A case of idiopathic condylar resorption

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Background: Idiopathic condylar resorption (ICR) is a well-documented but poorly understood condition, characterised by the progressive degeneration of the temporomandibular joint without an identifiable cause. ICR most commonly presents as a spontaneous, self-limiting episode of bilateral condylar resorption in the absence of pathology, injury or systemic disease. The condition most commonly affects physically active adolescent females, and has a strong predilection for individuals with a Class II skeletal relationship. Some ICR patients exhibit symptoms such as discomfort and/or functional limitations during the active phase of resorption, though some may be completely asymptomatic.
Aim: This case report describes the ICR management of a 20-year-old female with asymptomatic bilateral condylar resorption, resulting in a rapid development of mandibular retrognathia. The retrognathic appearance was of concern to the patient and treatment was desired for this reason.
Methods and results: Combined orthodontic-orthognathic treatment was undertaken. Pre-surgical orthodontics was completed following ICR stabilisation, and surgical correction consisted of a mandibular advancement plus a genioplasty. The resorbed condyles were accepted given the temporomandibular joints remained asymptomatic and there were no functional limitations. Following treatment, the patient remained asymptomatic, functioning without restrictions, and satisfied with the aesthetic outcome.
Conclusion: The combined orthodontic-orthognathic approach addressed the aesthetic and functional concerns of the patient. The condyles were not reconstructed with prostheses and were left in their resorbed anatomical form and relationship. This illustrates the adaptive capabilities of the mandible and associated musculature, and that more complex condylar restorative procedures are not always required.
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Introduction

Idiopathic condylar resorption (ICR) is a well-documented but poorly understood condition, characterised by the progressive degeneration of the temporomandibular joint (TMJ) without an identifiable cause. ICR most commonly presents as a spontaneous episode of bilateral condylar resorption in the absence of pathology, injury or systemic disease.[1-3] The condition most commonly affects physically active adolescent females, and has a strong predilection for individuals with a Class II skeletal relationship.[1-4] Some ICR patients exhibit symptoms related to discomfort and/or functional limitations during the active phase of resorption, although some may be completely asymptomatic.[2-4] As most cases present as a single, self-limiting occurrence, ICR can be reactivated at a later stage, although rarely over 40 years of age.[2]

Diagnosis

The diagnosis of ICR can be challenging and is often made following the exclusion of all other known causes of condylar resorption. Condylar resorption has been documented following orthognathic surgery, traumatic injuries and infections.[5] Congenital jaw abnormalities and arthritic conditions including osteoarthritis and rheumatoid arthritis also have clinical findings that overlap with ICR. Clinical examination, imaging of the TMJ and its surrounding structures, and rheumatology input can all aid in the diagnosis of ICR.

Clinically, individuals often present with profound facial and occlusal changes with typical characteristics of a Class II skeletal appearance. The changes may include a retrognathic mandible, an increase in overjet, open bite, and high occlusal and mandibular plane angles.[1,4-6] Unilateral condylar involvement can also lead to facial asymmetry, a midline shift and a lateral open bite.[4]

Imaging provided by conventional radiographs, tomograms, computed tomography, magnetic resonance imaging (MRI), and nuclear medicine bone scans may all be used to diagnose and monitor the progression of the disease. Changes in condylar morphology related to cortical bone loss, changes in condylar height, volume and shape, and the presence of sclerosis, can be expected.[6-8] MRIs may show anterior disc displacement and the presence of a thick, amorphous-like hyperplastic synovial tissue between the condylar head and mandibular fossa.[3,9]

Nuclear medicine bone scans and serial cephalometric tomograms can be used to determine whether ICR is active or has stabilised.[7-10] An increased uptake of a radioactive material (Technetium-99) can confirm areas of bone activity.

Pathogenesis

The pathogenesis of ICR is unclear, and further research is required to fully understand the aetiology of the disease. It has been proposed that an increase in mechanical loading of the TMJ (from factors linked to parafunction, trauma, and internal derangement), exceeding its adaptive capacity, can cause dysfunctional remodelling and trigger ICR.[1,4,6] However, evidence for a causal relationship between related factors and condylar resorption is weak, highlighting the idiopathic nature of the condition. Additional theories, such as avascular necrosis, suggest that ICR is due to pathological compressive forces which impinge on circulation to the condyle.[11] As ICR predominantly affects adolescent girls and young women, it has also been proposed that ICR is hormonally mediated, with environmental factors contributing to the development and progression of the condition. Oestrogen and its receptors are thought to play a role in the biomechanical changes and metabolic activity within the TMJ.[12,13] However, a systematic review by Nicolielo et al.[14] revealed limited evidence for a link between oestrogen and ICR, due to contradictory findings and a lack of well-designed randomised controlled trials. Previous authors have also suggested that ICR may affect airway dimensions due to the retruded mandibular position; however, there is a lack of evidence to either confirm or refute this opinion.[2,9]

Management

Various theories exist regarding the best management of ICR, and treatment may range from conservative (occlusal splint therapy), to complex (TMJ surgery with prosthetic joint replacement). Currently, it is unknown which treatment modality can provide long-term condylar and occlusal stability. Occlusal splint therapy may be considered during active condylar resorption to relieve discomfort; however, little evidence exists to suggest that splints help prevent ICR progression.[9]

Surgical management of ICR (with or without orthodontic treatment) is often considered after active condylar resorption to manage the end stage malocclusion and dentofacial deformity. However, there is no clear consensus as to the optimum time to operate. The most reported surgical intervention for ICR is combined orthognathic and TMJ surgery (during which the TMJ is either reconstructed, and/or the articular disc is repositioned or stabilised).[1,2,15-17] Reconstruction of the TMJ is usually performed using costochondral grafts or alloplastic materials, the latter of which is more predictable.[18-20] Orthognathic surgery alone, although less invasive, has been reported to result in higher relapse rates, either from a surgical perspective or from the re-activation or additional progression of ICR.[2,9,15-17] Hence, any surgical intervention should be performed following the cessation of ICR activity, as this may reduce the risk of post-treatment change. Orthodontic treatment alone can also be considered for limited aesthetic improvement, but will not correct any skeletal discrepancies caused by ICR.

The present case is unusual as, despite the presence of severely resorbed condyles, the patient did not present with an anterior open bite, and the TMJs remained asymptomatic and functional without restrictions. Case management therefore involved orthodontics and orthognathic surgery alone, without TMJ reconstruction.

Case report

The present report describes a case of ICR in a 20-year-old female patient who first sought orthodontic treatment in May 2007 for moderate upper and lower arch crowding. The extraction of four premolars and routine fixed appliance therapy was completed in December 2009, when aged 14 years and 2 months (Figures 1 to 3).

The patient re-presented six years following initial treatment, aged 20 years and 7 months, concerned with her 'disappearing chin' and a sudden change in facial profile occurring over a period of six months (Figure 4). She reported difficulty eating due to an increase in overjet, yet was asymptomatic and had no limitations in jaw movement. Radiographic imaging showed diminutive condyles, and severe skeletal

Figure 1. Intra-oral and extra-oral photographs following initial orthodontic treatment; December 2009.

Figure 2. Post-treatment lateral cephalogram following initial orthodontic treatment; January 2010.

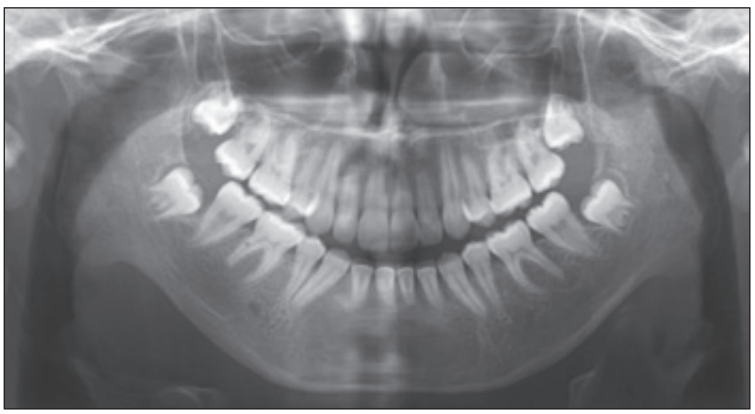

Figure 3. Post-treatment orthopantomogram following initial orthodontic treatment; January 2010.

retrognathia (Figures 5 and 6). Her clinical findings are outlined in Table I.

The cephalometric findings are shown in Table II. The measurements in 2007 and 2010 relate to the pre- and post-orthodontic treatment variables. The 2016 measurements relate to the presentation with ICR, whilst the 2019 measurements were taken following orthognathic surgery.

A nuclear medicine bone scan using 99m technetium hydroxydiphosphonate tracer (99mTc-MDP) was carried out in July 2016. The scintigraphic appearance was consistent with ongoing bone activity in bilateral mandibular condyles as demonstrated by the increased uptake of the radioactive tracer (Figure 7).

The patient was referred to a rheumatologist to exclude underlying inflammatory or systemic conditions. Inflammatory markers, immunoglobulin levels and an autoimmune screen were unremarkable and the patient was advised that no anti-resorptive therapy was warranted.

Figure 4. Intra-oral and extra-oral photographs following ICR presentation; May 2016.

Figure 5. Lateral cephalogram following ICR presentation; June 2016.

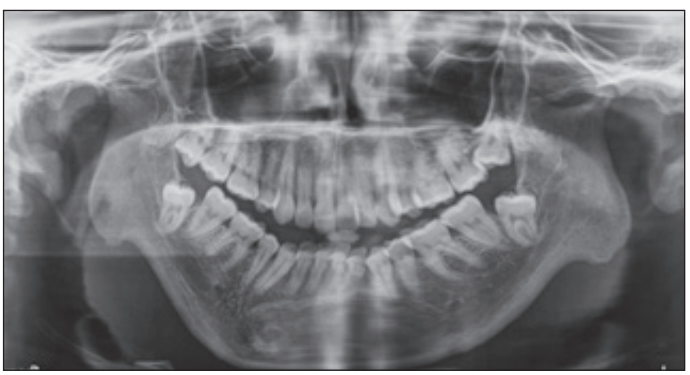

Figure 6. Orthopantomogram following ICR presentation; June 2016.

The clinical features and investigations led to a diagnosis of ICR with no systemic features. Prior to treatment, clinical monitoring supported by photographs and imaging, as well as a further nuclear medicine bone scan were prescribed to determine whether the resorptive process had ceased (Figure 8). Additionally, disease progression was not anticipated given that the condyles had almost resorbed down to the level of the mandibular notch, as shown radiographically.

Treatment

Following the cessation of ICR, treatment options were discussed as outlined in Table III. If the patient had no concerns, no treatment but ongoing observation would have been a reasonable option. However, the patient desired treatment to address her aesthetic concern relating to her 'disappearing chin'. Conservative management by orthodontic treatment alone was not suitable as it would not address this concern. Hence a combined orthodontic-orthognathic approach was taken. Multiple surgical opinions were obtained related to the surgical plan; in particular, the need for TMJ reconstruction. An assessment and opinion by an external surgeon with a subspecialty interest and experience in TMJ surgery and joint replacement was also obtained. The consensus was that TMJ reconstruction was not required at this time, as the patient was asymptomatic with no functional limitations. However, this treatment option should be kept as a contingency plan if required later due to further unexpected change. As she did not have an anterior open bite and her vertical maxillary excess

Table I. Extra- and intra-oral assessment with ICR presentation in May 2016.

Extra-oral	Intra-oral
- Skeletal pattern: Class II – severe mandibular retrognathia	- Dental pattern: Incisors: Class II div 1, overjet 12 mm
- Convex mesiofacial pattern	- Dentition: previous removal of four premolars as part of orthodontic treatment
- Growth considerations: negligible growth	- Third molars: present
- Soft tissue patterns: competent lips, deep labiomental fold	- Upper crowding: 12, 11 mildly imbricated with the distal edge of 11 being more labial
- Vertical maxillary excess	- Lower crowding: minimal
- Mandibular function: asymptomatic with no limitations	

Table II. Serial cephalometric measurements.

Cephalometric variables	09/01/2007	11/01/2010	02/06/2016	11/04/2019
SNA (°)	79	78	76	79
SNB (°)	75	72	67	74
ANB (°)	4	6	9	5
LFH (ANS-Xi-Pm) (°)	48	55	57	54
MP Angle (MP-FH) (°)	33	37	41	36
Pog to Nasion Perp (mm)	6	8	20	6
Mandibular Length (Co-Gn) (mm)	102	106	78	90
Lower incisor to MP (°)	98	97	94	97
Lower Lip to E Plane (mm)	+1	0	+2	-3
Nasolabial Angle (°)	110	111	113	113
Overjet (mm)	6	4	12	2
Overbite (mm)	6	3	3	2

SNA, Sella - Nasion - A Point Angle; SNB, Sella - Nasion - B point Angle; ANB, Difference between SNA and SNB; LFH, Lower Facial Height, Anterior Nasal Spine - Xi Point-Protuberance Menti Angle; MP, Mandibular Plane, Menton - Gonion; FH, Frankfort Plane, Porion - Orbitale; MP Angle, MP - FH Angle; Pog – N Perp. Pogonion to Nasion Perpendicular referenced to FH; Mandibular length, Condylion – Gnathion; E Plane, Esthetic Plane, Nasal Tip - Soft Tissue Pogonion. Condylion refers to the most posterior superior point on either the condyle or the residual condylar process.

Table III. Treatment options for the patient.

1)	No treatment (given no TMD like symptoms)
2)	Conservative management (occlusal splints, orthodontic treatment alone)
3)	Mandibular surgery with genioplasty
4)	Bi-maxillary surgery with genioplasty
5)	Orthognathic surgery with bilateral TMJ autogeneous or prosthetic joint reconstruction

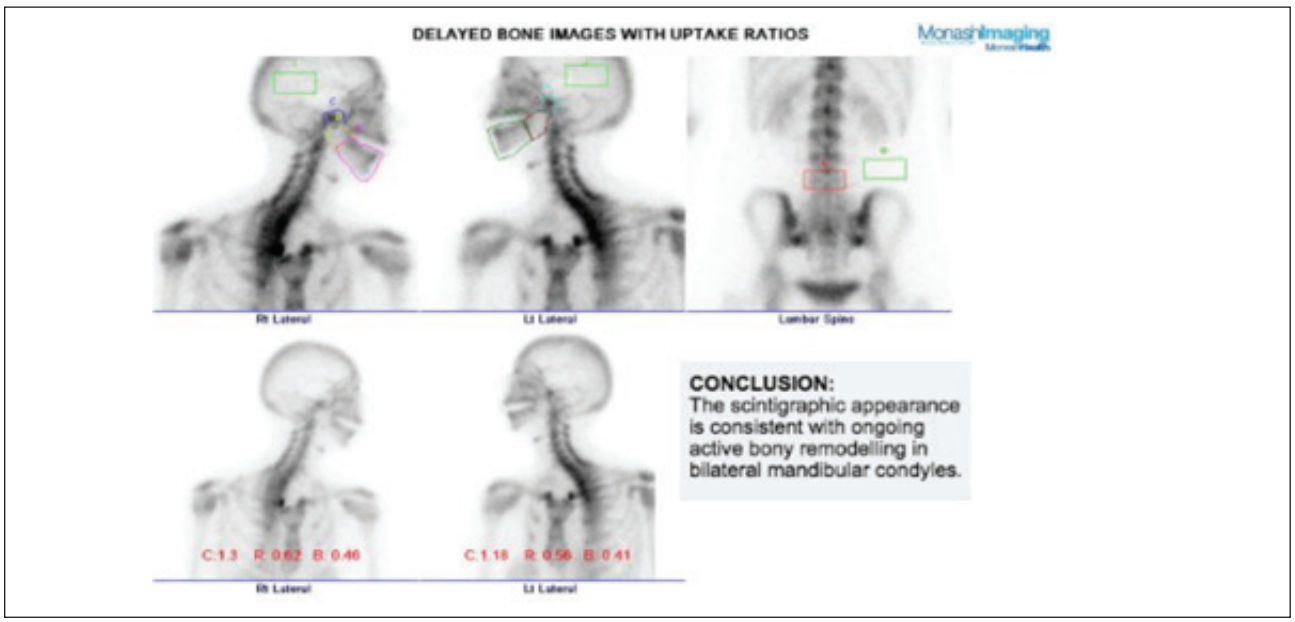

Figure 7. Nuclear medicine bone scan showing active bony remodelling; July 2016.

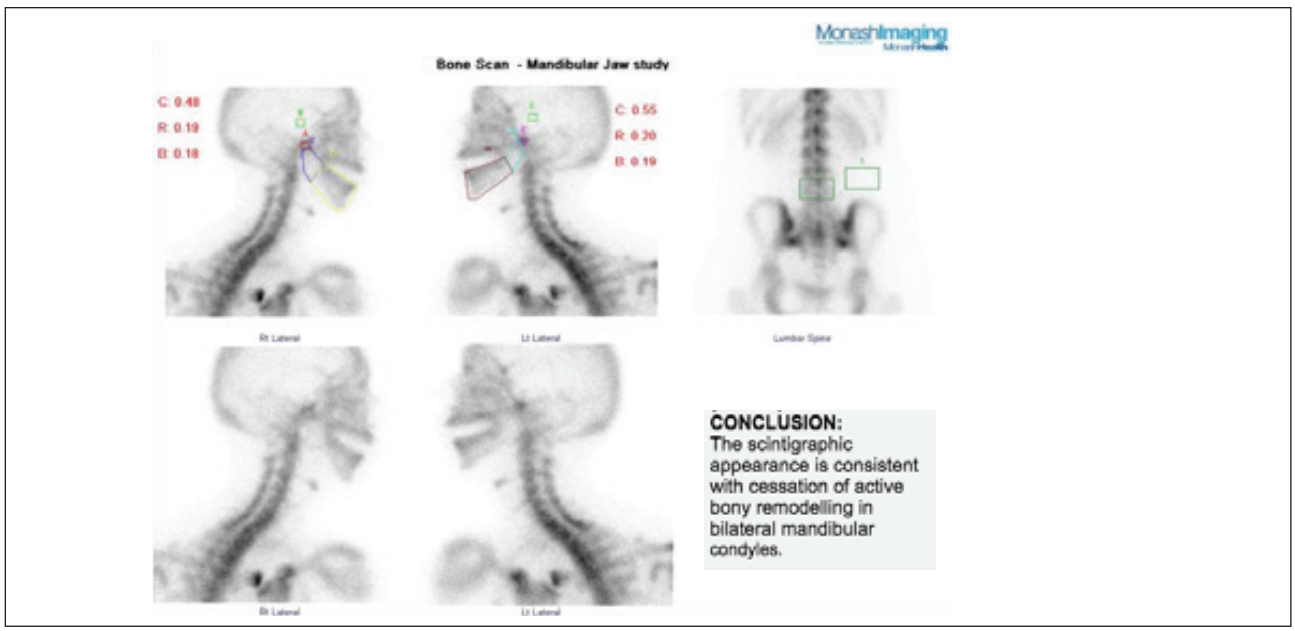

Figure 8. Nuclear medicine bone scan showing cessation of bony remodelling; July 2017.

was not a concern, mandibular advancement with a genioplasty alone was selected, rather than a bi-maxillary osteotomy.

Treatment progress

Pre-surgical orthodontics was commenced after her second bone scan did not detect areas of bone activity. In January 2019, the patient had a bilateral sagittal split osteotomy (BSSO) mandibular advancement of 10 mm together with a genioplasty advancement of 8 mm. The upper and lower third molars were also removed uneventfully. At the six-week review, overbite and overjet both measured 1.5 mm and the midlines were co-incident. The surgical movements are presented in Figures 9 and 10. The orthodontic appliances were removed in June 2019, approximately six months after the surgery. The patient was happy with the outcome and is currently being reviewed on a regular basis (Figure 11).

Figure 9. Lateral cephalogram post-orthognathic surgery; April 2019.

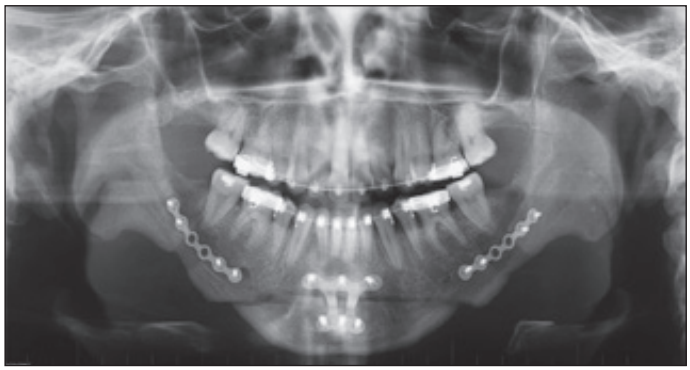

Figure 10. Orthopantomogram post-orthognathic surgery; April 2019.

Figure 11. Intra-oral and extra-oral photographs following orthognathic surgery and four months after the removal of appliances; October 2019.

Discussion

This case report presents a conservative surgical treatment approach for the management of ICR. The patient's retrognathic appearance was of most concern and, following ICR stabilisation, treatment was desired for this reason. If aesthetics was not of concern, interventional treatment may not have been required due to the absence of symptoms and functional limitations.

Earlier radiographs in 2010 showed unusual condylar morphology, a small condylar neck height and antegonial notching. Surgical intervention was not indicated at this time as there was no significant active condylar resorption adversely affecting the facial aesthetics. In addition, the Class II skeletal pattern was not severe enough to be of concern to the patient. The initial treatment was aimed at addressing the patient's chief complaint, which was her dental malocclusion.

Past studies have suggested that orthognathic surgery should be undertaken earlier in ICR but this aggressive approach for asymptomatic patients is based mainly on expert opinions. Evidence that surgery performed at an earlier age is protective against the development or progression of ICR is lacking. Had orthognathic surgery been performed at this early time, the patient may have required a second orthognathic procedure to correct aesthetic issues as a result of the ICR progression. Surgical intervention is undertaken on the basis of the presenting signs and symptoms, rather than for condylar morphology correction and halting the progress of the disease. Had earlier surgery been undertaken, the ICR may have been falsely attributed to the earlier procedure.

The diagnosis of ICR was made through a combination of history, clinical findings, imaging and medical investigations. It was important to exclude any serious underlying pathology, as the clinical manifestations of ICR can mimic those of osteoarthritis, rheumatoid arthritis and other rheumatic and inflammatory diseases.

In the present case report, the patient reported a sudden change in facial profile over a relatively short period of time prior to presentation. Between 2007 and 2010, as seen in Table II, there were cephalometric changes consistent with a mild increase in the Class II pattern and an increase in vertical facial height. Despite these changes, the profile was acceptable and the vertical dimensions were such that the mouth was positioned one-third of the way between the base of the nose and the chin, consistent for well-proportioned faces**.** Although orthodontic treatment cannot be ruled out as a risk factor for condylar resorption, it is unlikely that the initial orthodontic treatment completed four years earlier was the cause. A risk factor for this patient was most likely related to the pre-existing condylar morphology. At the present time, no accurate predictive tools are currently available to determine which cases will develop the relatively rare condition of ICR. It is also of note that ICR most commonly affects young females, being the same population group that are well represented in having had, or in currently undergoing, orthodontic treatment. This suggests that orthodontic treatment may well be coincidental rather than causative for ICR. Taking into account all presenting factors, the diagnosis for the present case was consistent with ICR.

Relapse is common following ICR management due to the ongoing nature of the disease. Hence, it is important to distinguish between active and inactive condylar resorption prior to commencing treatment. This case report demonstrates the use of multiple nuclear medicine bone scans to determine the progress of ICR. Moreover, in the 18 months prior to orthognathic surgery, there was no detectable clinical progression of the disease. It has been proposed that orthognathic surgery may reactivate condylar resorption; however, it is thought that this risk decreases with the increasing patient age.[4,15] This was not such a concern in the current case as the mandibular condyles had nearly resorbed down to the mandibular notch. However, there is potential for surgical relapse due to the lack of a stable posterior stop during the proximal segment BSSO advancement in the absence of condyles.

Currently there is insufficient research to determine factors that may account for the progression or cessation of ICR. A systematic review based on 17 articles documenting 178 cases of ICR found that orthognathic surgery alone showed the highest rate of relapse, particularly associated with mandibular advancements greater than 8 mm.[8] However, limitations were noted related to the lack of randomised control trials, inadequate review periods, and a wide geographical spread of treating institutions. It could not be determined that these cases had appropriate pre-operative management and imaging to assess the state of condylar resorption.

Further research is required to investigate the most appropriate treatment for ICR. To date, there are no published randomised clinical studies that compare the outcomes of different treatment modalities. Current studies often have major limitations highlighted by inadequate follow-up periods and inappropriate pre-operative management to ensure ICR stabilisation prior to treatment.[8]

Orthognathic surgery alone as a treatment modality should be considered in cases in which mandibular function has been preserved following ICR.[15,17] Despite the abnormal condylar morphology, the present patient had no limitations in overall jaw function, and therefore orthognathic surgery without TMJ reconstruction was considered to be suitable. Mandibular surgery alone was performed rather than a bi-maxillary osteotomy, as the patient did not have an anterior open bite and chose not to address the vertical maxillary excess. This approach, involving a mandibular BSSO and genioplasty, was chosen as

a more conservative and less aggressive treatment option with possible superior long-term stability compared to a bi-maxillary osteotomy. This treatment still successfully addressed the patient's concerns, and offered less surgical risk, less post-operative pain and a shorter recovery. The placement of cosmetic onlays on the mandibular ramus as a treatment option could also have been considered had there been a related aesthetic concern. It was discussed with the patient that, should the correction relapse, the placement of joint prostheses would need to be reviewed.

Other treatment modalities that have been utilised in ICR are distraction osteogenesis or repositioning and stabilisation of the articular disc using a mini anchor placed in the condylar head. The latter protocol was introduced in 1999 by Wolford and Cardenas[2] and is implemented independently of whether condylar resorption is active or inactive. Although numerous treatment options exist for ICR, contemporary practice should aim for an evidence-based patient-driven approach.

In summary, combined orthodontic and orthognathic management of this case aimed to correct the patient's presenting concern of a 'disappearing chin', and to restore incisal function. Despite extensive condylar resorption, the patient retained functional bilateral mandibular movements without a history of pain or other symptoms. This questions the need for prosthetic condylar replacement as a component of ICR management.

Conclusion

The present case report illustrates the interdisciplinary management involving orthodontics and orthognathic surgery in the treatment of an adult patient presenting with ICR. An aesthetic and functional outcome was achieved with mandibular advancement surgery to address the principal complaint of severe retrognathia. ICR pathogenesis is not completely understood and the patient will require regular monitoring to assess for any signs of change.

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Conflict of Interest

The authors report no professional nor financial conflict of interest in relation to this case report.

The patient provided permission for the publication of her clinical data and photographs.

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