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Odontogenic Myxoma of the Maxilla: A Rare Case Report

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Abstract

Odontogenic Myxoma [OM] is a relatively rare neoplasm which is almost exclusively seen in tooth bearing areas. It has annual incidence of 0.07 per million and constitute around 3-6 % of total odontogenic tumours. OM occurs commonly in mandible; however their presentation in maxilla is relatively uncommon. It is an asymptomatic lesion that shows an infiltrative growth pattern. It histologically presenting spindle-shaped, stellate and round cells within loosely arranged myxomatous tissue stroma. OM originates from the dental papilla, follicle or periodontal ligament with an exclusive location in the tooth-bearing areas of the jaws, association with missing or unerupted teeth. There are currently no clear surgical management guidelines for odontogenic myxoma, and a variety of approaches may be used. This article presents a rare case of an odontogenic myxoma occurring in the maxilla of 21-year-old male patient. which was managed conservatively by surgical excision.

Keywords

odontogenic myxoma; ectomesenchyme; myxoma; maxilla

Introduction

The term myxoma was first used by Virchow in 1871 [1].Myxomas are benign but locally invasive neoplasms that rarely appear in the skeleton [2]. The rates for prevalence and incidence of odontogenic myxoma are not available [3].The term OM is often applied when the tumor occurs in the jaws to reflect its odontogenic origin and thought to be derived from the mesenchyme of a developing tooth or periodontal ligament [4,5]. According to Lucas, the classification of the OM as an odontogenic tumor has been justified by its frequent occurrence in adolescence; its association with missing or unerupted teeth; and the sporadic presence of odontogenic epithelium within the neoplastic, myxomatous tissue [6]. Adekeye et al. supposed that the rarity of the OM in any extragnathic bone could be the only firm reason for suggesting odontogenic origin and considered dental papilla, dental follicle and periodontal tissues as possible germ centres for OM [7]. OM is a benign non metastasizing tumour characterised grossly by mucoid or gelatinous grey-whitish tissue that replaces the cancellous bone and expands the cortex [8].

Case Report

A 21-year-old male patient reported with a painless, diffuse swelling on the left side of the face since 6 months. Past medical history was found to be insignificant and patient was apparently alright 6

months back. The patient gave past dental history of extraction of grossly carious permanent maxillary left first and second molar, following which, he noticed a small swelling from the alveolar socket of the extracted permanent maxillary left first and second molar region, which gradually increased to present size.

Extra-oral examination showed a diffuse swelling resulting in facial asymmetry on the left side extending superoinferiorly from the infraorbital ridge to 2.5 cm above the inferior border of the mandible and anteroposteriorly from the left corner of the mouth to 1.5 cm anterior to the tragus. Skin overlying the swelling was normal and no sinus tract or fistula was observed. The patient also gave history of mild paresthesia in the same region. No regional lymphadenopathy was noted and the patient's vision was unaffected.

Intra-oral examination revealed a sessile, firm, non-tender soft tissue growth measuring about 4 x 3 cm in size, extending from the maxillary left second premolar to maxillary tuberosity region obliterating the left buccal vestibule. The overlying mucosa was reddish pink in colour with no history of pus discharge, sinus tract or fistula (Figure 1). Based on the clinical findings, pyogenic granuloma, giant cell lesion and salivary gland neoplasm were considered as the differential diagnosis.

Orthopantamogram revealed a unilocular expansile radiolucent lesion in left posterior maxillary region eroding the floor of the maxillary sinus. Computed tomography showed an expansile lesion measuring 3.9 x 4.1 cm in the left maxillary posterior region obliterating the left maxillary sinus. The wall of lesion showed discontinuity on the buccal aspect (Figure 2).

Multiple incisional biopsies of the intraoral lesional tissue and the left maxillary sinus were performed under general anaesthesia and subjected for histopathological examination. Light microscopic examination of hematoxylin and eosin stained sections of both the areas showed monotonous loose myxoid stroma with interspersed delicate network of collagen fibrils and numerous stellate to plump spindle shaped fibroblasts with small rounded nuclei. The histopathological diagnosis of OM was made on the basis of the features suggested in World Health Organization (WHO) Classification of tumors [9] (Figure 3).

A left maxillary surgical enbloc resection under general anaesthesia was planned for treatment, however the patient refused to undergo extensive surgery inspite of explaining the high chances of recurrence in case of incomplete surgical removal of the lesion. The patient opted for a conservative surgical excision of the intra-oral soft tissue mass under general anaesthesia. Grossly, the lesion was greyish gelatinous mass, oval in shape, firm in consistency having glistening mucoid appearance (Figure 4). Histopathological features of the excised tissue were consistent with that of incisional diagnosis of OM. The postoperative healing was uneventful. The patient was asked to report for a recall one month post-surgery. Unfortunately, the patient failed to report and was lost to follow up.

Discussion

The word myxoma, which describes a benign mesenchymal tumor of stellate and spindle cells within a polysaccharide rich avascular stroma, is derived from the Greek word “myxo”, meaning mucus or slime, first used by Virchow, for a group of tumors that had histological resemblance to the mucinous substance called Wharton's jelly of the umbilical cord. Myxomas were recognized as distinct lesions by

Stout in 1948 [10]. Myxoma can be found in the heart, skin, subcutaneous tissue and bone, the heart being the most commonly affected [11, 12]. However, myxoma affecting the jaws are relatively infrequent.

OM of the jaw was first described by Thoma and Goldman in 1947 [8]. According to WHO (2005) histological classification of odontogenic tumors, OM is categorized as a benign odontogenic tumour arising from mesenchyme and/or odontogenic ectomesenchyme with or without odontogenic epithelium [9].

Microscopically, OM mimics the dental pulp or follicle connective tissue. Various studies including Gunhan et al. [13] and Regezi et al.[14] reported a higher incidence of OM in women (64–95%) than in men. Mandibular myxomas accounted for 66.4% as compared to 33.6% in the maxilla [11]. In juxtaposition to the above reports, our case presented a lesion in the maxillary posterior region of a male patient which is a rare site for a myxoma to occur.

OM generally presents as a painless, slowly enlarging expansion of jaw with possible loosening or displacement of the teeth and they behave in a locally aggressive fashion. Tumors of maxilla tend to enlarge and often fill the maxillary sinus [15]. Similarly, in our case, the lesion spread into the sinus before presenting as an intraoral swelling; which made swelling unnoticed by the patient before extraction of the teeth. As teeth were extracted, the site provided a passage for the lesion to grow outwards and become visible in the oral cavity.

OM shows various radiographic features ranging from small unilocular lesions to large multilocular lesions. Multilocular lesions are described as 'honey comb', 'soap bubble' and tennis racket' appearance [16]. The radiographic appearance of unilocular OM resemble odontogenic cyst and unilocular ameloblastoma, however presence of fine bony septae differentiates OM from the latter two. Unilocular lesions are rare as compared to multilocular lesions and they are most commonly found in the mandibular posterior region [17]. Our case presented a unilocular radiolucent lesion in the left maxillary posterior region expanding into the maxillary sinus causing erosion of floor of the sinus and discontinuity on the buccal aspect of the lesion.

Macroscopically, OM usually shows greyish-white glistening or gelatinous mass with minimal true encapsulation. Histopathologically, all OM are mainly composed of spindled or stellate shaped cells in a mucoid rich intercellular matrix, but histological variations do exist. The majority of the tumours are monotonous and hypocellular. Cytoplasmic processes of the cells are often long and anastomose with other cell process. Nuclei are small, inconspicuous and hyperchromatic [2]. In addition, Martinez MG et al. found epithelial islands are a very rare entity in OM [18]. The presence of odontogenic epithelial islands is not mandatory for the diagnosis of OM [4]. Our case also showed similar histopathological features with absence of epithelial islands.

The current recommended treatment modality for OM depends on the size, nature and behavior of the lesion ranging from curettage to radical excision. Due to infiltrative behavior of the myxomatous tissue surgical removal using curettage and peripheral ostectomy alone is not a desirable treatment option for OM. Incomplete removal of the OM has a recurrence rate ranging from 10 to 33%. Reconstruction can begin immediately following thesurgical procedure or delayed until an adequate disease free period has past. Due to high recurrence rate, delayed reconstruction along with lifetime

follow up is the recommended treatment of choice. Small bony defects (smaller than 5 cm) can be reconstructed using buccal fat pad (maxilla), or using corticocancellous iliac crest bone graft. Larger defects (more than 5 cm) usually require primary prosthetic reconstruction (obturator) followed by a final obturator. Prosthetic reconstruction in maxillary lesions is highly advisable due to postoperative morbidity and almost immediate phonetic and masticatory function [19].

Conclusion

Clinical and radiographic characteristics of the odontogenic myxoma are variable, thus it should be always considered in the differential diagnosis of mixed and radiolucent lesions in the maxilla and a histopathological examination is compulsory to make the right diagnosis.

Variations in clinical and radiographical presentation make its differential interpretation challenging, as these features may overlap with those of other benign and malignant neoplasms. Thus, a correlation of clinical, radiological and histopathological findings is a prerequisite for accurate diagnosis. In addition, a proper surgery with clear margins and long term follow-up of these patients is essential to treat the patient successfully.

Figures

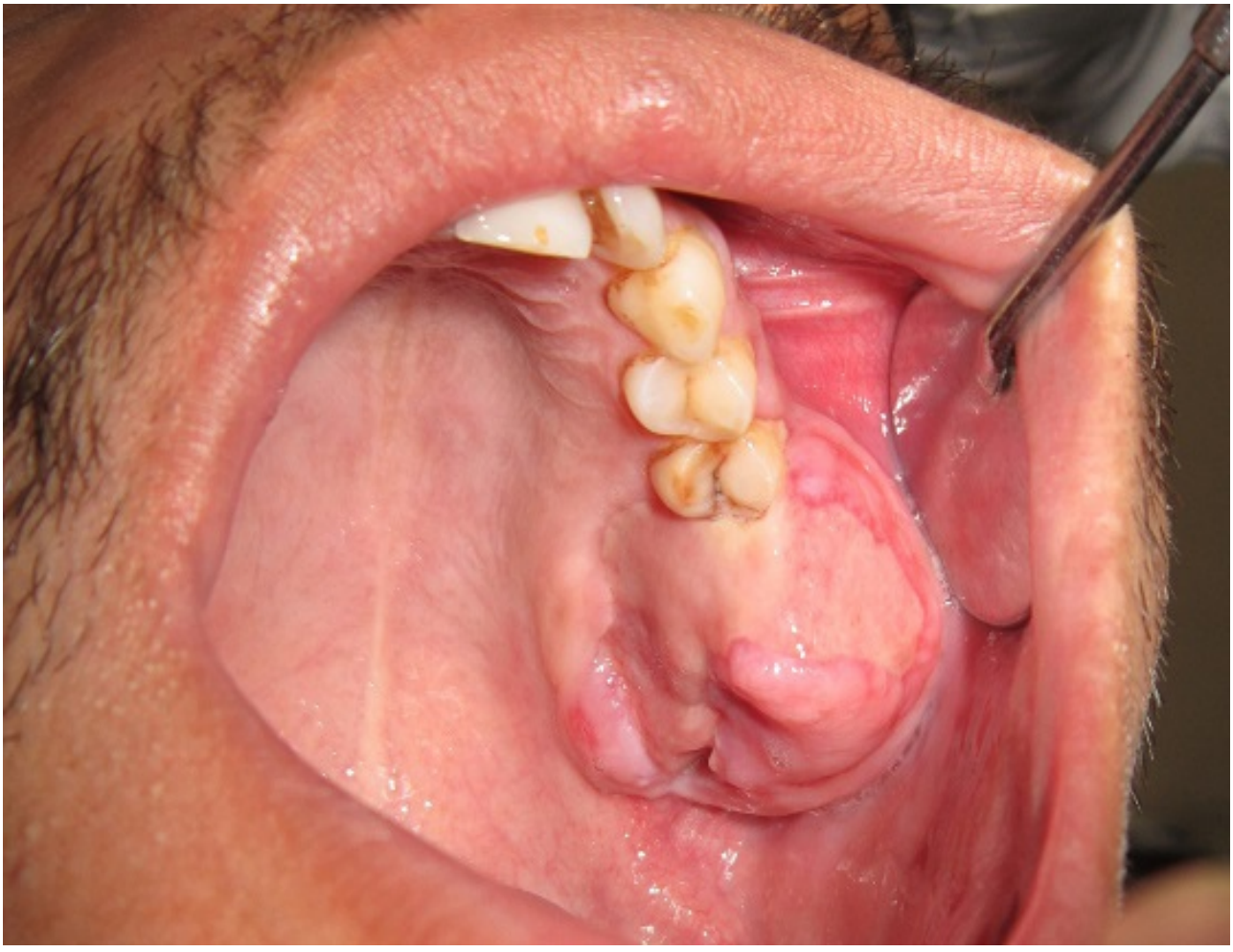

Figure 1: Clinical photograph showing a sessile, firm, soft tissue growth in maxillary left posterior region

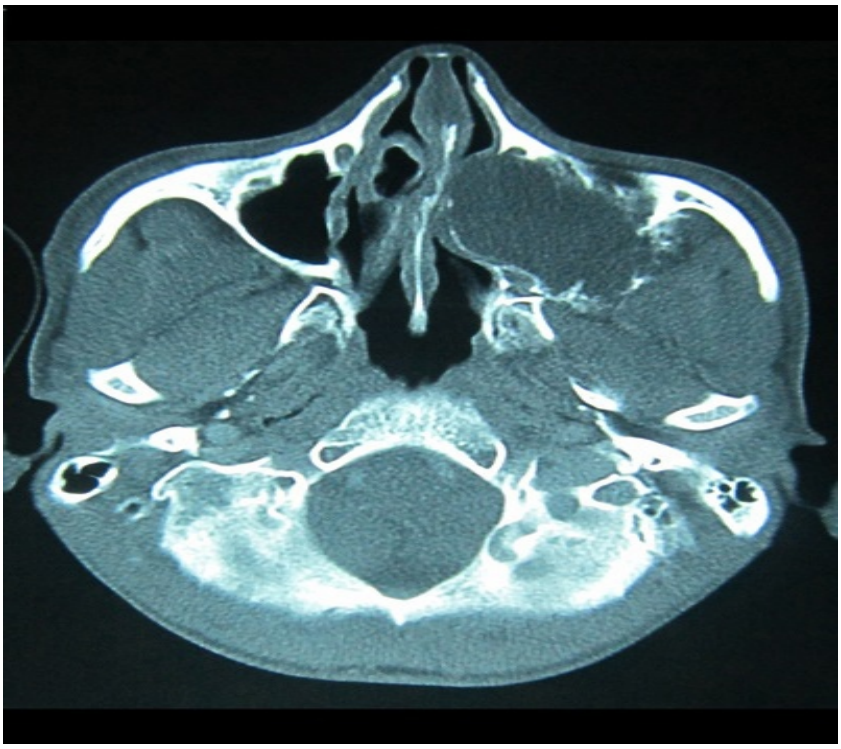

Figure 2: Computed tomography scan showing expansile lesion measuring 3.9 x 4.1 cm in the left maxillary posterior region obliterating the left maxillary sinus.

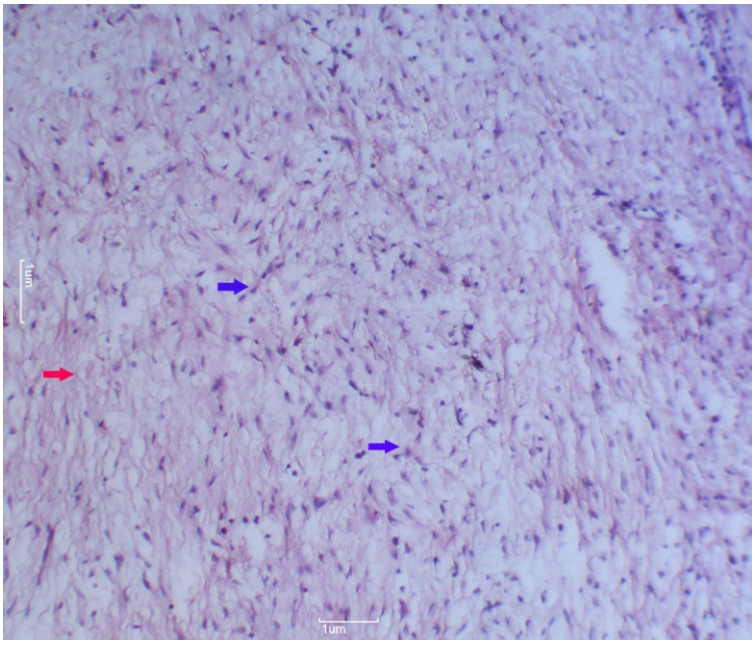

Figure 3: Photomicrograph showing evidence of monotonous loose myxoid stroma with interspersed delicate network of collagen fibrils (Red arrow) and numerous stellate to plump spindle shaped fibroblasts with small rounded nuclei (Blue arrow). (Haematoxylin-eosin, 10x)

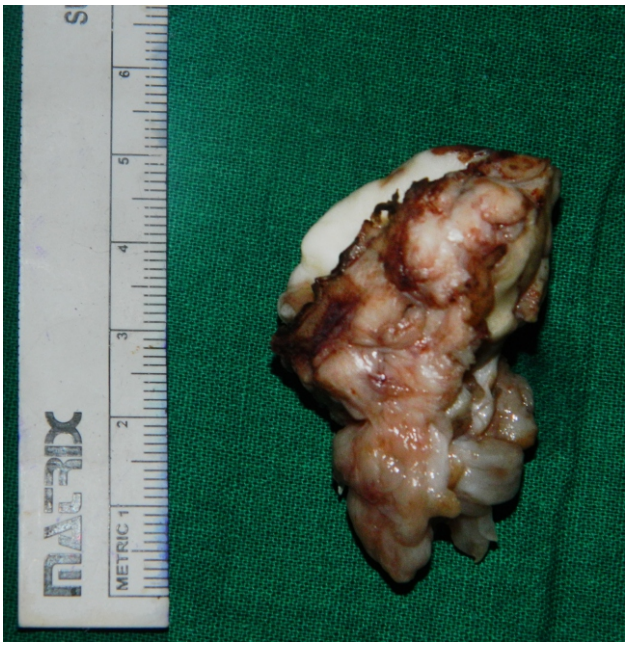

Figure 4: Gross specimen showing greyish-white, oval mass with glistening mucoid appearance

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