recent report, we showed a successful comprehensive treatment of condylar hyperplasia and mandibular crowding with the use of the customized self-ligating lingual braces combined with the 2-jaw surgery.[28]

Our treatment objectives were to eliminate the TMJ pathology and provide an excellent functional and esthetic outcome for the patient. To achieve our goals

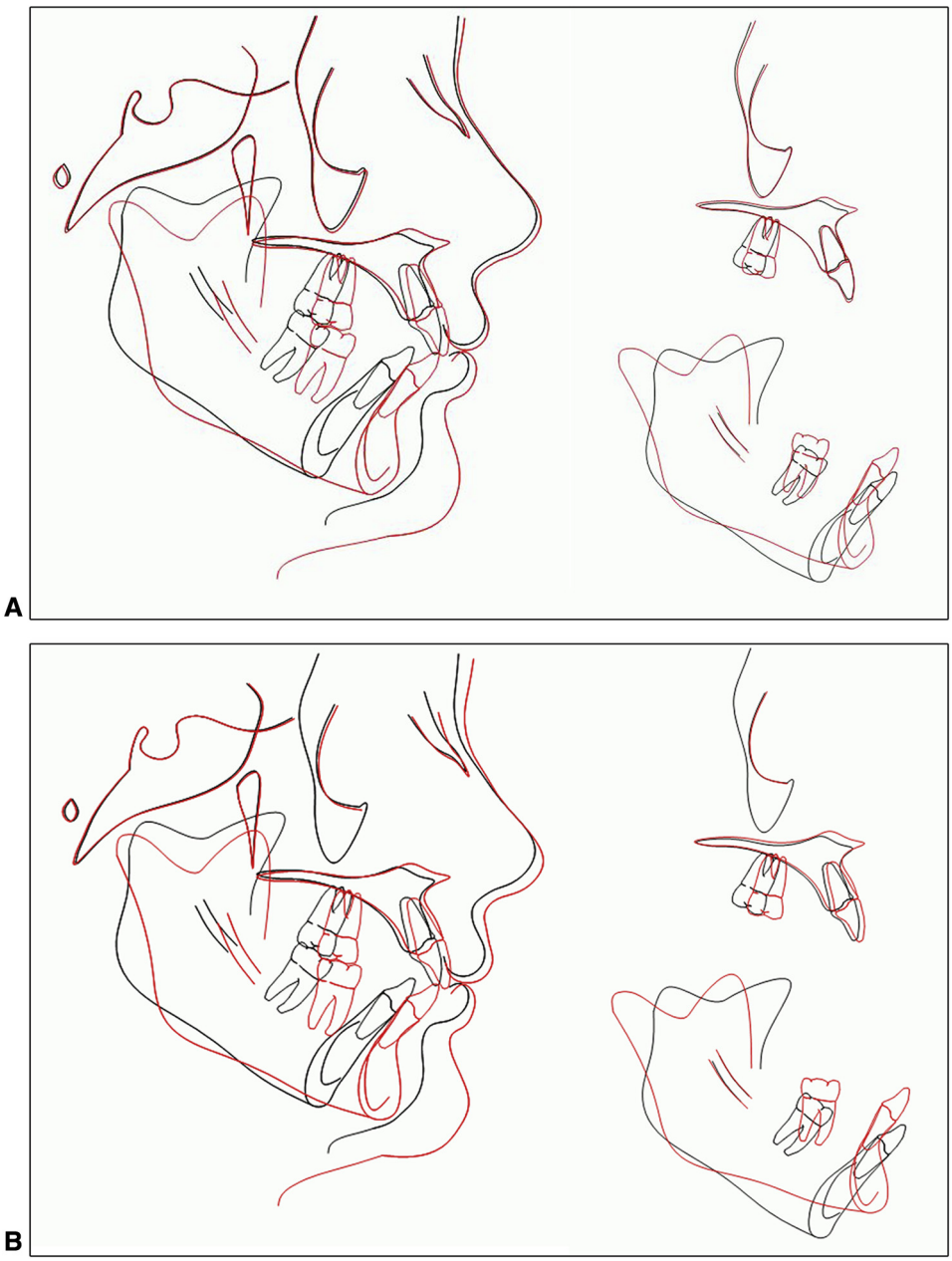

Fig 9. A, Superimposition of initial and middle-postsurgical lateral cephalometric radiographs. **B,** Superimposition of initial and final lateral cephalometric radiographs.

and the patient's demands, it was essential to combine orthodontic treatment with a proper surgical planning and expertise. Therefore, the cooperation between the 2 specialties was extremely important.

CONCLUSION

Studies have suggested that a large number of patients with ICR remain unrecognized and undiagnosed in the orthodontic clinic owing to the poorly understood underlying etiology of the disease and lack of diagnostic tools. ICR often causes occlusal and skeletal changes, TMJ dysfunction, pain, and maxillofacial deformities. Therefore, precise diagnosis of the disease and proper treatment plan are essential. In this case report, we showed a comprehensive treatment approach to achieve the patient's esthetic and functional demands with the

Fig 10. Final extraoral and intraoral photographs.

use of customized self-ligating lingual braces and total joint prostheses by means of VSP and orthognathic surgery.

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