Case Report

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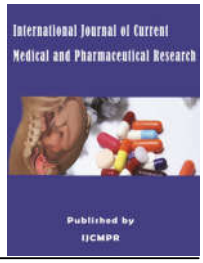

ODONTOGENICMYXOMA: RADIOGRAPHIC FINDINGS KEY TO DIAGNOSIS

PoojaSiwach*., GirishPatil and Arush Thakur

Department of Dentistry, Government Medical College and General Hospital,
Dhule, Maharashtra, India 424001

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ABSTRACT

Odontogenicmyxomas (OM) are locally aggressive, non-encapsulated and non-metastasizing neoplasms that infiltrate bone marrow spaces. The frequency of OM varies in different parts of the world between 3-20% of all odontogenic tumors. Histopathologically, the OM is characterized by loose, abundant mucoidstroma that contains rounded, spindle-shaped, or angular cells. The radiographic features of the OM are variable, ranging from small unilocular lesions to large multilocular neoplasms. Herewith, we present a case of odontogenicmyxoma in left mandibular body region in a 35 year old female with distinct radiographic presentation.

INTRODUCTION

In 1947, Thoma and Goldman first described myxomas of the jaws. Since then the odontogenicmyxoma (OM) has been a subject of continuous scientific debate. Histogenesis, pathogenesis and therapy of this benign non-encapsulated odontogenic neoplasm have been discussed avidly. In the 1992 WHO classification, the term "myxoma" is used along with odontogenicmyxoma and myxofibromaas alternative terms. Odontogenicmyxomas are locally aggressive, non-encapsulated, non-metastasizing neoplasms that infiltrate bone marrow spaces. Histopathologically, the OM is characterized by loose,abundant mucoidstroma that contains rounded, spindle-shaped, or angular cells.[1]Formerly, some investigators made a distinction between odontogenicmyxomas (derived from odontogenic mesenchyme) and osteogenicmyxomas (presumably derived from primitive bone tissue). However, most authorities in orthopedic pathologic practice do not accept that myxomas occur in the extragnathic skeleton, and all myxomas of the jaws are currently considered to be of odontogenic origin.[2]

Case Report

A 35 year-old female patient reported with the chief complaint of a painless gradually progressive swelling in the lower left side of jaw, which was initially noticed as a small swelling in the lower left vestibule and grew to the present size within a span of one year. There was no history of trauma. Past medical and dental histories were non-contributory. Extra-oral examination revealed a slight diffuse, non-tender, bony hard swelling of the left body region of mandible. [Figure 1]. No local rise in temperature or change in color of the overlying skin was seen. No lymphadenopathy was observed.

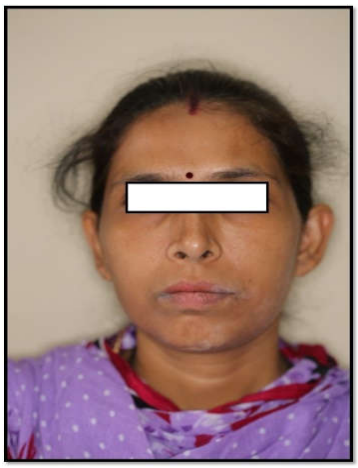

Figure 1 Extra-oral examination revealed a slight diffuse, non-tender, bony hard swelling of the left body region of mandible.

On intra-oral examination, a well-defined, bony hard, non-tender swelling extending from mesial of 34 to distal of 36, obliterating buccal and lingual vestibule. Lingual cortical expansion was more as compared to the buccal cortex.34 and 35 were grade 1 mobile and root-piece of 36 was seen superficially with buccal displacement. There was no

Corresponding author:* **PoojaSiwach
Department of Dentistry, Government Medical College and General Hospital, Dhule, Maharashtra, India. 424001

erythema, ulceration or pus discharge in the overlying mucosa [Figure 2].

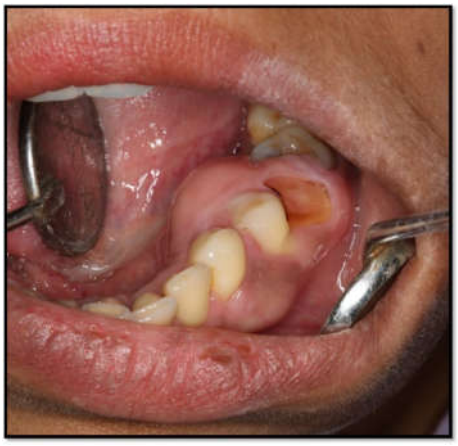

Figure 2 On intra-oral examination, a well-defined, bony hard, non-tender swelling extending from mesial of 34 to distal of 36, obliterating buccal and lingual vestibule.

Based on history and clinical examination, provisional diagnosis of ameloblastoma was given while odontogenicmyxoma, keratocysticodontogenic tumor and central giant cell granuloma were considered in differential diagnosis. Radiographically, the Orthopantomograph (OPG) showed a well-defined, multilocular radiolucency, extending from distal of 34 to mesial of 37 with intact inferior border of mandible. Internal of the lesion showed fine trabeculae, some intersecting at right angles. These thin, straight septae resembled tennis racket or stepladder pattern. [Figure 3].

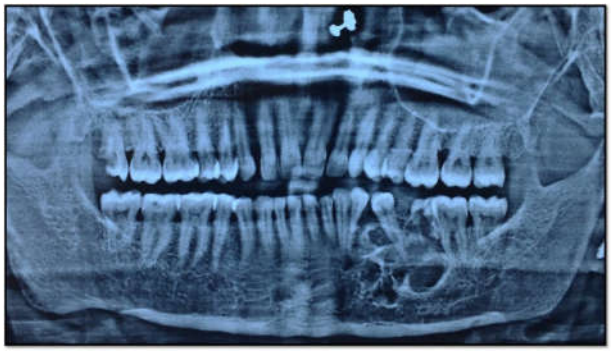

Figure 3 OPG showed a well-defined, multilocular radiolucency, extending from distal of 34 to mesial of 37 with intact inferior border of mandible

To clearly define the tumor margins and to ensure that the true extent of the tumor is visualized, cone-beam computed tomography (CBCT) was conducted. CBCT also showed multiple thin, straight septae within the radiolucent lesion. Expansion and perforation of both buccal and lingual cortical plates was also seen. [Figure 4]

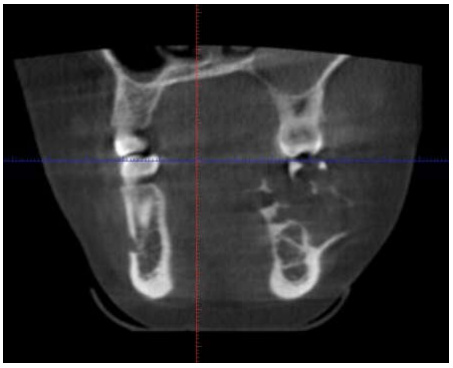

Figure 4 CBCT showed multiple thin, straight septae within the radiolucent lesion.

As, straight septa is a characteristic radiographic feature for odontogenicmyxoma, radiographic diagnosis of the same was given. For confirmation, incisional biopsy was done. Gross specimen was smooth, glistening white, soft tissue mass, suggestive of myxomatous tissue. On microscopic examination, H and E stained section revealed loosely arranged stellate, spindle and round shaped cells within homogenous myxoidstroma. (Figure 5 and 6]. Based on the history, clinical, radiological, and histopathological examinations, the final diagnosis of odontogenicmyxoma of the left-side mandible was given.

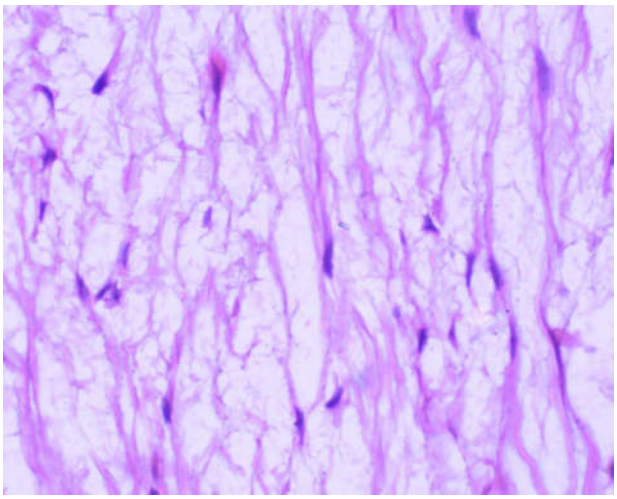

Figure 6 H & E (x40) stained section showed spindle shaped cells in myxoidstroma.

Segmental resection of the left-side mandible was performed under general anesthesia and fixation was achieved with titanium plates. The patient has been put on regular follow-up and responded well to the treatment without recurrence.

DISCUSSION

WHO 2005 defined Odontogenicmyxoma (OM) as an intraosseous neoplasm characterized by stellate and spindle-shaped cells embedded in an abundant myxoid or mucoid extracellular matrix. When a relatively greater amount of collagen is evident, the term myxofibroma may be used. In WHO classification of odontogenic neoplasms (2005), the myxoma appears under the subheading ‘‘odontogenicectomesenchyme with or without included odontogenic epithelium’’. The frequency of Odontogenicmyxoma (OM) varies in different parts of the world between 3-20% of all odontogenic tumors. In most studies, OM is the third most frequent odontogenic tumor (after odontoma and ameloblastoma).[3] The age range varies from 1-73 years, with a mean age of 30 years. The majority is diagnosed in the 2nd-4th decades. OM is slightly more common in females.[3,4]

Radiographic features

The radiographic features of the odontogenicmyxoma are variable, ranging from small unilocular lesions to large multilocular neoplasms, which often displace teeth or, less frequently, resorb roots. Most multilocularmyxomas are greater than 4.0 cm; unilocularmyxomas tend to be smaller. Only 5% of myxomas are associated with unerupted teeth. Most investigators report myxomas to be radiolucent; however, Kaffe *et al* found that 12.5% of myxomas were mixed radiolucent and radiopaque, and 7.5% were radiopaque.

Because myxomas do not produce calcifications, the mixed radiographic appearance was attributed to residual bone within the neoplasm.[5] Residual bone trapped within tumor remodels into curved and straight, coarse or fine septa. The presence of these septa gives the tumor a multilocular appearance. A characteristic septum identified with this tumor is a straight, thin, etched septum. These septa have been described as making a tennis racket-like or stepladder-like pattern, but this pattern is rarely seen. In reality, most septa are curved and coarse, but the finding of one or two of these straight septa helps in identification of this tumor. This tumor has a tendency to grow along involved bone without the same amount of expansion seen with other benign tumors; however, when achieving a large size, there may be considerable expansion. The finding of characteristic thin straight septa with less than expected bone expansion is very useful in differential diagnosis from other multilocular lesions.[6] Similarly, in our case, straight septae were seen within the radiolucent lesion, which helped us to arrive at diagnosis of odontogenicmyxoma. Jawaid *et al* (2016) also described similar findings in their case report.[7]

However, it is still not known why internal of the lesion show fine and straight septae in case of odontogenicmyxoma while coarse and curved in case of ameloblastoma. Possible reason could be that the ameloblastoma frequently has internal cystic components, causing these septae to remodel into curved shapes providing honeycomb or soap bubble pattern.[6] While, odontogenicmyxoma is consisting of more of mucoid-like material and do not show microcystic degeneration, septae are at right angle to each other. Kheir *et al* (2013) in their study reported the imaging characteristics of odontogenicmyxoma and compare the different imaging (conventional radiographs, computed tomography (CT) and magnetic resonance imaging (MRI)) modalities used. They found that, despite the many limitations of conventional radiography, it is still easily accessible, feasible, affordable, and easy to interpret, making it a basic and essential tool in the investigation process. Whereas, CT and MRI can accurately reveal the true margins and extent of tumors, and greatly aid in diagnosing the tumor and differentiating OM from other tumors with similar presentation.[8]

Histopathology

Histologically, the myxoma is bland in appearance and is composed of loosely arranged, evenly dispersed spindle-shaped, rounded, and stellate cells with a lightly eosinophilic cytoplasm in a mucoid-rich (myxoid) intercellular matrix.[5] The microscopic differential diagnosis include hyperplastic dental follicle, myxoid nerve sheath tumours, chondromyxoid fibroma, low-grade myxoidfibrosarcoma and other myxoid sarcomas.[3]

Infiltrative and Recurrent behaviour of OdontogenicMyxoma

The cells in Myxoma are spindle shaped or stellate with long cytoplasmic processes. They probably give rise to the abundant stroma, which is composed of acid muco-polysaccharides and rich in hyaluronic acid and chondroitin sulfate. This gives the tumor its gelatinous consistency and may be responsible for its infiltrative nature.[9] It pervade surrounding bone by expansion rather than cellular growth, possibly as a result of the large content of hyaluronic acid.[5] Using the argyrophilicnucleolar organizer region technique, Martins *et al* (2001) suggested that the infiltrative and recurrent features of the odontogenicmyxoma probably were not related to its cell proliferation index. Bast *et al* (2003) found that the tumor cells of odontogenicmyxomas have a low proliferation rate as determined by Ki67 staining; however, in their study, myxoma tumor cells showed increased expression of antiapoptotic proteins (Bcl-2 and Bcl-X) and of the matrix metalloproteinase MMP-2, which could contribute to their growth.[5]

Treatment and Prognosis

The tendency of odontogenicmyxoma to permeate into marrow spaces makes effective enucleation and curettage difficult. Small lesions have been successfully treated in this way but larger lesions may require complete excision with free margins. Recurrence rates from various studies average about 25% but in spite of this, the prognosis is good. This high rate may be explained by lack of encapsulation of tumor, its poorly defined boundries, and extension of nests or pockets of myxoid tumor into trabecular spaces, where they are difficult to detect and remove surgically.[6] Recurrence usually follows incomplete removal within two years but may occur much later. Death may ensue due to cranial base extension.[3]

CONCLUSION

Radiographs play an important role in tapering down the differential diagnosis of oral lesions while confirmatory diagnosis is given by histopathology only. But careful examination and evaluation of radiographic findings can, sometimes, be helpful in early diagnosis and thus, early treatment. Thereby, we discussed a case report where the characteristic radiological findings helped to arrive at the diagnosis of odontogenicmyxoma which was further confirmed by biopsy.

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