

Resection of Mandibular Tumor of Odontogenic Myxoma Type, A View on Modern Surgery: Case Report

Diagnóstico y manejo quirúrgico de mixoma odontogénico tempomandibular utilizando CAD/CAM: Reporte de caso

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ABSTRACT

Introduction: Odontogenic myxoma (OM) is a benign, locally invasive neoplasm that affects the maxillomandibular complex and has a high recurrence rate. It accounts for an average of 8.3% of odontogenic tumors. Its clinical course is usually asymptomatic in the early stages; however, it may present with pain, paresthesia, tooth mobility, and limitations in mastication and swallowing in more advanced stages. Diagnosis includes imaging studies such as computed tomography (CT) and magnetic resonance imaging (MRI), which typically reveal multilocular radiolucent lesions described as having a "soap bubble" or "honeycomb" appearance. Due to the asymptomatic nature of the lesion, diagnosis is often delayed.

Case report: We report the case of a 15-year-old male patient with no relevant medical history, presenting with a space-occupying lesion in the right mandibular body and ramus. Imaging studies raised the diagnostic suspicion of a possible odontogenic myxoma. Histopathological examination revealed a low-grade spindle cell tumor with extensive myxoid transformation, focal atypia, and no necrosis. Given the extension of the tumor, age of the patient, and degree of invasion, a wide surgical resection with safety margins was performed, followed by mandibular reconstruction using a custom-designed prosthesis based on three-dimensional imaging and a vascularized free fibula flap. **Discussion:** Extensive mandibular myxomas require a multidisciplinary approach and meticulous, individualized preoperative planning aimed at achieving optimal functional, aesthetic, and oncologic outcomes. **Conclusion:** This case highlights the importance of applying three-dimensional imaging technologies in the reconstructive surgery of complex maxillofacial lesions.

Keywords: Myxoma, imaging, three-dimensional, maxillomandibular neoplasms, case report.

RESUMEN

Introducción: El mixoma odontogénico es una neoplasia benigna, localmente invasiva, que afecta el complejo maxilomandibular y presenta una alta tasa de recurrencia. Representa en promedio el 8,3 % de los tumores odontogénicos. Su curso clínico suele ser asintomático en las etapas tempranas, aunque puede manifestarse con dolor, parestesia, movilidad dental, limitación para masticar y deglutir en etapas avanzadas. El diagnóstico incluye estudios de imagen —tomografía

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axial computarizada y resonancia magnética nuclear—, que evidencian lesiones radiolúcidas multiloculares descritas como “pompas de jabón” o “panal de miel”. Debido a la naturaleza asintomática de la lesión, su diagnóstico suele ser tardío. **Presentación del caso:** Se reporta el caso de un paciente masculino de 15 años, sin antecedentes médicos relevantes, con lesión ocupante de espacio en cuerpo y rama mandibular derecha. Los estudios imagenológicos sugirieron la sospecha diagnóstica de posible mixoma odontogénico, y el estudio histopatológico reveló lesión tumoral fusocelular de bajo grado con extensa transformación mixoide y con atipia focal, sin necrosis. Dada la extensión tumoral, la edad del paciente y el grado de invasión, se realizó resección quirúrgica amplia con márgenes de seguridad y reconstrucción mandibular utilizando prótesis diseñada por imagenología tridimensional y colgajo libre de peroné vascularizado. **Discusión:** Los mixomas mandibulares extensos requieren un abordaje multidisciplinario y planificación prequirúrgica meticulosa individualizada, para lograr adecuados resultados funcionales, estéticos y oncológicos. **Conclusiones:** Este caso resalta la importancia de aplicar tecnologías de imagenología tridimensional en la cirugía reconstructiva de lesiones maxilofaciales complejas.

Palabras Clave: mixoma, imagenología tridimensional, neoplasias maxilomandibulares, reporte de caso.

1. Introduction

Odontogenic myxoma (OM) is a benign locally invasive maxillomandibular neoplasm with a high recurrence rate. It primarily affects women in the third and fourth decades of life, although pediatric cases have also been reported. In most cases, it is located in the posterior mandibular region, followed by the incisor area, maxilla, and, in rare cases, the mandibular condyle [1,2].

OM accounts for approximately 2.2% to 17% of odontogenic tumors, with an average frequency of 8.3%. It is the third most common odontogenic tumor, following ameloblastoma and odontoma [3,4]. Its rarity and invasive behavior encourage further research to better understand its biological characteristics and improve clinical management.

Initially asymptomatic, OM can present symptoms such as pain, paresthesia, tooth mobility, and ulceration as it progresses, which can lead to limitations in mastication and swallowing. These manifestations are often associated with bone perforation and soft tissue invasion [5,6].

Diagnosis relies on imaging techniques such as computed tomography (CT) and magnetic resonance imaging (MRI), which typically reveal multilocular radiolucent lesions described as having a “soap bubble” or “honeycomb” appearance. Less frequently, unilocular images with well-defined or poorly defined borders may be observed. OM may also be associated with an unerupted tooth and, upon cortical bone perforation, may present a peripheral “sunray” appearance—characteristic of bone sarcomas—thus hindering differential diagnosis [3,7,8,9].

Diagnosis is confirmed through histopathological examination. The tumor, of mesenchymal origin, is composed of round, angular, spindle-shaped, and stellate cells in a loose myxoid stroma with scarce collagen fibers. Mesenchymal markers such as vimentin, muscle-specific actin, and, to a lesser extent, the S-100 protein are generally positive. In isolated cases, positivity for CK19, a marker of normal odontogenic epithelium, may be observed [10].

Treatment consists of total surgical resection; however, the surgical approach remains controversial. Some authors suggest enucleation and curettage due to the benign nature of the tumor, while others recommend radical surgery with primary reconstruction because of its high recurrence rate. In this case, managed at Clínica Bonnadona, radical surgery with primary reconstruction supported by CAD/CAM (Computer-Aided Design/Computer-Aided Manufacturing) technology was conducted to ensure comprehensive treatment [11,12].

2. Case Presentation

This case report was prepared in accordance with the SCARE guidelines.

A 15-year-old male patient, with no significant medical, familial, or genetic history, high school graduate, of mixed race, was admitted to our institution presenting a space-occupying lesion located in the right mandibular body and ramus. The patient was hemodynamically stable with no other significant findings on physical examination.

The maxillofacial surgery service requested a biopsy and histopathological identification of the lesion. The report described a low-grade primary spindle cell tumor with extensive odontogenic myxoid changes, focal atypia, and no necrosis. Immunohistochemistry showed focal S100 protein reactivity in tumor cells.

Based on these findings, it was decided to perform a hemimandibulectomy + condylectomy + replacement of the right temporomandibular joint + microvascularized flap + virtual planning of a custom-made prosthesis for the temporomandibular joint, right mandibular body and ramus, and mandibular angle, followed by the placement of a three-dimensional reconstructive plate.

A computed tomography (CT) scan of the paranasal sinuses (PNS) and face in three-dimensional projection was performed, which revealed a large expansile osseous mass in the right mandible, with extension into the soft tissues of the ipsilateral hemiface, apparent involvement of the masseter muscle and the medial pterygoid muscle, and infiltration of the ipsilateral maxillary sinus (Figure 1A).

The tumor margins were defined and projections were made for reconstruction with a vascularized fibula graft and the design of a prosthesis with an occlusal splint to ensure proper mandibular positioning (Figure 1B).

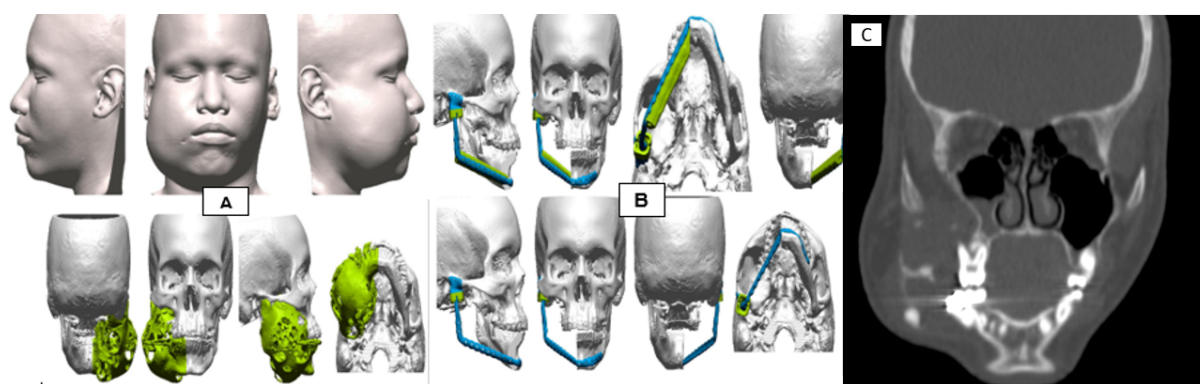


Figure 1. A. Three-dimensional imaging reconstruction of the tumor. **B.** Implant design. **C.** Simple coronal slice of paranasal sinuses (PNS).

Source: Clínica Bonnadona Prevenir, patient case report.

The surgical procedure was performed using a multidisciplinary approach by a highly specialized team composed of an oral and maxillofacial surgeon, a head and neck surgeon, a vascular surgeon, a pediatric surgeon, and a plastic surgeon. All members of the team had more than 15 years of experience in the management of oncologic patients, which allowed for efficient and precise coordination within a single surgical session.

Tracheotomy and gastrostomy were performed under general anesthesia as measures to secure the airway and ensure enteral nutrition. Subsequently, resection of the maxillomandibular neoplasm was carried out through a right submandibular and preauricular approach, extending to the submandibular gland. The ascending ramus of the mandible was approached, and subperiosteal dissection was performed, followed by in bloc resection of the lesion involving the symphysis, body, ramus, and mandibular condyle, ensuring safety margins as per the virtual surgical plan (Figures 2 and 3). Satellite lymph nodes were also resected and sent for pathological examination.

For the reconstruction, a microvascularized osteomyocutaneous flap was positioned and fixed with a mandibular plate on the left mandible and aligned using an occlusal splint. A vascularized fibula graft was harvested (Figure 4A) and transferred as a microvascularized flap to the hemimandibulectomy defect, allowing for the obliteration of the dead space and improved vascularization through an end-to-end microvascular anastomosis between the facial artery and the superior thyroid vein. After confirming adequate perfusion, the surgical wound was closed in layers (Figure 4B), and the procedure was completed without complications. The patient's family was informed that the procedure was well-tolerated with no adverse events, and the patient would be transferred to the pediatric intensive care unit (PICU) as part of the established protocol for this type of surgery.

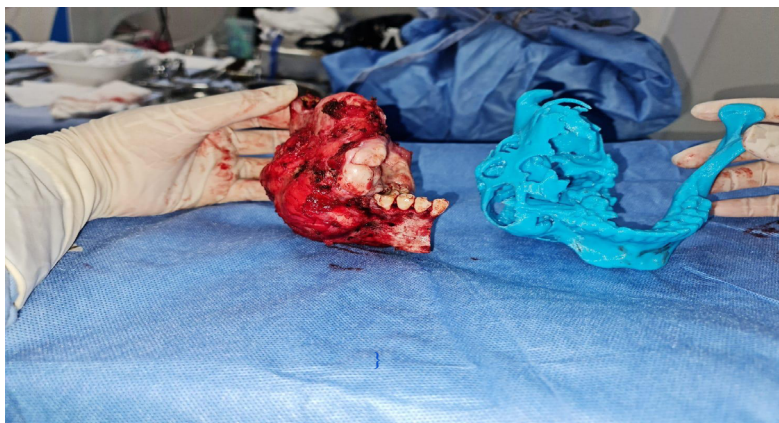


Figure 2. Comparison between the resected tumor and three-dimensional imaging.

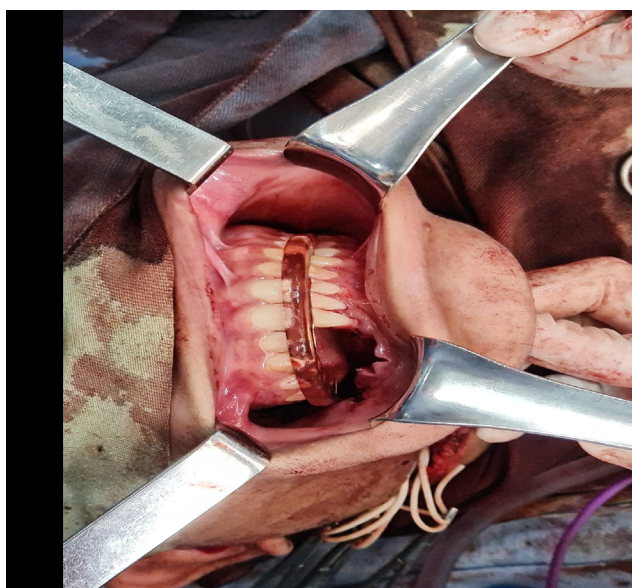


Figure 3. Hemimandibulectomy

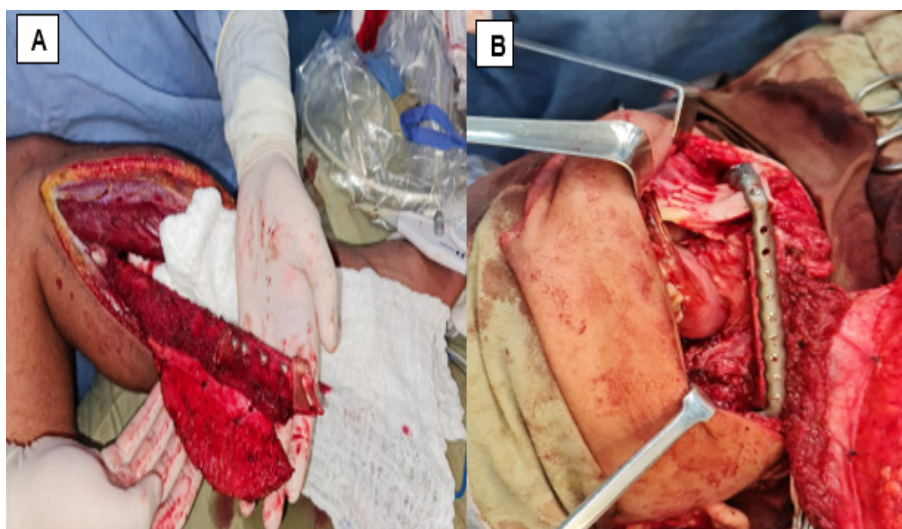


Figure 4 A. Vascularized fibula graft **B.** Fixation of the graft to the mandible.
Source: Clínica Bonnadona Prevenir, patient case report.

During his hospital stay, the patient developed a collection in the right side of the neck, accompanied by febrile spikes and a positive culture for Gram-negative bacteria, which required broad-spectrum antibiotic therapy and surgical reintervention for lavage, debridement, and drainage.

Following clinical improvement, the patient was transferred to the general ward under multidisciplinary management. After an adequate recovery, he was discharged to continue his outpatient rehabilitation.

The outpatient follow-up performed four months post-surgery showed a favorable evolution, with no signs of infection or recurrence (Figures 5B and 5C).

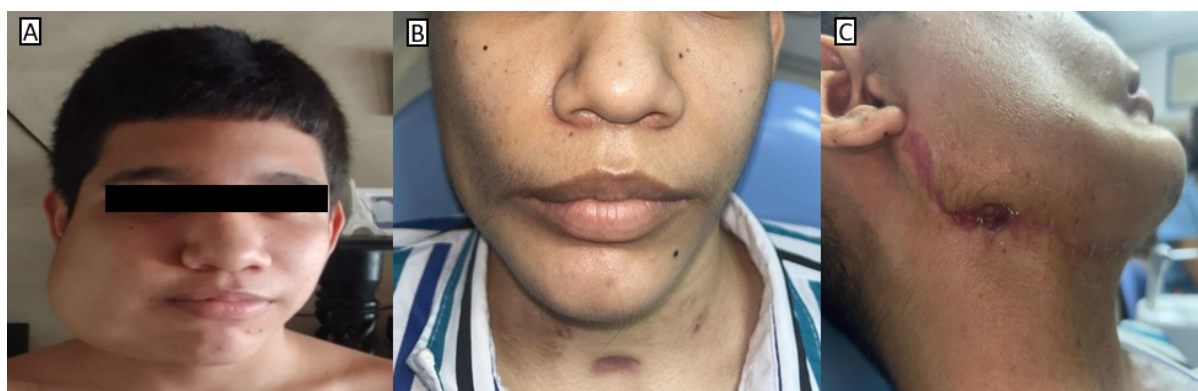


Figure 5 A. Preoperative view **B.** Postoperative result, frontal view **C.** Postoperative result, lateral view
Source: Clínica Bonnadona Prevenir, patient case report.

3. Discussion

Odontogenic myxoma (OM) is a benign, locally invasive odontogenic tumor with a high rate of local recurrence, although it is generally not associated with malignant transformation or metastasis [13]. It is a rare lesion and is scarcely documented in the literature [14]. These maxillomandibular neoplasms are considered radioresistant. Suárez Condez described a case in which radiotherapy was indicated prior to surgical resection, but the poor clinical progression of the disease resulted in the patient's death [15].

The participation of an experienced pathologist is essential because diagnosis can guide the treatment approach, whether surgical or conservative. Additionally, follow-up with imaging studies is crucial to detect early recurrences. In cases of extensive resections, reconstructive procedures should be postponed until a disease-free period is confirmed [13].

Surgical options include conservative management, such as enucleation and curettage—especially in pediatric patients—or non-conservative approaches. Tapia Contreras and colleagues emphasize the importance of considering factors such as age, growth, function, and craniofacial aesthetics when selecting the appropriate treatment [11,12].

Conservative management is associated with a recurrence rate of 19%, compared to 6% in those treated with resection [14]. In radical treatment, vascular grafts or microvascularized flaps using fibula or iliac crest are employed to maintain both aesthetics and functionality [11].

In the case presented here, considering the tumor size, the patient's age, and the degree of invasion, a non-conservative approach was chosen and a complete resection with appropriate safety margins was carried out. Reconstruction was achieved using a custom-designed prosthesis based on three-dimensional imaging and a vascularized fibula graft. The radical treatment resulted in satisfactory aesthetic and functional outcomes, with no evidence of tumor recurrence up to four months postoperatively.

4. Conclusion

Odontogenic myxoma, despite its benign nature, presents a therapeutic challenge due to its infiltrative potential and tendency for recurrence. In this case, the maxillomandibular neoplasm was located in the right mandibular body and ramus and required extensive surgical resection. Through a multidisciplinary approach and meticulous preoperative planning, complete tumor excision was achieved, along with immediate reconstruction using a custom-made prosthesis designed with three-dimensional imaging and a vascularized fibula graft. This therapeutic strategy not only preserved the patient's masticatory function but also successfully restored facial aesthetics. This case highlights the importance of comprehensive assessment and individualized treatment planning to achieve optimal functional, aesthetic, and safe outcomes for each patient.

5. Declarations

5.1 Ethical Considerations

The case report was approved by the Research Ethics Committee of OCBP, with prior consent from the patient's family.

5.2 Conflict of Interest

The authors declare that there are no conflicts of interest related to this case report. All authors independently participated in the development, analysis, and interpretation of the data, as well as in the article writing.

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5.4 Disclaimer

The authors assume full responsibility for the opinions and conclusions presented in this article.

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