Temporomandibular Joint Total Bilateral Replacement due to Ankylosis. A case report

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Case Report

Maxillofacial Surgery

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Background

Introduction: Temporo-mandibular joint (TMJ) ankylosis is a degenerative joint condition that is commonly associated with trauma, infections, systemic diseases and congenital diseases[1]. It can occur extrarticular[2] or intrarticular[3]. In the case that occurs, the ankylosis of both TMJ has a mixed origin, since it is the condition of severe mandibular hypoplasia as part of the Pierre Robin sequence (PRS), and in sequelae of previous treatments with unfavorable results. Therefore, the total replacement of both TMJ was decided, with favorable final results.

Objective: Presentation of the case of a patient with PRS and bilateral TMJ ankylosis, as a result of complications in previous management of severe mandibular hypoplasia condition.

Methodology: The recruitment of the patient is recorded, and the different treatments carried out during 14 years of follow-up are mentioned. This publication is focused on treatment of both TMJ total replacement, with titanium and ultra-high molecular weight polyethylene (UHMWP) implants.

Results: Replacement of both TMJ, despite their complications, constituted the success of the fourth and final stage of treatment.

Keywords: Pierre Robin sequence; Ankylosis; Total Replacement of Temporo-Mandibular Joints.

The case presented herein is the third case of Pierre Robin sequence (PRS) treated over the last 23 years in the Maxillofacial Surgery Service of the North Central Hospital of Petróleos Mexicanos (*Hospital Central Norte de Petróleos Mexicanos*, HCNPM), and it is the only case of PRS with bilateral TMJ ankylosis, as a complication of a previous treatment of mandibular distraction osteogenesis (MDO) in the neonatal period. It is the case of a female patient with PRS consisting of severe mandibular hypoplasia, cleft soft palate and glossoptosis, lacking other syndromic features.

OBJECTIVE

Surgical treatments performed on the patient during the last 14 years are discussed and the resolution of the bilateral TMJ ankylosis complication by means of total TMJ replacement is described.

MATERIALS AND METHODS

A typical hospital clinical case is presented, which is closed due to the certainty of its diagnosis.

1. PATIENT'S BASELINE INFORMATION

The patient was introduced in 2001, 30 days after her birth. Phenotypically she showed the typical characteristics of PRS and had external mandibular osteogenic distraction (MOD) elements, which were not placed at the Petróleos Mexicanos North Central Hospital. All further contact was lost.

In 2006, she was re-admitted into the HCNPM's Maxillofacial Surgery Service and was diagnosed with Pierre Robin sequence (severe mandibular hypoplasia, cleft soft palate and glossoptosis), and ankylosis of both temporomandibular joints (TMJ).

2. SURGICAL TREATMENTS PRIOR TO TEMPOROMANDIBULAR JOINT TOTAL BILATERAL REPLACEMENT

For its management, a short, medium and long-term stage-based treatment plan was established. Year 2006. Tracheotomy. Release of bilateral ankylosis of TMJ left mandibular condylectomy, right TMJ gap arthroplasty.

Year 2006. Palatoplasty of soft palate fissure.

Year 2010. Tracheotomy. Right condyle ankylosis relapse. Right mandibular condylectomy and mandibular osteotomies and intraoral mandibular distractor placement.

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Figure 1. Year 2019. Clinical signs of mandibular hypoplasia are quite evident. Central image shows hypertonicity of the chin to force labial competence. Lateral images show a lack of cervical distance.

Year 2011. Tracheotomy. Removal of mandibular distractors. Bone grafting and left side mandibular angle/body rigid fixation.

Year 2014. Removal of rigid fixation material from left side mandibular angle/body. Orotracheal intubation with direct laryngoscopy and guidance.

Year 2020, February 10. Temporomandibular joint total bilateral replacement.

3. TMJ TOTAL BILATERAL REPLACEMENT

Due to the patient's change of address, all contact with her was lost for four years and seven months. At the age of 17, the patient was showing very severe mandibular hypoplasia (Fig. 1). Imaging studies showed that there was no ankylosis, and yet mandibular mobility was once again limited. This condition was determined by two situations: 1) bilateral absence of mandibular condyles, and 2) mandibular rotation downward and backward, at the chin level, caused by the traction of the suprahyoid muscles, and upward and forward, at the mandibular angles, caused by the masticatory muscles, such that the left coronoid process was being blocked when opening the mouth due to contact with the maxillary tuberosity (Fig. 2).

The severity of mandibular hypoplasia and the absence of both mandibular condyles were driving factors for choosing total replacement of both TMJs as the treatment approach.

A. DESIGN AND CONSTRUCTION OF PROSTHETIC IMPLANTS FOR BOTH TMJs.

The first step was designing the prosthesis, a task supported by TechFit (Sampedro Sistemas de Fijación Ósea/TechFit Medellín Cra 47# 100 sur 40 Bodega 14 Centro Ind. Portal del Sur. La Estrella, Antioquia, Colombia), (Tech Fit: 1511 Aviation Center Pkwy, Ste 220H, Daytona Beach, FL 32114 USA), using computer-aided design and computer-aided manufacturing technology (CAD/CAM)

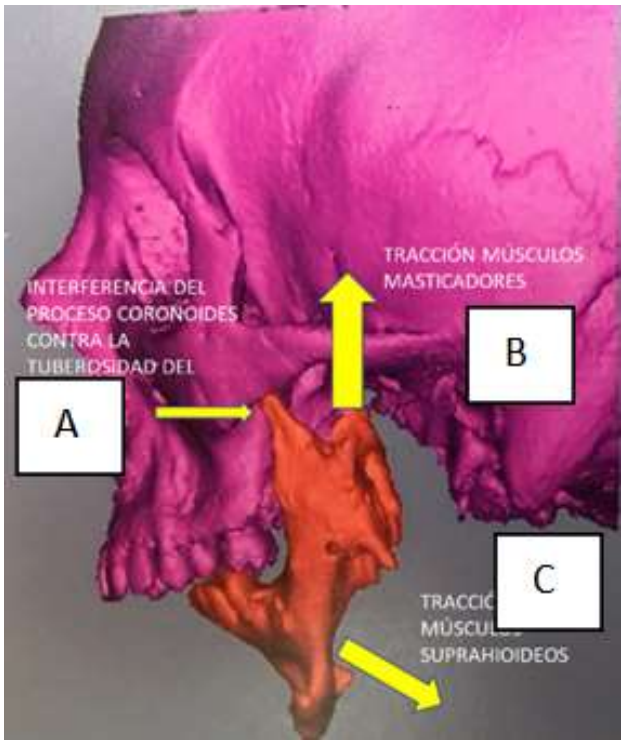

Figure 2. This image shows the status of the masticatory and suprahyoid muscle action and their contribution to the blocking of the coronoid process of the mandible against the maxillary tuberosity on the left side. A. Interference of the coronoid process against maxillary tuberosity. B. Masticatory muscle traction. C. Suprahyoid muscle traction.

(DICOM: Mimics, 3-matic / powermill Autodesk, Autodesk Inc San Rafael CA, USA). This included processing of computed axial tomography (CT) images of the skull and maxillae in 3D rendering, as well as scanning of the dental study models, thus reproducing the real existing conditions of the relationship of the jaw to the skull, of both maxillae to each other, and of the maxillae to the teeth (Fig. 3).

Scanning of study models and determination of the trans-operative occlusal guidance summarize the prediction of mandibular advancement and interocclusal space for future prosthetic rehabilitation. Incorporation of dental study model scanned images into the three-dimensional CT reconstruction model provides an invaluable tool to verify that the proposed mandibular movement will be symmetrical while obtaining the best possible dental occlusion, or whether it should be modified (Fig. 4).

Resulting prediction was that the maxilla would have to be moved about 20 mm forward with almost 45-degree counterclockwise rotation, which means that the final maxilla position, intended to be totally passive, would be difficult to obtain by surgery, while still favoring a sufficiently harmonious dental relationship (Fig. 5).

Verification is achieved through three-dimensional printing of the skull, maxillae and prosthetic elements corresponding to the skull and maxillae themselves, in plastic resin; lastly, metal

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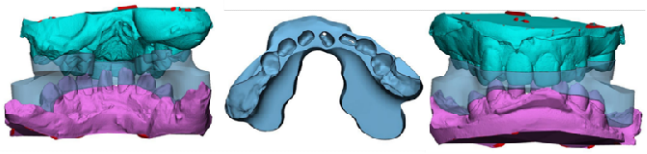

Figure 3. Dental study models scanned with the proposed final dental occlusion, captured in the final occlusal guide (central image).

implants (mandibular rami) and plastic/metallic implants (fossae replacements) are manufactured.

Each substitute used to repair fossa consists of a metallic cranial component and a metal-integrated articular portion made of ultra-high molecular weight polyethylene (UHMWPE). Different plastic test impressions of all these elements (Stratasys F370, fused deposition modeling, FDM) were obtained to determine their accuracy with respect to the planning of the required dental occlusion. The project was accepted and the sintering of all the metallic elements (Arcam EBM electronic beam melting) of titanium-aluminum-vanadium alloy (Ti6Al4V) replacements for metallic elements and metal-integrated joint cavities in UHMWPE[4] was carried out (Fig. 6).

Our pre-operative protocol consisted of an updated medical history record, routine pre-operative laboratory testing, ensuring the availability of hemo-components, and assessment by the anesthesiology and otorhinolaryngology services. The patient was prepared by shaving 4cm^2 of hair from the temporal regions and general control of her hair. Admission 12 hours in advance and administration of antibiotics: clindamycin 600mg/6h IV[5,6]; ceftriaxone 1g/12h IV[7], and dexamethasone 16mg IV[8], for pre-incision.

Antisepsis: For this type of surgery to be successful, it is essential to maintain aseptic conditions during the whole procedure. Every time there was contact with the oral cavity, no matter how minimal, the sterile fields used to cover the surgical sites and the sterile gloves were changed.

Pre-auricular approach[9].

Skin and subcutaneous cellular tissue incision along the entire length of the above-mentioned approach design. Anterior skin flap dissection extending 2 centimeters. Dissection of the anterior wall of the external auditory canal (EAC). Division of the superficial temporal fascia sheet at a 45-degree angle from the zygomatic arch to where the mandibular condyle should be, gaining access to the articular eminence and the glenoid cavity of the temporal bone. Complete dissection of the articular surface of the temporal bone to allow seating of the cranial portion of the joint replacement.

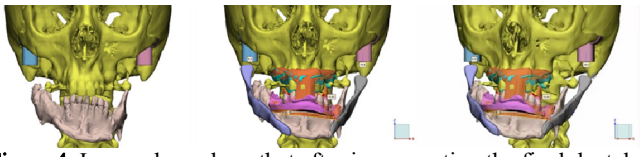

Figure 4. Images here show that after incorporating the final dental occlusion, mandibular movement is asymmetric and must be corrected.

Submandibular approach[9].

Skin and subcutaneous cellular tissue incision along the entire length of the above-mentioned approach design. Division of the platysma muscle. Two cm-wide carving of superior myocutaneous flap. Division of the superficial sheet of the deep cervical fascia and ligation of facial blood vessels. Division of the pterygoid-masseter hammock and periosteum. Dissection of the entire lateral side of the mandibular body, angle and ramus.

Connection of both approaches by tunneling, performed with blunt scissors and periosteal elevator. Verification of passivity in mandibular manipulation.

Placing of replacements[10]: Testing of the cranial and mandibular components in place. Verification of the passive seating of these elements separately, without attempting to articulate them. Occlusal guide placement and application of inter-dento maxillary fixation as tight and rigid as possible.

Fixation of the cranial prosthetic element on one side by placing the 1.5mm diameter screws one by one according to the pre-established depth in the prosthetic design. Next, fixation of the mandibular element following the same procedure with 2.5mm diameter screws. Direct visual verification to ensure there are no gaps between bone surfaces and prosthetic elements. Verification that the mandibular element was placed in the center of the articular cavity of the cranial element, in a latero-medial direction and that it made full contact with the posterior edge of the cavity, in a passive way. This process was performed bilaterally (fig. 7).

Removal of the inter-dental maxillary fixation locks. Verification of absolute passivity in mandibular mobilization. Verification, through direct observation, of the mandibular element stability in the new articular fossa, on each side.

Thorough cleaning of the oral cavity, removal of pharyngeal packing, removal of the occlusal guide, and placement of elastics for soft inter-dental maxillary fixation.

Thorough cleaning of surgical approaches and placement of negative pressure drains by counter-opening from the anterior end of submandibular approaches. Hemostasis assessment and suturing on three planes of each approach. Activation of drains and placement of compression facial dressings.

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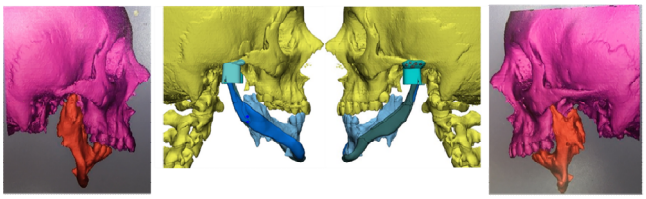

Figure 5. Central images show the design of the cranial and mandibular joint replacement elements, and the required advancement and rotation of the maxillae compared to the initial mandibular position are displayed (images on the sides).

Completed.

In-hospital stay was 11 days long, during which medications administered were continued clindamycin-based antibiotic prophylaxis 600mg/6h IV[5,6], and ceftriaxone 1g/12h IV[7,11]. Analgesia is achieved with infused buprenorphine 300mcg/24h IV[12], paracetamol 600mg/6h IV[13], and ketorolac 30mg/8h IV[14]. Anti-thromboembolic therapy with enoxaparin 40mg/24h SC[15] was administered. Additional medication included dexamethasone 8mg/12h IV[8], ondansetron 8mg/8h IV [16], and omeprazole 40mg/24h IV[17].

Soft inter-dento maxillary fixation was maintained and feeding was based on blended and strained food. The patient was discharged home after 11 days of in-hospital stay. Antibiotic medication at home was clindamycin 300mg/8h orally[5], and moxifloxacin 400mg/24h orally[8], for one week.

C. RESULTS

From the study and preparation of the case for total TMJ replacement, the result obtained was that the mandible could be moved approximately 20mm forward with almost 45 degrees counterclockwise rotation, thus privileging a sufficiently harmonious dental relationship.

Surgery involved wide mandibular movements, but it was achieved passively and in a stable manner. Results were verified by X-ray and clinically (Fig. 8 and 9).

Asymmetry of the mandibular body was managed as well as possible since final dental occlusion was preferred.

Discussion

TMJ ankylosis is a degenerative joint condition commonly associated with trauma, infections, systemic diseases, and birth disorders[1]. It may involve extra-articular manifestations in which case treatment focuses on coronoidectomy procedure and placement of some interpositional material to prevent recurrence[2].

In the case of intra-articular ankylosis, methods most commonly used to treat it include gap

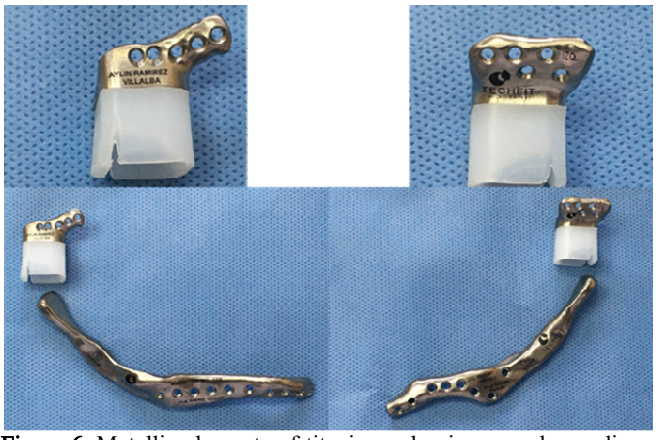

Figure 6. Metallic elements of titanium, aluminum, and vanadium (Ti_6Al_4V) alloy and UHMWPE replacements finished and ready for packaging and sterilization.

arthroplasty and TMJ arthroplasty with inter-positional material[19]. At the same time, it should be considered that on one side there may be osseous ankylosis and on the opposite fibrous ankylosis[19]. Performing mandibular condylectomy and leaving the patient with an open mouth for several weeks was also suggested as a preventive measure against recurrence of ankylosis[20], while others more often rely on the benefits of costochondral grafting with inter-positional material, mainly in children and adolescents[21].

Based on the fact that as interposition material had not been placed and the right TMJ developed new ankylosis, the decision made was to release the right TMJ ankylosis (year 2010) at the same surgical time when mandibular distraction osteogenesis (MDO) devices were placed bilaterally[22]. However, results were not favorable. Another option considered was to perform bilateral sagittal split osteotomy (BSSO) of mandibular rami with the goal of attaining advancement[23], but it was discarded since a very large advancement was required.

The major benefit to the airway was clearly obtained after total TMJ replacement surgery, due to the anterior traction effect on the tongue, and the sudden increase in intraoral space caused by mandibular advancement[16]. The patient can now remain for long periods in a supine position (including sleeping) without the need to remove the trainer tamponade from her tracheostomy tube and without experiencing breathing difficulty or snoring.

Findings

Temporomandibular joint ankylosis is the most serious complication of mandibular osteogenic distraction in newborns. In turn, complications in its management negatively alter the individual's quality of life, because as described, by not being able to open the mouth, the airway is dangerously compromised, feeding is difficult due to the inability to chew, oral hygiene becomes inefficient despite the efforts of the

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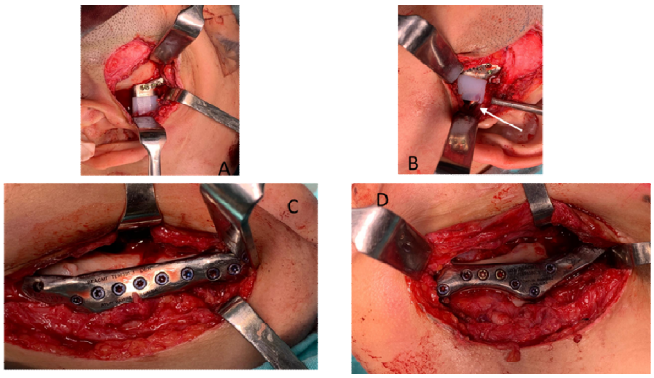

Figure 7. A. Right cranial element; fixed. B. Left cranial element, fixed. The arrow points to the left mandibular element condyle in its correct place in the proper acetabulum. C and D. Right and left mandibular elements in their fixation process.

patient and the integral dentist and speech is unintelligible.

As to total TMJ replacement planning, implant design using CAD/CAM technology ensures accuracy at the time of surgery. However, the surgeon must be extremely aware of the design processes because they often require certain movements of bone structures and soft tissues that are not always possible to perform passively and with stable results.

Conflicts of interest

The authors declare no conflict of interest.

Figure 8. Immediate post-operative X-ray monitoring. It shows the correct position of the implants placed on the sites chosen at the design stage.

Figure 9. Pre- and post-operative clinical comparative. Chin projection and new mandibular angles are evident.

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