

texto, la maxilectomía subtotal 2B, como tratamiento radical con abordaje Weber-Ferguson, previene la recidiva en al menos dos años postquirúrgicos, lo cual mejora la estética facial del paciente.

Clinical case

Surgical treatment of maxillary odontogenic myxoma. A clinical case report

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ABSTRACT

Odontogenic myxoma is a tumor that usually appears in the oral cavity. It is a locally aggressive, slow-growing tumor occurring especially in the mandible. Radiographically it is observed as unilocular or multilocular radiolucency similar to a honeycomb. This report describes the case of a 26-year-old woman who presented to the Maxillofacial Service of the Puebla City General Hospital with swelling of the left cheek. Imaging showed a delimited lesion affecting the left-side maxillary bone and malar and orbital floor. The surgical treatment consisted of subtotal maxillectomy class 2B, resulting in a favorable long-term prognosis (two years) for the patient, without recurrence.

Keywords: Maxilla, tumor, odontogenic myxoma, maxillectomy.

BACKGROUND

Myxomas are defined as benign, non-metastatic neoplasms. They occur in different body locations including skin, subcutaneous tissue, heart and bone, and are uncommon in head and neck. Two forms of this tumor have been identified: (1) myxoma derived from the bone tissue of the facial massif (previously subdivided into true osteogenic and odontogenic myxoma), and (2) myxoma derived from the soft tissue of the perioral region, parotid glands, ear, and larynx.¹

The odontogenic myxoma (OM) is classified as a benign neoplasm of ectomesenchymal and/or mesenchymal origin, with or without the presence of odontogenic epithelium.² The OM is an intraosseous dental tumor, relatively rare, but locally invasive. It is often asymptomatic and is clinically characterized by a progressive increase in its volume, causing changes

in facial appearance and interfering with occlusion, with the displacement of the tooth and the expansion of the cortical bone. This leads to alterations such as ulcers of the underlying tissues.^{3,4} The radiographic characteristics of odontogenic myxomas are variable, they may appear as small unilocular radiolucent lesions or in more serious cases as large multilocular neoplasms (> 4.0 cm), which often displace teeth or less frequently reabsorb roots. The multilocular trabecular pattern has been described as having a «honeycomb» or «soap-bubble» appearance, with fine trabeculae.^{5,6}

The OM accounts for 3 to 6% of all dental tumors and can occur at any age, mainly from 10 to 50 years, with a maximum incidence in the third decade of life.⁷ Despite its benign character, the OM can be locally aggressive and has a relapse rate of 25%.⁸ Regarding treatment, surgical resection remains the procedure of choice.⁹ However, because of its percentage of relapse, it usually returns if treated conservatively,⁷ so its long-term follow-up is crucial both for the patient and for the specialist. The goal of the present work is to report a case of a OM in the left posterior maxilla, with a follow-up of two years after performance of subtotal maxillectomy class 2B.

CASE REPORT

A 26-year-old woman presented to external consultation of the maxillofacial surgery service at the North Zone General Hospital of Puebla complaining of a swelling in the left posterior maxillary region (*Figure 1 A*). Antibiotic treatment was administered but no improvement was observed. The medical history revealed no data relevant to her condition. On physical examination the patient showed normocephalic skull without sinking or exostosis, unaltered frontal region, asymmetrical orbital region due to slight left-sided proptosis, preserved eye movements and vision, full, symmetrical auricles; no exit of pathological secretions through external auditory canals, straight nose with open nostrils, swelling on left zygomatic region (painless, non-hyperemic, non-hyperthermal, of solid-consistency, with poorly delimited edges, and normal skin color), oral opening of approximately 3.5 cm, mucous membranes of adequate color and hydration, usual and stable occlusion, volume increase in left upper vestibular sulcus, intraorally at molar region level (*Figure 1 B*) (painless at palpation, non-hyperemic, non-hyperthermal, firm, of slightly soft consistency), cylindrical neck with central and movable trachea, without adenomegalies; normal thorax with well-ventilated pulmonary areas, heart sounds of good tone and intensity, soft, depressible abdomen without data of peritoneal irritation; deferred genitalia, and extremities in

good condition, symmetrical with distal capillary refill of three seconds.

An incisional biopsy was performed under local anesthesia and the specimen was sent to histopathological examination. The study revealed an OM, so a surgical resection under general anesthesia was programmed, requesting preoperative and imaging studies (axial, sagittal, and coronal CT cuts and 3D reconstruction (*Figure 1 C*), in which a lesion covering left maxilla, orbital floor, and teeth of left posterior sextant was observed.

The patient entered the operating room in decubitus supine position. Orotracheal intubation, asepsis and antisepsis, placement of surgical fields, and incision marking for Weber-Ferguson approach were carried out (*Figure 2 A*). Infiltration anesthesia with lidocaine and epinephrine was done for hemostatic purposes. Subtotal maxillectomy class 2B was performed (*Figures 2 B and C*); subsequently, a rib bone graft was applied (*Figure 2 D*) and fixed to the infraorbital margin using the 2.0-mm osteosynthesis system and a 0.6-mm titanium mesh (*Figure 2 E*). The surgical wound was closed by planes and the procedure was finished without any incident or accident (*Figure 3 B*). Lastly, the surgical specimen was sent for definitive histopathological study.

After the surgical procedure, the patient's progress was monitored through outpatient consultation. Postoperative imaging showed adequate placement of the osteosynthesis system and mesh on orbital floor (*Figure 3 A*). At two years of maxillectomy, facial symmetry was restored, and proptosis decreased (*Figure 3 C-E*).

The histopathological study confirmed an OM (*Figure 4*). Sections dyed with hematoxylin-eosin showed a destructive benign neoplastic lesion composed of an abundant proliferation of immense spindle-shaped cells with angular contours in a stroma of loose myxoid tissue with thin collagen fiber zones randomly arranged and infiltrating the mature bone of the area, destroying and perforating cortical bone. The bone was observed in abundant quantity and also exhibited areas of necrosis. There were also some zones of dense collagen fibers arranged randomly and zones of medium-caliber vessels mixed and arranged in the myxoid stroma. Areas near the bone trabeculae showed dense collagen and bone edge with peripheral osteoblasts. Large tumor areas separated by a pseudo-capsule of fibro-collagen tissue were observed in the periphery.

DISCUSSION

The OM is a benign intraosseous dental neoplasm characterized by being a slow-growing, asymptomatic,

locally invasive and aggressive tumor with a high rate of recurrence.^{2,4} The OM makes up about 3 to 6% of all dental tumors and its usual onset is between 10 and 50 years of age, with a maximum incidence in the third decade of life. Several reports have shown the lesion to be more common in the premolar mandibular area, while others found an equal incidence in both maxilla and mandible.¹⁰ However, its most common location is the posterior mandible, although other locations such as the incisive region, maxilla, and mandibular condyle¹¹ should be considered. The overall prevalence rate is between 0.04 and 3.7%. The marked female predilection is the most common feature reported in several studies.¹² In our case, a female patient with an OM in the left posterior maxillary region is reported, whose surgical treatment involved an intraoral and extraoral approach. Generally, the OM is painless, and the displacement of teeth and paresthesia are rare clinical characteristics; thus, it reaches a considerable size before being detected.^{10,13} Although no symptomatology was found, the patient presented to consultation because of swelling in the left side of her face.

The particular characteristics of the OM make a computed tomography (CT) scan essential for both surgical planning and differential diagnosis with lesions that could contraindicate biopsy.^{8,12} In the present case, a CT scan was performed to know the location and area of bone destruction caused by the tumor, so that we could mark the area of resection and surgical margins, finally confirming the OM with the histopathological study. Histologically, the OM has an encapsulated gelatinous consistency, flexible structure, spindle-shaped and stellate cells submerged in an extracellular mucoid matrix or abundant myxoid stroma with minimal cellularity and variable increase in collagen, as well as nests of odontogenic epithelial tissue. Histochemical studies show mucopolysaccharide acid, hyaluronic acid, and chondroitin sulfate¹⁰ as a fundamental substance. In our case we observed abundant proliferation of immense spindle-shaped cells with angular contours in a stroma of loose myxoid tissue with areas of thin collagen fibers arranged randomly and infiltrating the mature bone of the area, destroying and perforating the cortical bone. However, the histological differential diagnosis of the OM should include mesenchymal neoplasms and mesenchymal non-odontogenic neoplasms with presence of myxoid changes, such as myxoid neurofibroma, myxoid lipoma and chondromyxoid fibroma, ameloblastoma, intraosseous hemangioma, aneurysmal bone cyst, and giant cell central granuloma.^{4,8,10}

In imaging studies, the OM has a variable appearance. It can look as a multilocular or unilocular radiolucent zone or a mixture of radiolucent and radiopaque areas, showing well-defined or poorly defined corticated margins or diffuse edges.^{10,12} In our patient, 3D imaging studies showed a lesion spanning the left maxilla, orbit floor, and posterior teeth sextant. On the other hand, complete surgical enucleation of the OM as a conservative treatment is often difficult because unlike other benign neoplasms this tumor does not have a capsule, so the myxoid tissue infiltrates adjacent bone tissue without evidence of immediate bone destruction. Thus, timely diagnosis and treatment is difficult.^{4,8,10} Additional curettage after enucleation has no impact on recurrence rates compared with enucleation alone, but the performance of an additional peripheral osteotomy after enucleation may decrease the likelihood of recurrence.¹² Most authors recommend extensive removal given the possibility of performing primary microsurgical reconstructions with optimal outcomes from the esthetic and functional point of view.¹⁴ The rib bone graft used in our patient provided a good bone length; however, its diameter is limited, so sometimes the graft sits at different height in relation to the remaining bone tissue.

These tumors are radio resistant and hence radiation therapy is never a treatment option. The recurrence rate is attributed to the tumor's ability to infiltrate the surrounding bone, while incomplete removal of the tumor is responsible for recurrence rather than biological behavior.¹⁵ Currently, there are several supportive techniques available, such as cryotherapy, peripheral osteotomy, and chemical cauterization with Carnoy solution.¹⁶ Lastly, our patient was given the indications to follow after surgical treatment, and favorable results have been observed at two years of clinical and imaging follow-up. No relapse has been observed, proptosis decreased, and facial symmetry was restored. The patient recovered her normal quality of life.

CONCLUSIONS

The OM is a benign intraosseous dental neoplasm. It is characterized by being a slow-growing, asymptomatic, but locally invasive and aggressive tumor with a high rate of recurrence. Because the clinical and radiographic characteristics of the OM are inconclusive, a histopathological study should be done to confirm diagnosis. Also, the size and degree of expansion of the tumor over adjacent tissues should be considered to effectively treat the patient to

obtain a favorable prognosis. In this regard, subtotal maxillectomy (class 2B) as a radical treatment with a Weber-Ferguson approach prevents recurrence in at least two post-surgical years, improving the facial esthetics of the patient.

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