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Case Report

Placid odontogenic myxoma- A case report with literature review

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ABSTRACT:

Odontogenic Myxoma is a rare benign, locally aggressive and non-metastasizingneoplasm which is believed to arise from odontogenic ectomesenchyme and bear a close microscopic resemblance to mesenchymal portion of tooth germ. It represents 3 to 6 % of all odontogenic tumors and its etiology remains controversial.These tumors areoften slow growing and asymptomatic in its early stage, making it challenging to diagnose. Here wereportthe case of a placid odontogenic myxoma in right mandibular third molar region in a 25- year- old female patient.

Keywords: Odontogenic Myxoma, Benign, Ectomesenchyme

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INTRODUCTION

Odontogenic Myxoma is a rare benign tumor that originates from the embryonic mesenchyme of the dental apparatus and the myxomatous component is gelatinous in nature[1].Leiser in 2009 described odontogenic myxoma as a benign, locally invasive and aggressive,non-metastasizing neoplasm of the jaw bones[2]. Later in 2013, WHO defined Myxomas, histologically as benign odontogenic neoplasms of ectomesenchymal origin consisting of rounded and angular cells embedded in an abundant myxoid stroma with few collagen fibrils probably originating from either the dental papilla, follicle or the periodontal ligament[2].

There are two proposed theories regarding the origin of odontogenic myxomas. The first theory suggests that they develop due to the myxomatous degeneration of the fibrous stroma. The second one proposes that they originate from the mesenchymal component of the tooth germ, such as the dental papilla, follicle, or periodontal ligament[3].

Most of the odontogenic myxomas reported were young adults affected mostly in their second and third decade of life with marked female predilection (Manne et al., 2012)[4]. Odontogenic myxoma is generally depicted as slow growing tumor with the potential to attain considerable size without noticeable signs and symptoms. The molar and ramus regions of the mandible are most frequently involved, whereas the premolar–first molar region is the site of predilection in the maxilla (Noffke et al., 2007[4]. Thesetumors are usually slow growing, although they may be locally aggressive in maxilla[1].

Its slow growing nature and nonspecificsymptoms making it challenging to diagnose. And due its high recurrence rate, periodic followup is necessary. Here we present a case where the patient is totally asymptomatic on the involved site.

CASE REPORT

A 25-year-old female patientreported to the department of Oral Medicine and Radiology for oral prophylaxis. She reported that she was allergic to penicillin drug and hermedical, family and personal history were non - contributory. No relevant findings on extra oral examination, but on intraoral examination, mild swelling and inflamed gingiva noticed on the distal aspect of lower right second molar and unerupted third molar region. But the patient was totally asymptomatic in that region. And

on palpation, abrupt expansion noted on the posterior-inferior aspect of lingual cortical plate in relation to lower right third molar region,which was firm in consistency and non-tender in nature. No evidence of buccal cortical plate expansion and nobleeding and pus exudation noted from the site and aclinical diagnosis of benign odontogenic cyst like dentigerous cyst,odontogenic keratocyst were considered.

Panoramic radiograph showed a mixed radiolucent–radiopaque lesion surrounding the horizontally impacted lower right third molar extending from the distal aspect of lower right second molar till 2.5 cm away from the posterior border of ramus and thetooth was lying approximately 0.4 cm away from the inferior border of mandible. Cone beam computed tomography images showed awelldefinedmultilocular hypodense lesion of appropriate size 30.58 x 16 x 26.5 mm with faintseptae and horizontally impacted lower right third molar lying inferiorly and inclose relation to the lingual cortical plate causing an expansion, thinning and discontinuity of the lingual cortical plate along with distal root resorption of lower right second molar.And in theradiographic diagnosis odontogenic keratocyst,odontogenic myxoma, central giant cell granuloma and ameloblastoma were considered.

Aspiration cytology performed (with 18G Needle) was negative. After appropriate work up, the patient was referred to OMFS Department, where they did extraction of lower right second, third molar, followed by enucleation, chemical cauterization of the cystic lesion and peripheral ostectomy,which was done under GA and sent for histopathological analysis. On the macroscopy, cut section was light brown in colour.Microscopy showsfibromyxoid tissue partly lined by stratified squamous epithelium with foci showing lymphocytic infiltration and areas of calcifications. The lesion was cellular with many spindle and stellate cells embedded in myxoid stroma, with bony spicules/ osseous foci and calcific areas were seen within and diagnosed as Odontogenic myxoma.

Currently the patient is on regular follow-ups and appears healthy both on clinical and radiographic review after 6 months.

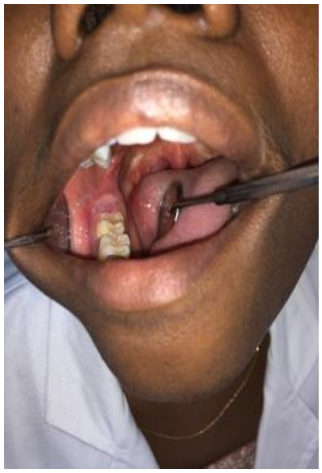

Figure 1: Intraoral photographs showing mild swelling noted distal to 47

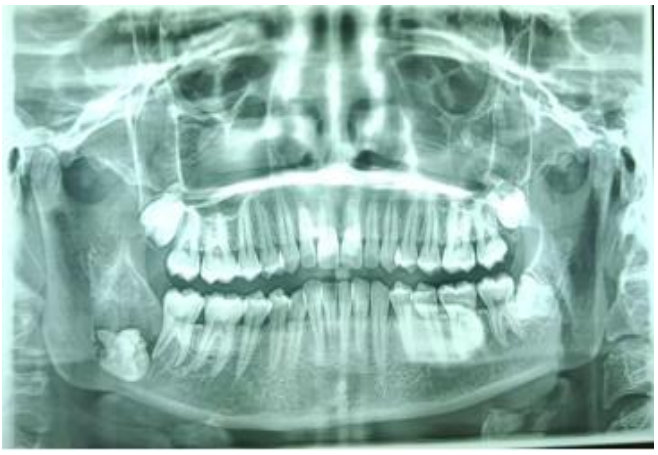

Figure 2: Panoramic radiograph showing a mixedradiolucent-radiopaque lesion surrounding the horizontally impacted 48

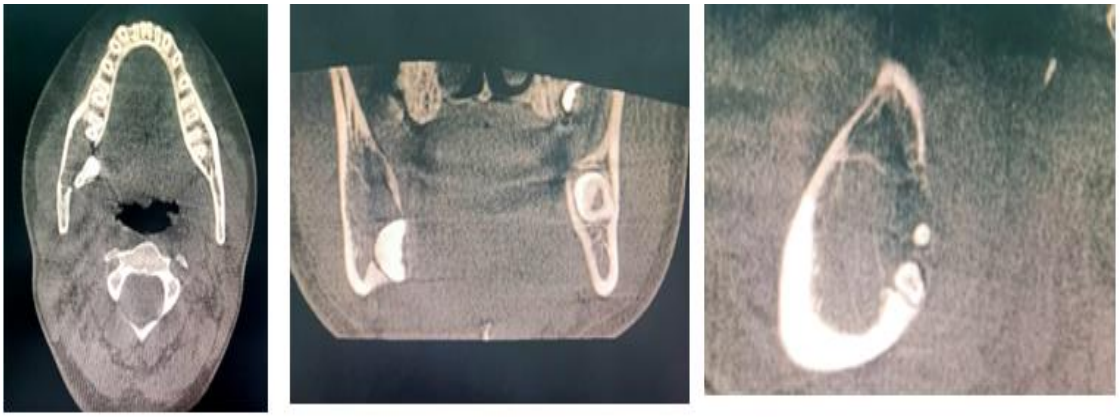

Figure 3 (a,b,c): CBCT images (Axial,Coronal and Sagittal) showing multilocular hypodense lesion with faint septa ; impacted 48 with thinning and discontinuity of lingual cortical plate noted.

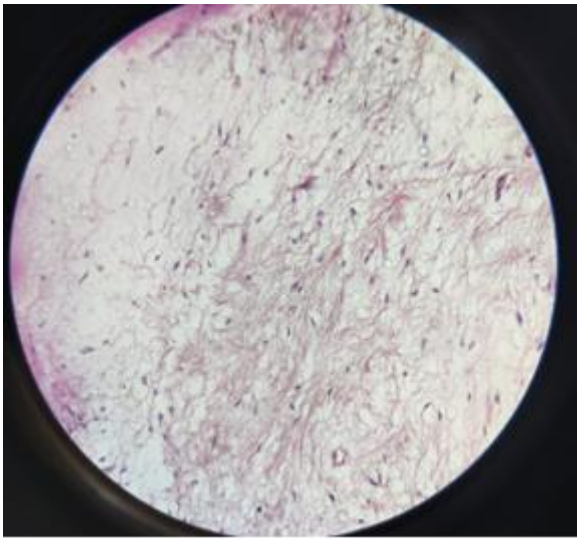

Figure 4: Histopathological view showing proliferation ofstellate or spindle shaped cells in a loose myxoid connective tissue stroma.

DISCUSSION

Odontogenic Myxoma is a rare benign odontogenic tumor of ectomesenchymal origin comprising 3 to 6 percentage of all odontogenic tumors. First described by Goldman and Thoma in 1947[5]. It was termed initially as 'myxofibroma' by Rudolf Virchow in 1863 owing to its histologic similarity to the mucous substance in the umbilical cord[6]. The prevalence of OM in Asia, Europe and America is 0.5%-17.7%, with the global prevalence is slightly lower at about 0.04%-3.7%[2].

It's unclear where this tumor came from. It has historically been believed to have odontogenic origin, which is backed by the following evidence: However, research by Goldbat and Slootweg[7]suggests that the development of myxoma may be from nonodontogenic ectomesenchyme as it is observed to develop within the sinonasal tract or non-tooth bearing regions of the jaws. It almost exclusively occurs in the tooth bearing areas of the jaws, is frequently seen in younger individuals, and is commonly associated with an unerupted tooth or developmentally absent tooth. They suggested that in the lack of odontogenic epithelium, a diagnosis of bone myxoma may be established. In contrast, McClure and Dahlin[7] found in their analysis of 600 bone tumors that genuine myxomas are limited to the mandible.

According to independent reports from Kaffeand Farman et al[8], these tumors are often more common in the second and fourth decades of life. According to Kaffe et al[7]study, 75% of cases happened in the second and third decades of life, and 7% happened in the first decade. However, cases might occur in any age group. All of the patients treated at our facility were in their second or third decade, with the exception of one 45-year-old patient.

Dysregulated apoptosis may potentially contribute to the abnormal growth of odontogenic myxoma, as suggested by the increased expression of the anti-apoptotic proteins Bcl-2 and Bcl-xL in these cells. Furthermore, the presence of gelatinase-type matrix metalloproteinase, MMP-2, in 90% of tumor cells indicates its potential involvement in the neoplastic growth[8].

Mazabraud syndrome is a condition characterized by the coexistence of intramuscular myxomas and fibrous dysplasia of bone, while Carney complex is a familial multiple neoplasia syndrome characterized by various features, including pigmentation, endocrine abnormalities, cutaneous and cardiac myxomas. In patients with Carney complex, dysregulation of the cAMP signaling pathways, resulting from alterations in the PRKAR1A gene, has been linked to tumorigenesis[9].

Mandibular posterior region is frequently involved than maxilla with slight female predilection. During its early stage, this tumor typically does not produce any symptoms and is often detected incidentally through routine radiographic examination. However, in later stage it can lead to painless cortical plate expansion with possible loosening, migration of teeth and with occasional root resorption. And sometimes it produces bosselated surface due to perforation of the cortical plates.

Radiographically, Odontogenic Myxoma can present with several patterns including unicystic, multilocular, pericoronal and as mixed radiolucent-radiopaque lesions. Frequently, multilocular pattern presents with straight septa which are thin, etched and this linear appearance of septa often described as the characteristic tennis racquet or step ladder pattern[5]. However, a unilocular appearance is more often in children and in anterior part of jaw[9]. In the tooth bearing areas, the tumor is often scalloped between the roots and root resorption may occur, thus giving a false appearance of OKC[3].

Zhang et al[11]examined 41 cases Odontogenic Myxoma and classified with 6 radiographic appearances; Type I – unilocular, Type II –

multilocular (honeycomb, soap bubble, tennis racquet) Type III – involvement of local alveolar bone, Type IV – involvement of maxillary sinus, Type V – osteolytic destruction, Type VI – a mix of osteolytic destruction and osteogenesis. Both CT and MRI play complimentary roles in the imaging evaluation of the tumor with CT providing information about hard tissue components and bone involvement and MRI offering detailed information about the soft tissue components and involvement of adjacent structures.

To effectively treat odontogenic myxomas, an accurate diagnosis is crucial in determining the most suitable surgical and adjuvant treatments. As this type of tumor is locally aggressive and not responsive to radiation, surgery is the primary treatment approach. Conservative techniques like enucleation, curettage, and cryotherapy offer several benefits such as reduced morbidity, shorter hospital stays, intra-oral accessibility, absence of donor site, lower cost, and minimal impact on facial growth in pediatric patients[2]. However, due to the high rate of recurrence, many researchers advocate for radical surgery involving partial or complete resection along with liquid nitrogen cryotherapy and subsequent reconstruction using vascularized free tissue transfer from sources like the iliac crest, radial forearm, or fibula. Ayranci et al. have proposed a protocol suggesting conservative measures like enucleation and curettage for lesions smaller than 3 cm, while segmental resection with immediate reconstruction should be considered for larger lesions[2]. The characteristic appearance of surgical specimens resembling a pale brown, glistening, gelatinous texture, often compared to a tender coconut, is considered a definitive sign[11], and odontogenic myxosarcoma is an extremely rare malignant variant.

Histologically, it has a mucoid matrix with uniformly distributed, loosely organised spindle-shaped and rounded cells. J.D. Harrison[13]has described "cavities" bordered by weakly defined fibers and epithelial islands. For immunohistochemical investigation, the cells in Sivakumar G's[12] case report differentiated into hyaline, stellate, and spindle-shaped cells. Ferritin, transferrin, alpha-1-AT, alpha-1-ACT, S-100 protein, and vimentin were all positive in spindle cells. Stellate cells reacted well to vimentin, alpha-1-AT, transferrin, and S-100 protein. Alpha-1-ACT and alpha-1-AT responded with hyaline cells. Antibodies did not react with the Myxomatous matrix. These results pointed to a myofibroblastic genesis for the tumor.

Following the removal of an odontogenic myxoma, Nardy Kasap[14]has detailed the usage of a surgical navigation system for implant surgery. Autogenous grafts or prosthetic devices can be used to reconstruct the deformity. According to published research, defects after myxoma excision are repaired utilizing vascular free fibula graft for mandibular defects and endoscopically harvested temporalis flap for maxillary defects.

Recurrence rates are reportedly high at around 25%, especially when a more conservative approach is taken[15]. Insidious local invasion to cancellous bone beyond the radiographically visible margins and absence of encapsulation are considered as reasons for high recurrence. In addition, its hypocellular matrix and mucoid ground substance of glycosaminoglycans and chondroitin sulphate can also account for its rapid growth and recurrence. MMP – 9 induced high invasiveness of the tumor in to the neighbouring tissue makes the surgical method a crucial determinant of recurrence[11]. Additionally, the recurrence rate may be influenced by the location and size of the tumor, as well as specific treatment approaches used. Regular follow up and monitoring are mandatory to detect any recurrent or residual tumors.

CONCLUSION

Diagnosis of odontogenic myxoma can be challenging due to its diverse clinical and radiographic presentations, often mimicking other lesions. Therefore, a comprehensive assessment involving clinical evaluation, radiographic imaging and histopathologic examination is crucial for accurate diagnosis. Don’t let the benign and placid appearance of lesions deceive us, maintain a watchful eye in order to catch any subtle shifts that may signal a more concerning pathology.

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