# Surgical treatment of odontogenic myxoma and facial deformity in the same procedure

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# Abstract

Odontogenic myxoma (OM) is an uncommon benign tumor with aggressive and invasive behavior. Predominant symptoms are usually slow and painless swelling, sometimes resulting in perforation of the cortical borders of the affected bone. In this paper, a case report of a patient with an OM on the right maxillary sinus and a vertical excess of maxilla will be presented. The treatment chosen was tumor resection in association with orthognathic surgery with biomodels assessment for surgical planning. A 3-year follow-up showed disease free and stability of the new position of maxilla. The international literature is evaluated to discuss this case report.

Keywords: Facial pathology, odontogenic myxoma, orthognathic surgery

## Introduction

Odontogenic myxoma (OM) of the jaw was first described by Thoma and Goldman in 1947.<sup>[1]</sup> It is a benign, locally invasive and aggressive, non-metastasizing neoplasm of the jaws. The origin of OM is believed to be the mesenchyme of a developing tooth or the periodontal ligament. It is the second most common odontogenic lesion with incidence of approximately 0.07 new cases per million people per year.<sup>[2]</sup> In Asia, Europe and America, OM frequencies between 0.5% and 17.7% of all odontogenic tumors have been reported.<sup>[3]</sup>

Predominant symptoms are usually slow and painless swelling. The tumor occurs more often in the mandible, especially in the molar region. However, some investigators reported an almost equal frequency in the mandibular and maxillary areas. When located in the maxilla, OM often involves the maxillary sinus.<sup>[4]</sup> Despite the benign nature of these lesions, there is a high rate of local recurrence after

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curettage alone and in certain cases it requires a resection of the surgical area.<sup>[5]</sup>

The purpose of this paper is to present a case of OM treated with orthognathic surgery techniques.

#### **Case Report**

A 25-year-old female patient was referred to our department with a chronic headache. Patient's first diagnosis hypothesis was sinusitis and she had previously been referred to an otolaryngologist. Waters' radiograph was performed, which demonstrated a limited radiopacity area into the right maxillary sinus. The patient's medical history was unremarkable. On physical examination, the patient was healthy, without swelling. The mucosa overlying the area of the lesion was the same color and texture as the surrounding mucosa. The sensory examination of the maxillary branch of the trigeminal nerve was normal bilaterally. Furthermore, she had aesthetic concerns about her high smile. Facial and cephalometrical examination revealed an excessively facial height and an excessive incisal and gingival display upon smiling. She was diagnosed with vertical maxillary excess and Class I malocclusion [Figure 1]. Initially, the patient was treated for third molar extraction 7 years ago; in this panoramic radiograph, was observed a discrete radiopacity area in the left maxillary sinus. It was probably an early stage of the lesion.

A biopsy was taken with a Caldwell-Luc approach; histological result show abundant myxoid stroma with collagen fibrils presenting a diagnosis of OM.

The treatment planned was Le Fort I osteotomy with down-fracture to remove the tumor with segmental maxillectomy and 5 mm superior maxillary repositioning for better occlusion and facial esthetic.

Surgery was performed under general anesthesia and nasal endotracheal intubation. Pre-orthognathic surgical models,

radiographs, computed tomography face, photographs and biomodels were obtained before the surgery [Figure 2]. When the maxilla was down fractured, the tumor mass was visualized in the right maxillary sinus with four teeth involved in the lesion, which were removed. After the segmental maxillectomy, maxillary segment was stabilized with plate and screw in the anterior area on the right side and fixation on the anterior and posterior maxillary buttress on the left side. No intermaxillary fixation was used and it was not necessary turbinectomy.

The 3 years follow-up show satisfaction for the occlusal and esthetic result and the patient is disease-free [Figure 3]. A removable dental prosthesis without functional or aesthetic compromise is used [Figure 4].

## Discussion

OM is regarded as a locally invasive tumor that does not metastasize and presents slow and asymptomatic expansion,



Figure 1: Facial analysis showing a maxillary vertical excess with over exposition of gum



**Figure 3:** Three years post-operative computed tomography scan showing no signs of the tumor and good repair without sinus invasion

sometimes resulting in perforation of the cortical borders of the affected bone. Previous studies mention the peak of the incidence in the third decade of life and the majority of cases between 10 and 40 years old.<sup>[2]</sup>

The presence of pain, paresthesia, ulceration and dental mobility has been referred in literature. In the case described, the only complaint of the patient was headache.

Histopathologically, these benign neoplasms were classified by the World Health Organization, 1992, as benign odontogenic neoplasms of ectomesenchymal origin consisting of rounded and angular cells embedded in an abundant myxoid stroma with few collagen fibrils probably originating from either the dental papilla follicle or the periodontal ligament.<sup>[6]</sup>

The recommended treatment of choice for OM is radical surgery or conservative excision depending on tumor size.<sup>[2]</sup>

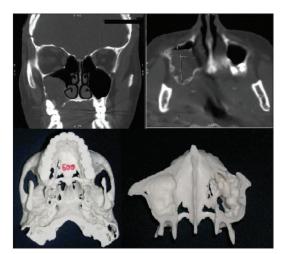


Figure 2: Pre-operative computed tomography scan (coronal and axial image) showing right sinus invasion and biomodels utilized to the planning of surgery; was clear the bone alteration in the right maxillary sinus



Figure 4: Facial image of the patient 3 years after surgery with stability of movement and adequately gum exposition

Since it is a locally aggressive tumor with the potential to cause extensive bone destruction and high recurrence rate with a reported average of 25%, segmental resection of the jaw may be required for large lesions.<sup>[7]</sup> In our case, the lesion was extended along the entire right maxillary sinus and involved some teeth. As the patient has a vertical maxillary excess and Class I malocclusion, the treatment chosen was resection associated with a Le Fort I osteotomy and superior maxillary repositioning.

The temporary mobilization of the upper jaw was first described by Cheever in 1867 for the removal of a nasopharyngeal tumor. In 1927, Wassmund introduced the Le Fort I osteotomy to correct an anterior open bite. Nowadays, we can use this surgical procedure to remove tumors and to correct various deformities of the maxilla simultaneously, with good results.<sup>[8]</sup>

The use of biomodels was an important step in the planning of the surgery [Figure 3]. They have been used by surgeons for patient education, diagnosis and operative planning. In a study to attempt an assessment of biomodel usage in surgery, the authors concluded that biomodels in combination with the standard imaging data have greater utility in the surgical management than the standard imaging data alone.<sup>[9]</sup>

The resection allows only one plate to be placed on the right side. However, good stability was obtained because the superior maxillary repositioning is the most stable movement in orthognathic surgery and in association with that; the chewing forces were diminished because of the absence of teeth.

Patient should be followed closely for at least the first 2 years because this is the time, which the tumor is most likely to recur, although sometimes recurrence may appear much later.<sup>[10]</sup> Hence, we should keep patient in touch for long-period.

In this case report, the patient has been followed-up for 3 years and has remained disease free. Literature recommends a minimum follow-up of 4-5 years to establish disease free status in order to move to the final reconstructive phase.<sup>[5]</sup>

The Le Fort I osteotomy is a versatile surgical technique that allows the treatment of the tumor and at the same time, correction of facial deformities. Good planning has allowed the patient to be referred to only one surgical procedure with satisfactory results to treat pathology, function and esthetic.

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