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CASE

Conservative Treatment of Odontogenic Fibromixoma in maxilla with 11-year follow-up. Case report.

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ABSTRACT

Introduction: Odontogenic mixomas (OMs) are a locally infiltrating slow-growing intraosseous nonmetastasizing tumors of the maxilla and the mandible that have the potential for bone destruction and cortical expansion, showing high recurrence rates. Their frequency varies around the world, accounting for 3-20% of all odontogenic tumors, ranking third among odontogenic tumors. They predominantly affect young adults, but may occur in various age groups. Report: A 37-year-old female patient who in August 2005 sought treatment at the Maxillofacial Dental Unit at Hospital Higueras, Talcahuano, Chile, for a left maxillary bone lesion. An increase in volume was observed in the left maxillary region in the clinical analysis of the oral cavity. The neoplasm was sessile and painless, measuring approximately 3 cm, with a greater diameter in the vestibule, firm on palpation and without signs of gangliopathy. Computed cone beam tomography imaging showed an extensive infiltrating osteolytic lesion in the left maxillary sinus, with no involvement of the orbital bone structure. Analysis of incisional biopsy yielded the diagnosis of Odontogenic Fibromixoma. It was proposed to perform the conservative treatment of the lesion, consisting of enucleation and surgical curettage, obtaining excellent postoperative results and absence of relapse after 11-year follow-up. Conclusion: The present case report provides evidence that supports the conservative surgical approach for the treatment of odontogenic myxomas, which contributes to a better postoperative quality of life for the patient.

Keywords: myxoma, fibromixoma, maxillary, conservative treatment, neoplasia, infiltrating.

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INTRODUCTION

Odontogenic Mixomas *(OMs)* are an intraosseous lesion characterized microscopically by the presence of stellate spindle cells immersed in an abundant myxoid extracellular matrix *(WHO, 2005)*. The macroscopic analysis shows a firm gelatinous mass without a capsule. Although its etiology is unclear, OM is thought to arise from odontogenic ectomesenchyma due to its location in alveolar processes and its microscopic similarity to the developing dental follicle *(Neville et al., 2016)*. If the amount of collagen tissue immersed in its epithelium is large, it may be called Odontogenic Fibromixoma or Mixofibroma *(WHO, 2005; Neville et al., 2016)*.

OMs predominantly affect young adults, but may occur in various age groups, average age ranging from 25 to 30 years, with no sex prevalence. It can be found in almost any region of the maxillae and the mandible, more commonly affecting the posterior regions of the mandible *(Neville et al., 2016; Limdiwala & Shah, 2015)*. It is a locally infiltrating slow-growing intraosseous tumor that has the potential for bone destruction, cortical expansion and recurrence rates of approximately *(Rashid & Bashir, 2015)* 25% without metastasis *(WHO, 2005; Neville et al., 2016; Limdiwala & Shah, 2015; Rashid & Bashir, 2015; Chaudhary et al., 2015; Subramaniam et al., 2016; De Souza et al., 2014; Simon et al., 2004)*. Small lesions are often asymptomatic and described as radiographic findings on routine imaging tests; however, larger lesions are often associated with pain, hyposthesia, displacement or reabsorption of adjacent teeth, expansion, and cortical bone perforation.

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Radiographically OMs are observed as a unilocular or multilocular radiolucent lesions of well defined and corticalized margins: although in some occasions they may be diffuse and poorly defined. OMs may contain fine trabeculae and residual bone inside them. Large lesions show imaging patterns similar to "soap bubbles" or "honeycomb", making a differential diagnosis with multicyst ameloblastoma *(WHO, 2005; Neville et al., 2016; Limdiwala & Shah, 2015; Rashid & Bashir, 2015; Chaudhary et al., 2015; Subramaniam et al., 2016; De Souza et al., 2014; Simon et al., 2004).*

Small OMs are generally treated by enucleation and curettage, but careful periodic follow-up is required for at least 5 years *(Neville et al., 2016; Limdiwala & Shah, 2015; Rashid & Bashir, 2015; Chaudhary et al., 2015; Subramaniam et al., 2016; De Souza et al., 2014; Simon et al., 2004).* In larger lesions, resection with safety margins has been documented as the treatment of choice for recurrences. Recurrence is usually associated with incomplete excision of the lesion *(Neville et al., 2016; Limdiwala & Shah, 2015; Rashid & Bashir, 2015; Subramaniam et al., 2016; Simon et al., 2004).*

The aim of this article is to report the diagnosis, conservative treatment and 11-year follow-up of a case of Odontogenic Fibromixoma.

CASE REPORT

A 37-years-old female patient, who on August 23rd, 2005, sought treatment at the Maxillofacial Dental Unit at Hospital Higueras, Talcahuano, referred from a public medical center in Penco, Chile, for "noticing a swelling in her face a month ago". The patient had a history of maxillary sinusitis in 1992, root canal treatments in teeth 1,1-2,1 performed in 1995, renal lithiasis in 1998, and left ocular lesion in 2002 *(Figure 1).*

Clinical analysis showed an increase of volume in the left maxillary region. The neoplasm was sessile and painless, measuring approximately 3 cm, with a greater diameter in the vestibule, firm on palpation and with no previous inflammatory signs. The patient had complete teeth, which were in good condition.

Computed cone beam tomography imaging revealed an extensive infiltrating osteolytic lesion in the left maxilla, which extended from suborbital foramen through cephalic foramen to interradicular bone of teeth 2.5-2.6 in the caudal direction. Towards medial direction it invaded nasal fossa; lateral border represented by sinus wall. The lesion was aggressive and had destroyed bony walls of the nasal cavity, hard palate, and external cortex of the upper jaw, causing exteriorization and soft tissue involvement with an evident increase of volume in the genial region reaching the corresponding vestibule. The tumor had some areas with bordering calcifications, which seemed characteristic of expansionary growth. The initial diagnosis was of an infiltrating tumor of the upper left jaw *(Figure 1).*

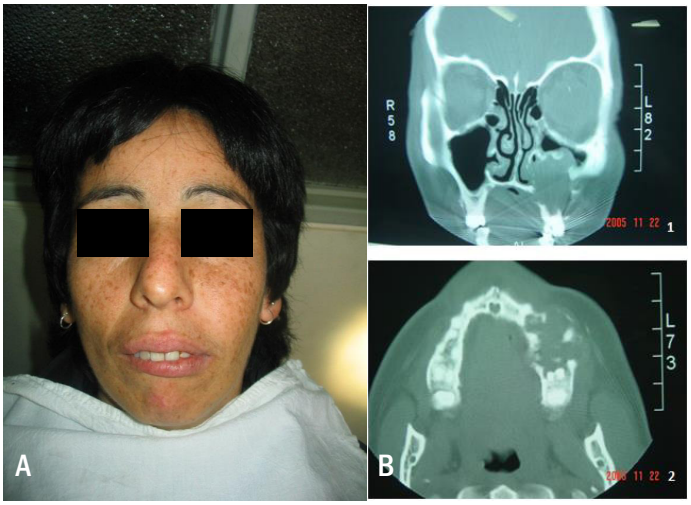

Figure 1: A Initial examination shows facial asymmetry associated with the left genial region, B 1. CT coronal section, infiltrating tumor in the left maxillary sinus with no involvement of left suborbital ridge is observed, 2. CT coronal section, infiltrating lesion with irregular borders, internal cortical involvement and external cortical destruction.

On August 30th , 2005, an incisional biopsy of the lesion was requested. It revealed a fragment of connective tissue with stellate spindle cells with lax, myxoid, and non-atypial stroma, and two small thin trabeculae coated by osteoblasts. It was concluded the lesion was compatible with odontogenic fibromixoma, but also with a myxoid area suggestive of osteofibrous dysplasia *(Figure 2).*

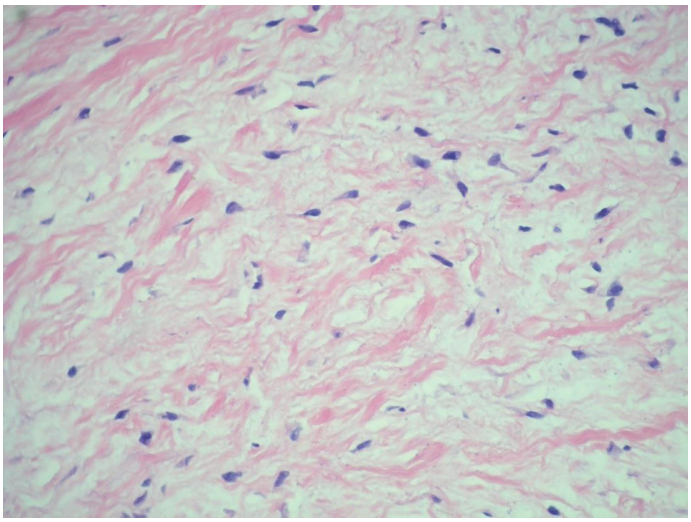

Figure 2: Histopathological sample: stellate cells in myxoid matrix.

Given the histopathological and imaging results, after the completion of root canal treatments in teeth of the left maxillary hemiarch, given the possible damage in the vasculonervioso package *(Figure 3)*, surgical treatment of the lesion was proposed. The Hospital Surgical Committee decided to carry out a conservative treatment of the tumor process, due to the size of the lesion and eventual facial mutilation that would cause radical surgical resection *(maxilectomy)*.

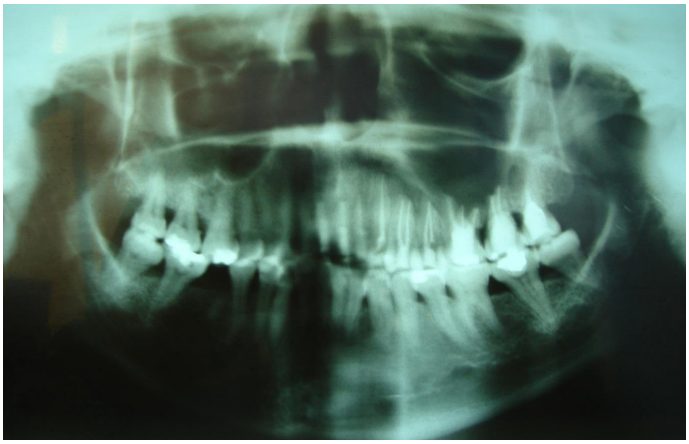

Figure 3: Previous root canal treatment of teeth involved in surgical area.

On November 22nd, 2005, in the surgical ward and under general anesthesia, excision of tumor lesions was performed with Weber-Ferguson flap. The procedure also included mucosal displacement and tumor mass exposure, blunt and shear dissection with moderate difficulty, infraorbital nerve was respected and displaced, tumor enucleation, rapid biopsy, surgical curettage, hemostasis control, saline cleansing solution, Merocel plug *(Medtronic, USA)* in the left nostril, flap replacement, resorbable suture, Vicryl 3.0 orbicular, Dafilon 4.0. *(Figure 4)*. Drug treatment consisted of Sodium Penicillin 2 million IU EV every 6 hours, Gentamicin 160 mg per day, both for 5 days, and Metamizol Sodium 1 g every 8 hours for 3 days. Positive postoperative reviews showed no signs of infection or painful symptoms. Continuous postoperative reviews without signs or symptoms of relapse were conducted until April 14, 2011 *(Figure 5)*.

In May 2017 the patient is referred to annual review requested by the surgical committee. Soft tissues and bone component of the treated area were normal. The patient reported no painful symptoms, no changes in her bite, no loss of function and adequate aesthetics, expressing satisfaction with the proposed treatment. The surgical team proposed to carry out a histopathological study of the treated area by means of an incisional biopsy to confirm the positive result of the treatment and the absence of recurrence that is usually present and documented in this type of benign neoplasms.

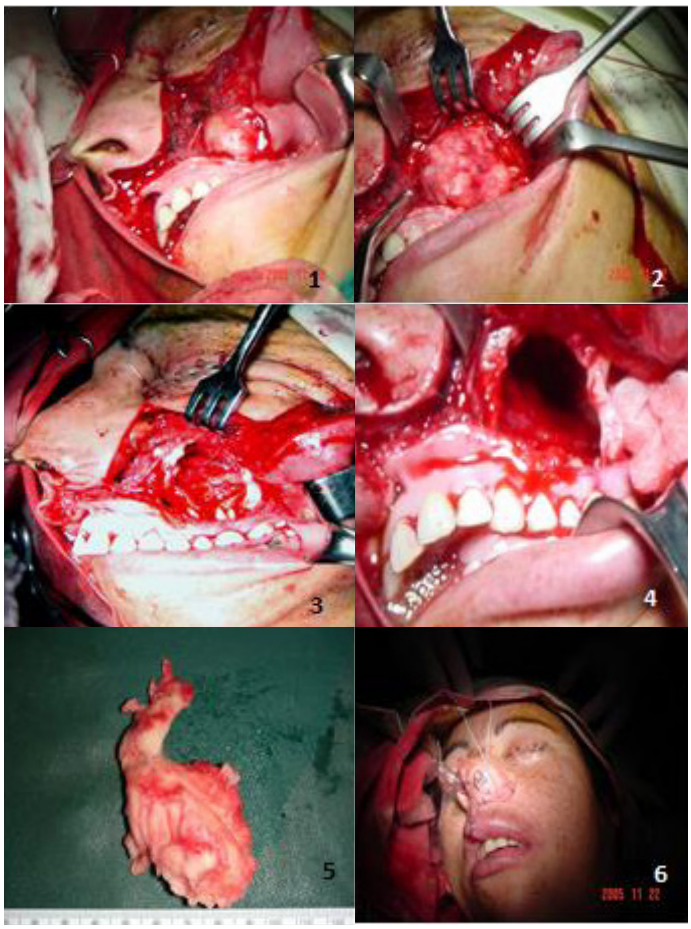

Figure 4: 1. extraoral approach by Weber-Ferguson flap, note external cortical bone involvement and infraorbital nerve 2. lesion with defined borders 3. total lesional excision, note preservation of teeth in the lesion area 4. postoperative surgical area after rotary curettage. Note apicectomy of involved teeth 5. sample of surgical specimen 6. flap replacement

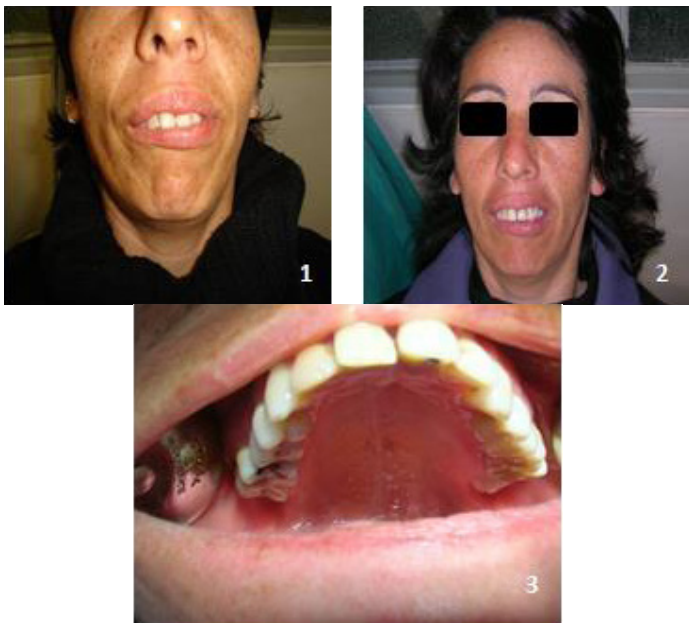

Figure 5: 1-2. Minimal aesthetic involvement caused by surgical flap. 3. rehabilitation status (checkup after 1 year).

On May 31st , 2017, under local anesthesia and in the ambulatory ward at Hospital Las Higueras, a surgical incisional biopsy of the area was performed, which was later sent to the pathology unit yielding the following result: scarring connective tissue within normal appearance *(Figure 6)*. In this way the absence of relapse in 11 years was confirmed histopathologically, corroborating, at the same time, the positive result of the conservative surgical procedure and the behavior of Odontogenic Fibromixoma as a less aggressive and less infiltrating form of neoplasm.

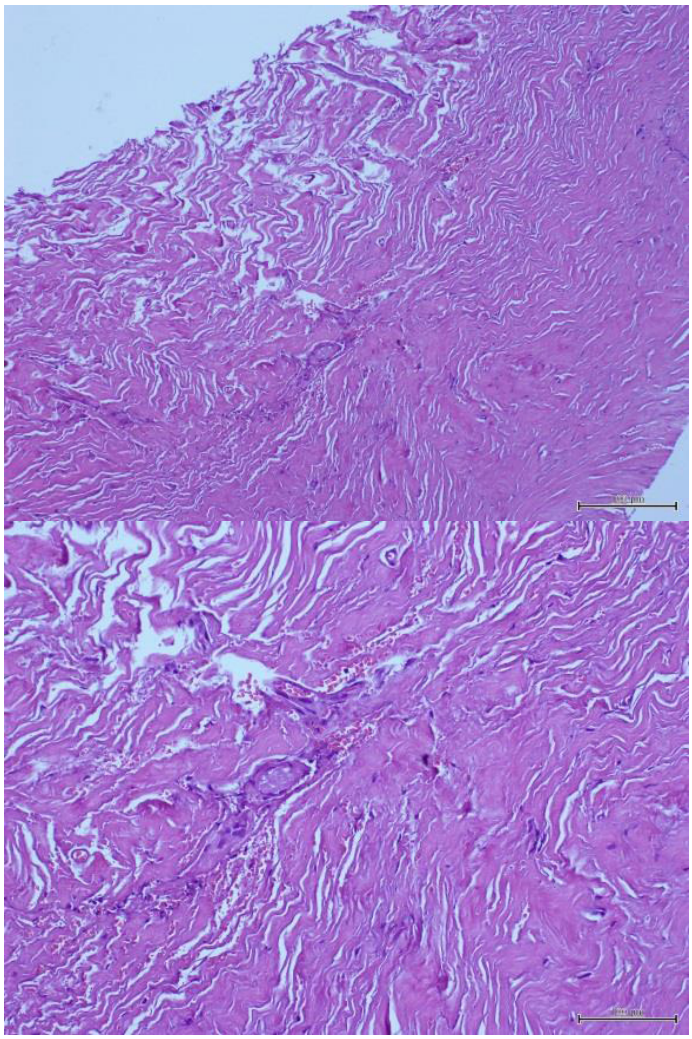

Figure 6: Histological study of incisional biopsy of the surgical area during checkup. Fibroconective scarring tissue is observed with absence of myxoid matrix.

DISCUSSION

OM is an intraosseous neoplasm *(WHO, 2005; Neville et al., 2016; Limdiwala & Shah, 2015; Rashid & Bashir, 2015; Chaudhary et al., 2015; Subramaniam et al., 2016; De Souza et al., 2014; Simon et al., 2004)*, described by Rudolf Virchow in 1863, first mentioned in the literature by Thoma and Glodman in 1947. In 1948 Stout defined its histological diagnostic criteria characterizing it as a true neoplasm, excluding components of other mesenchymal tissues recognizable as chondroblasts, lipoblasts and rhabdomyoblasts *(Rashid & Bashir, 2015)*.

Its frequency varies across the world, ranging from 3-20% of all odontogenic tumors. It is the third most frequent odontogenic tumor *(after odontoma and ameloblastoma) (WHO, 2005)*. It is found predominantly in young adults, but may affect a large age group. Recent studies show a higher incidence in females, reaching a ratio of 1:1.5 to 1: 4 *(WHO, 2005; Limdiwala & Shah, 2015; Rashid & Bashir, 2015; Chaudhary et al., 2015; Simon et al., 2004)*.

Small lesions are often asymptomatic and described as radiographic findings on routine imaging tests. Higher volume lesions are often associated with pain, mobility, reabsorption *(Melo et al., 2008)* and loss of teeth, bone cortical expansion and perforation, and facial deformity *(WHO, 2005; Neville et al., 2016; Limdiwala & Shah, 2015; Rashid & Bashir, 2015; Chaudhary et al., 2015)*.

The present case report is about an odontogenic fibromixoma of the left maxillary sinus of 5.5 cm in diameter, with images showing infiltration in the ipsilateral nasal compartment without involvement of infraorbital ridge. In spite of its size, its histopathological characteristics and recurrent behavior, the surgical medical committee at Hospital Las Higueras Talcahuano opted for a conservative treatment of the lesion to avoid excessive facial damage, as described in the literature when using more radical treatments.

The most interesting aspect of the present case is its treatment. Lesions exceeding 3cm require extensive resection with safety margins *(WHO, 2005; De Souza et al., 2014; Rocha et al., 2009; Kawase-Koga et al., 2014; Melo et al., 2008; Lahey et al., 2013)* of 1-2 cm *(Simon et al., 2004)* and a periodic long-term review, according to literature. However in the present case long-term positive results were obtained having opted for a conservative treatment. This was confirmed by 11-year follow-up and reviews in which the patient had no clinical or imaging signs of relapse, confirming conservative treatment as new treatment alternative for Odontogenic Fibromixomas.

It should be mentioned that the neoplasm described in this case corresponds to the fibrous variant of the Odontogenic Mixoma, which is characterized by the amount of collagen fibers in its epithelium. In the literature, only 24 cases of Odontogenic Fibromixoma have been documented. There is a third type of variant with only three documented cases in the world, which presents intrinsic epithelium *(Lahey et al., 2013)* and received radical treatment. It could be used as an inclusion criterion at the time of deciding the type of treatment.

The results of this case display a new type of behavior and confirm a new treatment to Odontogenic Fibromixomas. This may be due to the greater amount of collagen immersed in its epithelium, which results in a less aggressive and less relapsing form with respect to other types of mixomas; subsequently, a conservative care approach could be used to treat these lesions.

The commonly proposed treatments for these type of lesions are usually radical, regardless of the stage of growth and development of the lesion affecting the patient *(Murphy et al., 2016)*; and although radical treatments greatly affect their quality of life *(Rashid & Bashir, 2015; De Souza et al., 2014)*, they produce a decrease in the rate of relapse and provide better life expectancy at the expense of affecting the patient's aesthetics *(Lahey et al., 2013)*. The conservative treatment of Odontogenic Myxofibroma has shown excellent postoperative results, as has been documented in the literature, with recurrence rates below 10% *(Meleti et al., 2015)*.

Given the choice of conservative treatment, it is important to emphasize that there should be adequate postoperative follow-up. The recurrence of early diagnosed lesions does not imply treatment failure and the lesion can be removed by a relatively simple procedure without resorting to mutilating surgical interventions that make the rehabilitation of the patient more difficult. However, checkups and reviews are crucial to confirm that the lesion has healed, and periodic clinical and radiographic follow-up should be maintained indefinitely *(Rocha et al., 2009; Lahey et al., 2013)*. In this case follow-up has been successful after 11 years.

Conservative treatments offer several advantages over more radical treatments. They are substantially less invasive, can be achieved using an intraoral surgical approach, preserve function and aesthetics, and have a shorter hospitalization time, being more cost-efficient and reducing the economic burden for health systems. However, the risk of recurrence after conservative surgery is greater because ototogenic myxoma is not an encapsulated lesion and its myxomatous tissue infiltrates the surrounding bone tissue without causing immediate destruction. The latter may explain the high rates of recurrence *(10-30%)* after conservative surgical treatment *(Kawase-Koga et al., 2014)*.

CONCLUSION

The present case report provides evidence that supports the conservative surgical approach for the treatment of odontogenic myxomas, which contributes to a better postoperative quality of life for the patient.

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