Custom-made Total Joint Replacement After Segmental Resection of Recurrent Follicular Ameloblastoma

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Abstract

Design: Clinical case report.
Objective: To present custom-made total joint replacement as a suitable option after resection of extensive odontogenic tumors.
Background: Ameloblastoma is the most common odontogenic tumor. Clinical presentation is characteristic as it presents with a slow growth rate, but highly expansive and recurrent. Ample resection with safety margins is imperative to allow for a functional and predictable reconstruction.
Methods: We present the case of a 51-year-old male diagnosed with a recurrent follicular ameloblastoma and was treated with segmental resection and subsequent reconstruction with custom-made total joint replacement.
Results: Follow-up at 14 months after surgery showed satisfactory quality of life and proper functionality.
Conclusions: Total custom joint replacement is an ideal option after extensive resections involving the articular region.

Keywords

custom joint replacement, ameloblastoma, mandibular reconstruction

Introduction

Ameloblastoma is a benign epithelial odontogenic tumor, according to the World Health Organization Classification of Odontogenic Tumors (WHO 2017) is divided into 4 types: Ameloblastoma (formerly known as Common, Solid, Multicystic), Unicystic, Peripheral and Metastatic. The first being the most prevalent and aggressive variant. Diagnosis is based on the histopathological study and correlation among clinical findings and imaging studies.1,2 Ameloblastoma was treated for years conservatively, consisting of enucleation and curettage and marsupialization, with recurrence rates close to 70%, while radical treatment, such as segmental or marginal resection with safety margins of 1 to 2 cm, lowered recurrence rates to less than 20%. The size of the lesion (each 10 mm increase in the size of the lesion represents a 1.26% increase in the risk of recurrence), the radiographic presentation (multilocular lesions presented a risk of recurrence 78% lower than the unilocular lesions), and treatment (enucleation was associated with a radius of danger of recurrence 24.46% greater than radical treatment) represent variables to consider to determine the risk of recurrence.3,4 A few cases have been...
documented of patients with BRAFV600E mutation and with ameloblastomas with advanced local extension, who have been treated with a BRAF inhibitor (dabrafenib) with encouraging results; however, more series of cases are required to demonstrate the effectiveness of this method as an adjunctive treatment to surgery.\textsuperscript{5} Block resection is proposed as the treatment of choice for ameloblastoma;\textsuperscript{6-8} however, it requires reconstruction with vascularized or nonvascularized bone grafts, bone morphogenic protein, distraction osteogenesis or alloplastic total joint replacement to resolve functional and cosmetic problems. Involvement of the temporomandibular joint (TMJ) adds another level of complexity since a functional and stable joint must also be reconstructed.\textsuperscript{9}

**Case description**

We present the case of a 51-year-old male patient with a past medical history of controlled arterial hypertension, who after the extraction of a retained third molar, showed during radiographic testing, a radiolucent lesion in the mandibular ramus which was treated by enucleation and curettage somewhere else on 09/11/2008 (Figure 1). The histopathological examination yielded a diagnosis of unicystic ameloblastoma. The patient had clinical and radiographic follow-up for 2 years. He visited our department on 04/26/2018 with intense pain (VAS 8/10) to the touch, increase in size + / +++, paresthesia in the right mandibular region, and a burning sensation once opening of the mouth (Figure 2).

An orthopantomograph was requested, in which we observed a radiolucent lesion, 30 mm in diameter, unicellular, with well-defined edges, located in the right mandibular ramus that respected the anterior and posterior edge. These radiographic characteristics were confirmed when performing maxillofacial CT scan, finding that the cortical cells were expanded, without perforation. An incisional biopsy was performed and the histopathological diagnosis of multicystic ameloblastoma with a follicular pattern was made (Figure 3).

Based on the extension and recurrence of the lesion, it was decided that a segmental resection with safety margins of 1.5 cm (respecting the integrity of the molars adjacent to the lesion) and reconstruction with custom-made total joint replacement would be performed. This was planned in conjunction with the Ortho Baltic Medical equipment manufacturer (Kaunas, Lithuania) using computer-aided design/computer-aided manufacturing technology in keeping with TMJ CONCEPTS (Figure 4). While waiting for the prosthesis, the patient presented a pathological fracture of the anterior edge of the right mandibular branch. Since occlusion was not unstable, and movement of the segments was conserved, it was decided not to treat the fracture. On 06/21/2019, surgery was performed under balanced general anesthesia and nasotracheal intubation, and with prior placement of Erich’s bar arches, using a modified Blair approach with submandibular extension.

The resection of the jaw and the articular fossa were performed with the use of cutting guides, followed by mandibular disarticulation. Afterward, the adjustment of the prosthesis is guaranteed by using the replicas of the mandibular components and the glenoid cavity, which were then removed. Dentomaxillary fixation is performed with 18 gauge steel wire. After extensive antisepsis with chlorhexidine and after a change of gloves, the final prosthesis of the glenoid cavity is placed and secured with system 2.0 osteosynthesis material with bicortical screws, followed by the placement of the mandibular prosthesis secured with bicortical screws of system 2.3 (Figure 5), the surgical approach was closed with anatomical planes. The surgery was completed without complications, meeting the criteria for success of total joint replacement of WB Saunders (1991).

With a surgical time of 340 minutes, the patient did not require intensive care in the immediate postoperative period, the patient recovered in a surgical ward and was adequately tolerating oral feeding without the use of maxillomandibular fixation. The patient was discharged after 72 hours, after postoperative orthopantomography and maxillofacial CT scan (Figure 6).

Currently, he is in a 14-month follow-up period, with an oral opening of 40 mm, stable occlusion and preserved facial expression, absence of pain, normal mandibular function in relation to the preoperative function, without dietary restriction and an adequate quality of life. The patient reports symptomatology consistent with Frey’s syndrome, which has been treated successfully with Botulinum toxin (Figure 7).

**Ethical considerations**

The patient signed a free and informed consent form for the use of his image and his procedure for publishing a clinical case report in scientific journal. Approval from an Ethical Committee was not necessary for the nature of the study. This disclosure is justified by the clinical and research relevance resulting from the presentation of data from careful observation of a single case.
Discussion

Reconstruction of the temporomandibular joint may be required in cases in which the pathology to be treated has produced or will produce, after resection, complex mandibular defects involving the articular region. In this situation, the reconstruction options are limited to autogenous grafts and alloplastic joint replacements. The first has 2 variations: free costochondral graft and microvascularized fibular graft. Both can work adequately to cover in defects of the body and mandibular ramus with the possibility of simultaneous placement of osseointegrated dental implants. However, they have certain limitations and negative aspects such as increased surgical and recovery time, complexity of treatment, comorbidity of the donor site and restriction on the three-dimensional conformation of the graft. In addition, subjective problems such as limitations on diet and asymmetric facial appearance. These aspects are related to a higher rate of complications and the need for a reintervention, especially when compared with reconstruction with alloplastic material.

Total temporomandibular joint replacement (TMJ TJR) was devised as a treatment option in cases of advanced joint degeneration, in which the objectives are restoration of the function and shape of the joint. As a secondary objective, pain reduction is paramount. There are two types of TMJ TJRs: stock (adjusts the prosthesis to the implantation site) and custom (manufacture is personalized in order to find an ideal fit for each case). Customized TMJ TJRs present better subjective and objective results in relation to stock TMJ TJRs, obtaining an improvement in quality of life in up to 85% of cases. The success criteria of Petty W. for total joint replacement coupled to the temporomandibular joint (Table 1), provide a rationale for the manufacture and use of these prostheses.

Predictable results, immediate return to functionality and the elimination of the need for a donor site are benefits that have extended the use this method to bone ankylosis, congenital disorders, condylar fractures, failure of autogenous grafts, and joint reconstruction due to
extensive tumor resections, among others. However, the economic burden and sensitive technique have limited the number of cases that can be treated with this type of reconstruction.17-20

The reconstruction of the TMJ due to tumor resections represent a major challenge. Multiple variables have to be consider: resection with safety margins, condyle position, stability in the occlusion, dental implant placement, and
others. Virtual surgical planning enables the surgeon to make clear diagnosis and treatment planning, optimize osteotomy planes and deal with multiple surgical scenarios in a safe computerized environment. With the final purpose of design and manufacture surgical guides, including drilling holes and cutting templates, that represent effective devices, with an almost perfect concordance, to transfer the preoperative plan in the operating room during surgery.21

This represent a tendency that alloplastic reconstruction, in the era of 3D printing, may replace autogenous flap reconstruction within the tree of reconstruction choices for full-continuity mandibular defects. This paradigm shift leaves the autologous replacement as a salvage procedure for implant failure.22

In the specific case of post-resection reconstruction of an ameloblastoma with custom-made total joint replacement, our case is part of the few cases reported in the literature until 2020.23

Conclusion
Given the need to treat ameloblastoma with a block resection, to avoid its recurrence, long-term predictable reconstructive options should be considered. Options that allow adequate function and aesthetics with minimal morbidity. Total custom joint replacement is an ideal option in these cases.

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Author Contributions
Ismael Vejar Alba: He was the first surgeon in this case, made the diagnosis of the patient and made the treatment plan of the total joint replacement. Guadalupe Jacinto Arias: She was the second surgeon in this case, made the research of the background and the redaction of the paper. Emmanuel Guerrero Sot: he was one of the third surgeons in this case, made the research of the background and the redaction of the paper. Sara Itzel López Zenteno: she help with the traduction to the English language and with the distribution of references.

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