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CASE REPORT

Odontogenic myxoma in the mandible - a diagnostic dilemma

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Odontogenic myxomas are slow-growing and invasive tumors comprising of 3-6% of all Odontogenic tumors, that can become very large and distend the maxilla or mandible. Here we report a case of 19 year old female patient who came with a complaint of swelling in the left side of the jaw with occasional pain since two months.

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INTRODUCTION

Myxomas can be found in various sites in the body including the skin and subcutaneous tissue, heart (mainly in the left atrium), and also in various sites of the head and neck.[1] Odontogenic myxoma (OM) is the second most common odontogenic lesion with an incidence of approximately 0.07 new cases/million /year.[2] World Health Organization has classified OM as a benign tumor of ectomesenchymal origin with or without odontogenic epithelium. It appears to originate from the dental papilla, follicle or periodontal ligament. Myxomas are non-encapsulated benign locally aggressive tumors which may occur in both the soft tissues and bone. In 1863 Virchow was the first man to describe about myxomas.[3]

CASE REPORT

A nineteen-year-old female patient had visited our department with the chief complaint of swelling on the left side of the mandible since two months with occasional dull aching pain. History revealed that the swelling was small and gradually increased in size. Patient also gave a history of extraction in the same region one year back. Medical history was non-contributory. On Extraoral examination, no gross facial asymmetry was noted. On intraoral examination there was mild expansion of the buccal cortical plates in the left mandibular premolar region (Fig 1) with no vestibular obliteration. 35 was missing. Restorations were present in 33, 34 and 36. No tenderness and parasthesia were present. No lymph node enlargement was present. Based on the history and clinical examination, Provisional diagnosis of Residual cyst was given. To confirm the diagnosis, certain radiographs were advised. Mandibular occlusal radiograph revealed multilocular radiolucency extending from 32 to 36 region with expansion of the buccal cortical plates (Fig 2). Panoramic radiograph revealed multilocular radiolucency with angular septations extending from 36 to 42. Obturating material present in 33 is extending into the radiolucency. Root resorption in seen with respect to 34,36 (Fig 3). Cone beam computerized tomography revealed irregular multilocular bone destruction with angular septations. (Fig 4)

Fine needle aspiration was negative. Differential diagnosis of Odontogenic keratocyst, Central giant cell granuloma and Odontogenic myxoma were given. For further confirmation incisional biopsy was advised. Gross sections revealed greyish white glistening, gelatinous mass (Fig 5). Heametoxylin and eosin sections showed stellate cells loosely arranged in a fibrillar stroma which confirmed the diagnosis of Odontogenic myxoma. (Fig 6). Treatment plan of segmental mandibulectomy followed by reconstruction using free autogenous graft was planned. However, the patient was lost to follow up.

Figure 1: Mild expansion of the buccal cortical plate irt 34,36 region

Figure 2: Mandibular occlusal radiograph revealed multilocular radiolucency extending from 32 to 36 region with expansion of the buccal cortical plates.

Figure 3: Orthopantomograph reveals multilocular radiolucency with angular septations extending from 42- 36. Obturating material present in 33 is extending into the radiolucency. Root resorption in seen wrt 34,36.

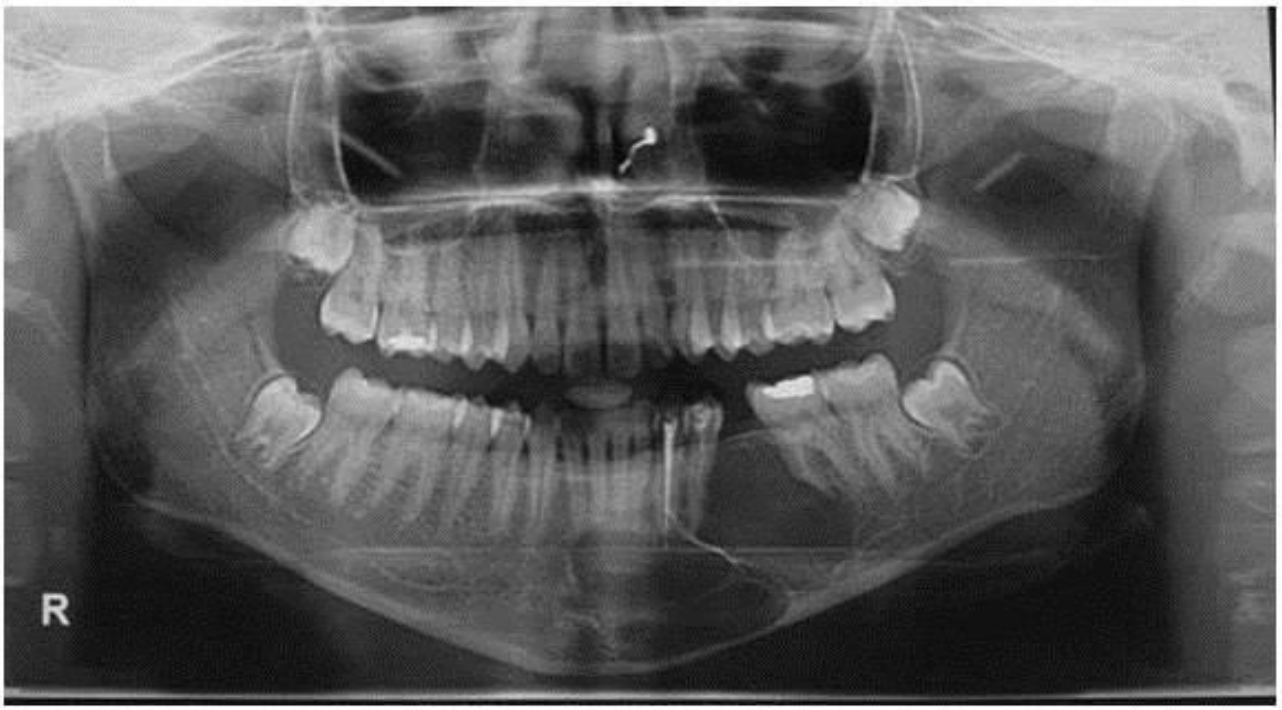


Figure 4: Cone beam computerized radiography reveals Cortical expansion, angular septations and obturative material in 33 in axial and sagittal sections respectively.

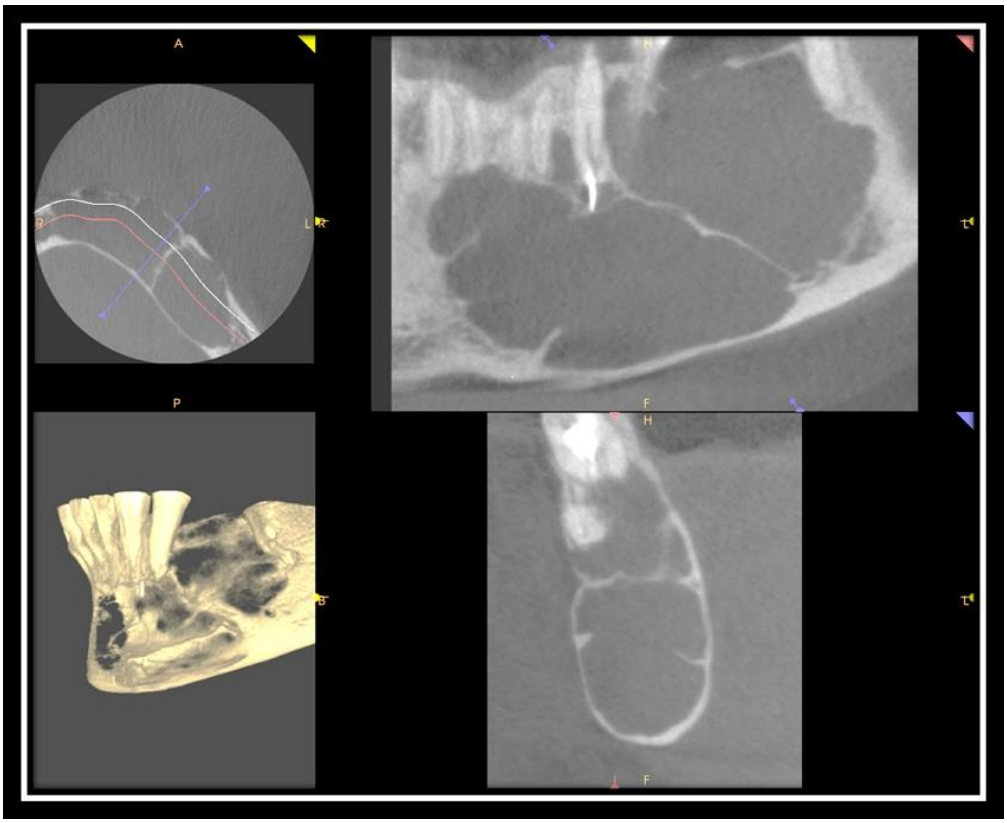
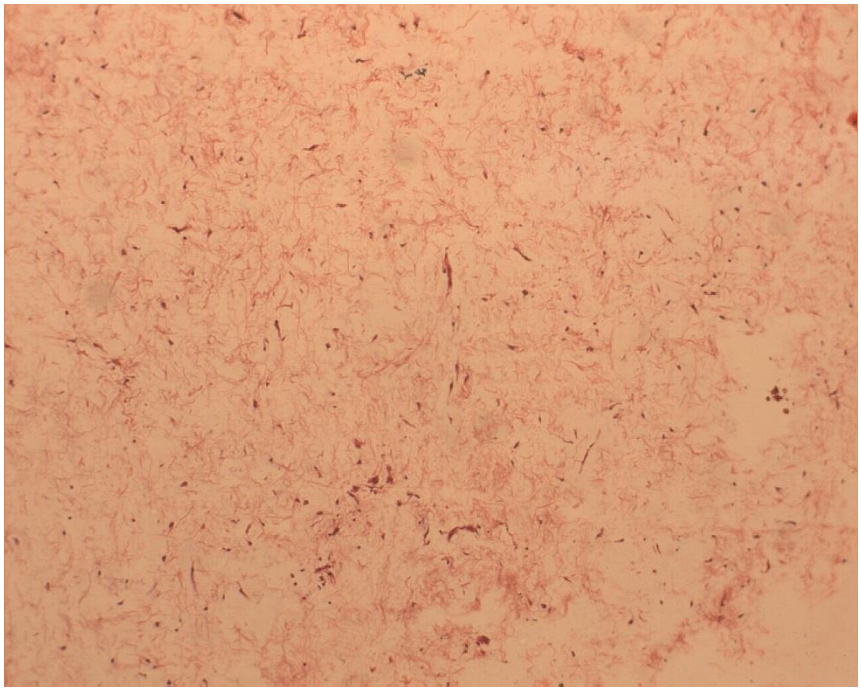


Figure 5: Gross specimen showed Whitish grey glistening gelatinous mass.

Figure 6: Haematoxylin and eosin stain showing stellate shaped cells loosely arranged in myxoid stroma.



DISCUSSION

Rudolph Virchow was the first to describe the histologic features of myxofibroma in 1863, although the lesions of jaws were not particularly mentioned. He described that these tumors resemble the mucinous substance of the umbilical cord. These tumors are most frequently seen in heart muscle. Myxomas of the mandible and maxilla are very rare. In 1947, Thoma and Goldman first described myxomas of the jaws. Since then odontogenic myxoma has been a subject of continuous scientific debate.[4]

Odontogenic myxomas (OM) are rare tumors derived from embryonic mesenchymal elements of dental anlage.[5] World Health Organization has classified OM as a benign tumor of ectomesenchymal origin with or without odontogenic epithelium.[6] It appears to originate from the dental papilla, follicle or periodontal ligament.[5]

In Asia, Europe and America relative frequencies of occurrence of odontogenic myxomas are between 0.5 and 17.7% have been reported.[7, 8-13]

Odontogenic myxomas are asymptomatic. Pain, paresthesia or asymmetries occur only when they take on larger sizes. Their growth is usually slow; however, they are locally aggressive. They may cause divergence of root, resorption, tooth shifting or movement. When involving maxilla, OM can expand inside the maxillary sinus and are then diagnosed later only after having grown to larger sizes.[14]

Radiological appearance of OM is usually a unilocular or multilocular radiolucency having well-defined or diffused margin. The size of the lesion and locularity are interrelated. OM lesions which are larger than 4 cm follow a multiloculated presentation, whereas smaller lesions seem to be unilocular in appearance and the presentation is often defined as soap bubble, honeycomb, ground-glass, or tennis racket pattern.[15] Kaffe et al reported mixed appearance in 12.5% and radiopaque appearance in 7.5% cases.[16]

On gross examination, odontogenic myxoma appears as a grayish white, nodular heterogeneous mass of variable consistency,[1] with a glistening gelatinous cut surface,[17] which was obvious in our case.

In a clinicopathological study conducted on 25 cases of odontogenic myxomas by Li et al (2006), it was seen that histologically, all odontogenic myxomas were mainly composed of spindled or stellate-shaped cells in a mucoid-rich intercellular matrix.[18]

In a review conducted by Farman et al (1977) on histochemical findings in odontogenic myxoma it was seen that the ground substance of odontogenic myxomas consist of about 80% hyaluronic acid and 20% chondroitin sulfate. Tumor cells showed slight alkaline phosphatase activity. It was also seen that the myxoid intercellular matrix stains positively with alcian blue, but PAS staining may be negative.[19]

The current recommended therapy depends on the size of the lesion and on its nature and behaviour and can vary from curettage to radical excision. Due to its locally invasive nature, OM of the jaws tends to be treated by bone resection including peripheral ostectomy and segmental mandibulectomy.[20,21] These techniques remove a circumferential margin of bone around the tumor. The lesion is not encapsulated and its myxomatous tissue infiltrates the surrounding bone tissue, accounting for a high recurrence rate of 25% with conservative treatment.[22-23] Therefore, radical resection including a margin of 15–20 mm of healthy bone seems to be the best option to prevent recurrences, especially in rapidly expanding or locally destructive lesions with cortical bone perforation.[11] These

characteristics may explain the high rate of recurrence of myxomas which ranges from 10 to 33%.[24-25]

CONCLUSION

Odontogenic myxomas are rare tumors with ambiguous behavior. Odontogenic myxoma share common features with many other odontogenic tumours which may lead to a diagnostic dilemma. The use of plain radiography and advanced imaging modalities such as Computed Tomography and Cone Beam Computed Tomography can help in determining the extension of multilocular lesions like odontogenic myxoma and its effect on the surrounding structures. Proper initial clinicoradiological diagnosis followed by histopathological examination helps in arriving at an appropriate diagnosis and helps in formulating a treatment plan with minimal recurrences.

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