

Odontogenic fibromyxoma of left maxilla - A case report

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Abstract

Odontogenic myxoma is a locally invasive benign mesodermal tumour found exclusively in the jaws comprising 3-6% of all odontogenic tumors. It usually occurs in the second and third decade of life commonly involving the mandible as compared to maxilla. Its variable clinical and radiological appearance makes the diagnosis difficult.

Here we present a case report of a 30 year old female with a swelling in the left maxilla. The clinical findings made us suspect a case of radicular cyst with the carious molar. However, the radiological and subsequent histopathological examination established a final diagnosis of odontogenic fibromyxoma.

Keywords: Myxoma, Odontogenic tumor, Multilocular radiolucency, Fibromyxoma

Introduction:

Odontogenic myxoma is classified as a benign tumor of ectomesenchymal origin with or without odontogenic epithelium. It is defined as “a locally invasive neoplasm consisting of rounded and angular cells lying in an abundant mucoid stroma” according to the World Health Organization (WHO) 1992 (1).

Odontogenic myxoma is an uncommon, benign neoplasm which occurs in the maxillofacial skeleton comprising 3-6% of all odontogenic tumors. It is slow growing but locally destructive having a tendency to occur in second to predil fourth decades of life with a slight female ection

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(2). It is commonly seen in the mandibular posterior region (3). Clinically it presents as a slow growing asymptomatic swelling, however pain and paresthesia can occur in advanced stages. Facial asymmetry may occur due to large lesions. Displacement and mobility of teeth are relatively common. It may be associated with unerupted teeth. Cortical expansion can occur and large lesions can cause perforation (2). Radiographic appearance may vary from unilocular or multilocular radiolucent lesion to a mixed radiolucent–radiopaque lesion (4). The odontogenic myxoma has complete myxomatous tissue. However, varying proportions of myxomatous and fibrous tissue have also been reported and this entity is designated as fibromyxoma or myxofibroma (5).

In view of its rarity, the present case of odontogenic fibromyxoma of the left maxilla in a 30 year female involving the maxillary sinus is herewith reported.

Case report:

30 year old female patient reported to the Department of Oral Medicine and Radiology with a chief complaint of swelling in the left middle third of face since 4 months. The swelling had gradually increased to the present size in the last 4 months and was associated with pain from the last 15 days.

On extraoral examination, there was a diffuse swelling measuring about 2x2 cms in the left maxillary region with obliteration of the left nasolabial fold (Figure 1). The skin over the swelling appeared normal and there was no local rise in temperature. On palpation, the swelling was firm to hard in consistency. Intraoral examination revealed a swelling obliterating the buccal vestibule extending from left upper canine to left upper first molar region (Figure 2). The swelling was also evident on the palate with similar extensions. The swelling was firm in consistency and tender on palpation. There was expansion of buccal cortical plate and egg shell crackling was evident in 25, 26 region. The associated teeth i.e 24, 25 were found to be grade I mobile and 26 was Grade II

Mobile. Deep carious lesion was seen with 26, 27, 16 and 17.



Figure 1: Extraoral picture showing a swelling in the left infraorbital region obliterating the left nasolabial fold.



Figure 2: Intraoral swelling w.r.t 23, 24, 25, 26 region.

Based on the clinical features and history, provisional diagnosis of Radicular Cyst with 26 was given.

The patient was subjected to the following investigations:

Aspiration was done and was found to be non-productive. The patient was further subjected for radiographic investigations: Intraoral periapical (IOPA) radiograph w.r.t 24, 25, 26, 27 (Figure 3). IOPA radiograph revealed displacement with 25, 26 and loss of lamina dura with 24, 25. Maxillary cross-sectional occlusal radiograph (Figure 4) and

orthopantomograph (Figure 5) were also taken. The radiographs revealed an ill-defined mixed radiolucent–radiopaque lesion extending from distal aspect of 23 to mesial aspect of 27. Occlusal radiograph revealed expansion of the buccal cortical plate. Panoramic radiography also showed haziness in the left maxillary sinus.



Figure 3: IOPA radiograph showing displacement with 25 and an ill defined area of bone rarefaction.

Incisional biopsy was performed and the specimen was sent for histopathological examination which revealed a partially encapsulated myxomatous area composed of dense collagen fibres with few calcified structures and islands of odontogenic epithelium suggestive of ODONTOGENIC FIBROMYXOMA (Figure 6).

Advanced imaging:

Ultrasound examination (Figure 7) revealed fairly well defined predominantly hypoechoic mass measuring 3.1x 2.2x 2.0 cms confined to the left maxillary region with internal areas of calcification and cystic changes and underlying bony irregularities.

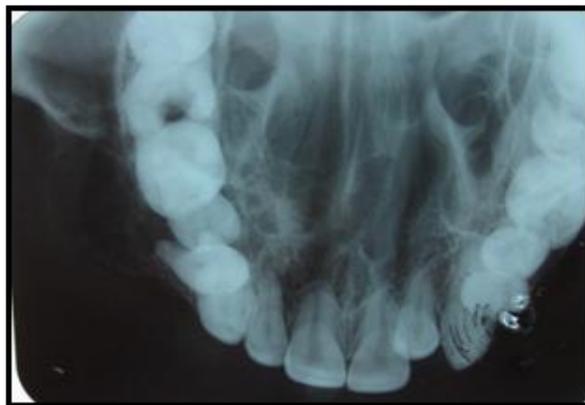


Figure 4: Maxillary occlusal cross-sectional view showing buccal expansion wrt 24, 25, 26 region.

A CT scan (Figure 8) was carried out at this stage to delineate the true extent of the lesion. The CT image showed a hypodense, irregular ill-defined expansile lesion measuring 3.5x 3.0 x 2.6 cms arising from the alveolar arch of left maxilla and extending into the maxillary sinus and nasal cavity of left side resulting in deviation of nasal septum towards right side. The lesion was seen to erode the anterior and medial wall of maxillary sinus, hard palate and floor of orbit on left side. There was evidence of linear calcification within the lesion.



Figure 5: Panoramic radiograph revealing haziness of left maxillary sinus.

Routine hematological and serological investigations of calcium, phosphorus, alkaline phosphatase were found to be normal.

Treatment: The tumor was surgically excised and segmental maxillectomy was carried out from 23-27 region. The excised specimen was

sent for histopathological examination which confirmed the diagnosis of ODONTOGENIC FIBROMYXOMA (Figure 9). Thereafter, an obturator was fabricated to cover the surgical defect and help in feeding (Figure 10).

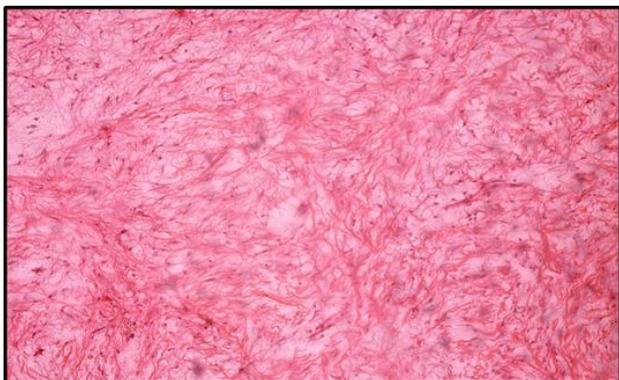


Figure 6: Photomicrograph showing histological features (Hematoxylin-Eosin stain X 10x)

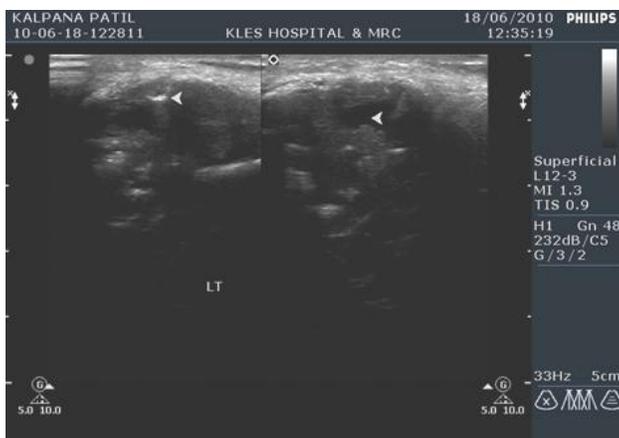


Figure 7: Ultrasonography showing a predominantly hypoechoic mass confined to the left maxillary region with internal areas of calcification and cystic changes.

Discussion:

Virchow (1) in 1863 coined the term myxoma for a group of tumors that had histologic resemblance to the mucinous substance of the umbilical cord. In 1947, Thoma and Goldman (6) first described myxomas of the jaws.

Odontogenic myxoma has been thought to originate from primitive mesenchymal structures of the developing tooth

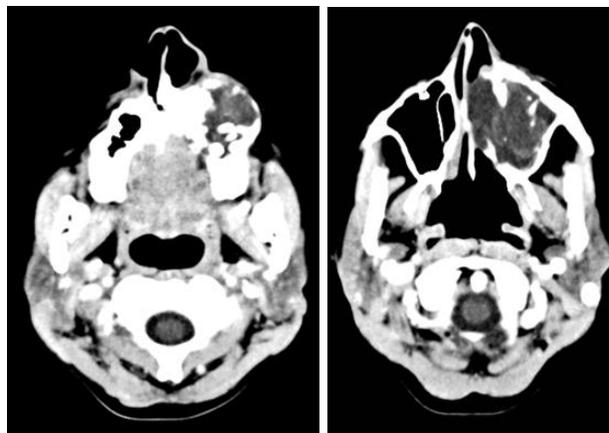


Figure 8: A CT scan showing a hypodense, irregular ill-defined expansile lesion of left maxilla and extending into the maxillary sinus and nasal cavity of left side.



Figure 9: Excised specimen.



Figure 10: Post operative picture with obturator.

(follicle/papilla/periodontal ligament) as an inductive effect of nests of odontogenic epithelium on mesenchymal tissue or as a direct myxomatous change in fibrous tissue, hence called odontogenic myxoma (7,8).

Odontogenic myxoma almost exclusively occurs in the jaw bones, comprising around 3-6% of all odontogenic tumors (4). We have reported an odontogenic myxoma involving the maxillary sinus in a 30 year female which is in conformity with age range reported in literature (4, 8-9). The mandible is involved more frequently than maxilla with ratios of 3:1 (10). The female to male ratio is 1.5:1 (2) although some reports state no sex predilection and equal frequencies in maxilla and mandible (11).

Odontogenic myxoma of the maxilla is less frequent than mandible but is more aggressive (8, 11-13) than that of the mandible, as it spreads through the maxillary sinus as was seen in our case.

WHO classified it as multiple radiolucent areas of varying size, separated by straight or curved bony septa with poorly defined borders (1). Odontogenic myxomas appear as multilocular or unilocular radiolucencies. Unilocular are more frequently found in the anterior region of the jaws, while multilocular lesions occur mainly in the posterior region (12). In multilocular lesions gracile or rough trabeculations can be found expressing a 'honeycombed,' 'soap bubble,' or 'tennis racket' appearance sometimes with diffuse calcifications. Displacement of teeth is a relatively common finding, root resorption is rarely seen and in tooth bearing areas the tumor is often scalloped between the roots (9, 14). When unilocular and without trabeculae, the tumor closely resembles periapical, lateral, periodontal and traumatic bone cysts. When multilocular, it must be distinguished from ameloblastoma, central hemangioma, central giant cell granuloma and odontogenic keratocyst and certain non-neoplastic lesions (fibrous dysplasia) (12, 14).

Grossly, myxomas are characterized by mucoid or gelatinous grayish-white tissue. It primarily consists of a myxomatous ground substance with widely scattered undifferentiated spindled mesenchymal cells (8). It is distinguished by the presence of sparse cords and islands of inactive odontogenic epithelium but these are not essential for diagnosis (15). Farman et al (9) reviewed the histochemical findings in odontogenic myxomas. The ground substance of odontogenic myxomas has been shown to consist of about 80% hyaluronic acid and 20% chondroitin sulfate (14). Odontogenic myxoma tumor cells are mesenchymal in origin and express vimentin and muscle-specific actin (16). Recently, advanced imaging modalities such as CT and MRI are being applied to this tumour which possess the special predominance in detecting whether the adjacent bone and soft tissues are involved or not, and the exact lesional extent (17-19).

The controversy has mainly been on therapeutic management with recommendations varying, depending on the size of tumor, from simple enucleation and curettage or wide excision to segmental bone resection (20). Slootweg and Wittkamp (10) comment that site of the myxoma should be taken into account when deciding on management plans.

The tumor is not radiosensitive, and surgery is the treatment of choice (9). The lack of a capsule and infiltrative growth pattern is responsible for high recurrence rate (upto 25%) when conservative enucleation and curettage are performed (21). Recurrence is minimized with extensive partial or total resection procedures, and this method of treatment is particularly indicated in the maxilla due to the proximity of vital structures (10). Hence, surgical excision with segmental maxillectomy was performed in our case. Prognosis of myxomas of the jaw is generally good. Recurrence typically occurs during the first 2 years after removal although recurrence has been described over 30 years after original surgery (21).

Conclusion:

There is a wide variety in clinical and radiologic appearance of odontogenic myxomas, being the most common form of presentation as an asymptomatic expansion in the jaw and a multilocular radiolucent image. Due to its variable clinical and radiological appearance it should be considered in the differential diagnosis of radiolucent and mixed radiolucent-radiopaque lesions of both jaws in all age groups.

Correlation of clinical, radiological and histopathological features is essential when trying to diagnose lesions which lack the characteristic appearance.

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