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Case Report

Odontogenic myxoma of the maxilla: A rare case report and review of the literature

Hicham Ngham, Zineb Elkrimi *, Walid Bijou, Youssef Oukessou, Sami Rouadi, Redallah Larbi Abada, Mohamed Roubal, Mohamed Mahtar

ENT Head and Neck Surgery Department, Ibn Rochd University Hospital, Faculty of Medicine and Pharmacy, Hassan II University, Casablanca, Morocco

A R T I C L E I N F O

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A B S T R A C T

Introduction: Odontogenic myxoma (OM) is an uncommon benign odontogenic tumor arising from the jaw bone. The diagnosis poses a challenge because its clinical features overlap with those of other benign and malignant neoplasms. Although surgery is usually the choice treatment, there is still some controversy concerning surgical techniques and proper indications.

Case report: We present the unusual case of an odontogenic myxoma involving the maxilla, diagnosed in a 31 years old patient presenting to our department for facial swelling through computed imaging and pathological analysis. After careful consideration, the patient was treated with conservative surgery, with a satisfying end result.

Discussion: Because of its slow growth, odontogenic myxoma is often asymptomatic. The diagnosis is based on clinical, radiological and histological caracteristics. Complete surgical excision is the treatment of choice, but it can be challenging because of the tumor's indistinct margins.

Conclusion: Though there are still no clear guidelines for the management of OM in the head and neck region, the general consensus is that the surgical excision should be complete, and patients treated in a conservative manner should benefit from regular follow-ups.

1. Introduction

Odontogenic myxomas (OM) account for 3–6% of all odontogenic tumors [1]. According to the latest World Health Organization classification of odontogenic tumors in 2005, odontogenic myxomas are rare, benign which tumors of ectomesenchymal origin with or without odontogenic epithelium [2].

The diagnosis can be made on clinical and radiological data. Though benign and slow-growing, odontogenic myxomas have the potential to be locally aggressive; this often leads these tumors to be confused mainly with malignant lesions, the importance of histological diagnosis [3]. The main challenge in surgical management is trying to balance preservation of vital structures with complete tumor resection [4].

In this article, we describe an unusual case of OM located in the maxilla, and we review the clinical, radiologic, and histologic characteristics of this case.

This work has been reported in line with the SCARE 2020 criteria [5].

2. Case report

A 31-year-old man was referred to our department by his family physician with a 2-month history of an increasing prominence over his left maxilla, and swelling of his palate. He reported that he had a long history of tooth sensitivity to cold since childhood. No other significant medical, surgical, toxic history was found, and there was no similar case in his family. The patient was of north African ethnicity, was married and worked as an accountant in a private enterprise.

Aside from the tumefaction of the left jaw, the patient reported no associated symptoms, such as pain or bleeding or hypoesthesia.

Clinical examination found an enlargement of the alveolar process in the left maxillary molar region, with a swelling of the hard palate on the same side (Fig. 1).

The mucous membrane covering the tumor mass was regular and normal in color and appearance; there were no signs of a fistula or an ulceration. On palpation, the mass was of hard consistency, painless, and seemed attached to the bone, allowing for no mobility.

* Corresponding author. 6, Rue Lahcen Al aarjoune, Casablanca, 20250, Morocco.
E-mail address: zineb.elkrimi@gmail.com (Z. Elkrimi).

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A CT of facial bones was performed, which revealed an expansive soft tissue mass in the left maxilla measuring 28 mm in its biggest axis, with thinning and breaking of the cortical bone all around it (Fig. 2). The lesion involved the ipsilateral maxillary sinus and the hard palate.

Differential diagnosis included malignant as well as benign tumors.

The patient underwent a biopsy of this lesion under local anesthesia. Histological examination found spindle-shaped mesenchymal cells in a loose myxoid matrix, which are pathological signs indicating an odontogenic myxoma (Fig. 3).

The diagnosis and proposed treatment were discussed with the patient, who had no objections to surgery.

A left partial maxillectomy was carried out using a transoral approach aided by an upper vestibular incision to allow adequate exposure of tumor; Per-operative inspection found a highly friable tissular formation (Fig. 4). The excision was thorough and made in fragments, with a satisfying local control at the end of surgery. The surgical procedure was performed under general anesthesia, by a senior head and neck surgeon, who has ample experience in surgery of the maxilla.

The patient was put under oral antibiotics and analgesics after surgery. There was no need to use a feeding tube; however, the patient did need to follow a strict liquid diet for 15 days, to permit correct healing of the incision.

The pathological examination of the resected tumor confirmed the diagnosis of odontogenic myxoma; the quality of the resection margins, however, could not be determined because of the fragmented nature of the resection.

The first follow-up was done at 15 days after surgery. After clinical examination showed no sign of local complications, the patient was allowed to return to his normal diet and activities. A 2 months follow-up found the patient in good health, with no sign of a local recurrence.

At this time, 6 months after surgery, the patient has undergone two more follow-ups at 2-month intervals. There was no sign of recurrence at either examination. We plan to continue with regular follow-ups every 3 months for the next two years at least, before eventually having a check-up every 6 months.

3. Discussion

Odontogenic myxoma (OM) is a rare benign, locally aggressive tumor generally observed in young patients, that occurs in tooth-bearing areas of the maxilla or the mandible [6–8]. A worldwide estimated incidence of OM ranges from 0.5 to 17.7% from all odontogenic tumors of the jaw [7]. In the majority of the cases, these tumors occur in the second, third and fourth decade [9,10]; this concurs with our patient, who was 31 years old. The exact prevalence of OM in children is deemed to be under 10%, but is still considered higher than that of other aggressive tumors [9,11]. The preferred localization of OM seems to be the anterior maxilla, according to Vasconcelos' series of 85 patients [12].

OM is a slow growing tumor; as a result, it is often asymptomatic, discovered fortuitously on a radiograph, or results in a painless facial swelling or deformation, increasing regularly in volume, as was the case for our patient [8]. The maxillary sinus and the hard palate can be infiltrated by the process [13]. Other symptoms include dental mobility, abnormalities in dental development, disturbance of mastication or speech, pain, and paresthesia [3,10].

Different imaging techniques can be used to explore the extension and the origin of OM. Panoramic radiograph is usually performed at first, though it is not the best imaging technique available. The radiographic aspects of OM in the literature vary from small unilocular radiotransparent lesions, to large multilocular lesions, possibly displacing the corresponding teeth [10,14,15]. The specific diagnosis of OM can be suspected in the presence of fine, angular septa, visible on the panoramic radiograph, and realizing an aspect of a "soap bubble", a "honeycomb" or "tennis racket strings" [6,8,16–18].

While Computed Tomography (CT) imaging is non-specific in the diagnosis of OM, it remains of great topographic interest, as it allows the assessment of the extent of the lesion as well the presence of cortical perforation, which is essential in the planning of surgery [10,19–21].

While certain authors find that panoramic radiography and CT imaging are sufficient to evaluate OM [22], other consider Magnetic Resonance Imaging (MRI) more efficient and superior to CT imaging in the positive diagnosis of OM, especially in the evaluation of soft tissue involvement and in the differential diagnosis between OM and ameloblastoma [10,14,23]. OM generally appears as a well-defined, expansive and multilocular mass, lobulated, containing small crevices and septas, characterized by a low signal intensity on T1-weighted imaging, and high signal intensity on T2-weighted imaging, with a variable and homogeneous enhancement after Gadolinium injection [10,23]. However, while this aspect is highly suggestive of OM, it is not pathognomic, and the real objective and certain diagnosis of OM can only be made histologically.

Histologically, the OM is bland in appearance and is composed of loosely arranged, evenly dispersed spindle-shaped, rounded, and stellate cells, with a lightly eosinophilic cytoplasm in a rich mucoid intercellular matrix [24,25]. Although some degree of mild nuclear pleomorphism or hyperchromatism may exist, including an occasional mitosis or binucleate cell, there is no proven correlation between the presence of these particularities and the recurrence of OM [24].

The differential diagnosis includes odontogenic tumors, such as ameloblastoma, odontogenic fibroma, dentigerous cyst, odontogenic keratocyst, and non-odontogenic tumors, such as central giant cell granuloma, ossifying fibroma, haemangioma, fibrosarcoma, chondrosarcoma and osteosarcoma [10,14,19,23,26,27].

To this day, there is no consensus for the management of OM. According to several authors, the recommended treatment is surgery, either by a radical resection (segmental or block resection, hemimandibulectomy), or a conservative approach (enucleation, curettage, and marginal resection), depending on the size of the tumor [6–8,10,22,

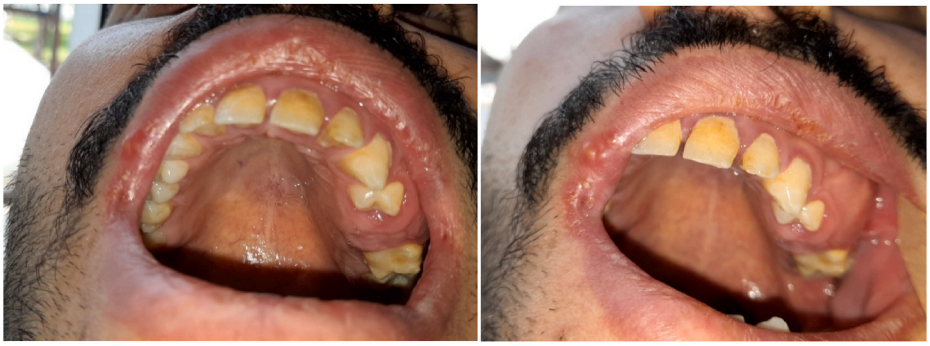

Fig. 1. Image of the enlargement of the alveolar process in the left maxillary associated with a swelling of the hard palate.

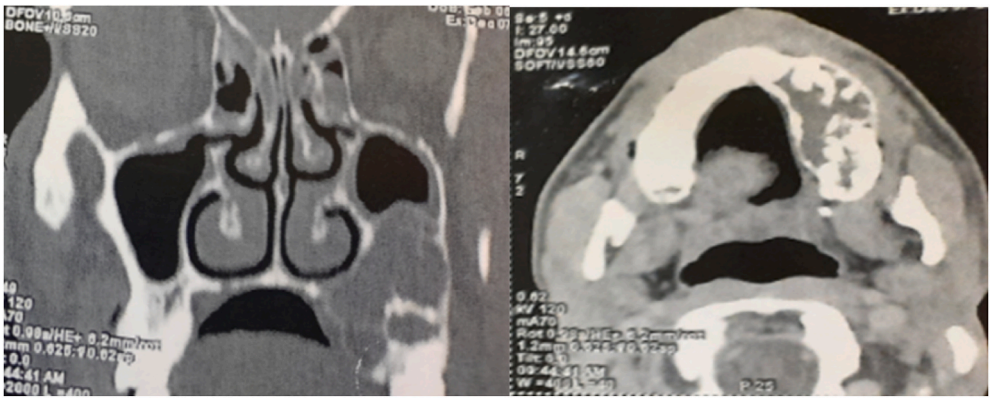

Fig. 2. Pre-operative coronal and axial CT scan shows an expansive soft tissue mass in the left maxilla. The lesion has expanded into the alveolar ridge and maxillary sinus.

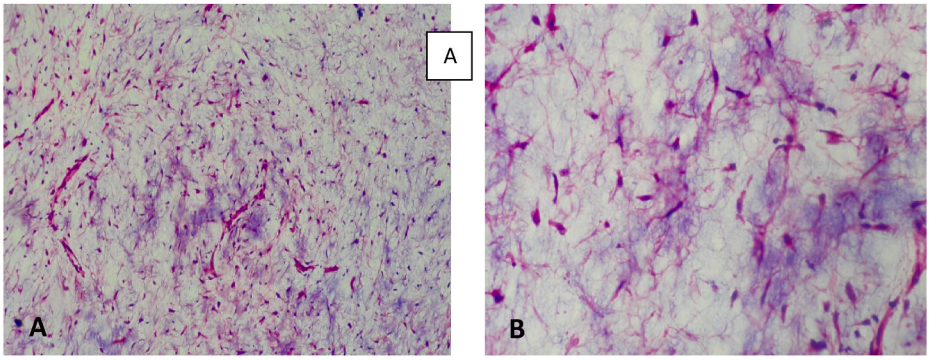

Fig. 3. Microscopic images showing the stellate and spindle-shaped mesenchymal cells in a loose myxoid matrix, with different magnification levels (A: original magnification ×10; B: original magnification x40).

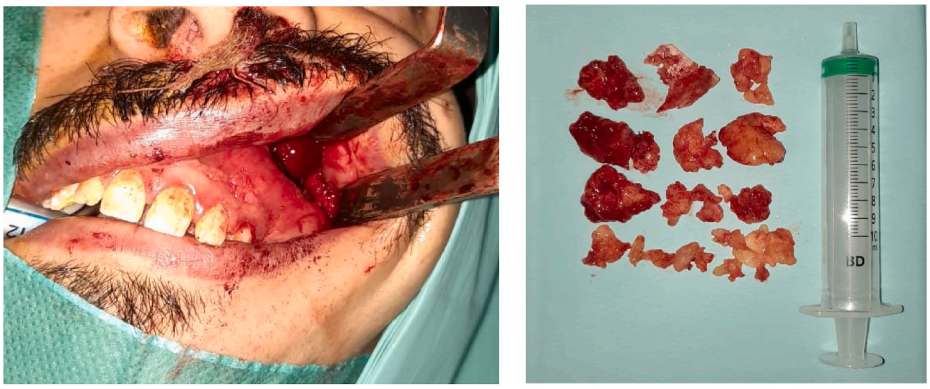

Fig. 4. Intraoperative images of the intraoral surgical approach (left) and the resected tumor fragments (right).

28,29]. Radical surgical resection includes a 1.5–2 cm of healthy bone margin, to reduce the risk of recurrence [7]. Some authors, however, have suggested that a margin of healthy bone tissue might not be necessary to avoid recurrence [30]. Conservative treatment by enucleation and curettage is recommended for tumors with a diameter less than 3 cm, but since myxomas are not encapsulated and tend to infiltrate the surrounding bone, a more extensive resection than curettage and peripheral osteotomy is often required for larger lesions [6,8,10].

Other therapeutic options include radiotherapy and cryotherapy, but these are markedly less effective than surgery [10,31,32].

The recurrence rate of OM seems to be largely related to the treatment method, rather than an inherent behavior of the tumor [8,33]. Although conservative treatments are less invasive and better tolerated by the patient, they present a high risk of recurrence of 10–30%, because complete resection of the myxomatous tissue can be difficult to achieve [8,28]. Maxillary OM is also more likely to recur compared to mandibular OM [34].

A clinical and radiographic surveillance is recommended for at least 5 years after treatment, with a close follow-up during the first two years after surgery, when OM is most likely to reappear [8,10,28,35].

4. Conclusion

Maxillary myxoma is a benign but locally aggressive tumor; positive diagnosis is founded on histologic specimen analysis. Because of its rarity, there are still no clear guidelines as to the management of OM.

Early detection and intervention with careful periodic evaluation may help avoid aggressive treatment methods and their morbidity.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

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