

# Odontogenic Fibromyxoma of Maxilla: A Rare Case Report

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## ABSTRACT

Odontogenic myxoma is a slow growing, locally invasive, non-metastasizing neoplasm of the jaws. In this article, we present a case of odontogenic myxoma occurring in the left maxilla of a 22 year-old otherwise healthy young adult. This case report presents the rarity of fibromyxoma of maxilla, and the importance of a meticulous enucleation in order to prevent recurrence.

**KEYWORDS:** Enucleation, Maxilla, Odontogenic fibromyxoma

## INTRODUCTION

Odontogenic myxomas are benign neoplasms of the jaws, almost exclusively found in dentulous areas.<sup>1,2</sup> It is a slow growing tumour consisting of an accumulation of mucoid ground substance with little collagen, the amount of which determines if it is a fibromyxoma. Myxomas in represents 2.3% to 17.7% of all odontogenic tumors. Fibromyxomas represent a small number of all myxomas.<sup>3</sup> Odontogenic fibromyxoma represent a rare slow-growing benign neoplasm, which usually occurs in the 2<sup>nd</sup> and 3<sup>rd</sup> decades of life and rarely in children or adults over 50 years of age.<sup>4,5</sup> Main sign is the swelling of the affected

region and the displacement of dentition with pain occurring less frequently.<sup>6</sup> Myxomas can occur anywhere in the jaws but have a predilection for molar and premolar regions of the mandible and maxilla. Cortical expansion and perforation are common. Maxillary myxomas may also extend into the maxillary sinus.<sup>7</sup> In this article, we present a case of odontogenic myxoma occurring in the left maxilla of a 22 year-old otherwise healthy young adult. The aim of this case report is to present the rarity of a fibromyxoma of the maxilla and the importance of a meticulous enucleation in order to prevent recurrence.

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## CASE REPORT

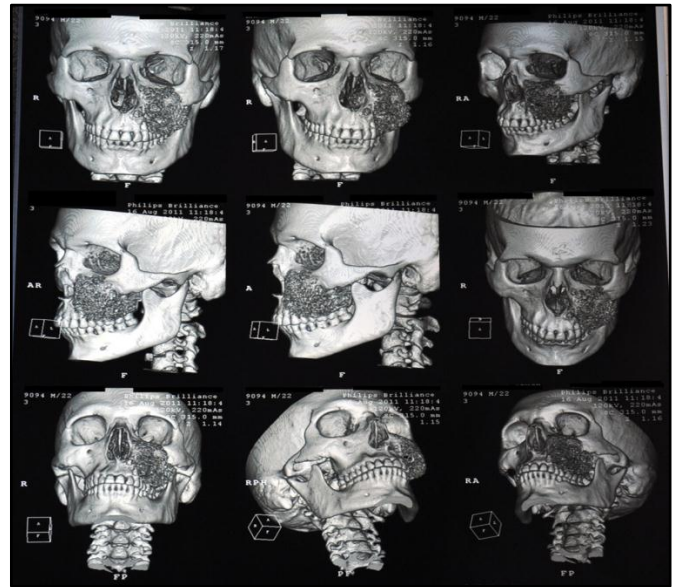
A 22 year old male patient reported to us with the chief complaint of swelling over left side of the face. The swelling had appeared one year back and had shown marked increase in size since the last six months, along with watery discharge from the left nostril and eye since one month. There was no associated pain, tooth mobility or numbness of adjacent areas.

On examination the swelling was diffuse, extending superio-inferiorly from the left orbital rim to the vermilion border of the lip, and medio- laterally from the bridge of the nose to an imaginary line through the outer canthus of the eye. Intraorally the swelling extended mesio-distally from left maxillary central incisor region to left maxillary first molar with complete obliteration of the vesibular sulcus. The overlying skin and mucosa were intact but showed blanching in various places. The swelling was bony hard, non-tender and was not fixed to the overlying skin. There was no associated lymphadenopathy.

The orthopantomogram revealed root displacement of maxillary left premolars and first molar along with apical resorption. Water's view showed complete obliteration of the left maxillary sinus. A CT scan with 3-D reconstruction of the maxilla revealed an expansile cystic mass measuring approx. 55×53×50 mm reaching antero-medially up to the naso-maxillary suture and laterally up to the zygoma (Figure No.1). The left orbital floor and the hard palate were intact.

The lesion was approached surgically through transoral maxillary vestibular incision. The lesion was exposed to its entirety (Figure No.2) and complete enucleation and curettage of the mass was carried out (Figures No.3, No.4) followed by peripheral osteoectomy. Inferior nasal antrostomy was performed and nasal and

antral packing was done with iodoform gauze. The histopathological examination of the specimen revealed randomly placed spindle and stellate shaped cells in an abundant myxoid stroma suggestive of fibromyxoma.



**Figure No.1** 3-D CT scan of the patient



**Figure No.2:** Lesion exposed through trans-oral maxillary vestibular incision

One and a half year postoperative the patient shows no signs of recurrence. The rehabilitation period was uneventful except for some minor degree of sinusitis for first two weeks and he gained complete function and aesthetics soon after surgery.



**Figure No.3:** Lesion being excised



**Figure No.4:** Full lesion excised, size being compared to a standard 5 ml syringe

## DISCUSSION

Odontogenic myxoma is a rare odontogenic tumour, non-encapsulated and non-metastasizing with a potential for local invasion. Though there are reports showing it to be the second commonest odontogenic tumour in many countries, only 0.5 to 17.7% of them have been reported in Asia, Europe, and America.<sup>6</sup> The histologic similarity to stellate reticulum of a developing tooth, its association with a missing or unerupted tooth. Odontogenic

epithelium may be present in a minority of cases and the fact that it rarely appears in other parts of skeleton often supports its odontogenic origin. In a recent immunohistochemical and ultrastructural study, Moshiri et al. Supports the odontogenic origin of myxomas by suggesting that fibroblasts that compose the tooth germ undergo modification to give rise to odontogenic myxoma.<sup>8</sup>

According to Dutz and Stout, the term myxoma was first used by Virchow in 1863, but the term fibromyxoma was described by Marcove et al. in 1964 who reported extragnathic locations of fibromyxoma.<sup>9,10</sup> Fibromyxoma is classified as a specific type of myxoma with a higher fibrous / myxoid tissue ratio than myxoma. In a recent study by Dietrich et al, the maxilla was reported to be a rare location of a fibromyxoma with only 30 cases reported till date.<sup>11</sup>

The lesion can be diffused or well defined, unilocular or multilocular. It is characterized by a mucous or gelatinous grayish-white tissue that replaces the spongy bone and displaces the cortical plates of the jaws.<sup>1</sup> Most odontogenic myxomas are first noticed as a result of a slowly increasing swelling or asymmetry of the affected jaw. Lesions are mostly painless. Ulceration of the overlying oral mucosa only occurs when the tumor interferes with dental occlusion. When the maxillary sinus is involved, the lesion often fills up the entire maxillary antrum. In severe cases, nasal obstruction or exophthalmus may be present.<sup>12</sup> Root displacement and resorption may be present.<sup>1</sup> Although exophthalmus was not noticed in the present case but some nasal obstruction in left nostril as well as displacement of teeth and resorption of root of teeth were evident.

Radiographically, it is a bone destroying lesion and has ill-defined borders. Conventional

radiographs present varying radiographic appearances, which are divided into six types: Type I- unilocular; Type II- multilocular (honeycomb, soap bubble and/or tennis racquet patterns); Type III- involvement of local alveolar bone; Type IV- involvement of the maxillary sinus; Type V- osteolytic destruction and Type VI- a mix osteolytic destruction and osteogenesis.<sup>13</sup> Radiographic examination in our case depicted a multilocular expansile cystic mass with complete obliteration of the left maxillary sinus along with tooth displacement and root resorption of maxillary left premolars and first molar.

Histopathological characteristics of the myxoma/fibromyxoma are the hypocellularity; presence of stellate, spindle-shaped cells in a loose myxoid extracellular matrix with cells presenting with thin, long cytoplasmic projections, that give to the tissue characteristics of immature mesenchyme.<sup>14</sup> In case of fibromyxoma, the amount of collagen in the mucoid stroma is more prominent.

Pathological conditions that should be included in differential diagnosis are ameloblastoma, central haemangioma, fibrous dysplasia, odontogenic cysts, aneurysmal cysts, central giant cell granuloma, metastatic neoplasms etc.<sup>11</sup>

## CONCLUSION

While generally considered a slow-growing neoplasm, odontogenic myxomas can also be infiltrative and aggressive, with high recurrence rates. Treatment of odontogenic myxomas vary from local excision, curettage and/or enucleation to radical resection. The recurrence can be avoided by careful complete resection of the lesion. The patient should be monitored for at least two years post operatively due to the higher rate of recurrence during this period.<sup>11</sup>

Myxomas/Fibromyxomas show a recurrence rate upto 43%.<sup>1</sup> This is strongly related to the nature of the lesion, presenting without a sheath, thus making the complete removal difficult. It is recommended that complete resection and peripheral osteotomy should be the primary mode of treatment depending on the size and behaviour of the tumor as well as age of the patient.

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