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Case Report

Bilateral alloplastic temporomandibular joint reconstruction with orthognathic surgery: a case report of idiopathic condylar resorption

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ABSTRACT

Idiopathic condylar resorption (ICR) also known as cheerleader's syndrome, progressive condylar resorption, idiopathic condylysis, and condylar atrophy can be defined as chronic ongoing changes of condylar shape and decrease in mass. It is characterized by an excessive physical stress to the articular structures of the temporomandibular joint (TMJ) that exceeds the normal adaptive capacity. Consequently, leading to decrease condylar head volume, decrease ramus height, progressive mandibular retrusion in adults, or decreased growth rate in adolescents. A case report of a 30-year-old female based on the history, clinical, and radiographic examination has class II skeletal relationship secondary to bilateral ICR, receded chin, vertical maxillary excess (VME) and canting. Surgical treatment plan was established using 3D Systems, Inc., ("3DS") VSP® and case was managed by bilateral total alloplastic temporomandibular joint reconstruction (BATMJR) with orthognathic surgery to do LeFort I, bilateral condylectomy, genioplasty, and inferior border osteotomy. The case report includes one-year follow-up which shows stable outcome, emphasizing long-term success of comprehensive treatment strategy in addressing skeletal class II malocclusion associated with ICR.

Keywords: Idiopathic condylar resorption, Temporomandibular joint replacement, Temporomandibular joint reconstruction, Orthognathic surgery, Case report

INTRODUCTION

Idiopathic condylar resorption (ICR) also known as cheerleader's syndrome, progressive condylar resorption, idiopathic condylysis, and condylar atrophy can be defined as chronic ongoing changes of condylar shape and decrease in mass. It is characterized by an excessive physical stress to the articular structures of the temporomandibular joint (TMJ) that exceeds the normal adaptive capacity. Consequently, leading to decrease condylar head volume, decrease ramus height, progressive mandibular retrusion in adults, or decreased growth rate in adolescents.[1,2] According to the term "Idiopathic" it is of an unknown etiology and has no local or systemic diseases that could cause dysfunctional TMJ remodeling leading to condylar resorption. Local factors include osteoarthritis, reactive arthritis, avascular necrosis, infection, or traumatic injuries. Furthermore, systemic connective tissue or autoimmune diseases include rheumatoid arthritis, scleroderma, systemic lupus erythematosus, Sjogren's syndrome, or ankylosing spondylitis.[3] Diagnosis of ICR can be established based on patient history, clinical and radiographic evaluation, and appropriate exclusion of known local and systemic factors.[4]

Clinical features of patients with ICR usually presents in the worsening of their occlusion and aesthetics, and whether they also have TMJ symptoms or related pain. Condylar involvement is often bilateral and symmetrical, albeit unilateral examples rarely occur. In bilateral condylar involvement, clinical findings are symmetrical posterior shift of the jaw (deteriorating class II relationship), anterior open bite development, condylar flattening, condylar height reduction, and loss of posterior

facial height. In contrast, unilateral condylar involvement usually results in a shift of the dental midline and tip of the chin towards the affected side, an ipsilateral Class II malocclusion, a cross bite, as well as early occlusal contacts that result in a lateral open bite on the contralateral side.[5]

Certain image modalities such as panoramic, lateral cephalometric, and cone beam computed tomography (CBCT) scans or CT scans can be used in the diagnosis of ICR. Furthermore, bone scans can be used to assess the active nature of ICR to distinguish between progression and arrest phase. Moreover, magnetic resonance imaging (MRI) can be used to identify altered disc position in ICR. The most common radiographic findings of ICR were loss of condylar volume, anterior open bite, high mandibular plane angle (MPA), decreased ramus height, increased overjet, deep bite, class II skeletal relationship, and decreased sella–nasion–B-point (SNB) angle. Other less common radiographic findings included increased radionuclide uptake in the condyle, disc derangement, pseudocystic lesions in the condyle, excessive joint space due to hyperplasia of the synovial tissue, and decreased oropharyngeal space.[4]

Management of ICR depends on condylar activity and the surgeon's ability to preserve the disk and mandibular condyle. Consequently, surgical treatment can either be orthognathic surgery alone or orthognathic surgery with TMJ surgery. In TMJ surgery, it can either be by disc preservation and stabilization, or by TMJ replacement whether it is autogenous or alloplastic.[6]

In this article, we will discuss a case report of bilateral alloplastic temporomandibular joint reconstruction (BATMJR) with orthognathic surgery to do LeFort I, bilateral condylectomy, genioplasty, and inferior border osteotomy.

CASE REPORT

A 30-year-old female with no medical conditions or allergies had a chief complaint of being unhappy with her smile. She began orthodontic treatment in 2013 and was scheduled for orthognathic surgery, but experienced gradual bilateral condylar resorption. The surgery had to be postponed multiple times due to active changes in the condyles. The patient was then referred to the oral and maxillofacial surgery department at King Saud University Medical City for management of the condylar resorption.

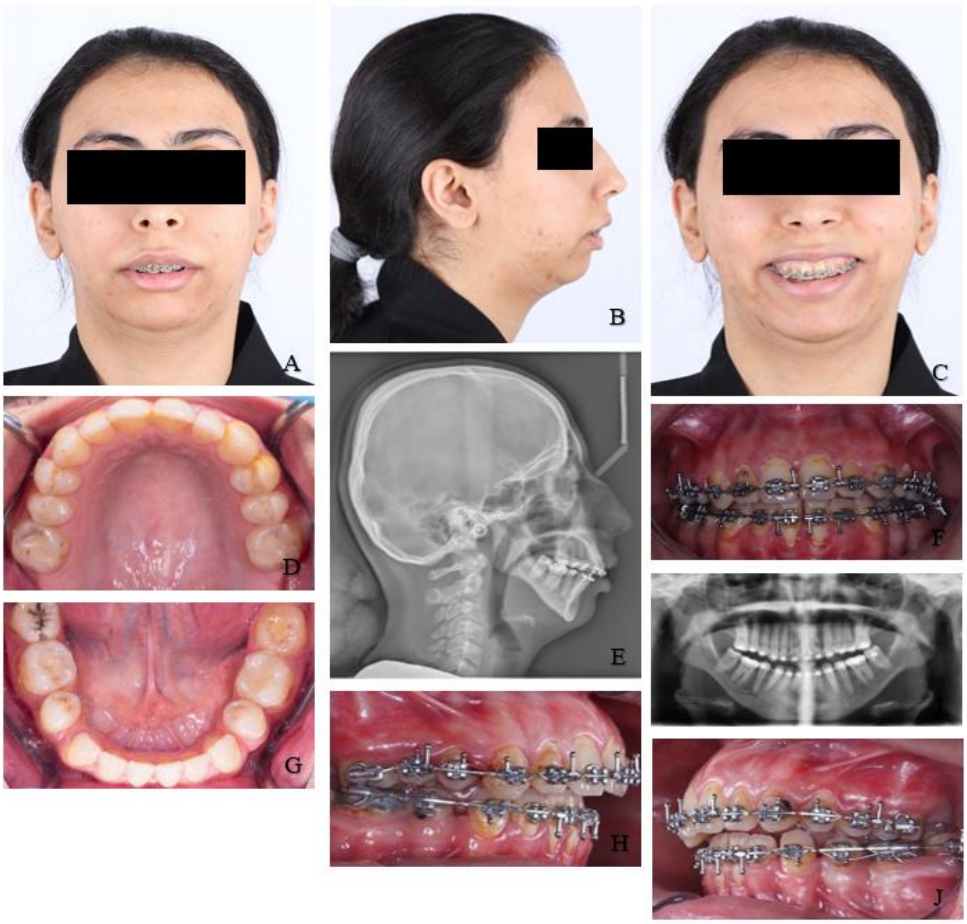

Figure 1: Shows pre-operative images (A) frontal view at rest (B) lateral right view at rest (C) frontal view while smiling (D) occlusal view of maxilla (E) lateral cephalometric radiograph (F) frontal view of occlusion (G) occlusal view of mandible (H) lateral right view of occlusion (I) panoramic X-ray (J) lateral left view of occlusion.

Table 1: Summary of pre-operative cephalometric analysis.

Measurement	Norm	Value
Sagittal (apical bases and chin)		
SNA	82°±2°	80.3
SNB	80°±2°	70.5
ANB	2°±1.7°	9.8
Vertical divergency		
MMPA	27°±5°	39.3
Dental (incisor position)		
UI-NA	22.8°±5.7°	20
LI-NB	25.3°±6°	37.9
Interincisal angle	130°±6°	128.0
Soft tissue		
Upper lip to E line	-4 mm±2	0.4
Lower lip to E line	-2 mm±2	3.5

Abbreviations: SNA, Sella-Nasion-Point-A angle; SNB, Sella-Nasion-Point-B angle; ANB, Point-A-Nasion-Point-B angle; MMPA, Maxillary-Mandibular Plane Angle; UI-NA, Upper-Incisor to Nasion-Point-A angle; LI-NB, Lower-Incisor to Nasion-Point-B angle; E, Esthetic.

Table 2: Summary of VSP movement.

Point	Name	Anterior/posterior	Left/right	Up/down
ANS	Anterior nasal spine	0.23 mm anterior	1.53 mm left	6.77 mm up
A	A point	1.36 mm anterior	1.25 mm left	6.48 mm up
ISU1	Midline of upper incisor	7.00 mm anterior	0.00 mm	7.00 mm up
U3L	Upper left canine	6.20 mm anterior	0.08 mm left	4.92 mm up
U6L	Upper left anterior molar (mesiobuccal cusp)	4.98 mm anterior	0.16 mm left	0.61 mm up
U3R	Upper right canine	6.99 mm anterior	0.03 mm right	6.28 mm up
U6R	Upper right anterior molar (mesiobuccal cusp)	5.52 mm anterior	0.14 mm left	2.79 mm up
ISL1	Midline of lower incisor	11.06 mm anterior	0.57 mm left	4.43 mm up
L6L	Lower left anterior molar (mesiobuccal cusp)	9.61 mm anterior	0.50 mm left	1.09 mm down
L6R	Lower right anterior molar (mesiobuccal cusp)	10.37 mm anterior	0.47 mm left	0.90 mm up
B	B point	14.64 mm anterior	0.39 mm right	2.58 mm up
Pog.	Pogonion	21.67mm anterior	1.14 mm right	1.88 mm up

Abbreviations: VSP, virtual surgical planning.

During the clinical examination, the patient displayed moderate bilateral tenderness in the preauricular region, bilateral TMJ clicking, and myofascial pain. Other findings included a limited mouth opening of 21 mm, a mesocephalic and symmetrical face, incompetent lips with a 9 mm inter-labial gap, a 7 mm tooth display at rest, a 4 mm gingival display when smiling, a 2 mm downward occlusal right maxillary cant, a convex profile, a slightly obtuse Nasio-Labial angle, a deep Labio-Mental fold, a receded chin, and an obtuse throat-neck angle. These observations can be seen in Figures 1 (A-C). Additionally, the maxillary dental midline coincided with the philtrum of the upper lip, while the mandibular dental midline was shifted 1 mm to the left of the maxillary dental midline. The patient exhibited a 1 mm overbite, bilateral canine and molar class II malocclusion, and a 7 mm overjet. These findings can be observed in Figures 1 (D, F-H, J). A panoramic X-ray revealed decreased condyle height bilaterally, flattened condyles, and loss of condyle volume (Figure I). Cephalometric radiograph analysis based on the lateral cephalometric radiograph showed a Class II skeletal relationship due to a retrognathic mandible, hyperdivergent mandible, protruded lower incisors, and protruded upper and lower lips (Table 1 and Figure 1E).

Rheumatoid factor was negative and all other laboratory tests within normal ranges. Therefore, ICR has been diagnosed. Based on the history, clinical, and radiographic examination, the patient has class II skeletal relationship secondary to bilateral ICR and receded chin. Furthermore, patient has vertical maxillary excess (VME) and canting.

The surgical treatment plan was established using 3D Systems, Inc., ("3DS") VSP® Orthognathics to do LeFort

I, Bilateral Condylectomy, Genioplasty, inferior border osteotomy, and reconstruction with bilateral total alloplastic temporomandibular joint prosthesis. Furthermore, Table 2 shows the list of bony and occlusal anatomical landmarks and their summarized movements from preoperative position to simulated postoperative position.

Moreover, 3D construction of pre-operative, intra-operative, and post-operative can be seen in Figure 2. 3D construction of intermediate and final splint can be seen in Figure 3. Furthermore, 3D construction of transparent soft tissue analysis can be seen in Figure 4.

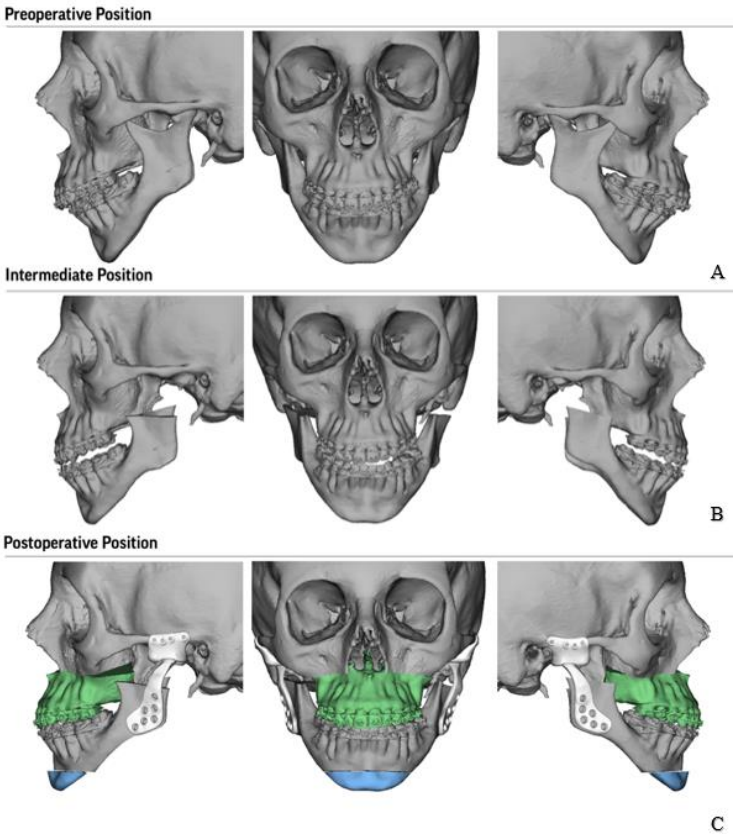

Figure 2: Shows 3D construction using 3D systems VSP orthognathics (A) pre-operative position of 3D construction (B) intra-operative position of 3D construction (C) post-operative position of 3D construction.

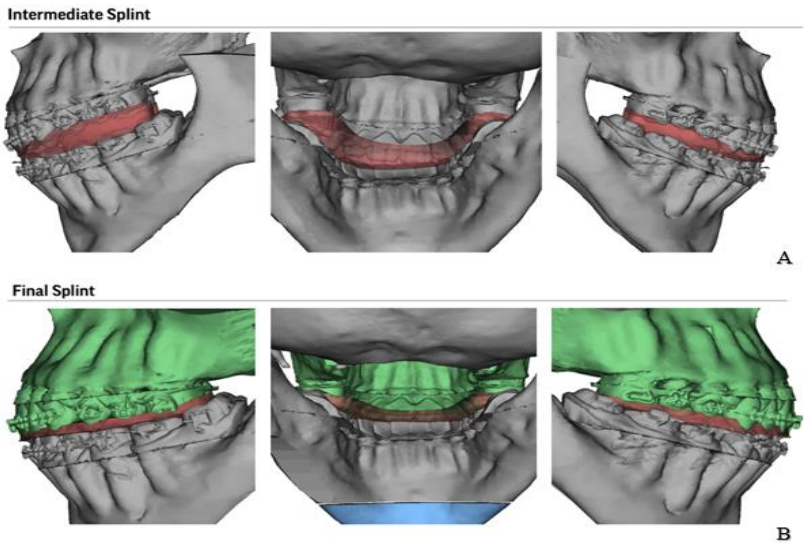

Figure 3: Shows 3D construction using 3D systems VSP orthognathics for splint (A) intermediate splint of 3D construction (B) ffinal splint of 3D construction.

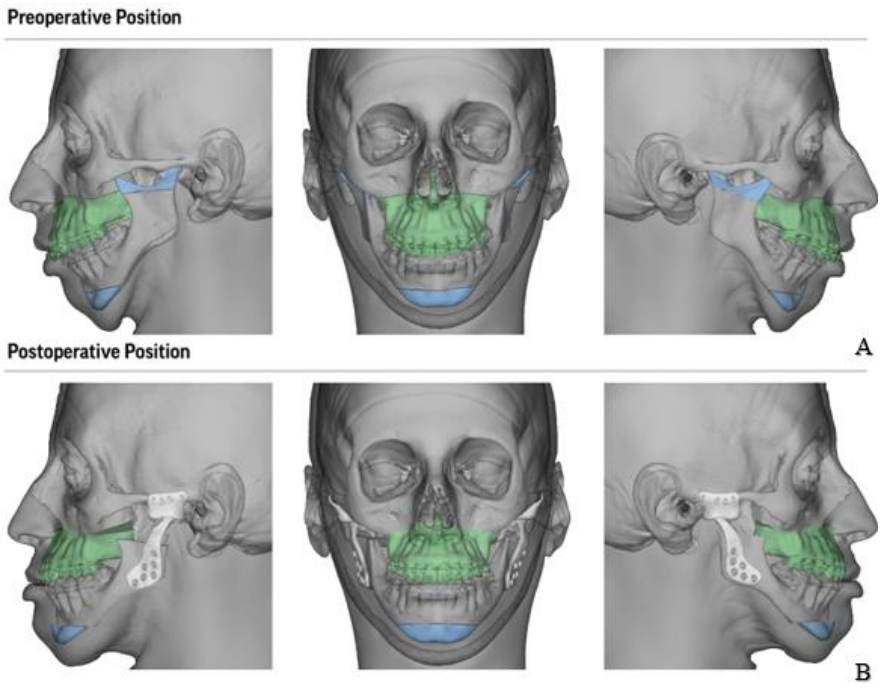

Figure 4: Shows 3D construction of transparent soft tissue analysis (A) pre-operative (B) post-operative.

The patient was brought to the operating room (OR), awake and oriented in supine position, monitors connected, general anesthesia induced, and intubation performed. Furthermore, the patient was prepared and draped in a sterile manner exposing the face, the preauricular area and the neck bilaterally, while the ear was plugged with a cotton gauze peanut, and the mouth and the nose were covered and shut with Ioban drape. Moreover, surgical marking was done as can be seen in Figure 5A.

The preauricular area was done using endural incision to access the TMJ space, utilizing the skin incision through the skin fold on the preauricular region from the upper helical to inferior of targus through the skin. Moreover, subcutaneous dissection was done to the temporoparietal facia, then temporalis facia through the avascular plane, then continued down to the joint capsule. Afterwards, zygomatic arch was identified and subperiosteal dissection was done to protect the temporal branch of facial nerve.

The retromandibular approach by incision which was done through the skin, subcutaneous, and then subplatysmal dissection reaching the pterygomandibular sling to access the ramus. Tunneling was done to have communication with the two spaces for prosthesis installation.

Condylectomy and coronoidectomy was done as planned using cutting guides through the intra-oral approach. Furthermore, the condyle head was released and freed from the lateral pterygoid muscle, fossa was debrided, and all soft tissues was removed. bleeding was encountered from the right internal maxillary artery and controlled by ligation clips. Resectioned right and left condyle and coronoid process with 3D printed template can be seen in Figure 5 (B and C).

All extraoral surgical sites were covered with sterile drapes after temporary closure with silk sutures. The intermediate splint was fitted and secured by intermaxillary fixation (IMF) then re-draped with tegaderm and ioban applied over the mouth.

The preauricular and retromandibular was accessed again with gentamycin templates of the mandibular fossa and the ascending ramus tried in with screws in place. Inferior border osteotomy was completed in the left inferior border using cutting guide plate as can be shown in Figure 5D, and the resectioned inferior portion of the mandible can be seen in Figure 5E. Final artificial bilateral joint prosthesis delivered with no issues as can be shown in Figure 5 (F and G).

Surgical site closed with vicryl 3.0 in layered fashion, monocryl 5.0 used to close the skin. The extra-oral surgical sites were covered with new drapes and the mouth accessed and occlusion checked.

The Le Fort I orthognathic surgery then commenced the injection of lidocaine 2% with epinephrine 1:80,000 in all surgical sites in the maxilla. Kirschner wire was applied in the nasion area as an external reference point, then a measurement was done from central incisors, lateral incisors, canines, and molars bilaterally. Mucosal incision was made 5 mm above the mucogingival junction from premolar to premolar followed by subperiosteal dissection and full thickness flap reflection exposing piriform rim, infraorbital nerve, zygomaticomaxillary buttresses, and pterygomaxillary junction. Moreover, nasal mucosa was reflected from anterior nasal septum and nasal floor. Internal bony reference points were drilled and measured. Afterwards, osteotomy was done 5 mm above teeth apices

from piriform rim to pterygoid plates. Separation of the pterygomaxillary junction and nasal septum was done. Subsequently, down fracture of maxilla was completed, and nasal mucosa was released. Bony interferences were removed, and maxilla was mobilized. Nasal mucosa was intact and descending palatine was observed without need of ligation. Impaction of 3 mm to the right side was achieved to correct the cant and then even impaction by 2 mm was completed. Surgical final splint used, and IMF elastics applied. Fixation using 1.5 mm synthes L-shaped plates with 5 mm screws in the maxilla. Classic alar cinch suture with Prolene 4.0 and V-Y closure with vicryl 3.0 were done.

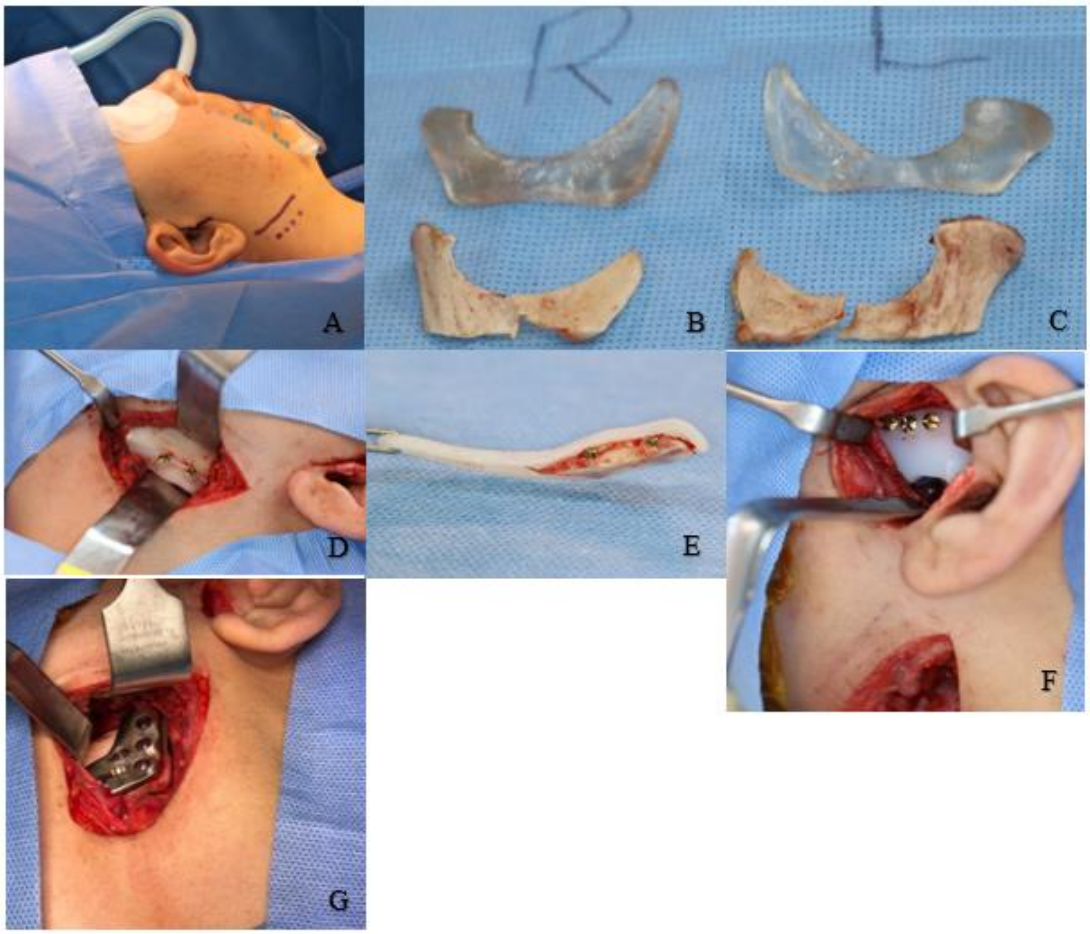

Figure 5: Shows clinical images (A) pre-operative image after draping and intubation with surgical marking (B) resectioned of right condyle and coronoid process with 3D printed template (C) resectioned of left condyle and coronoid process with 3D printed template (D) intra-operative image after osteotomy of left inferior border of the mandible with marking guide plates fixed with mini-screws (E) left resectioned inferior portion of the mandible (F) intra-operative image after fixation of artificial joint prosthesis in fossae (G) intra-operative image after fixation of artificial joint prosthesis in mandible.

The genioplasty orthognathic surgery then was initiated by injection of lidocaine 2% with epinephrine 1:80,000 in all surgical sites in the mandible. Mucosal incision was done 5mm below the mucogingival junction from canine to canine, followed by subperiosteal dissection and full thickness flap reflection without any communication of the artificial Joint bilateral. The mental nerve bilaterally was identified and protected. Furthermore, midline was marked with fissure bur 5 mm below the apices of the teeth. Subsequently, osteotomy was done and chin advanced 4 mm and fixed with plate and mini-screws. Closure using vicryl 3.0 in layered fashion was completed.

Patient was handed to anesthesia team and then shifted to intensive care unit (ICU) for post-operative airway monitoring due to prolonged OR lasting around 14 hours. Blood loss and urine output was estimated to be 600 ml and 2.4 l, respectively. Patient received 2 units of blood transfusion during operation.

After 24-hours in ICU, patient was extubated and shifted to general ward for 3-days then discharged home after total hospital course of 4-days in a stable condition. During in-patient general ward care, post-operative Computed tomography (CT) scan was requested which showed correct position of artificial joint prosthesis as planned. As seen in Figure 6.

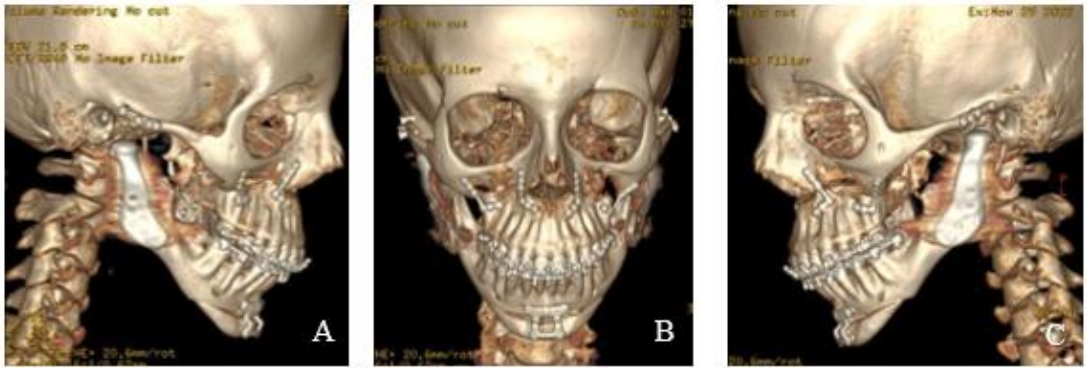

Figure 6: Shows 24-hour-post-operative 3D reconstruction of CT facial bone scan (A) right lateral view (B) frontal view (C) left lateral view.

Patient was seen in multiple follow-up visits during first year post-operatively, one week, one-three-six months and one year, respectively.

In the one-year post-operative follow up visit, clinical examination showed no tenderness in the preauricular region nor myofascial pain, symmetrical face, slightly obtuse Nasio-Labial angle, competent lips, no maxillary cant, straight profile, shallow Labio-Mental fold, no paresthesia noted, normal and symmetrical facial expression muscles (House-Brackmann grade I). As can be seen in Figure 7 (A-C). In addition, mouth opening is 33 mm, maxillary dental midline is coincident with philtrum of upper lip, mandibular dental midline is shifted 1 mm to the left of maxillary dental midline, bilateral class III molar relationship, right and left canine class II and I relationship, respectively, overjet is 3 mm, overbite is 2 mm. As can be seen in Figure 7 (D, G-I).

Radiographic examination showed improved SNB and ANB, improved incisors position, improved MMPA, and improved upper and lower lip to E-line. Additionally, well-integrated and seated artificial fossa and mandible segments, as can be seen in Figure 7 E and Table 3.

Figure 7: Shows clinical 1-year-post-operative images (A) frontal view at rest (B) lateral view at rest (C) frontal view while smiling (D) occlusal view of maxilla (E) lateral cephalometric radiograph (F) frontal view of occlusion (G) occlusal view of mandible (H) lateral right view of occlusion (I) lateral left view of occlusion.

Table 3: Summary of post-operative cephalometric analysis.

Measurement	Norm	Value
Sagittal (apical bases and chin)		
SNA	82°±2°	81.4
SNB	80°±2°	79.1
ANB	2°±1.7°	3.3
Vertical divergency		
MMPA	27°±5°	32.8
Dental (incisor position)		
UI-NA	22.8°±5.7°	18.5
LI-NB	25.3°±6°	28.7
Interincisal angle	130°±6°	128.0
Soft tissue		
Upper lip to E line	-4 mm±2	-2.0
Lower lip to E line	-2 mm±2	-2.5

Abbreviations: SNA, Sella-Nasion-Point-A angle; SNB, Sella-Nasion-Point-B angle; ANB, Point-A-Nasion-Point-B angle; MMPA, Maxillary-Mandibular Plane Angle; UI-NA, Upper-Incisor to Nasion-Point-A angle; LI-NB, Lower-Incisor to Nasion-Point-B angle; E, Esthetic.

DISCUSSION

Management of ICR remains controversial as several treatment modalities were proposed to address ICR depending on whether ICR is active and if the condyle is salvageable.[6] Among these modalities is conservative treatment which includes occlusal splint therapy, which aims to unload the condyles to relieve discomfort and muscular pain.[5] When ICR isn't active, especially when the disfigurement of the facial soft tissue is mild and acceptable to the patient, this treatments modality can take place. Non-active and healed stable stage can be determined by taking CBCT scans annually to observe any change in condylar anatomy.[7]

Orthognathic surgery alone is not recommended for ICR due to high relapse rate compared with other treatments. Furthermore, long term results are not stable, and resorption may proceed or be triggered. Additionally, resulting in significant risk for redevelopment of functional and esthetic deformities, worsening TMJ symptoms and dysfunction, and pain.[8] Cowford et al reported 7 cases of ICR that underwent orthognathic surgery alone, the follow-up period exceeded one year for all cases, 5 cases reported relapse of surgical movement of B-point, and 2 out of the 5 cases reported unstable occlusion.[9] In cases where the condyle is salvageable and ICR is active, Wolford et al protocol can be utilized. Simultaneously, orthognathic surgery will also be performed to manage the consequences of jaw and occlusal discrepancies.[3] Moreover, Wolford et al, reported 12 cases of ICR underwent Wolford et al protocol with simultaneously orthognathic surgery, follow-up period exceeded one year for all cases, and results showed no reported relapse with excellent stability.[10]

If ICR is active and condyle cannot be preserved, alloplastic temporomandibular joint reconstruction (ATMJR) can be used as a treatment modality. Furthermore, ATMJR is indicated in cases of inflammatory arthritis involving the TMJ not responsive to other modalities of treatment, recurrent fibrosis and/or bony ankylosis, failed tissue grafts (bone and soft tissue), failed ATMJR, and loss of vertical mandibular height and/or occlusal relationship such as in cases of ICR. However, ATMJR is contraindicated in young growing patients, mentally unstable patients, uncontrolled systemic disease, active infection at the implantation site, and allergy to the materials that are used in the devices to be implanted.[11] The utilization of orthognathic surgery in conjunction with TMJ reconstruction employing alloplastic prostheses has demonstrated notable efficacy, safety, and success in addressing both functional and aesthetic concerns arising from advanced TMJ disorders.[12] Furthermore, it is considered an effective procedure in the treatment of ICR that showed excellent stability of surgical movement over long-term course of follow-up. Mercuri et al, reported 11 cases of ICR, out of which 9 were treated with BATMJR alone, while 2 underwent BATMJR in conjunction with Lefort I surgery. Furthermore, the follow-up period exceeded one year for all cases, with no instances of relapse observed in mandibular function, facial form, occlusion, and joint pain.[7] Similarly, Mehra et al documented 21 cases of ICR, with 5 cases treated solely with BATMJR and 16 cases undergoing BATMJR along with Lefort I surgery. Moreover, the follow-up duration exceeded beyond five years for these cases, with no reported relapse in surgical movement.[13] Consequently, ATMJR is considered superior to other treatment modalities for ICR as it eliminates morbidities associated with second surgical site, no postsurgical remodeling, longevity of the prostheses with more than 20 years of follow up, lacks growth potential, predictable growth, and can be done along with mandible advancement movement with counterclockwise rotation without increasing loading in TMJ as the prosthesis does not remodel or resorb.[6]

CONCLUSION

In conclusion, this case report shows the treatment of skeletal class II malocclusion associated with bilateral ICR. Management aimed to improve both function and esthetic and was successfully implemented using BATMJR with simultaneous orthognathic surgery to do LeFort I, bilateral condylectomy, genioplasty, and inferior border osteotomy. One-year follow up shows stable outcome, emphasizing long-term success of comprehensive treatment strategy in addressing skeletal class II malocclusion associated with ICR.

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Ethical approval: Not required

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