

Rehabilitation of Dentofacial Asymmetry Secondary to Unilateral Temporomandibular Joint Ankylosis with Dual Distraction and Fixed Orthodontics – Stability at Three-year Follow-up

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Abstract: Optimal rehabilitation of asymmetric dentofacial deformity secondary to unilateral temporomandibular joint (TMJ) ankylosis is often a challenge. The purpose of this case series is to present an insight into esthetic, occlusal and functional rehabilitation of two patients with varying degree of asymmetric Class II dentofacial deformities secondary to long-standing unilateral TMJ ankylosis. The patients were treated with one-stage surgical protocol employing simultaneous dual distraction technique along with interpositional arthroplasty. Dual distraction technique entailed the simultaneous use of two distractors which allowed for proper control of proximal condylar segment during the course of distraction and lowering the risk of ankylosis recurrence. Thereafter, comprehensive fixed orthodontic mechanotherapy involving the use of temporary anchorage devices was instituted to align and level the compensated dentition. Post-treatment records showed significant improvements in skeletal disharmony and functional stability with good functional occlusion. At the three-year follow-up, the morphological and functionally acceptable results

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were reasonably well-maintained, with no signs of relapse. Through the two cases reported here, we would like to highlight that one-stage concurrent arthroplasty and dual distraction technique is a safe, stable, and reliable approach for surgical and functional rehabilitation of an adult asymmetric dentofacial deformity secondary to unilateral TMJ ankylosis. Meticulously executed comprehensive orthodontic manipulations involving use of acrylic bite-blocks, elastic traction, and temporary skeletal anchorage device play a crucial role in enhancing the final occlusal outcomes.

Introduction

In spite being one of the most prevalent and disfiguring craniofacial anomaly, asymmetric dentofacial deformity due to unilateral temporomandibular joint (TMJ) ankylosis often presents a complex therapeutic challenge (Zhu et al., 2013). Increased severity of muscular atrophy with laterognathism and retrognathism due to prolonged duration of associated hypomobility further compounds the challenging treatment process (Motta et al., 2007). Additionally, it becomes challenging to correct facial asymmetry among these patients by maintaining balance of optimum craniofacial health and cost-economic aspects to achieve satisfactory treatment goals (Zhu et al., 2013; De Roo et al., 2016).

Numerous treatment techniques, such as ramus osteotomy, high condylectomy, coronoidectomy, gap arthroplasty, interpositional gap arthroplasty, reconstruction of the joint using autogenous grafts, alloplastic materials or vascularised flaps, distraction osteogenesis, early mobilization, and aggressive physiotherapy, have been widely employed for the management of TMJ ankylosis and associated asymmetric skeletal deformities (De Roo et al., 2016).

One-stage surgery involving simultaneous gap arthroplasty and distraction osteogenesis has proven to be a viable option in the treatment of TMJ ankylosis and coexisting severe micrognathia in aptly selected cases (Dean and Alamillos, 1999; Yu et al., 2009; Giraddi et al., 2016; Sharma et al., 2016). Even so, an issue of concern to the surgeon is the potential for encroachment on the gap (arthroplasty) by the proximal segment (Srivastava et al., 2019). Moreover, the management of malocclusion after distraction osteogenesis is also fraught with challenges and is often not documented to a satisfactory degree in the literature.

The purpose of this case series is to present an insight into esthetic, occlusal and functional rehabilitation of two patients with varying degree of asymmetric Class II dentofacial deformities secondary to long-standing unilateral TMJ ankylosis. The patients were treated with one-stage surgical protocol employing simultaneous dual distraction technique along with interpositional arthroplasty. The long-term followed-up occlusal outcomes are detailed in which postdistraction orthodontic treatment following single-step simultaneous dual distraction and interpositional arthroplasty helped align and level the compensated dentition. Furthermore, the merits of dual distraction technique and factors governing stability of treatment results are also discussed.

Case report

Case 1

A 21-year-old adult male presented seeking treatment for facial asymmetry and chewing difficulties with reduced mouth-opening (Figure 1). He had a history of facial trauma due to fall at the age of 5 years.

On extraoral examination, the patient exhibited facial asymmetry involving the lower third of the face, with lateral deviation of the mandibular symphysis towards the left, a severely retrognathic mandible and limited interincisal opening of 10 mm.

Intraorally, an asymmetric sagittal occlusal relationship was observed: Class I molar and canine relations on the right side and Class II occlusion on the left side. Overjet

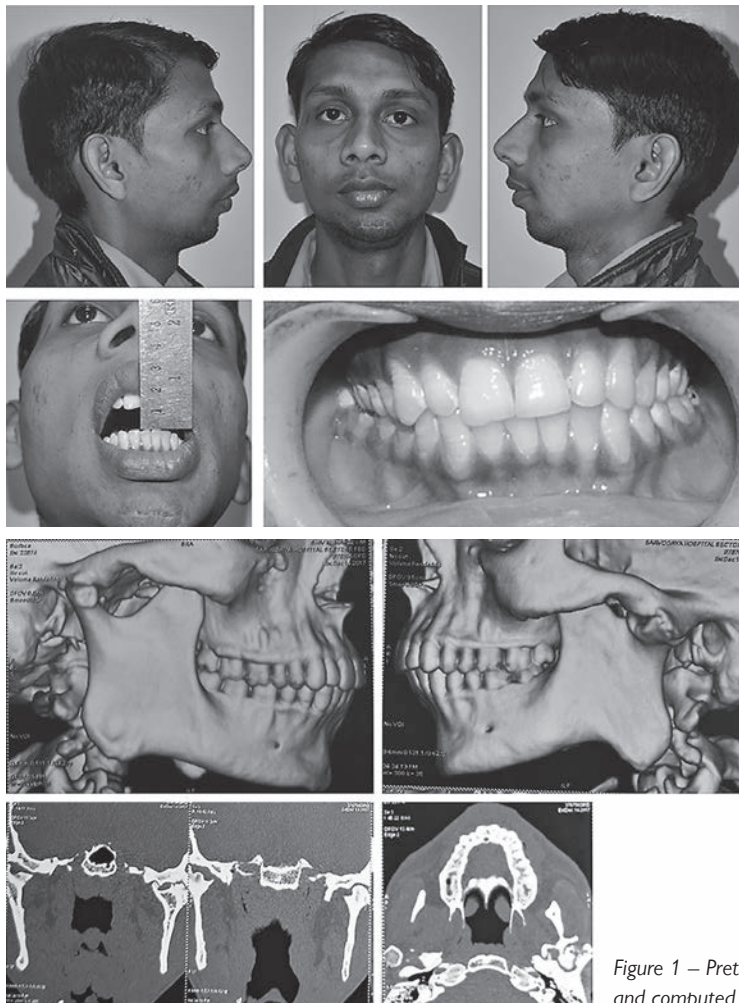


Figure 1 – Pretreatment photographs and computed tomography images.

and overbite were 3 mm and 4 mm, respectively. Mild crowding was noted in lower arch. The occlusal plane canted slightly superior on the left side and the mandibular dentoalveolar midline was deviated toward the left by 4.5 mm in relation to upper dentoalveolar midline. Pretreatment non contrast computed tomographic (NCCT) images revealed a bony ankylotic mass on the medial aspect of left TMJ, and a deformed condylar head, glenoid fossa and zygomatic arch on the right side (Figure 1).

The patient was diagnosed with an asymmetric Class II dentofacial deformity (with mandibular laterognathism and retrognathism) caused by unilateral TMJ ankylosis.

The treatment goals were to improve the diminished mouth opening by mobilizing the TMJ on the left side, improve the asymmetric facial appearance and mandibular position, and establish a normal intermaxillary relationship with a Class I molar occlusion.

Considering the history of ankylosis and long-standing severity of preoperative mouth opening limitation and low self-esteem caused by associated mandibular deformity, simultaneous execution of gap arthroplasty and dual distraction osteogenesis were contemplated to obtain adequate mouth-opening range (35 mm) and restoration of facial symmetry with stable jaw form and function. Thereafter, postdistraction orthodontic treatment was planned for levelling and aligning the dentition over the respective alveolar bases, guiding the selective eruption of teeth, correction of cross-bite and transverse inter-occlusal disharmony.

Treatment progress

The active treatment comprised of surgical phase involving simultaneous interpositional arthroplasty and dual distraction in phase 1 of treatment, accompanied by fixed orthodontic mechanotherapy in phase 2.

Initially, in a one-stage operation under general anaesthesia, interpositional arthroplasty with temporalis myofascial flap was performed to release left TMJ ankylosis (Figure 2). Concomitantly, in accordance with the protocol of Srivastava et al. (2019), two custom-fabricated extraoral monofocal/univector distractors were affixed at two different predetermined locations on the left side. Distractor A, with its vector oriented parallel to posterior border of the ramus, was used to maintain the gap created after removal of the ankylotic segment and, if indicated, also to increase the gap following activation. Another distractor B, positioned obliquely, was utilized to increase the ramus and body's vertical and the horizontal dimensions of the ramus and body, respectively, thus facilitating correction of the mandibular asymmetry (Figure 3). Following the completion of distractor B's distraction phase, distractor A was removed, and active physical therapy was initiated. During the consolidation phase, occlusal acrylic splints and elastics were used as internal fixators to counteract the vertical downward pull by supra-omohyoid group of muscles, mould the callus and obtain occlusal guidance. Following the radiographic appearance of the cortical outline on the callus during consolidation, distractor B was removed.

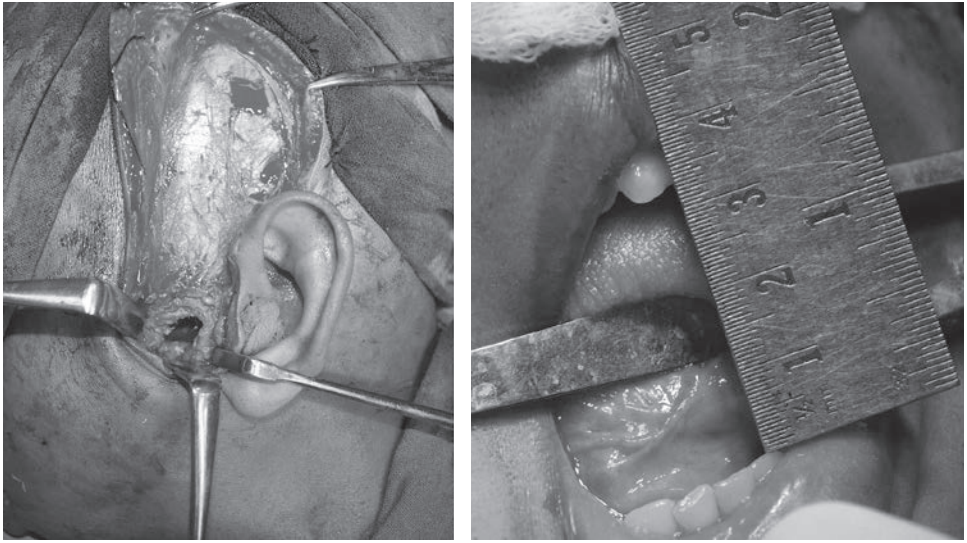


Figure 2 – Intraoperative photograph showing increased mouth opening achieved after arthroplasty.

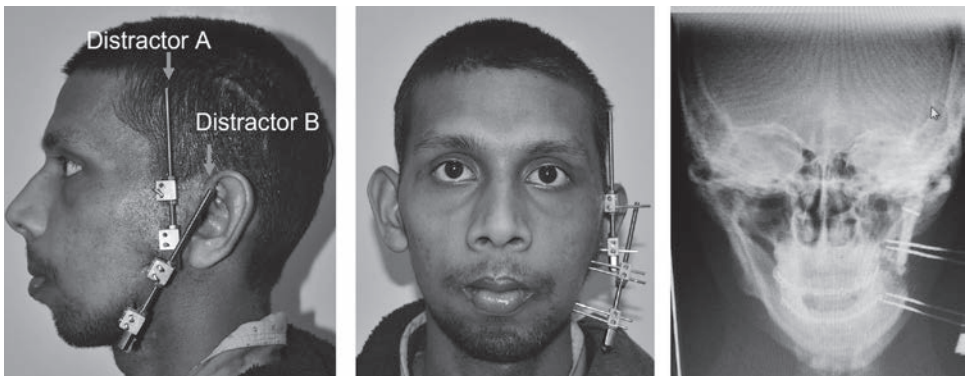


Figure 3 – Interdistraction photographs showing dual distraction and radiographs taken during consolidation phase after distraction.

A total of 12 mm of distraction was performed at a rate of 0.5 mm twice a day, followed by a 12-week consolidation phase. Post-distraction, mandibular symmetry was restored along with discernible reduction in facial convexity. Desired overcorrection of the mandibular midline was achieved with the appearance of Class III incisal relationship and reverse overjet of 2 mm (Figure 4).

At 3 months postdistraction, mandibular right central incisor was extracted and full fixed 0.022×0.028-inch preadjusted Edgewise appliances were placed in both arches in phase 2 of treatment. Alignment and levelling were performed with

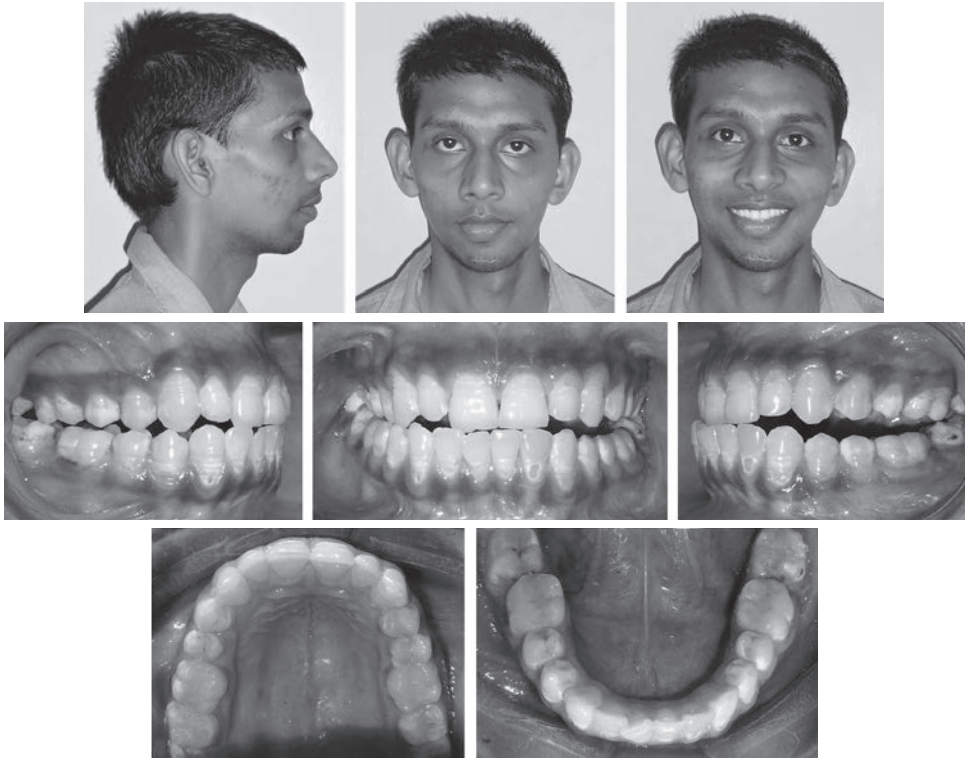


Figure 4 – Postdistraction extraoral and intraoral photographs demonstrating improved facial symmetry and appearance of incisal Class III relationship, posterior open bite on the left and crossbite on the right side.

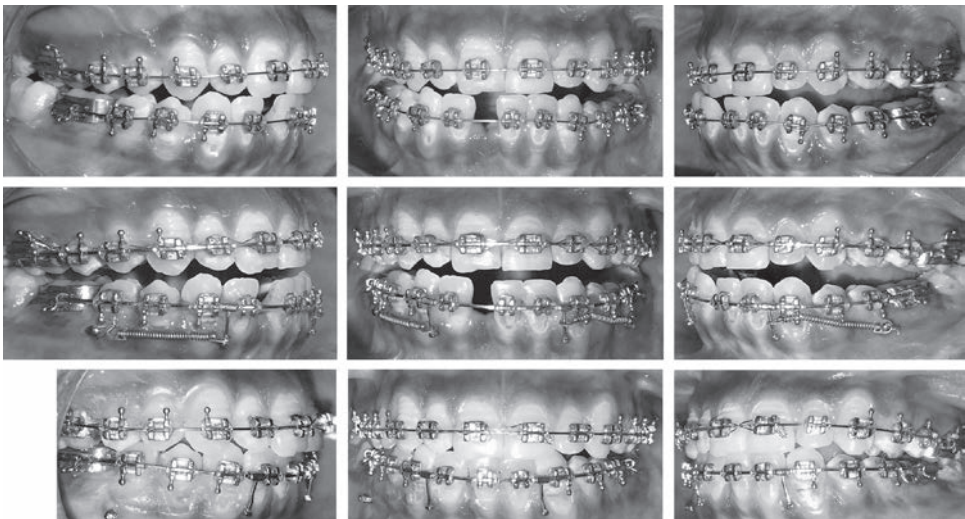


Figure 5 – Treatment progress photographs demonstrating levelling, aligning and miniscrew supported retraction of mandibular incisors during postdistraction orthodontic phase.

0.016-inch nickel-titanium (NiTi) and 0.017×0.025-inch superelastic NiTi wires. The lower incisors were retracted on a 0.019×0.025-in continuous stainless-steel (SS) wire using direct anchorage from temporary anchorage devices (TADs) i.e., mini-implants (6.0 mm long, 1.6 mm in diameter; Absoanchor – Dentos, Deagu, South Korea) inserted bilaterally into the interradicular attached gingiva between the mandibular second premolars and the first molars. 0.014-inch SS wires were used during detailing and settling phases (Figure 5). The overall treatment time including

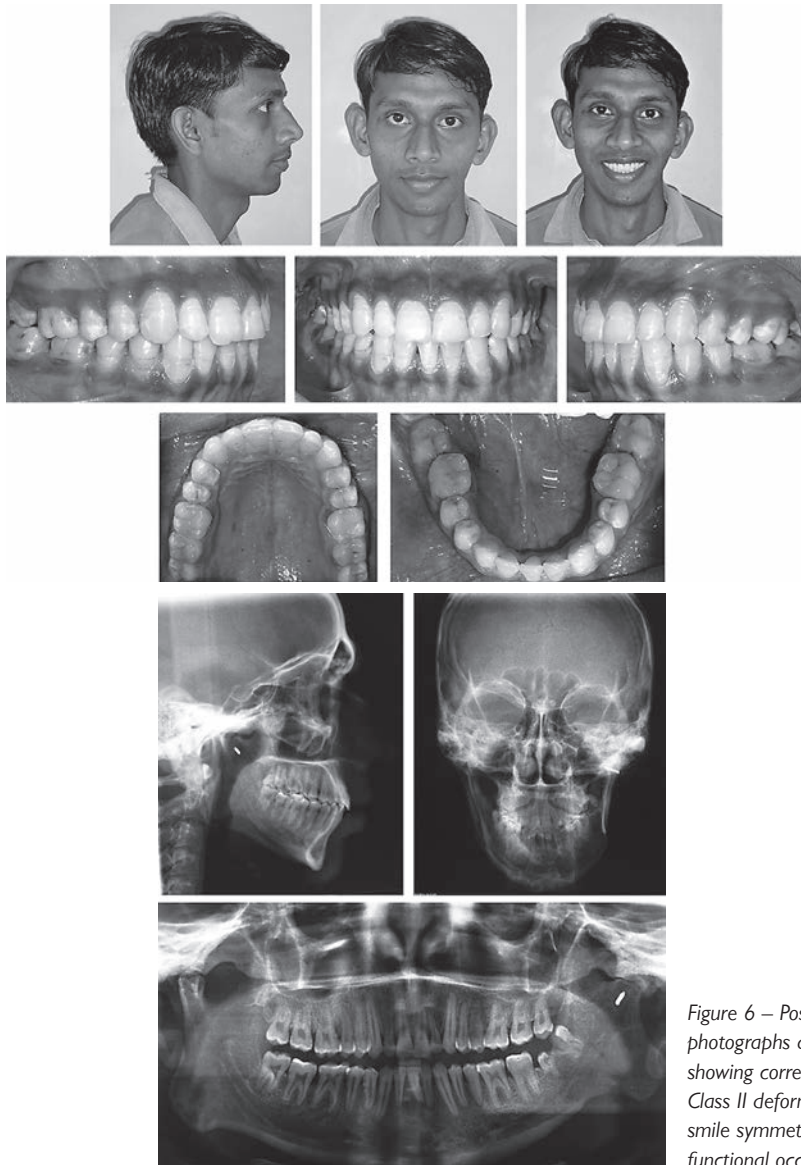


Figure 6 – Posttreatment photographs and radiographs showing corrected asymmetric Class II deformity with restored smile symmetry and Class I functional occlusion.



Figure 7 – Three-year follow-up extraoral and intraoral photographs depicting well-maintained stable results.

distraction osteogenesis (DO) was 20 months. After debonding, circumferential Hawley-type retainers were delivered for full-time wear for one year.

Treatment results

Post-treatment records showed significant improvements in facial aesthetics and profile, elimination of facial asymmetry, along with establishment of normal overjet, overbite and Class I canine-guided functional occlusion (Figure 6). The maxillary dentoalveolar midline coincided with the middle of the mandibular central incisor, without any aesthetic compromise. Panoramic radiograph demonstrated satisfactory root parallelism and no evidence of apical root resorption. After three years, the occlusion remained stable with well-maintained harmonious facial balance (Figure 7).

Case 2

The patient was a 24-year-old female with chief complaints of asymmetrical appearance, backwardly positioned chin and limited mouth opening. She exhibited inferiority complex with very low self-esteem. Her medical records revealed history of trauma to the mandible in childhood. Extraoral clinical examination revealed severe facial asymmetry with mandibular laterognathism to the left side and marked deficiency in the lower third of the face with severe mandibular retrognathism. The chin projection was inadequate, mimicking a bird face deformity. Facial atrophy was

observed on the right side of the chin. The maximum mouth opening was 11 mm due to the bony ankylosis of left TMJ. Intraorally, she exhibited a Class I malocclusion on the right and Class II on the left. Mandibular dentoalveolar midline deviation of 5 mm to the left side was observed (Figure 8).



Figure 8 – Pretreatment extraoral and intraoral photographs showing severe facial asymmetry with bird face deformity, limited mouth opening and malocclusion.

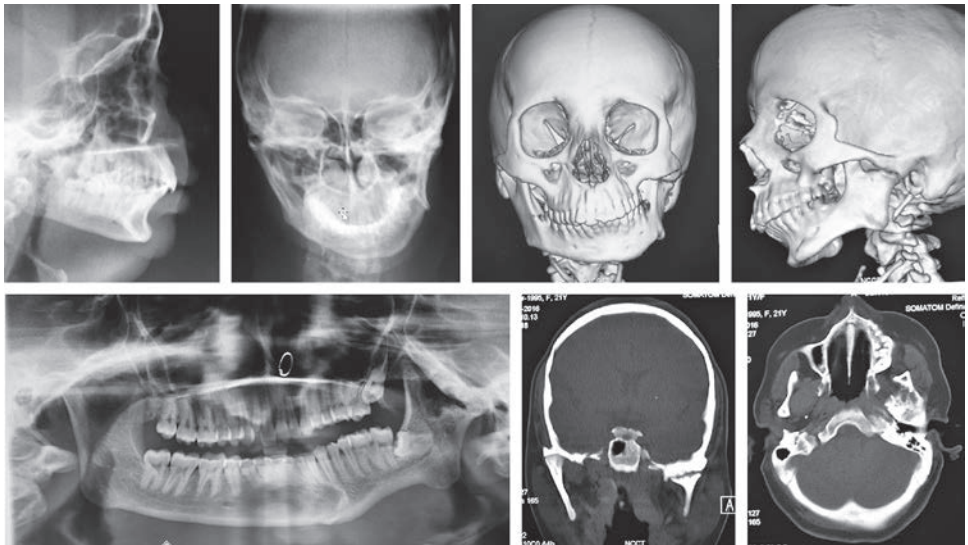


Figure 9 – Pretreatment radiographs and computed tomography images. Figure 9C (top row, third figure) is reprinted from *Journal of Oral and Maxillofacial Surgery*, 77/12, Dhirendra Srivastava, Payal Luthra, Sonal Mishra, Lokesh Chandra, Sarang Sharma, Harpreet Singh, *Technique of Dual Distraction for Correction of Unilateral Temporomandibular Joint Ankylosis with Facial Asymmetry: A Case Series*, 2555.e1–2555.e12. Copyright (2019), with permission from Elsevier.

Lateral cephalogram exhibited an obvious skeletal Class II relationship with severe mandibular retrusion. Dental compensations were observed in maxillary and mandibular dentitions. Postero-anterior cephalogram showed significant facial asymmetry with deviation of the chin point to the left side by 7 mm relative to the facial midline. The disparity in ramus sizes between the left and right sides, as well as impacted maxillary and mandibular left third molars, were evident on panoramic radiograph. NCCT of TMJ revealed deformed, flattened and mediolaterally widened left condylar process with irregular articular surface and reduced joint space, indicative of bony ankylosis of left TMJ (Figure 9).

Treatment objectives were to restore the joint function, correct the asymmetric retrognathic mandibular and chin deformity caused by left TMJ ankylosis, enhance her facial appearance and profile, and establish acceptable facial harmony. Orthodontic objectives included stabilization of post-surgical outcomes, harmonization of upper and lower arch forms, establishment of normal overjet, overbite and a stable functional occlusion.

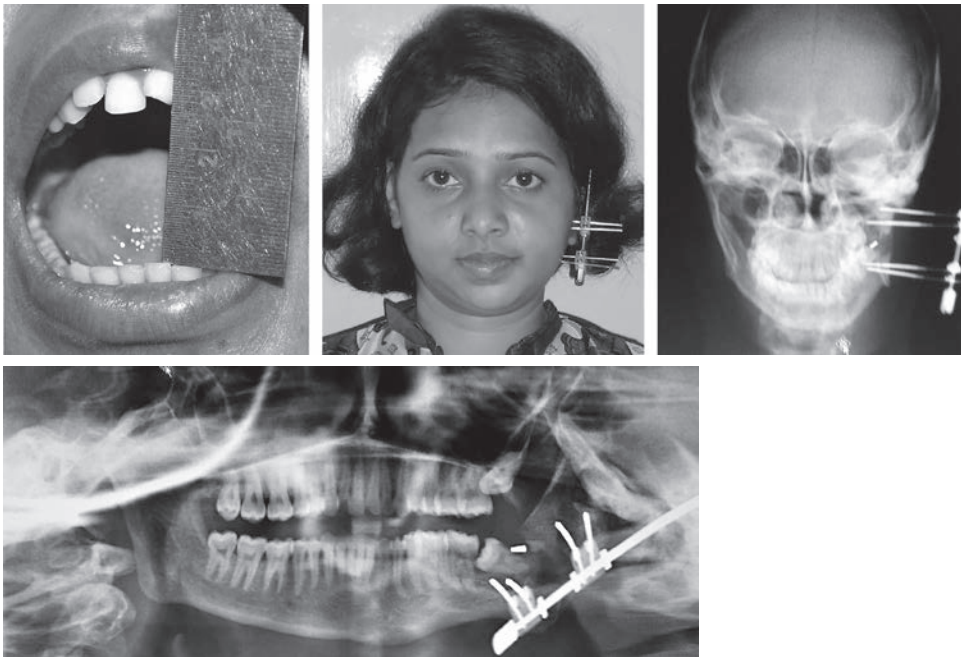


Figure 10 – Photographs showing improved mouth opening following arthroplasty, corrected mandibular asymmetry and radiographs during consolidation phase. Figure 10C (top row, third figure) is reprinted from *Journal of Oral and Maxillofacial Surgery*, 77/12, Dhirendra Srivastava, Payal Luthra, Sonal Mishra, Lokesh Chandra, Sarang Sharma, Harpreet Singh, *Technique of Dual Distraction for Correction of Unilateral Temporomandibular Joint Ankylosis with Facial Asymmetry: A Case Series*, 2555.e1–2555.e12. Copyright (2019), with permission from Elsevier.



Figure 11 – Postdistraction extraoral and intraoral photographs demonstrating improved facial symmetry, profile esthetics and appearance of Class III incisal relationship, left posterior open bite and right posterior cross bite. Figure 11B, D, E, F (top row middle figure; middle row all figures) is reprinted from *Journal of Oral and Maxillofacial Surgery*, 77/12, Dhirendra Srivastava, Payal Luthra, Sonal Mishra, Lokesh Chandra, Sarang Sharma, Harpreet Singh, *Technique of Dual Distraction for Correction of Unilateral Temporomandibular Joint Ankylosis with Facial Asymmetry: A Case Series*, 2555.e1–2555.e12. Copyright (2019), with permission from Elsevier.

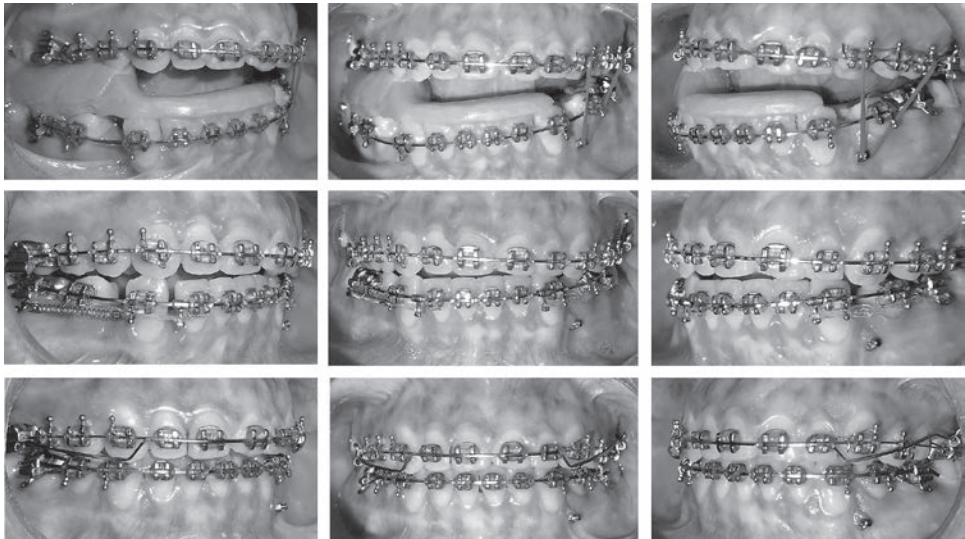


Figure 12 – Treatment progress photographs showing aligning of arches, and utilization of acrylic bite-block, elastic traction, and a temporary skeletal anchorage device for levelling of the cant of maxillary occlusal plane.

Treatment progress

At the outset, interpositional arthroplasty for release of TMJ ankylosis and establishment of adequate mouth-opening range, along with dual distraction for correction of asymmetric Class II deformity were performed simultaneously as single-step surgical procedures.

After achieving the desired ramal and mandibular lengthening, the distraction devices were maintained for 5 months until cortical outline was visible in

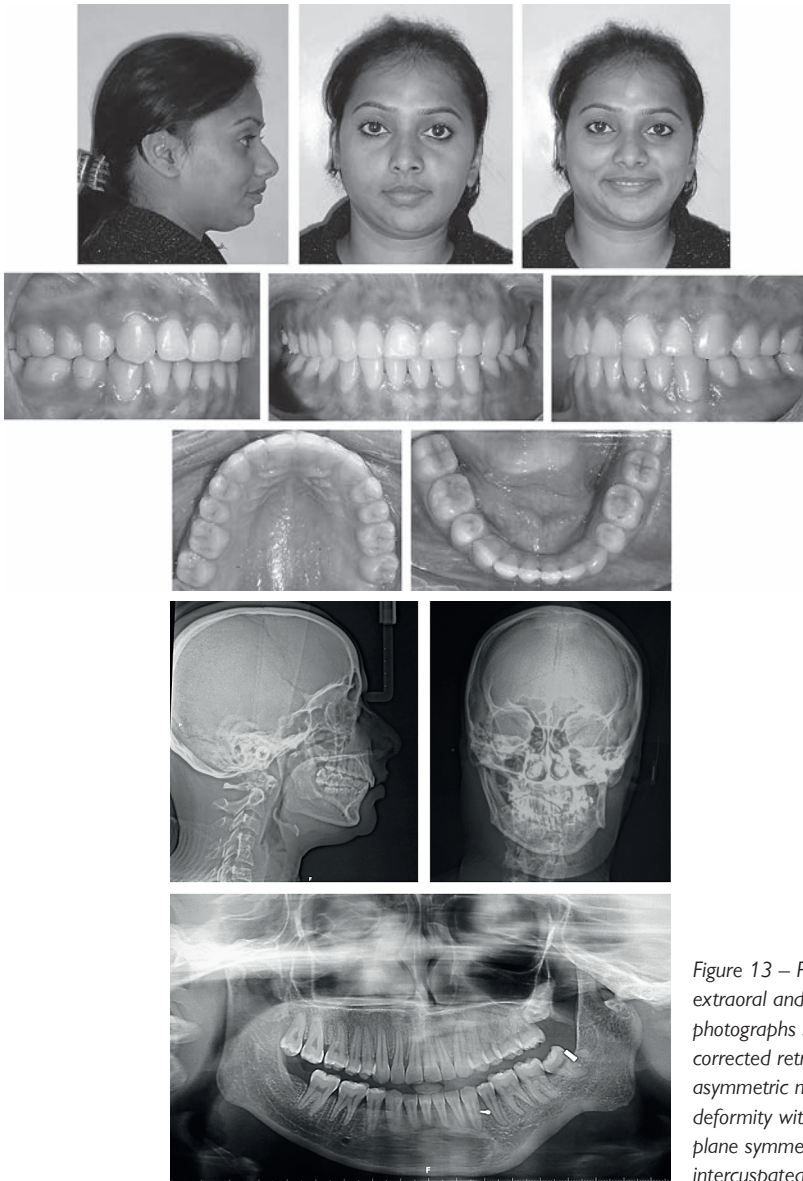


Figure 13 – Posttreatment extraoral and intraoral photographs showing corrected retrognathic and asymmetric mandibular deformity with restored occlusal plane symmetry and well-intercusped occlusion.

radiographs. A total of 13 mm of distraction was performed at a rate of 0.5 mm twice a day, accompanied by consolidation period of 16 weeks. Post-distraction, the mandibular retrusion considerably reduced and significant improvements in facial appearance were achieved (Figure 10).

However, intraorally, transient malocclusion developed with reverse overjet of 4 mm, posterior open bite on the elongated side and a crossbite on the contralateral side (Figure 11). Post-distraction orthodontic treatment was instituted following extractions of mandibular right and left first premolars and placement of 0.22×0.28-inch fixed appliances. Dental impressions were taken, and a lingual-arch supported acrylic bite-block covering the incisal edges of mandibular anterior teeth was cemented in the lower arch. To facilitate quick en-masse posterior levelling of the maxillary occlusal plane, sectional wires were hinged between the left canine and first molar, and vertical traction using elastics was applied by a mandibular miniscrew to maxillary left buccal segment. Following correction of the cant of maxillary occlusal plane, maxillary arch was stabilized using continuous 0.019×0.025-inch SS wire; and retraction of mandibular incisors progressed using continuous 0.019×0.025-inch SS wires (Figure 12).

After the removal of appliances, conventional maxillary Hawley retainer and a bonded 4-to-4 mandibular lingual retainer along with circumferential mandibular



Figure 14 – Three-year follow-up extraoral and intraoral photographs demonstrating stability of achieved results.

Hawley retainer were used for full-time retention for 6 months and then at night only for another 6 months.

Treatment results

Following 19 months of active treatment (including dual distraction), significantly improved facial symmetry and stable skeletal position with a well-settled symmetric functional occlusion was established (Figure 13). The upper dental midline coincided with the middle of the mandibular left central incisor, which did not cause any esthetic compromise to the patient. Three-year posttreatment records confirmed the stability of frontal and profile esthetics along with a tight functional occlusion (Figure 14).

Discussion

Facial asymmetry negatively impacts a patient's orofacial, psychosocial, and nutritional development by bringing forth inadequacies of a morphologic, aesthetic and stomatognathic nature. A tailor-made approach with treatment objectives determined by the patients' concerns and clinical diagnosis including malocclusion, dental compensations and concomitant sagittal and/or vertical jaw imbalance is therefore deemed most essential (Cheong and Lo, 2011).

For adults with severe asymmetric hypoplastic mandibular deformities, DO has proven to be a safe versatile technique with predictable and stable treatment results when compared to traditional bilateral sagittal split osteotomies (Al-Moraissi and Ellis, 2015). Additionally, gradual adaptation of the soft-tissue components to changes in mandibular length throughout the course of the distraction and consolidation periods represent the beneficial effects of the distraction forces on the surrounding soft tissues (distraction histogenesis) (Schreuder et al., 2007).

Dual distraction technique involving the placement of two distractors obviates the requirement for a second surgical procedure. The distractor A can be activated to increase the gap created by arthroplasty if it is deemed less than satisfactory. Patients with short ramus height, wherein increasing the gap by further bone removal could risk perforation into the middle cranial fossa (superiorly) or predispose injury to the inferior alveolar neurovascular bundle (inferiorly), may specially benefit from dual distraction approach (Srivastava et al., 2019).

Two distractors arguably allow for proper segment control by avoiding the upward and medial movement of the proximal bony segment as well as lowering the potential for impingement of the gap that was created by the arthroplasty, thus lowering the risk of recurrence of ankylosis (Srivastava et al., 2019). Oblique positioning of distractor B with angular osteotomy permits simultaneous elongation of both the mandibular ramus and corpus, thus enabling correction of mandibular asymmetry, eliminating the need of second surgery, and giving it an advantage over differential vertical and horizontal distraction.

Moulding of the generate at the end of the active period of distraction by intermaxillary elastic traction involving the use of acrylic bite-blocks and Erich arch bar helped counteract the vertical downward pull of supra-omohyoid group of muscles, close the anterior open bite and prevent inadvertent excessive increase in the mandibular plane angle. After distraction consolidation, intensive orthodontic treatment in both patients necessitated/entailed alignment and levelling of the compensated dentition over alveolar bone, establishment of symmetry within occlusal relationships between the maxillary and mandibular arches, correction of transverse maxillomandibular disharmony and controlled vertical closure of the unilateral posterior open bite (Hanson and Melugin, 1999). While performing dental decompensations, mandibular incisors were retracted taking into account the symphysis and alveolar housing's anatomical boundaries.

It has been observed that apart from mandibular deviation, the abnormal mandibular growth associated with TMJ ankylosis usually results in restricted downward maxillary growth on the affected side (Trahar et al., 2003). The resultant effect is canting of occlusal plane and decreased facial height on the affected side. Usually, additional surgery such as LeFort I osteotomy combined with bone grafts is necessitated to correct the maxillary occlusal cant and restore the normal vertical maxillary height on the affected side (Obwegeser, 1988; Molina, 1999). However, in Case 2, quick en-masse correction of the canted maxillary occlusal plane was achieved by camouflage orthodontic treatment involving the use of acrylic splint, elastic traction and miniscrew anchorage devices. Interocclusal gap created on the left side was used for selective extrusion of the maxillary left posterior dentition and alveolar process.

From a biological and biomechanical perspective, miniscrews have gained popularity for management of wide spectrum of discrepancies because forces can be applied directly from the bone-borne anchor unit. By offering the advantages of maintaining the integrity of the mandibular occlusal plane, reducing anchorage burden, preserving stability and optimizing time duration (Amm, 2012), utilization of TADs also provides biological, therapeutic and psychological advantages, as discernible in our case. Additionally, the complexity and potential risks associated with LeFort 1 osteotomy such as intraoperative and postoperative complications in terms of hemorrhage, pain, swelling, nerve injury, and infection were altogether obviated (Sousa and Turrini, 2012).

To further improve the lower one-third facial esthetics in Case 2, sliding genioplasty for centring of chin accompanied by volume augmentation on right side of face using medpore alloplastic reconstruction/fat graft, if required thereafter, was proposed as a secondary procedure. However, the patient refused to undergo another surgical intervention.

Postoperatively, the recovery period was uneventful in both patients with no detection of any relapse and neurosensory deficit. A major concerning factor commonly associated with the usage of extraoral distractor is the possibility of skin

scarring. In these circumstances, placement of the pins within the submandibular fold with minimum soft tissue strain and by squeezing the cheek skin between the thumb and index finger before inserting the second pin (to allow for stretching) helps reduce the length of the traction scar. Minor point-like external skin scarring following secondary healing did not result in esthetic compromise in present cases.

One-stage treatment, often requiring intermaxillary fixation limits the use of intensive mouth opening exercises, thereby increasing the risk of relapse (Zhu et al., 2013). Although the process of dual distraction involving concurrent fixation of gap arthroplasty avoids the need for intermaxillary fixation, early and aggressive postoperative mouth opening exercises cannot be performed owing to physical interference with the distraction process (with likelihood of disruption of callus). However, passive mouth opening exercises and regular physiological masticatory function are sufficient to preserve the achieved mouth opening during the distraction period, as observed in both patients. Following completion of consolidation period, aggressive physiotherapy exercises were employed to maintain functional integrity and prevent recurrence. In accordance with the patient-centred outcome, one-stage treatment protocol facilitating simultaneous correction of ankylosis, and facial deformities helped obviate the need for a second major surgery and provide more immediate resolution of the patients' chief complaints, thus uplifting their self-esteem. At 3-year follow-up review, both patients maintained satisfactory mouth opening, function, and esthetic appearance.

Stability considerations

A systematic review of the stability and the effects of mandibular DO on hard and soft tissues found variables such as high gonial angle and Jarabak ratio to be correlated with an increased tendency for significant skeletal relapse, but without significantly worsening the overall treatment results (Rossini et al., 2016). Considerable reduction in overjet at long-term follow-up has also been reported to support the long-term effectiveness of DO (Rossini et al., 2016), as was discernible in present cases.

Factors governing the stability of the surgical results included: effective surgical release of ankylosis, establishment of adequate mouth-opening range, improved control of the distraction vectors by employing two extraoral distractors and adequate consolidation phase. Above all, early institution of passive mouth opening and closing exercises during the distraction phase of distractor B, accompanied by active isometric and isotonic physiotherapeutic regimen (following removal of distractor A and completion of the distraction phase of distractor B) intensively for up to 2 years along with close follow-ups, also played a crucial role in achieving stable successful outcomes in terms of restoring physiological TMJ function and preventing recurrence of ankylosis. Occlusal acrylic splints and stable muscular forces also aid in preventing relapse and ensuring stability in peri-distraction phase (Chugh et al.,

2021). Postdistraction orthodontic manipulations involving elimination of occlusal compensations, adequate levelling and alignment of dental arches, restoration of proper overjet and occlusion, correction of transverse discrepancy by proper arch coordination, appropriate first and third order control and extended period of rigid retention protocol, combined altogether, contributed to long term stability of the occlusal results.

Conclusion

Correction of asymmetric dentofacial deformities present a complex therapeutic scenario to plan and accomplish. The optimal skeletal and functional enhancements along with harmonious esthetic outcomes demonstrate that one-stage concurrent arthroplasty and dual distraction osteogenesis is a safe, stable, and reliable approach for surgical rehabilitation of an adult asymmetric dentofacial deformity secondary to unilateral TMJ ankylosis. Comprehensive fixed orthodontic mechanotherapy which includes the use of acrylic bite-blocks, elastic traction, and temporary skeletal anchorage device is critical in the management of specific malocclusion caused by distraction osteogenesis. More research is warranted to determine the treatment's long-term efficacy and underlying remodelling processes.

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