Case Report

An aggressive odontogenic myxoma of the maxilla

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Abstract

Odontogenic myxoma (OM) is a relatively rare benign odontogenic tumor of mesenchymal origin. OM is more common in the mandible than in the maxilla. It is an asymptomatic lesion that shows an infiltrative growth pattern. When the maxillary sinus is involved, it often fills the entire antrum. Odontogenic tumors are uncommon in the maxillary molar area, which often leads to diagnostic dilemma as this region of the maxilla is in the vicinity of vital structures, and radiographic overlapping of structures is always present. We present a similar case of a 17-year-old male patient who reported with a swelling in the left maxilla that infiltrated the maxillary sinus in a short duration of time.

Key words: Maxillary sinus, myxoma, odontogenic tumor

INTRODUCTION

Odontogenic myxoma (OM) is considered to be a relatively rare benign tumor of mesenchymal origin. It is found in the skin and subcutaneous tissue, heart and also in various sites of the head and neck region.[1] Myxomas of the jaw bones have been traditionally considered to have an odontogenic origin due to the close relation to teeth. According to the literature, OM represents between 3% and 10% of all odontogenic tumors.[2] Histologically, it is composed of spindle- or stellate-shaped cells in an abundant mucous intercellular substance, with little collagen. Those cases with higher amounts of collagen may be termed as myxofibroma.[3] Radiographic appearance varies from unilocular to multilocular radiolucency. OM exhibits aggressive infiltration of the adjacent tissue as it is not encapsulated and complete surgical removal is difficult.[4,5] It has a high tendency to recur and can transform into malignant lesion; hence, radiographic and histopathological interpretation are important to establish appropriate surgical management. The treatment options can include curettage with peripheral ostectomy, segmental resection and radical resections for the more aggressive lesions.[5,6]

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CASE REPORT

A 17-year-old male patient visited us with a complaint of swelling in the left maxillary molar region, which enlarged to the present size within a span of 3 months. Extraoral swelling was evident in the left side of the maxilla [Figure 1]. Intraoral examination revealed swelling in the first molar region, obliterating the buccal vestibule. Multilocular radiolucency extending from the distal aspect of the canine to the maxillary tuberosity region was observed on panoramic [Figure 2a] and occlusal radiographs [Figure 2b]. The computed tomographic (CT) scan showed swelling with bony expansion and thinning of the cortical plates with strong enhancement of the mass lesion in the anterior maxilla [Figure 3]. Based on a clinical diagnosis of ameloblastoma, a biopsy was performed. The microscopic examination of hematoxylin and eosin (H and E)-stained section showed fine fibrillar mucoid stroma with evenly spaced spindle- and stellate-shaped cells, and a mild to moderate amount of collagen was observed [Figure 4]. The mucoid nature was confirmed with a positive reaction with alcian blue stain [Figure 5], and Periodic Acid-Schiff stain was negative. Subsequently, the lesion was diagnosed as OM and surgical resection followed by prosthetic reconstruction was proposed.

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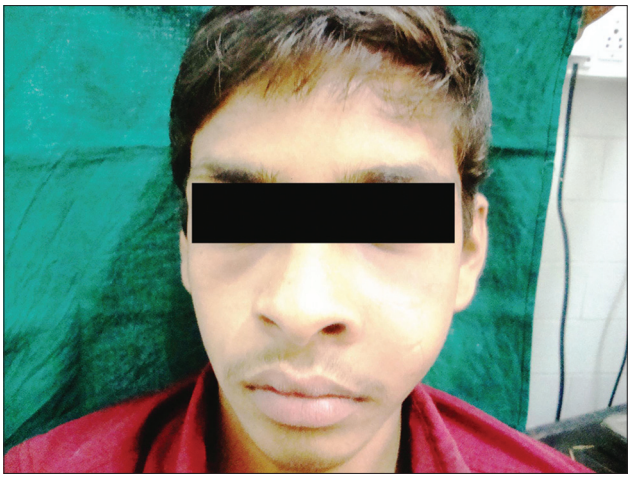

Figure 1: Swelling present in the left maxilla

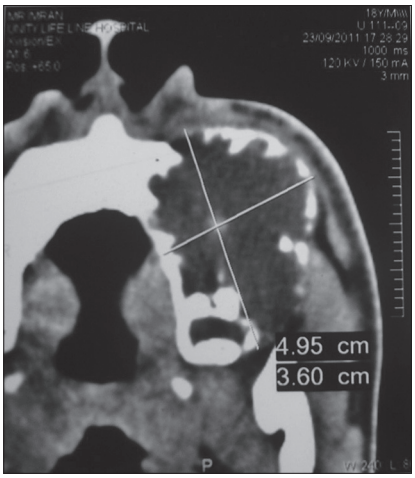

Figure 3: Computed tomography scan showing the extent of the lesion

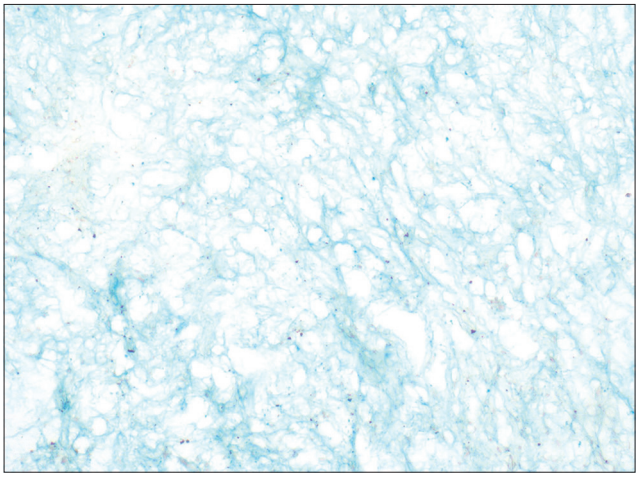

Figure 5: Alcian blue-positive reaction (x40)

DISCUSSION

Sivakumar *et al*.[3] suggested that OM is a tumor of a dual fibroblastic–histiocytic origin and the

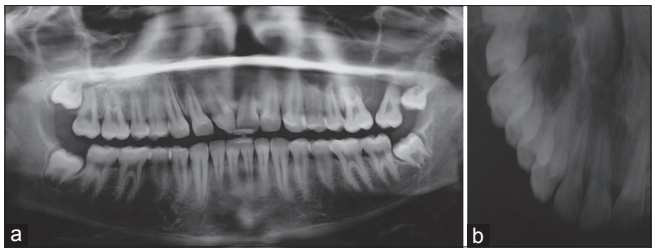

Figure 2: (a) Orthopantomogram showing multilocular appearance in the left maxilla, (b) Occlusal radiograph showing multilocular radiolucency extending from the distal aspect of the canine to the third molar region

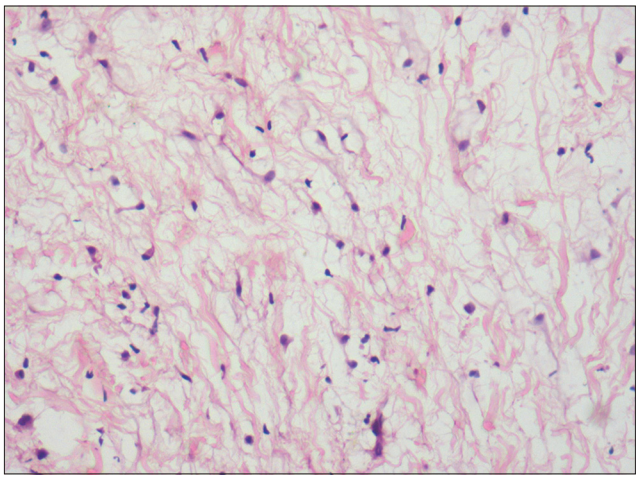

Figure 4: Hematoxylin and eosin section showing stellate-shaped cells in the fine fibrillar stroma (x100)

cells comprising odontogenic myxoma are of myofibroblastic origin. The histogenesis of OM is related to the odontogenic ectomesenchyme of a developing tooth or undifferentiated mesenchymal cells in the periodontal ligament. The odontogenic origin has been supported by the following reasons:

- Exclusive occurrence in the tooth-bearing areas of the jaws
- Association with an unerupted tooth or a developmentally absent tooth
- Frequent occurrence in young individuals
- Histological similarity between OM and pulpal ectomesenchyme
- Occasional presence of sparse amounts of odontogenic epithelium
- Its uncommon occurrence in other parts of the skeleton.[7]

The majority of cases are reported in the second and third decades of life. According to Kaffe *et al*.,[6] mandibular OM accounted for 66.4% and 33.6% in the maxilla. According to Lu *et al*.,[8] 52% were located in the mandible and 48% were located in the maxilla. Sixty-five percent of the mandibular cases were located in the premolar–molar region and 97% cases were seen in the same area of the maxilla.

OM is usually a central lesion, and the radiographic appearance is important to establish the diagnosis. The radiographic characteristics of OM are variable depending on its developmental stage. Multilocular radiolucency with either distinct or poorly defined margins is observed in adults and in the posterior part of the jaw. Unilocular appearance is seen in the anterior jaw of young children.[5,6,9] Zang *et al*.[5] examined the radiographic appearances of 41 OM that were divided into six types. These were Type I—unilocular; Type II—multilocular (including honeycomb, soap bubble and 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 of osteolytic destruction and osteogenesis. Kaffe *et al*.[6] in his radiographic study revealed an interesting correlation between size and locularity; unilocular lesions were smaller than 4 cm and multilocular lesions were larger than 4 cm. In the present case, the lesion was multilocular and larger than 4 cm. It is difficult to differentiate solid ameloblastoma, odontogenic keratocyst and OM using radiographs as all these lesions exhibit multiloculation. Dental radiographs are a bidimensional projection of a tridimensional structure, and therefore superimposition of anatomic landmarks can masquerade important findings.

Asuami *et al*.[10] examined the dynamic magnetic resonance imaging (MRI) features to differentiate these lesions; solid areas of ameloblastoma showed an earlier enhancement than the whole areas of the OM. These results indicated that the dynamic MRI features of the tumor substance of ameloblastoma differs from OM. Because of the scarcity of studies using MRI, the characteristics of the OM have not been established satisfactorily.[9]

Koseki, *et al*.[11] studied the CT characteristics of OM. They found that the tumor borders were generally well defined with a smooth margin both for bony and soft tissue structures. Cortical plate continuity was lost in numerous patients and intralesional trabeculations were observed. In the present case, bony expansion in the maxilla was present that measured 4.95 cm × 3.60 cm. Thinning and erosion of the cortical plates was present in the anterior and posterior regions of the maxilla and intralesional trabeculations were seen. MacDonald-Jankowski[12] suggested that both CT and radiographs should be used in the investigation of an OM. CT assesses perforation and pattern of septa while radiographs allow a better assessment of the degree of definition of the lesion's margins with the adjacent normal bone.

On gross examination, the specimen appears like an infiltrative mass of mucoid or slimy material. Microscopically, it is made up of loosely arranged spindle- and stellate-shaped cells, many of which have long fibrillar processes that tend to intermesh. In cases of myxofibroma, the amount of collagen in the mucoid stroma is more prominent.[13] The mucoid nature was confirmed with a positive reaction with alcian blue staining and negative Periodic acid-Schiff staining. Epithelial islands are not commonly observed in the myxomas of the jaws that do not play a significant role in OM. Akihiro Kimura *et al*.[14] reported a case of OM, in which the interesting feature was the presence of "active-looking" and irregularly proliferating epithelial islands with a microcystic appearance. Immunohistochemical positivity with CK 19 supports the odontogenic origin and high labelling index for Ki-67 indicates "active epithelium," which has never been reported. OMs are extensively described as case reports; however, the invasive behavior of these lesions has not been explained.

The tumor is not radiosensitive and surgery is the treatment of choice. Surgical procedures vary from currettage, enucleation, local excision and partial and total jaw resection. The lack of a capsule and infiltrative growth pattern is responsible for a high rate of recurrence when conservative enucleation and curettage are performed.[13] Boffano *et al*.[15] proposed the protocol to perform conservative surgery by enucleation and curettage when lesions were smaller than 3 cm, whereas a segmental resection with immediate reconstruction is preferred in patients affected by a bigger tumor.

Resection of the jaw was planned for this patient as the lesion in the maxilla is in close relation to vital structures; resection procedures minimize the risk of involvement of these structures and also reduce the recurrence rate. The patient is on follow-up and no sign of recurrence is noted.

CONCLUSION

OM is an uncommon tumor of uncertain behavior. OM and other odontogenic tumors share common features on conventional radiographs that lead to a diagnostic dilemma. In order to establish a treatment protocol, various radiographic modalities can be used to determine the extent of the lesion. Histopathological examination is essential to provide conclusive diagnosis and treatment planning.

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