



## Case Report

# Lichen Planus of the Jaw: Odontogenic Fibromyxoma – A Rare Entity in Mandibular Anterior Region

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## Abstract

*Odontogenic fibromyxoma in the jaws is infrequent, slow growing, benign, locally aggressive tumor of ectomesenchymal origin with or without odontogenic epithelium and accounting for only 3%-6% of odontogenic tumours. Due to its variable clinico-radiographic characteristics and propensity for recurrence, correct diagnosis is required for adequate management. We present a rare case of odontogenic fibromyxoma occurring in the mandible of a 58-year-old female patient with a brief review of clinical radiological and histopathological features. We also emphasize the usefulness of Cone Beam Computed Tomography in the diagnosis of such lesions. Along with histopathology, cone-beam computed tomography (CBCT) may prove extremely useful in clarifying the intraosseous extent of the tumor and its effects on surrounding structures.*

**Keywords:** *Odontogenic fibromyxoma, Lichen planus of the jaw, Cone beam computed tomography, Mandible, Myxoma.*

## Introduction

Odontogenicmyxoma (OM) is a rare, non-encapsulated benign but locally invasive odontogenic tumor.<sup>1</sup> Fibromyxoma is classified as a specific type of myxoma with a higher fibrous/myxomatous tissue ratio than myxoma.<sup>2</sup>

OM is an unusual benign tumor comprising 3–6% of all tumors of odontogenic origin.<sup>1,3</sup> It usually occurs during the second and third decades of life.<sup>1,3</sup> Females have predilection over males. The mandible is most commonly affected than maxilla and in the mandibular sites most often affected are molar and ramus region.<sup>3</sup>

The present report describes a rare case of fibromyxoma at unusual location in mandibular anterior region with unique radiographic pattern.

## Case Presentation

A 58 year old female patient came to the Department of oral medicine and radiology, CDSRC for gradually increasing swelling in the anterior part of lower jaw. The patient reported mild intraoral swelling for many years but she ignored it as there were no symptoms. Extraoral examination revealed no facial asymmetry. Intraorally, on inspection, single swelling was

present in the anterior mandible causing buccolingual expansion of approximately 2 cm x 4 cm and extending from 33 to 43 with extrusion of teeth 31 and 32. Overlying mucosa was normal in color. (Figure 1) On palpation, all the inspectory findings were conformed. The lesion was hard in consistency and non-tender.

The panoramic radiograph revealed, a well-defined multilocular lesion in anterior part of mandible with scalloped outline. There were scalloping between the roots. Mesial and distal border of radiolucency was well defined and corticated but superior and inferior margins were ill defined. In mandibular incisor region there were small locules forming reticular pattern and in some part thin straight septa were traversing the radiolucency. There was no displacement or resorption of teeth. (Figure 2)

CBCT revealed a multilocular lesion in mandibular anterior region (in axial and panoramic section), extending from 36 to 45 and from alveolar ridge in 42 to apical regions of all anterior and premolar teeth to lower border of the mandible. (Figure 3) There was superior displacement of 31 and 32 with lingual tilting of the crown. No marked root resorption was seen. Right mandibular canal seemed to be unaffected. Left mandibular canal was compressed and displaced in infero-buccal aspect.

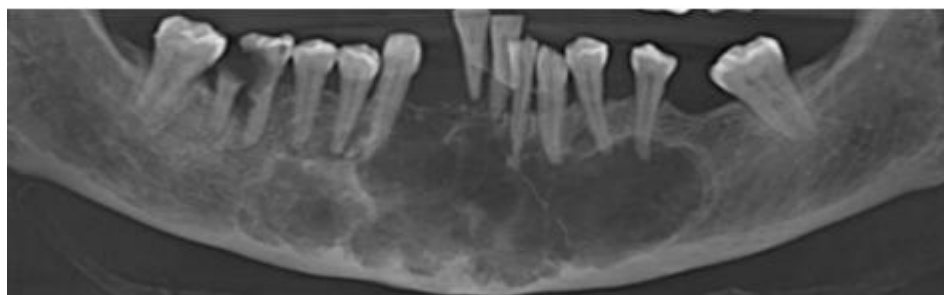
There was expansion of both labial and lingual cortices in mandibular anterior region with pronounce thinning of labial cortex and actual perforation in 31 and 32 region. The lesion showed large locules and straight thin septa throughout the lesion. However, there are small

locules with fine septa at right angles to each other at most superior aspect of the lesion in mandibular incisor region. Considering clinical features, OPG and CBCT findings and specially the pattern of reticular septa on OPG, clinico-radiographic diagnosis of odontogenic myxoma was made.

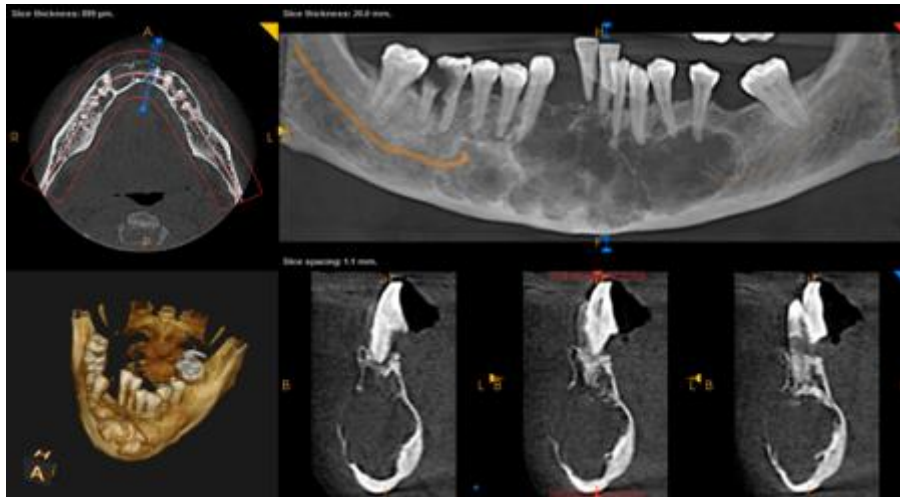
Histopathological examination revealed loose myxoidstroma interspersed with dense collagen bundles. Myxoidstroma contained stellate shape cells which were elongated. Collagen fibre bundles appeared hyalinised intermixed with myxomatous area. Surrounding stroma showed nodular proliferation of loose bland spindle-shaped cells with long, branching cytoplasmic processes. Nests or islands of odontogenic epithelium or residual bone trabeculae were not present. Inflammatory cells were seen. All cells were uninucleate. Granulation tissue and mixed inflammatory infiltrate were present. Considering all histopathological features, final diagnosis of odontogenic fibromyxoma was made. (Figure 4)



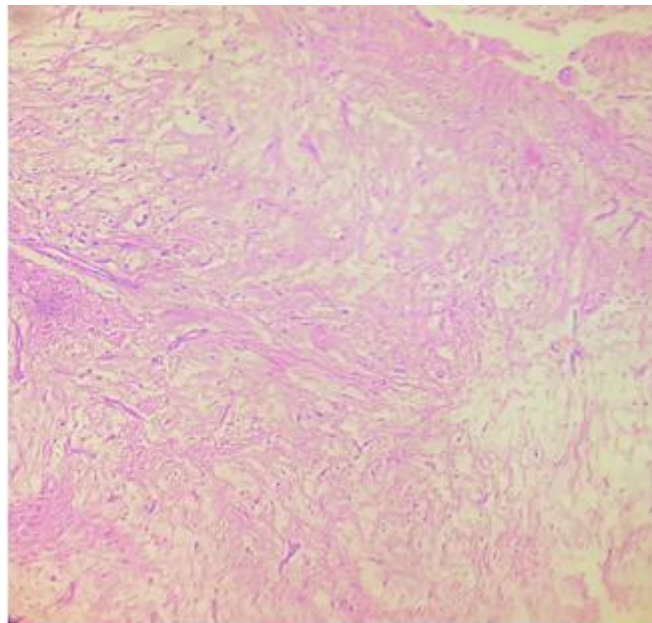
**Figure 1:** A swelling was present in the anterior mandible causing Buccolingual expansion and displacement of #41, #42



**Figure 2:** A multilocular lesion seen with scalloped outline. In incisor region small locules are forming reticular pattern and in some part there were thin straight septa.



**Figure 3:** CBCT revealed a multilocular lesion in mandibular anterior region (in axial and panoramic section), extending from #36 to #45 and from alveolar ridge in #42 to apical regions of all anterior and premolar teeth to lower border of the mandible.



**Figure 4:** H and E staining shows loose myxoidstroma interspersed with dense collagen bundles.

### Discussion

The term 'myxoma' was coined by Virchow in the first edition of *Die krankhaften Geschwulste* in 1863. OM of the jaw was first described by Thoma and Goldman in 1947.<sup>4</sup>

Odontogenicmyxoma (OM) is a rare aggressive intraosseous lesion.<sup>1</sup> Fibromyxoma is a benign lesion of ectomesenchymal origin with or without odontogenic epithelium.<sup>2</sup> Being a locally invasive benign tumor, OM is slow-growing, painless with well-defined pathologic characteristics and generally associated with tooth germ.<sup>1,5,6</sup> OM represents 0.04% to 3.7% of all odontogenic

tumors. In Asia, Europe, and America, relative frequencies between 0.5% and 17.7% have been reported.<sup>3,5,6</sup> OM commonly develops in patients between 10 to 50 years. It is rare in children and adults who are over 50 years of age.<sup>4</sup> According to some authors, female to male ratio is 1.5:1 to 4:1.<sup>7</sup> OM affects mandible more than the maxilla, with mandibular molar regions being the most common affected sites.<sup>3,6</sup> Hence the term "odontogenicmyxoma" (OM) is often applied when the tumor occurs in the jaws to reflect its odontogenic origin.<sup>5</sup>

The present case reported with 58 years old female patient, which is very rare according to literatures, this may be because of the patient had ignored for long. Moreover, the site of the lesion was anterior mandible which was also an unusual site.

Large lesions can cause marked asymmetry of the face. It causes expansion of bony cortices, displacement and loosening of teeth.<sup>1</sup>In the present case, the patient had bony hard swelling with the expansion of lingual and labial cortical plate with the displacement of lower anterior teeth 31 and 32. Halfpenny *et al.* stated that OM in most of the cases presents as an asymptomatic swelling and can grow to considerable dimensions before diagnosis, as was seen in our case.<sup>4</sup>

Radiographically, it is seen as multiple radiolucent areas of varying size, separated by straight or curved bony septa with poorly defined borders. On conventional radiographs, myxomas of the jaws may vary from a unilocular to multilocular radiolucencies which helps in distinguishing this entity from malignant tumors arising centrally within the jaw bones, because the later usually cause massive bone destruction without compartments by bony trabeculations or bony septa. The internal trabecular pattern has been described as “honey- comb”, “soap bubble” or “tennis racquet”.<sup>1,6</sup> Root resorption is rarely seen, and the tumor is often scalloped between the roots.<sup>6</sup> The latter appearance is characterized by angular or straight trabeculations forming square or triangular compartments and has been considered almost pathognomonic of OM. Eversole (1980) said that the internal configuration of the bony septa resembled lichen planus of the jaw bone.<sup>8</sup> In the present case also, panoramic radiograph revealed reticular pattern in mandibular anterior region.

Zhang *et al.*, classified radiographic appearances of OM into six types:

Type I: Unilocular well-defined radiolucency,

Type II (multilocular): Two or more compartments with multiple interlaced osseous

trabeculae described as honey comb, soap bubble or tennis racquet radiolucency

Type III: Lesion located in alveolar bone

Type IV: Lesion involving the maxillary sinus

Type V (moth eaten appearance): Larger radiolucent area with irregular borders

Type VI: Combination of bone destruction and bone formation giving sun ray appearance.<sup>1</sup>

Our case is thus classified as Type II.

Radiographic differential diagnosis must be done mainly with ameloblastoma, ameloblastic fibroma, odontogenic fibroma, central hemangioma or odontogenic keratocyst.

Histopathological characteristics of the myxoma/fibromyxoma are the hypocellularity, the presence of stellate, spindle-shaped cells into a loose myxoid extracellular matrix with cells presenting with thin, long cytoplasmic prolongations that give to the tissue characteristics of immature mesenchyma. Depending upon the pattern of differentiation, the histological nature of the tumor varies. It may be completely myxomatous tissue or varying proportions of myxomatous and fibrous tissue. Some studies show OM as a modified form of fibroma in which myxoid intracellular substance separates the connective tissue.<sup>5</sup> The fibromyxoid lesion may present loci of calcification or ossification and a higher amount of collagen fibres and vessels than a typical myxoma.<sup>9</sup> In the present case histopathological findings revealed that there was bands of collagen fibres but, odontogenic epithelium or residual bone trabeculae were not present.

According to the literature, myxomas in the maxillofacial area may be divided into three types: central, peripheral and soft-tissue myxomas. Different types have different characteristics.<sup>10</sup>

The treatment of the fibromyxoma is surgical and involves enucleation and curettage or block resection. Although small myxomas are generally treated by curettage, larger lesions require extensive resection. The avoidance of recurrence is strongly related to the complete resection of the lesion. The patient should be monitored for at least two years after the surgical intervention due

to the higher rate of recurrence during this period.<sup>5,9</sup>

### Conclusion

The present case and its histopathologic findings highlight a rare location of odontogenicmyxoma and varied radiographic pattern in panoramic radiograph. As odontogenicmyxomas are very rare tumors, they possessed a diagnostic and therapeutic challenge. Hence, correlation of clinical, radiographic and histopathologic findings are essential to diagnose the lesion.

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