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Osteogenic distraction and orthognathic surgery to correct sequelae of ankylosis of the temporomandibular joint: A case report

A 17-year-old female patient presented with sequelae to ankylosis of the temporomandibular joint, which included vertical maxillary protrusion, anterior open bite, labial incompetence, micrognathia, undefined neck angle, facial asymmetry, Class II molar relationship, and Class III canine relationship. She presented with the following cephalometric and soft tissue data: SNA angle = 78 degrees, SNB angle = 70 degrees, incisor-nasion-point A = 11 degrees, incisor-nasion-point B = 33 degrees, Frankfort-mandibular plane angle = 43 degrees, occlusal plane = 25 degrees, subnasale-stomion = 20 mm, stomion superius–stomion inferius = 9 mm, stomion inferius–soft tissue menton = 30 mm, neck angle = 144 degrees, and chin projection = 10 mm. Orthognathic surgery and mandibular osteogenic distraction were employed, specifically Le Fort I osteotomy to decrease a vertical excess of 12 mm, augmentation genioplasty of 17 mm, and bilateral extraoral distractors of bidirectional vector for a 14-mm augmentation of the mandible. The result was satisfactory with minimal adverse complications. (Int J Adult Orthod Orthognath Surg 2002;17:291–296)

Bony ankylosis of the temporomandibular joint (TMJ) results in immobilization of the mandible; when it occurs in children, it impairs mandibular growth and results in mandibular retrognathism. The ankylosis thus causes functional and esthetic disturbances as well as difficulties with nutrition and oral hygiene; this pathology can be considered incapacitating. A classification of ankylosis was proposed by Kazanjian in 1938[1] that divided the disorder into true and false varieties. True ankylosis was considered as any condition that produced fibrous or bony adhesions between the articulated surfaces of the TMJ. False ankylosis resulted from pathologic conditions outside of the joint, which resulted in limited mandibular mobility.

A variety of techniques for treatment of ankylosis have been described in the literature, but the most relevant is a management protocol proposed by Kaban et al in 1990.[2] The treatment of the sequelae of bilateral temporomandibular ankylosis is difficult, because these patients present with significant micrognathia, and it is impossible to use the conventional techniques for the mandibular augmentation[3]; in addition, the maxilla usually presents with vertical excess or more length than usual. In 1989, Ilizarov,[4] who has been engaged in clinical, biologic, engineering, and basic science research in orthopedics and traumatology, led the discovery of a general biologic principle that governs the stimulation of tissue growth and regeneration during distraction. This principle is called the *law of tension-stress*. Tissues subjected to slow, steady traction become metabolically activated, a phenomenon characterized by the stimulation of both proliferate and biosynthetic cellular functions. The application of this principle has allowed control, for the first time, of both osseous healing and the shaping processes of bone and soft tissues in many situations. Ilizarov[5] also describes

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Fig 1 *(left)* Frontal view, showing vertical excess of the maxilla.

Fig 2 *(right)* Lateral view of the patient, with the soft tissue landmarks indicated, as proposed by Epker et al.[21]

the optimum preservation of periosseous tissues, bone marrow, and blood supply at the time of osteotomy; stability of external fixation; and 1.0 mm per day of distraction in 4 steps. Osteogenesis within the distraction gap of an elongating bone takes place by the formation of a physislike structure, in which new bone forms in parallel columns, extending in both directions from a central grown zone. The growth plate that forms under the influence of tension-stress has features of both physeal and intramembranous ossification, yet is neither. Instead, the distraction regenerates bone; this technique has been used in clinical traumatology, orthopedics, maxillofacial surgery, and other medical disciplines. The quality of osteogenesis depends on the stability of the external fixation being used for distraction and the degree of preservation of the periosteal and marrow tissues and the nutrient blood vessels at the level of the osteotomy. Different daily rates of distraction and frequencies of distraction may have an effect on both bone and soft tissues under the influence of tension-stress. According to this principle, many authors have been utilizing this method to treat patients with severe craniofacial deformities, with great results.[6–19]

The purpose of this paper is to show how osteogenic distraction and orthognathic surgery can change the sequelae produced by the unsuccessful treatment of ankylosis of the TMJ.

Case report

A phototype II complexion female patient presented with sequelae to ankylosis of the TMJ, which had occurred in her infancy.

Two bilateral arthroplasties had been performed by another service, with no positive results; when she was 17 years old she came to the Stomatology Service, Department of Maxillofacial Surgery, National Pediatric Institute (Instituto Nacional de Pediatria, INP), where clinical and radiographic assessments were made. The diagnosis indicated bilateral bony ankylosis. A TMJ bilateral arthroplasty surgery was performed, followed by intense physical therapy[1] to provide her with a satisfactory oral aperture. At that time, she was 19 years old.

She then entered into the dentofacial deformity patient protocol, which began with presurgical orthodontic treatment. Several aspects were considered in planning her treatment: *(1)* clinical assessment; *(2)* radiographic assessment (lateral skull cephalometry, posteroanterior cephalometry, orthopantomography, TMJ lineal tomography); *(3)* study models; *(4)* photographic analysis; *(5)* cephalometric analysis according to Wolford et al[20]; and *(6)* soft tissue analysis according to Epker et al.[21]

Clinical data

The patient presented with the following (Figs 1 and 2): brachycephalic head type, euryprosopic facial type, a retrognathic facial profile, vertical maxillary excess, transverse maxillary plane alteration, maxillary protrusion, anterior open bite, labial incompetence, micrognathia, undefined neck angle, facial asymmetry, undefined labiomental angle, Class II molar relationships and Class III canine relationships, maxillary midline deviation, and absence of all third molars and the mandibular right first molar.

Cephalometric data

The patient's cephalometric measurements were as follows: SNA angle = 78 degrees, SNB angle = 70 degrees, ANB angle = +8 degrees, incisor-nasion-point A = 11 degrees, incisor-nasion-point A = + 4 mm, incisor-nasion-point B = 33 degrees, incisor-nasion-point B = +12 mm, interincisal angle = 130 degrees, incisor-Frankfort plane = 95 degrees, incisor-palatal-plane = 100 degrees, incisor-mandibular plane angle = 92 degrees, gonial angle = 118 degrees, FMA angle = 43 degrees, occlusal plane = 25 degrees, incisor-menton = 59 mm, soft tissue glabella-subnasale = 71 mm, subnasale-soft tissue menton = 71 mm, subnasale-stomion = 20 mm, stomion superius–stomion inferius = 9 mm, and stomion inferius–soft tissue menton = 30 mm (Fig 3).

Soft tissue assessment

Measurements for the nose were as follows (numbers in parentheses indicate normal measurements) alar base : alar base (34 ± 4 mm) = 25 mm; alar base-alar base : nasion-pronasale (0.60) = 0.74; nasal dorsum = convex; subnasale-pronasale : nasion-subnasale (40%) = 34%; nasal bridge (5 to 8 mm in front of glabella) = 10 mm; subnasale-columella : subnasale-superior lip (90 to 110 degrees) = 105 degrees; nasion-pronasale : nasion-glabella (132 ± 15 degrees) = 154 degrees; G-pogonion : nasion-pronasale (35 degrees) = 38 degrees; and columella = upward.

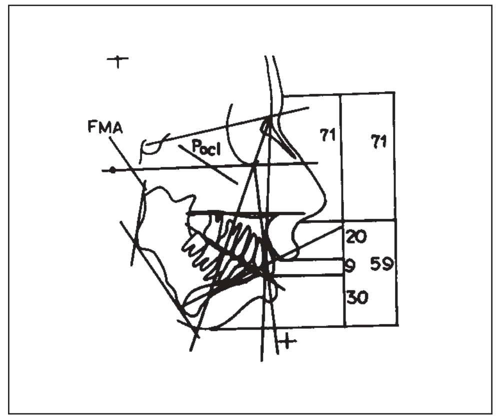


Fig 3 Cephalometric data, according to Wolford et al.[20] Pocl = occlusal plane.

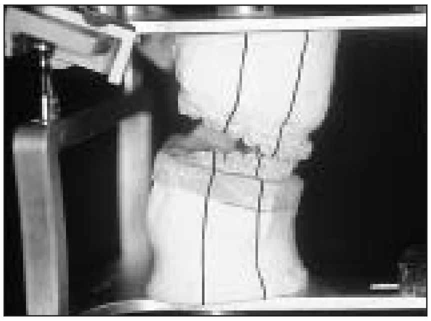
Assessment of the neck and submental area revealed the following: neck angle (110 degrees) = 144 degrees; chin length (50 mm from pogonion to neck angle) = 10 mm.

Treatment plan

The proposed treatment was divided in 2 phases according to Wolford et al[20]: surgical predictions and model surgery.

First phase. For the superior maxilla, the plan was as follows: Le Fort I osteotomy, anterior right side impaction of 12 mm, anterior left side impaction of 10 mm, posterior impaction of 3 mm, movement of midline 2 mm to the right, and resection of the inferior turbinate. For the mandible, we planned to use bidirectional extraoral distractors designed by Dr Fuente del Campo (González General Hospital/Hospital General González, in Mexico City) as well as a 17-mm advancement genioplasty and 3 mm of lowering. For soft tissue treatment, we planned an alar base cinch suture and V-Y sutures in the upper lip.

Distraction pins were activated 7 days after insertion, and given half a turn every 12 hours, for a total of 1 mm/day in each osteotomy. The right and left mandibular branches were activated for 14 days. After 18 days of right and left mandibular body pin activation, laterognathia and a rightward deviation of the mandibular midline were observed. A panoramic radiograph demonstrated incomplete osteotomy in the alveolar right portion. This was finished



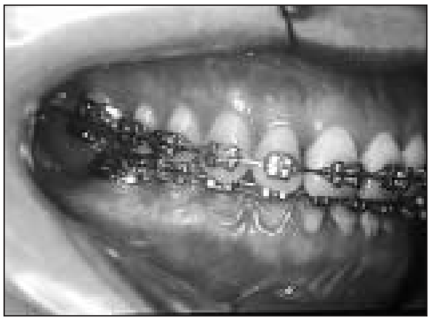


Fig 4 Study models showing posterior open bite after the end of the mandibular osteogenic distraction.

Fig 5 The posterior open bite is closed after the use of elastics.

with a hammer and chisel using local anesthesia. The mandibular second molar was extracted, as it presented root exposition (bone lost around the molar root). The right mandibular body pin was activated for 15 more days, and the left mandibular body pin was activated for 10 more days.

A month after the mandibular body pin distraction was interrupted, posterior bilateral open bite, 7 mm rightward deviation of the mandibular dental midline, and mandibular laterognathia were observed.

To correct the posterior open bite, the tension between the pins was lessened, soft elastics were applied (changed every third day), and the mandibular left second molar was extracted (since it interfered with a proper occlusion). After 2 months of treatment, the proper closing of the posterior open bite was achieved (Figs 4 and 5). The use of elastics was extended for more than 45 days to avoid relapse, after which the pins in the mandibular body were removed. The pins in the right and left mandibular branches were removed 3 months after the distraction was concluded.

Reconstruction of a light-bulb–shaped bone in the right mandibular body was verified after radiographic sampling; this probably occurred because the osteotomy was completed after the distraction process.

Dermatologic treatment was applied to treat acne and scars produced by the distractors. Washing of the face 3 times a day with neutral soap and the use of Adaferin gel 0.1% (Galderma) at night were prescribed to reduce sebaceous gland activity and thus improve the appearance of the scars. This was carried out for at least 3 months.

Second phase. To correct laterognathia and to improve the appearance of the mandibular and mentum angles, the following steps were proposed: Obwegeser sagittal split ramus osteotomy (SSRO) modified by Dal Pont for 1.5 mm augmentation and 5 mm right rotation; 12 mm augmentation genioplasty and 4 mm triangular impaction in the anterior portion to lift the chin and to improve the shape of labiomental angle; and the use of Medpore surgical implants (Interpore International) to define the mandibular angle and to correct light bulb shape in right mandibular body as well as to correct the verrucoid area on the upper lip.

During the second phase of the treatment, the patient faced pressing obligations; thus, a revision of the previous surgical plan took place. The SSRO modified by Dal Pont was not scheduled due to the patient's request, as she was unable to miss school for more than 5 days. Instead, a chin osteotomy was scheduled in the following manner: 12 mm augmentation using a 4-mm triangular impaction with 5 mm of rightward rotation, 3 mm of rightward transverse impaction, and a 3-mm leftward lowering. During surgery, only a 5-mm chin augmentation was possible because of poor stretching in the infrahyoid musculature.

The patient suffered a contusion to the right side of the mandible approximately 30 days after the surgery. Three days later she presented with suppuration, so it became necessary to remove the right prosthesis

Fig 6 Frontal view at the end of the treatment.

Fig 7 Lateral view showing the mandibular achievement with minimal scars.

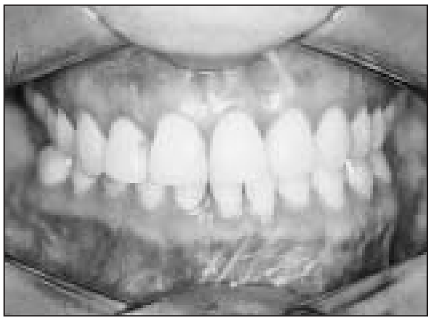


Fig 8 Final occlusion at the end of orthodontic treatment.

using local anesthesia; also, 300 mg clindamicin was prescribed (every 6 hours for 10 days). The left prosthesis was also removed due to facial asymmetry (Figs 6 to 8).

Discussion and conclusions

There is no doubt that osteogenic distraction becomes a great tool when dentoskeletal deformities require a considerable graft volume for their reconstruction but where grafting turns out to be an inappropriate treatment. Nevertheless, several aspects that are involved in the distraction process must be taken into account, including the appropriate selection of patients, meticulous treatment planning, and the required skills to deal with any unexpected situations during the process.

As osteogenic distraction is a relatively new form of treatment in maxillofacial surgery, we had to deal with some unexpected setbacks in the present case, for which we had to find solutions according to our knowledge and experience. The international literature consulted showed similar doubts, as well as similar solutions to our problems. Among these setbacks we can mention "jumping teeth"—those which migrate to one side of the osteotomy and change their position, affecting a proper occlusion. For these, the only possible solution was extraction. We also found a significant posterior open bite, which resulted from using bidirectional vector distraction. Fortunately, it was possible to correct this by lowering the tension in the pins and using elastics to encourage osseous reconstruction in the alveolar region (lateral to the distraction area). To avoid this kind of situation, and when maxillary surgery is required, we believe that posterior lowering would be indicated, so that the occlusal plane is correctly altered. The use of elastics in the osteotomy site before ossification is completed would be the best measure if only mandibular distraction is performed.

We highlight the issue that without the osteogenic distraction process, the results we achieved would not have been the same, since mandibular augmentation techniques and grafts are limited when a considerable soft tissue gain is required. Although it still represents a new kind of technique with several questions still unanswered, osteogenic distraction is the best indicated treatment in such patients.

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