

# Odontogenic myxoma

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## ABSTRACT

Odontogenic myxoma is a rare intraosseous neoplasm, which is benign but locally aggressive. It rarely appears in any bone other than the jaws. It is considered to be derived from the mesenchymal portion of the tooth germ. Clinically, it is a slow-growing, expansile, painless, non-metastasizing, central tumor of jaws, chiefly the mandible. Here we report the case of a typical odontogenic myxoma in a 26-year-old female patient, which had acquired large dimensions and involved the entire left half of the mandible including the ramus, resulting in a gross facial deformity, within a span of one and a half years.

**Key words:** Aggressive, mesenchymal, myxoid, myxoma, odontogenic

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## INTRODUCTION

Soft tissue myxomas are frequently found, but their intraosseous counterparts are rarely encountered, and those that are found are almost exclusively confined to the jaws.<sup>[1]</sup> Very few intraosseous myxomas have been found in locations other than the jaws, as reported in the clavicle by Stout *et al.*<sup>[2]</sup> or in ribs,<sup>[3]</sup> or femur,<sup>[4]</sup> but most of the investigators have found that the intraosseous myxomas are most commonly found in jaws and are odontogenic in origin.<sup>[2,5,6]</sup>

The odontogenic myxoma was first described by Thoma and Goldman in 1947.<sup>[1]</sup> Of all the biopsies submitted, odontogenic tumors comprise about 1.3%, while of all the odontogenic tumors, odontogenic myxoma contributes only 3–11%.<sup>[7,8]</sup>

Odontogenic myxoma most frequently occurs in second or third decades of life,<sup>[7,9]</sup> has a slight female predilection,<sup>[7,10]</sup> and involves the mandible more commonly than the maxilla.<sup>[5,9]</sup> Clinically, it is a slow-growing, expansile, painless tumor, which may cause root resorption, tooth mobility, bone expansion, cortical destruction and facial

distortion.<sup>[5,11]</sup> Radiographically, the classic presentation is that of a multilocular radiolucency, with well-developed locules, consisting of fine trabeculae, arranged at right angles, known as the 'Tennis-racquet' or 'Step-ladder' pattern. A 'sun-ray' or 'sun-burst' appearance has also been reported in the literature.<sup>[12,13]</sup> On gross examination, the surgical specimen is characteristically loose, slippery or gelatinous in nature. Histopathologically, the lesion consists of loosely arranged spindle, stellate-shaped or round cells, in an abundant myxoid stroma.<sup>[6,13]</sup>

We report this case in order to put forth a typical case of a rare benign tumor, i.e., odontogenic myxoma in a 26-year-old female, which grew to involve quite a large portion of the mandible, within a short span of one and a half years.

## CASE REPORT

A 26-year-old female patient reported with the chief complaint of a painless gradually progressive swelling in the lower left side of face, which was initially noticed as a small swelling in the lower left vestibule and grew to the present size within a span of one and a half years. There was no history of trauma. Past medical and dental histories were non-contributory.

Extra-oral examination revealed a diffuse, non-tender, bony hard swelling of the left body region of mandible, approx. 3-4 cm in size [Figure 1]. No local rise in temperature or change in color of the overlying skin was seen. No lymphadenopathy was observed.

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On intra-oral examination, a single well-defined localized swelling at left buccal as well as lingual vestibular region, extending from 34 to 37 region, with displacement of 35, 36 and mobility of 34, 35, 36 and 37, was observed. There was no erythema, ulceration or pus discharge in the overlying mucosa [Figure 2].

Radiographically, the Orthopantomograph (OPG) showed a multilocular radiolucency, extending antero-posteriorly from the midline to left ramus region, and supero-inferiorly from the alveolar process to the lower border of the mandible. Poorly defined radiolucent lesion causing slight cortical expansion, with fine trabeculae, some intersecting at right angles, were evident. Displacement of 34, 35, 36, root resorption of 34, 35, 36, 37, 38 as well as scalloping between the roots of the involved teeth, was also apparent [Figure 3].

The lesion was biopsied and submitted to the Department of Oral Pathology with the provisional diagnosis of ameloblastoma or odontogenic keratocyst (OKC).

On macroscopic examination, the biopsy specimen was in multiple pieces, 1–2 cm in size, whitish in color, transparent and slippery in nature and soft in consistency. On microscopic examination, H and E stained section revealed loosely arranged stellate-shaped cells with intermingled fibrillar processes in a homogenous mucoid ground substance with few collagen fibrils and capillaries [Figures 4 and 5]. Very few scattered odontogenic epithelial rests, areas of hemorrhage, and foci of bony trabeculae were visible. Odontogenic myxoma was the definitive diagnosis.

## DISCUSSION

Odontogenic myxoma is a rare benign intraosseous neoplasm, which is called 'locally malignant' on account of its exceptionally high local aggressiveness, high recurrence rate and non-metastasizing nature.

Zimmerman *et al.* reported that the average age for the odontogenic myxoma is 26.5 years, although majority of the investigators found that this lesion occurs in second or third decade of life.<sup>[2]</sup> Most of the reports suggest that there is a slight female preponderance, mandibular predilection, and the lesion has a silent locally destructive nature.<sup>[2,5,8,14]</sup> All these features were evident in our case. Our case, though a benign tumor, was a highly aggressive lesion, involving almost half of the mandible within a short span of one and a half years. Another interesting finding was that it did not cause much of a cortical expansion or facial deformity and appeared to be invading the bone antero-posteriorly, as well as displayed scalloping between the roots of the involved teeth, in much the same manner as an odontogenic keratocyst.



**Figure 1:** Extraoral view showing a diffuse swelling on the left body region resulting in a slight facial asymmetry



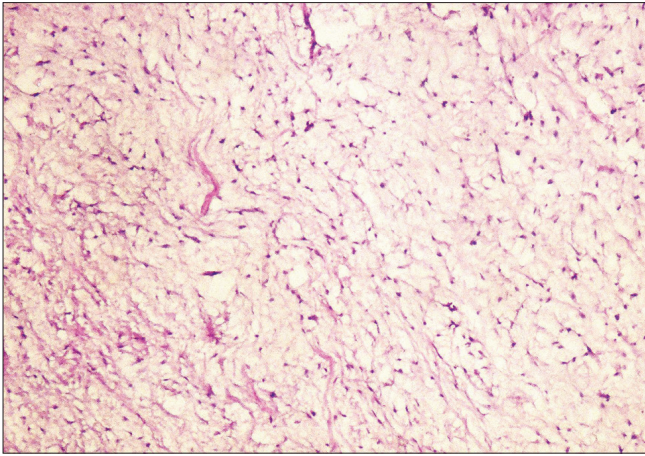
**Figure 2:** Intraoral view showing swelling at left buccal as well as lingual vestibular region, extending from 34 to 37 region, with displacement of 35 and 36



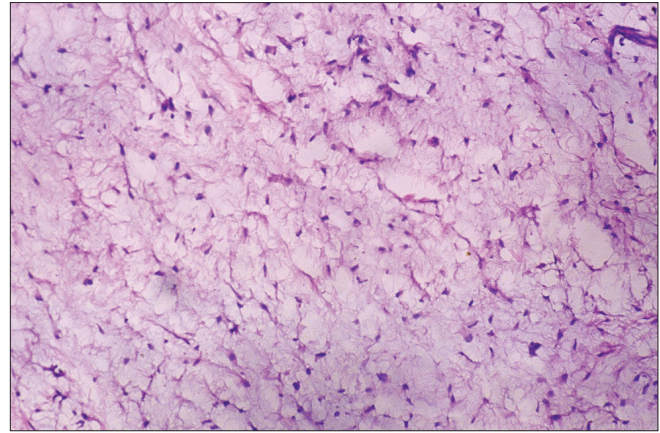
**Figure 3:** OPG showing a poorly defined multilocular radiolucency, extending from the midline to left ramus region causing cortical expansion and displacement of 34, 35 and 36 as well as root resorption of 34, 35, 36, 37 and 38

Radiographically, the appearance of an odontogenic myxoma may vary from a unilocular radiolucency to a multilocular lesion, with a well-defined to diffuse margin. However, a unilocular appearance is more common among children and in anterior parts of the jaws. In tooth-bearing areas, the tumor is often scalloped





**Figure 4:** Low-power photomicrograph illustrating loosely arranged stellate-shaped cells with few collagen fibrils and capillaries (H and E x10)



**Figure 5:** High-power photomicrograph showing loosely arranged stellate-shaped cells with intermingled fibrillar processes in a homogenous mucoid ground substance (H and E x40)

between the roots and root resorption may occur, thus giving a false appearance of an OKC.<sup>[15]</sup> However, the classical radiographic features of the odontogenic myxoma, where the bony trabeculae of a multilocular radiolucency intersect at right angles and the lesion causes root resorption, resulting in tooth mobility,<sup>[2,5,10]</sup> were readily identifiable in our case. The typical soft, slippery and gelatinous nature of the specimen on macroscopic examination<sup>[6,13]</sup> and the histopathological findings were also in accordance with the literature.<sup>[2,5,6,11]</sup> Thus our case was a prototype of an odontogenic myxoma.

However, in case of mixed clinical features, a differential diagnosis must be established with other clinically similar entities like simple cysts, ameloblastoma, OKC, intraosseous hemangioma and giant cell granuloma.<sup>[16]</sup>

The cause and histogenesis of the tumor remain controversial, but the tissue of origin is considered mesenchymal. Two theories of origin have been proposed. Firstly, the tumor results from myxomatous degeneration of fibrous stroma.<sup>[6]</sup> Secondly, it is derived from the mesenchymal portion of the tooth germ i.e., the dental papilla, follicle or periodontal ligament (PDL).<sup>[11,17]</sup>

## CONCLUSION

On the basis of its common site of occurrence (tooth-bearing areas of jaws), usual age of occurrence and gender predilection (second or third decade and female predilection), the striking histological resemblance to dental mesenchyme, and the sporadic presence of odontogenic epithelial islands, the WHO and many authorities consider this neoplasm to be of odontogenic mesenchymal origin.<sup>[1,9,11]</sup> Our case also seems to fall in the same category, as all the criteria are duly fulfilled.

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