A comprehensive treatment approach for idiopathic condylar resorption and anterior open bite with 3D virtual surgical planning and self-ligated customized lingual appliance

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Introduction: We report the successful treatment of a 38-year-old woman with bilateral idiopathic condylar resorption and anterior open bite. She had incompetent lips, a gummy smile, increased lower facial height, high mandibular plane angle, skeletal and dental Class II malocclusion with mild mandibular crowding, increased overjet, and mandibular midline deviation to the right. **Methods:** The treatment plan included: (1) presurgical alignment and leveling of the teeth in both arches; (2) jaw motion tracking (JMT) to detect mandibular movement; (3) 3-piece maxillary osteotomies with mandibular reconstruction and bilateral coronoidectomies; and (4) postsurgical correction of the malocclusion. The orthodontic treatment was performed with the use of custom lingual braces and clear brackets and the orthognathic surgery was planned with the use of virtual surgical planning. **Results:** The idiopathic condylar resorption and anterior open bite were treated, crowding was eliminated in the lower anterior segment, correction of skeletal and dental Class II malocclusion was obtained, mandibular plane angle was reduced, and facial profile improved. **Conclusions:** The results suggest that esthetic and functional results can be achieved with the cooperation of 2 specialties and with the use of state-of-the-art technology. (Am J Orthod Dentofacial Orthop 2019;155:560-71)

Idiopathic condylar resorption (ICR) is a progressive degenerative disease of the temporomandibular joint (TMJ), most commonly seen in female patients (female-male ratio ~9:1)[1] at 10-40 years of age, predominantly in teenagers during the pubertal growth phase.[2] ICR can cause skeletal and occlusal instability (mostly Class II malocclusion), dentofacial deformities,[3] TMJ dysfunction, and pain. Bilateral involvement of condyles creates symmetric posterior shifting of the mandible, high mandibular plane angle, and an anterior open bite. In unilateral cases, the mandibular midline and chin shift to the affected side, developing cross-bite and posterior occlusal prematurities. Although the disease is not well understood, there are several local and systemic factors that can cause and initiate ICR.[4,5] Studies showed that 25% patients with ICR have no TMJ symptoms.[6] Because this phenomenon is aggressive and a fast-moving form of degenerative joint disease, precise diagnosis and proper treatment is necessary. The diagnosis of ICR is often based on the patient's history, clinical evaluation, and imaging. Effective treatment planning includes presurgical orthodontics for arch alignment and leveling, orthognathic surgery with condylar prosthesis, and postsurgical orthodontics to correct the minor detail of occlusion. In the present case report, the comprehensive treatment of an adult patient with bilateral ICR and anterior open bite is presented and discussed.

DIAGNOSIS AND ETIOLOGY

In January 2015, a 38-year-old woman was referred by the oral surgeon to the Department of Orthodontics at the University of Alabama, Birmingham, with the

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Fig 1. Initial extraoral and intraoral photographs.

complaint of "difficulty in chewing and incising food." She was diagnosed with Crohn disease in 2010 and her third molars were extracted in 2012. Clinical examination revealed a convex high-angle facial profile, incompetent lips, gummy smile, increased lower facial height, and strained mentalis muscle in a skeletal Class II base. Her malocclusion was complicated by a 5-mm open bite (incisor), 10-mm overjet, and mild crowding (2 mm) in the mandibular arch. Maxillary dental midline coincided with the facial midline, but the mandibular midline was deviated 2 mm to right. She had no TMJ popping, clicking, or crepitation; her maximum incisal opening was 45 mm with right and left lateral excursions of 6 mm and 10 mm, respectively. Extraoral and intraoral photographs and upper and lower impressions were obtained at her second appointment, along with the lateral cephalometric and panoramic radiographs and a cone-beam computed tomorgraphy (CBCT) scan (Figs 1-3).

Dental casts showed an open bite from molars to molars with 5 mm open bite at the incisors. The first molars were in Angle Class I on the left side, and on the right side, they were in an end-on Class II relationship. The panoramic radiograph showed that all teeth were present except the molars. Right and left condyles were completely resorbed. Lateral cephalometric analysis revealed bilateral ICR, skeletal and occlusal Class II deformity with ANB 6.8°, anterior open bite, hyperdivergent mandible with increased mandibular plane angle (SN-MP 52°), decreased vertical height of ramus, and upper lip retruded 4.1 mm and lower lip protruded 4.0 mm from the Ricketts E-line. CBCT imaging showed the resorption of the condyles to the level of the sigmoid notch. SICAT (Bonn, Germany) jaw motion tracking (JMT) was performed, and the data provided visualization of true mandibular movements and predicted the condylar position without further radiation exposure (Fig 4).

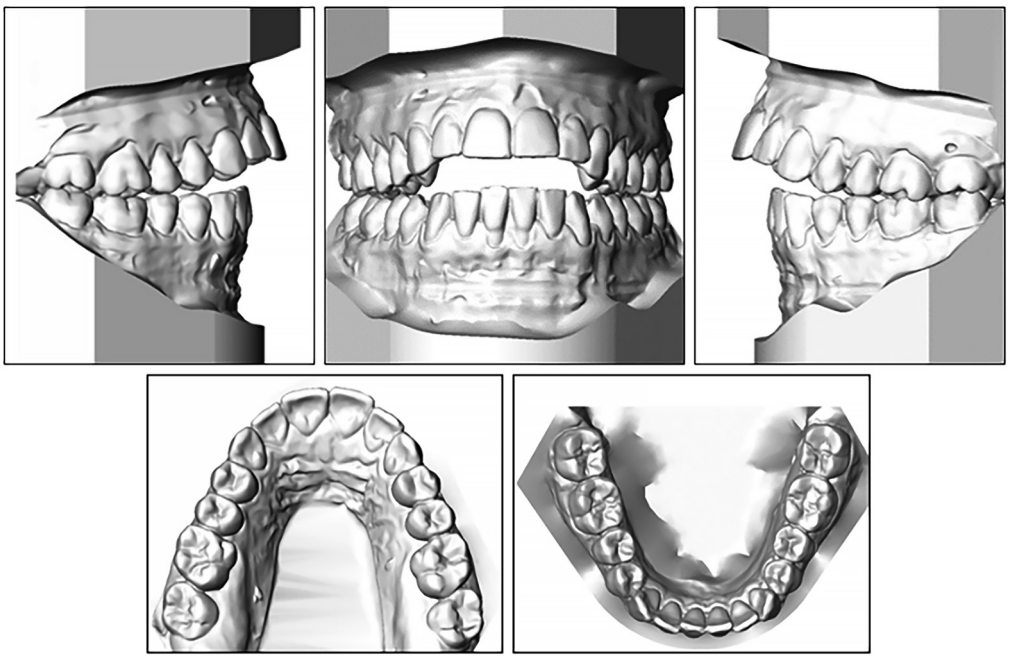

Fig 2. Initial orthodontic casts.

TREATMENT PLAN

The patient was diagnosed with bilateral ICR, Angle Class II malocclusion, anterior open bite, increased overjet, mild mandibular crowding, lower midline deviation, and gummy smile. The treatment objectives for this patient were: (1) dental: eliminate crowding in the mandibular anterior segment and correct the anterior open bite; (2) skeletal: decrease the high angle, reduce ANB by means of maxillary impaction, eradicate the TMJ resorptive phenomenon, and reconstruct the condyles bilaterally; and (3) soft tissue: correct the anterior and posterior gummy smile and improve the facial profile with lip competency.

After careful consideration of all treatment alternatives with the patient, a nonextraction approach for the maxillary and mandibular arches was selected, followed by Le Fort I 3-piece maxillary osteotomies to expand and impact the posterior maxilla and bilateral mandibular joint replacement with TMJ concept via facelift approach with bilateral coronoidectomies. Postsurgical orthodontic treatment would be used to correct minor discrepancies. Furthermore, the patient was given the option of esthetic brackets in this very complex case. Self-ligating fully customized lingual appliances (Harmony System, Paris, France) were used for the presurgical correction of dental alignment and leveling. SICAT JMT would be used to analyze mandibular movements. Preoperative virtual surgical planning was performed, and postsurgical interarch settling was accomplished with the use of clear brackets.

TREATMENT PROGRESS

Maxillary and mandibular high-accuracy polyvinyl siloxane (PVS) impressions were obtained and sent to Harmony. Customized self-ligating lingual appliances were fabricated with due consideration for how the final occlusion would be created. Initial leveling was started with the use of 0.014-inch NiTi (Fig 5). Wires were sequenced in 0.016 × 0.022. Dental open bite was maintained to facilitate the surgical procedure with 0.018 × 0.025 wire. Once the leveling and alignment were completed, CBCT and dental casts were taken for medical modeling by means of virtual surgical planning (VSP Orthognathics; 3D Systems, www.3Dsystems.com). Five months after the initial bonding, orthognathic surgery planning was discussed with the oral surgeon to determine the kind of maxillary movements during surgery and to fabricate the mandibular joint prosthesis (Fig 6; Table I).

During the presurgical phase of orthodontic treatment, the maxillary teeth were leveled segmentally (2-2, 3-6). To facilitate the osteotomies, the final

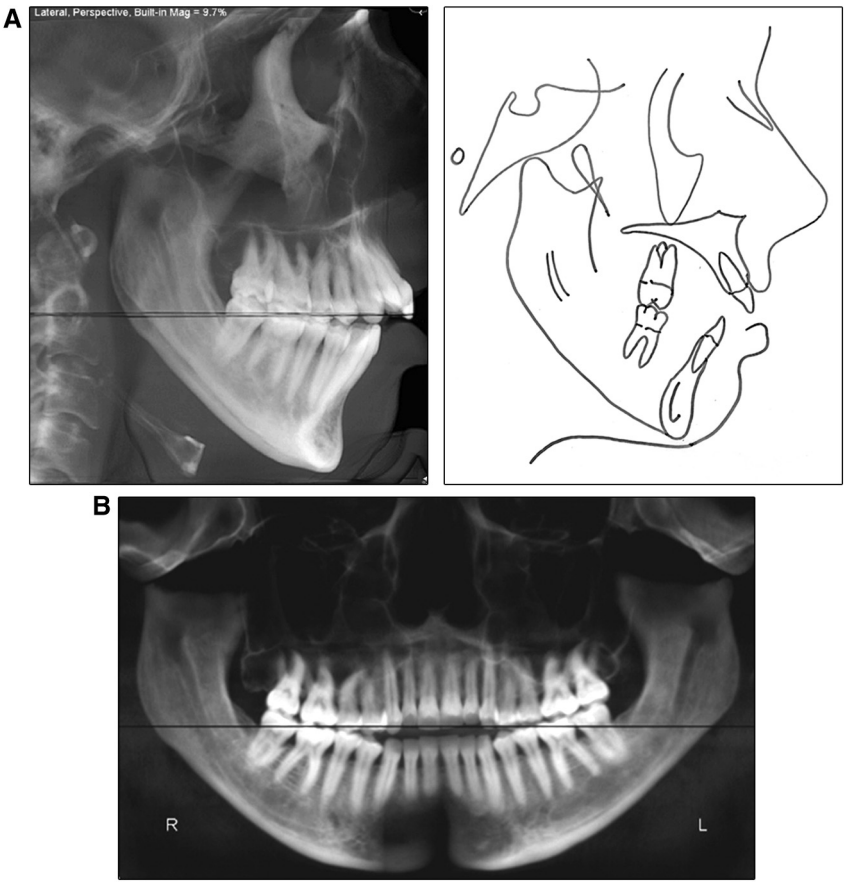

Fig 3. **A,** Initial lateral cephalometric radiograph with tracing. **B,** Panoramic radiograph.

maxillary archwire was cut between the lateral incisors and canines (Fig 7) to prevent relapse of the anterior open bite after surgery. A continuous archwire at the beginning of the postsurgical period would allow better control for final occlusal adjustments. Clear orthodontic brackets were placed on the facial axis point at the labial and buccal surfaces of the maxillary and mandibular arch to aid attachment for the occlusal splint and interarch elastic band setting.

The VSP workflow enabled us to visualize the expected final outcome with the intermediate and final position of the maxilla and mandible. VSP was carried out to correct the gummy smile. With the use of DICOM data, a stereolithic model was produced with the maxilla and mandible in the final position. The stereolithic model was sent to TMJ Concepts (Ventura, Calif) for the design, blueprint, and wax-up of the prostheses, and the surgeon approved the design. Then the custom-fitted total joint prostheses were manufactured. It took ~8 weeks to manufacture the total joint patient-fitted prostheses. The mesh framework on the underside of the custom-fitted titanium shell secures the polyethylene articulating portion of the fossa component, and the mesh on the superior surface of the fossa component allows osseous-integration with the fossa bone.

Thirteen months after the initiation of the orthodontic treatment, orthognathic surgery was carried out (Fig 8). Subperiosteal dissection was achieved to remove the residual condylar stump and sent for pathologic examination. Coronoid process was resected below the level of the sigmoid notch. The intermediate splint was placed between the dentition, and intermaxillary fixation was done with the use of 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 in the same manner on the left side and condylar

Fig 4. A, Merge of CBCT and JMT data and mandibular segmentation. **B,** Visualization of mandibular opening movement: condylar pathway. **C,** Incisor pathway.

replacement with TMJ concept custom fossa was inserted. Le Fort I osteotomy was performed from the posterior maxilla through the piriform rim. Interdental osteotomies were implemented between canines and lateral incisors. The pterygoid planes were separated and the maxilla was down-fractured. The final splint was placed and maxillary-mandibular fixation was done. Four L-shaped plates were incorporated in the maxilla, one on either side on the nasomaxillary buttress and one on either side along the pterygomaxillary buttress. Final occlusion was verified and found to be satisfactory. Light elastics were placed and the patient was extubated without any complication. The patient was transferred to the postoperative care unit and subsequently discharged after 2 days of postoperative observation and care. She was given Norco elixir, Zofran, Robaxin, and Guaifenesin as required for a standard orthognathic surgery.

Five weeks after the successful surgery, the splint was removed. Upper and lower arch alignment along with the minor occlusal adjustments were started with the use of 0.014 NiTi (Fig 9). Post setting was performed with the use of elastic bands. Three months after the surgery, lingual brackets were removed and occlusal settlements were done with the use of labial orthodontics.

Fig 5. Initial appliance placement.

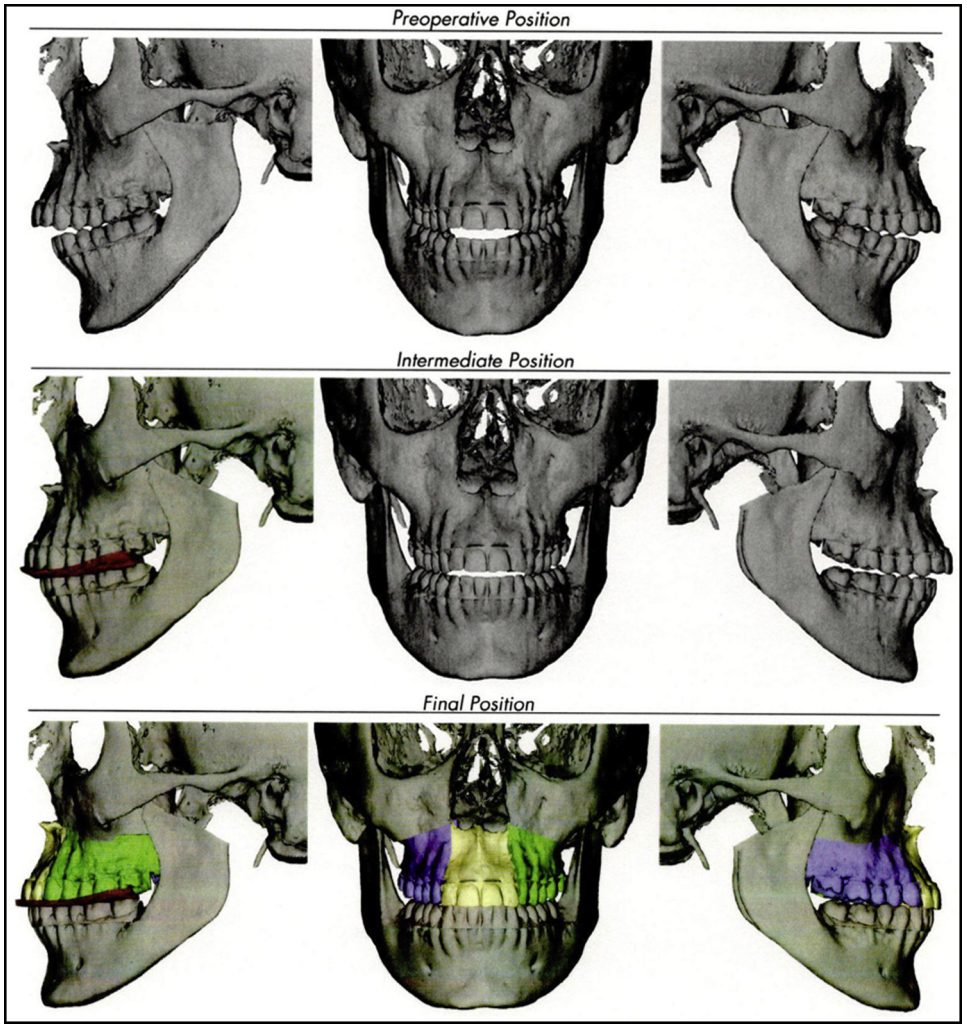

Fig 6. VSP workflow for Le Fort I maxillary osteotomies.

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

Point	*Name*	*Anterior/posterior*	*Left/right*	*Up/down*
ANS	Anterior nasal spine	1.11 posterior	0.15 right	8.82 up
A	A-point	0.78 posterior	0.10 right	1.73 up
ISU1	Midline of upper incisor	0.00	0.00	2.00 up
U3L	Upper left canine	0.07 posterior	0.90 left	3.95 up
U6L	Upper left anterior molar (mesiobuccal cusp)	0.25 anterior	1.77 left	2.76 up
U3R	Upper right canine	0.43 anterior	1.27 right	4.0 up
U6R	Upper right anterior molar (mesiobuccal cusp)	0.59 anterior	1.71 right	3.48 up
ISL1	Midline of lower incisor	4.98 anterior	0.45 left	6.97 up
L6L	Lower left anterior molar (mesiobuccal cusp)	4.07 anterior	0.12 left	2.52 up
L6R	Lower right anterior molar (mesiobuccal cusp)	4.73 anterior	0.10 left	2.34 up

Fig 7. Before surgery: upper and lower teeth with clear brackets.

TREATMENT RESULTS

The posttreatment records show a balanced facial profile and occlusion. ICR was treated and bilateral condylar reconstruction was accomplished exactly as estimated 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 reduced, and esthetically pleasant smile arc was achieved with lip competency and adequate gingival display. Skeletally, ANB angle was reduced 3.9° and SN-MP was reduced 14.8°. In addition, L1 to MP was increased 10.6°. Furthermore, soft tissue convexity was reduced 18.6 (Fig 10; Table II).

Twenty-two months after the initiation of orthodontic treatment, debonding was complete. Intraoral and extraoral photographs, panoramic and lateral cephalogram, and dental impressions were taken for final records (Figs 11 and 12). The final outcome of our comprehensive treatment approach satisfied both functional and esthetic demands. The gummy smile and lip incompetency were corrected.

DISCUSSION

Although the specific etiology of condylar degeneration has not been clearly understood, in a recent

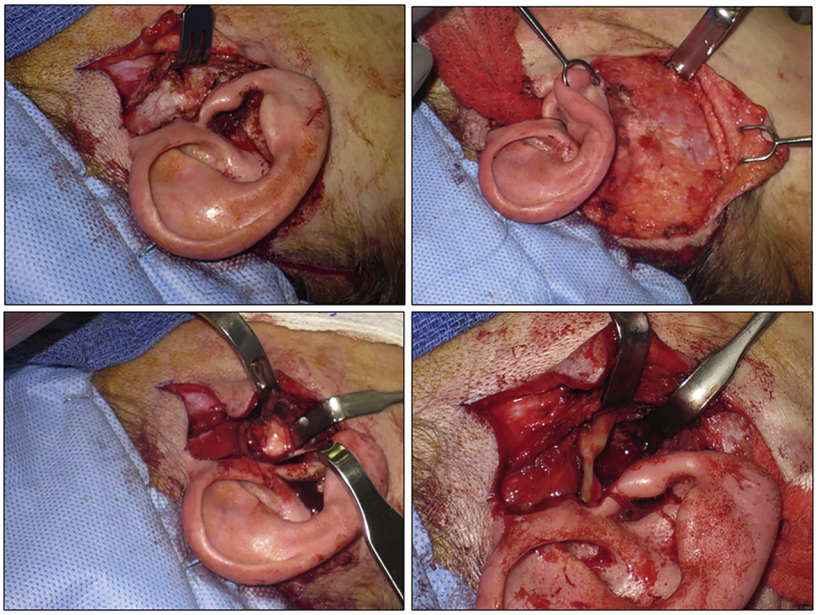

Fig 8. Orthognathic surgery with mandibular reconstruction.

Fig 9. Postsurgical orthodontic correction.

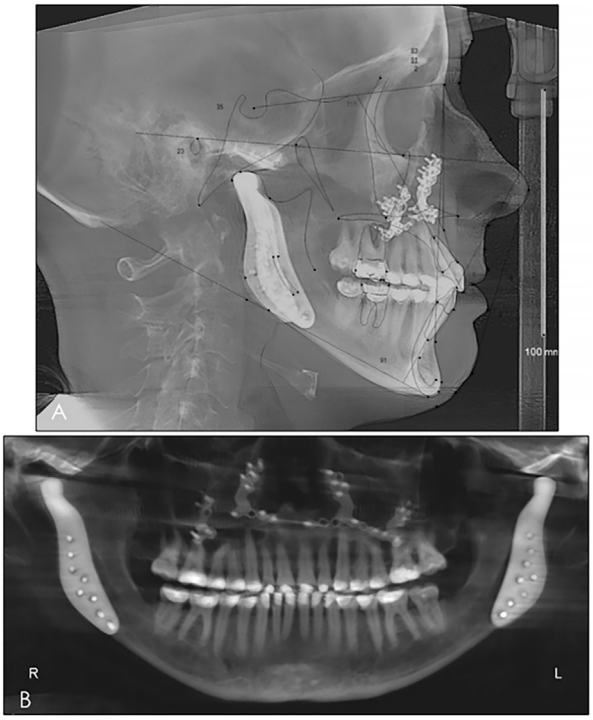

Fig 10. A, Postsurgical lateral cephalometric radiograph and tracing; **B,** panoramic radiograph.

Table II. Cephalometric measurements

Parameter	*Normal*	*Initial*	*Final*
SNA (°)	82	82.6	82.5
SNB (°)	80.9	75.8	79.6
ANB (°)	1.6	6.8	2.9
MP-SN (°)	33	52	38.8
Occ plane to SN (°)	14.4	15.1	12.8
U1-SN (°)	102.8	108.7	106.2
U1-NA (mm)	4.3	10.5	7.8
U1-NA (°)	22.8	26.1	23.7
L1-NB (mm)	4	7.7	7
IMPA (°)	95	73.8	84.4
Soft tissue convexity (°)	135	149.5	130.9

study, Sarver et al[4] reported that condylar changes might occur because of several local and systemic factors and as a sequel of postoperative factors. Local possible factors are osteoarthritis, reactive arthritis, traumatic injuries, and infection. Systemic disease includes rheumatoid arthritis, systemic lupus erythromatosus, connective tissue disease, Sjögren syndrome, and psoriatic arthritis.[4] In addition, parafunctional habits, third molar extractions, and previous orthodontic and orthognathic surgery might increase the mechanical load in the TMJ and facilitate the resorption phenomenon.[1] Scheerlink et al showed that orthognathic surgery of >10 mm mandibular advancement can lead to condylar resorption in 67% of cases owing to increased mechanical stress in the TMJ.[7] Previous studies reported that hormonal changes within the TMJ produce hyperplasia of the synovial tissue, which breaks down the ligamentous structure[6] and stimulates the active resorptive phenomenon around the head of the condyles until the mid 20s. Furthermore, Mônica et al showed the association of Crohn disease with TMJ dysfunction and pain.[8] In the present case report, the patient had a medical history of Crohn disease that might have led to inflammatory changes in the TMJ. On the other hand, removal of her third molars might also might have played an important role in exacerbating her ICR.

Precise analysis of the TMJ function with the use of condylar path tracing is a challenge in dentistry. Our previous study showed that the SICAT JMT function software can combine and merge the 3-dimensional CBCT and electronic JMT data to provide visualization of true mandibular movement.[9] It provides a way to predict the condylar position without further radioactive exposure. Therefore, we integrated the SICAT JMT system to evaluate the mandibular movement for this patient with ICR.

ICR often causes alteration of the maxillofacial morphology. Studies showed that 2D imaging for the treatment planning of 3D structures often leads to distortion of image and errors in magnification and landmark identification. However, cast mounting on a semiadjustable articulator is not only difficult, but also time consuming and it needs experienced skills. Therefore, a VSP system was incorporated with the CBCT to facilitate the implementation of the orthognathic surgery and overcome the above-mentioned errors and inconveniencies.[10,11] Thus, medical modeling was adopted in this case report to plan preoperative orthognathic surgery to achieve excellent esthetic and functional outcomes.

Several studies have suggested that the incidence of condylar resorption is 2%-5% for all orthognathic surgery patients but 20%-30% for patients with Class II high angle.[12-17] According to Wolford and Cardenas, the treatment planning for ICR patients includes pre- and postsurgical orthodontic treatment to reduce the mechanical load on the TMJ and orthognathic surgery consisting of disk repositioning and ligament repair with the use of Mitek anchor and bilateral mandibular ramus osteotomies together with the multiple maxillary osteotomies.[2,6] Furthermore, in 2006, Mercuri et al

Fig 11. Final extraoral and intraoral photographs.

reported that the replacement of the joint with an autologous costochondral graft or total alloplastic TMJ prosthesis provides postoperative stability.[18] In the present case report, however, we have introduced a novel surgical approach to manage ICR and achieve excellent facial profile and dentoalveolar correction through bilateral TMJ replacement with TMJ concept condyle and fossa via facelift approach and Le Fort I osteotomies with maxillary expansion and impaction.

Furthermore, in this study, a lingual appliance was offered to a patient with a complex case. The problems of working with lingual braces, such as limited visibility during working and short interbracket distance, complicating the attaching and detaching process,[19,20] were overcome with self-ligating customized brackets.[21] In a previous study, we also demonstrated a successful comprehensive treatment of condylar hyperplasia and mandibular crowding with the use of custom lingual braces and 2-jaw surgery.[22]

Our treatment objective was to eliminate the TMJ pathology and provide an excellent functional and esthetic outcome for the patient. To fulfil our goal and the patient's demand, it was essential to combine orthodontic treatment with proper surgical planning and expertise. Therefore, the cooperation between the 2 specialties was extremely important.

CONCLUSION

Studies suggest that patients with ICR remain undiagnosed and unrecognized in the orthodontic clinic owing to the poorly understood etiology of the disease and lack of diagnostic tools. ICR often causes occlusal and skeletal changes, TMJ dysfunction and pain, and maxillofacial deformities. Therefore, precise diagnosis

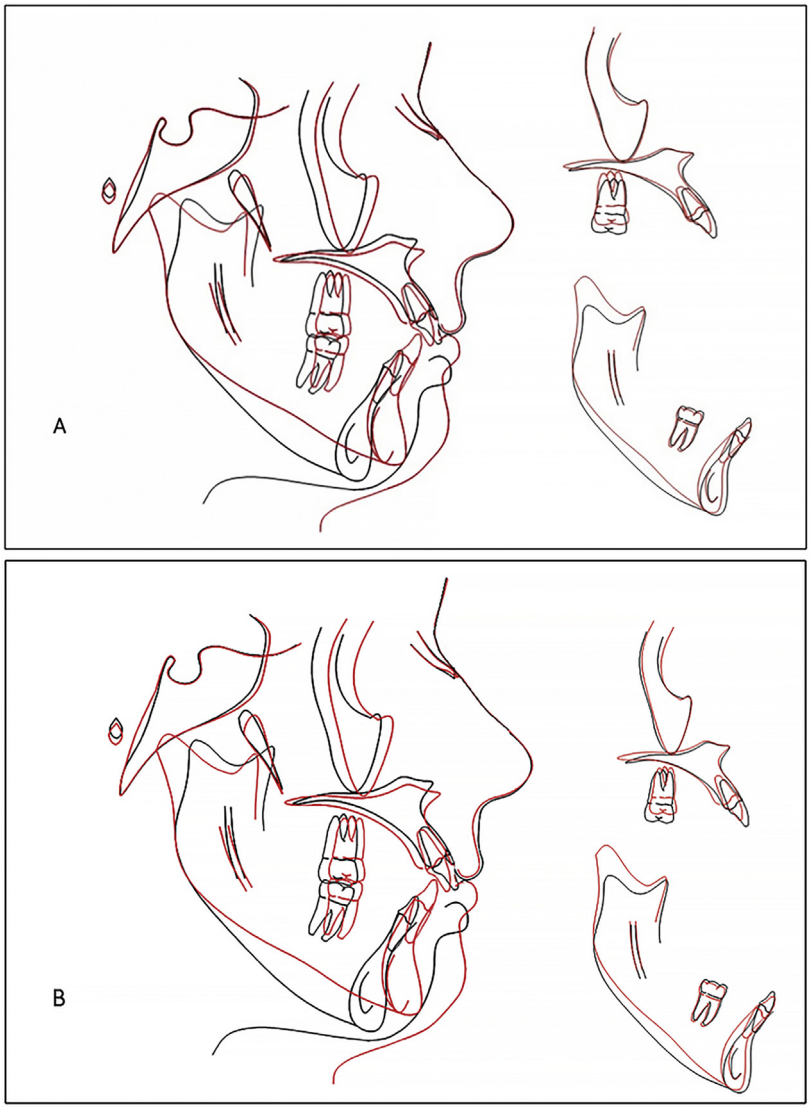

Fig 12. A, Superimposition of initial (*black*) and middle-postsurgical (*red*) lateral cephalometric radiographs. **B,** Superimposition of initial (*black*) and final (*red*) lateral cephalometric radiographs.

of the disease and proper treatment plan is essential. In this case report, we demonstrated a comprehensive treatment approach to achieve the patient's esthetic and functional demand with the use of customized self-ligated lingual braces and clear braces along with VSP and orthognathic surgery.

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