

Case Report

Central Odontogenic Fibroma (WHO type) mimicking Fibro-osseous Lesion- A Case Report

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Abstract:

The odontogenic fibroma is a relatively rare, slow-growing, benign odontogenic neoplasm of the jaws, with the potential to recur after excision. The World Health Organization (WHO) classified odontogenic fibroma into two variants: 1) WHO type (Epithelial-rich) 2) simple type (epithelial-poor type). It commonly occurs during middle age and has predilection for mandible. This article presents a rare case of Central Odontogenic Fibroma (WHO type) in a 24-year-old female demonstrated radiographically as mixed lesion involving right maxillary premolar – molar region. OF can mimic fibro-osseous lesions so, we recommend that vigilant comprehensive analysis of all the pertaining clinical, radiographic and histopathological aspects should be carried out with long term follow up after required treatment.

Key words: Odontogenic fibroma, Intra-osseous, benign.

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Introduction: Odontogenic fibromas (OFs) were defined in the latest classification of the World Health Organization (WHO) as a uncommon neoplasm characterized by an erratic quantity of inactive odontogenic epithelium in a rather-mature fibrous stroma.^[1] OF is considered to be an “Elusive and controversial tumor” due to its rarity and the uncertainty.^[2] It is a benign neoplasm, representing 1% to 2.5% of oral biopsy specimens.^[3] It occurs either as a

central (intra-osseous) lesion (COF) or as peripheral lesion (POF).^[3] POF is more common than its counterpart by ratio of 1.4:1.^[2] The age group for the occurrence of OF varies from 4 to 80 years, with an average of 40 years. It may affect both genders, but has slight female predilection.^[1] It has predilection to occur in the mandible and is generally asymptomatic except for the swelling of jaw.^[2] COF radiographically presents as uni- or multilocular radiolucencies

with well-defined borders. In some rare cases, it might present mixed radiolucent and radiopaque features and undefined borders.^[4] Root resorption is common and lesion located between teeth often cause root divergence.^[5] Microscopically, it is characterized by proliferation of cellular fibrous or fibromyxomatous connective tissue that exhibits variable amounts of odontogenic epithelium, mostly designated as epithelium-poor or epithelium-rich subtype. Foci of calcification in the form of dentinoid, cementicles, or bone may also be seen.^[3]

Here we presents a rare case of Central Odontogenic Fibroma (WHO type) in a 24-year-old female involving right posterior region of maxilla i.e premolar – molar region with emphasis on its differential diagnosis.

Case Report:

A 23 -year-old, female patient reported with a chief complaint of swelling in the upper right back tooth region of the jaw, since 4 months and also complains of pain in the same region since 1 month. Swelling was initially small in size and gradually increased to the present size. After 3 months patient developed pain, which was sudden in onset, mild, dull, intermittent in nature and non-radiating type. The patient's medical history was non-contributory.

Extra-oral examination revealed, a solitary diffuse swelling present on right side of the face extending superior- inferiorly from infra-orbital margin to the line joining corner of mouth to tragus of ear, medio-laterally from ala of nose to the 3cm away from tragus of ear. It measures approximately 3x3cm in diameter, oval in shape, smooth surface with normal color as that of adjacent skin. (Figure 1a). The swelling was firm in consistency and non tender on palpation. On intraoral examination, a solitary, well defined swelling was evident in right maxillary vestibule extending from distal aspect of 13 and to mesial aspect of 16. The swelling was oval in

shape, measuring 3x2cm in size, having smooth and shiny surface with normal overlying mucosa causing obliteration of buccal vestibule (Figure 1b). On the palatal aspect, intact swelling measuring 4 x 2cm was also seen on right side extends antero-posteriorly from mesial aspect of 13 to the distal aspect of 18 and medio-laterally from attached gingiva of 13 to 17 region 1cm away towards mid palatal suture (Figure 1c). On palpation, swelling was firm, non tender and compressible in nature with grade II mobility of 14, 15 and 16. Provisional diagnosis of Fibrous Dysplasia was given. The panoramic radiograph revealed a well-defined, unilocular, mixed radiopaque - radiolucent area with a ground glass appearance. Obliteration of right maxillary sinus was seen along with divergence of roots in relation to 14, 15 and 16 (Figure 2b). Correlating history, clinical examination and radiographic findings, a differential diagnosis of Fibrous dysplasia, Focal Cemento-osseous dysplasia and Ossifying fibroma was made. Incisional biopsy was done and the sample was sent for histopathological examination. The gross specimen consisted of a piece of tissue which was 2x1 cm in size, reddish brown in color and soft in consistency. The histological findings showed thin strands and small islands of odontogenic epithelium scattered within cellular, fibrous connective tissue stroma containing plump fibroblasts interspersed with bundles of collagen fibers. The stroma was vascular with few multinucleated giant cells and foci of calcification in it (Figure 3). Based on the clinical, radiographic and histologic findings, a diagnosis of central odontogenic fibroma was made and categorized as epithelium-rich or WHO type. Based on this diagnosis surgery was performed and tumor was enucleated followed by curettage and patient revealed uneventful recovery. Then prosthetic rehabilitation was initiated

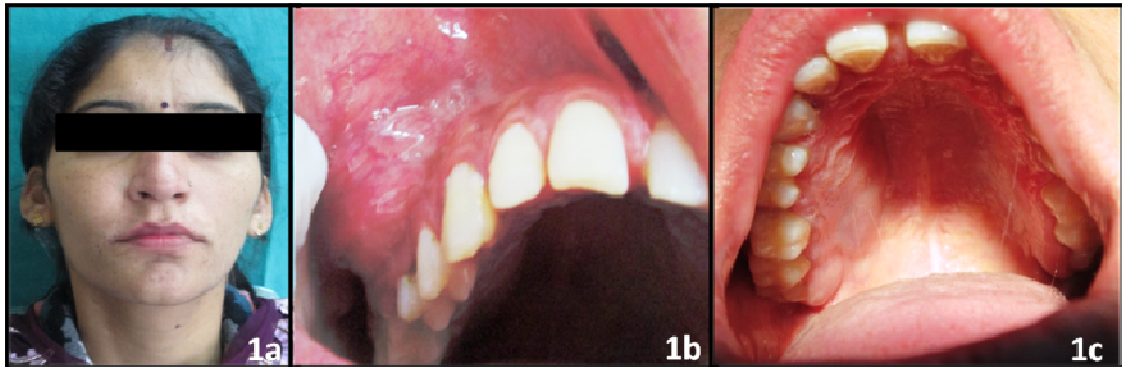


Figure 1: a) Extra-oral solitary diffuse swelling present on right side of the face; b) Swelling in relation to distal aspect of 13 to 16 causing obliteration of right maxillary buccal vestibule; c) Palatal swelling involving attached gingiva of 13 to 17 regions.

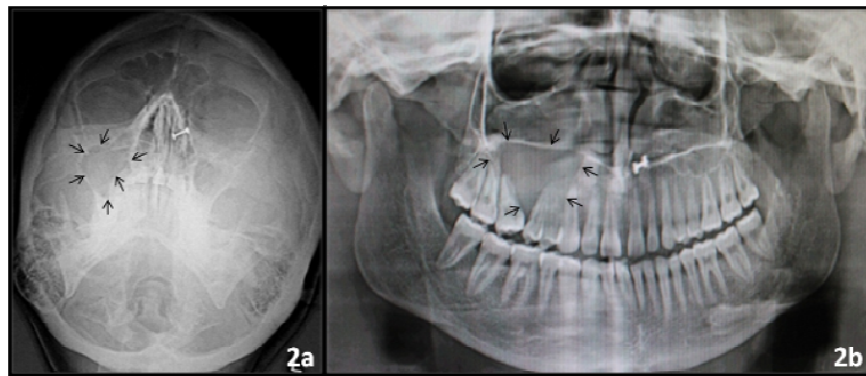


Figure 2: a) PNS view with obliteration of right maxillary sinus with areas of radiopaque structures in the centre of the lesion; b) Mixed radiolucency with divergence of roots in relation to 14, 15 and 16 with a ground glass appearance.

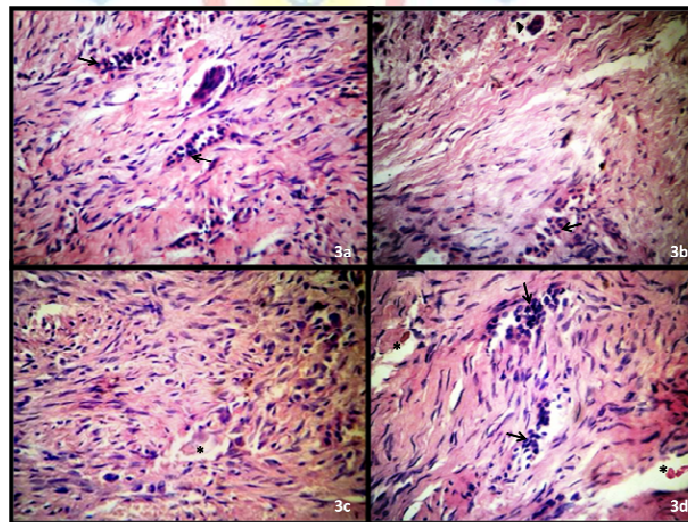


Figure 3: a) Photomicrograph showing numerous islands and strands of odontogenic epithelium (arrow); b) Few multinucleated giant cells (arrow head); c) & d) Foci of calcifications (asterisks) within cellular fibrous stroma. (H&E, 40x).

Table 1: Differential diagnosis: Odontogenic fibroma and similar fibrous jaw lesions

Features	Central odontogenic fibroma	Fibrous dysplasia	Cemento-osseous dysplasia (COD)	Cemento-ossifying fibroma/Ossifying fibroma
Etiology	Benign neoplasm of mesenchymal origin derived from remnants of the odontogenic apparatus	Somatic mutation in the GNAS1 gene	Derived from elements within the periodontal ligament space	True benign neoplasm of bone
Clinical findings	Age: b/w 4-80 years (Mean- 40 years)	Age: 2 nd - 3 rd decades	Age: 3 rd - 5 th decades	Conventional OF: 3 rd - 4 th Juvenile OF: Aggressive: < 15 years Psammomatoid: (5- 15 yrs) Trabecular: (8.5-12 yrs)
	Female predilection	Female	Female predilection	Female predilection
	Affects either mandibular (posterior) or maxillary(anterior) region	Affect maxilla more commonly than mandible.	Periapical COD (PA-COD)- Anterior mandible Focal COD(Fo-COD) and Florid COD (Fl COD Posterior mandible	Affects mandibular posterior region.
	Slow growing, painless swelling causing cortical expansion	Diffuse, painless jaw swelling may be mono stotic or polyostotic	Asymptomatic, mild discomfort and / or jaw expansion.	Slow-growth, no association with root apices, may cause roots displacement.
Radiologic findings	Solitary, unilocular or multilocular radiolucencies with well-defined borders. About 10% to 15% of tumors may be speckled with small radiopacities.	Poorly defined radiolucency in early stage and acquires more radiopacities with maturity. Typically shows orange peel or ground glass appearance. Margins blend with surrounding bone.	PA-COD: circumscribed, non-corticated radiolucency, with time may exhibit radiopacity Fo-COD: irregular radiolucency/mixed radiographic appearance with well-defined borders. Fl-COD: numerous, irregularly shaped, sclerotic radiopacities admixed with diffuse, ill-defined, mixed radiographic areas.	Unilocular lesions with sharply defined smooth, corticated borders Early lesions show radiolucency, whereas later-stage lesions may manifest as central radiopaque masses surrounded by a radiolucent rim.
Histology	Simple type: myxoid to densely hyalinised stroma, oval to spindle-shaped fibroblasts. Foci of odontogenic epithelial rests, small, round calcifications WHO type: A more cellular and collagenous stroma that contains cords and islands of odontogenic epithelium. Acellular, round or ovoid, cementum-like or droplet-like calcifications.	Hypercellular and cytologically uniform fibrous stroma with delicate and irregularly shaped trabeculae of woven bone resembling Chinese character & lack osteoblastic rimming. Pattern of mineralization is uniform throughout the lesion.	Replacement of bone with cellular fibroblastic tissue within which there is deposition of bony trabeculae and/ or rounded cementum like masses. Lack encapsulation, lesional tissue blends with adjacent bone Haemorrhage is uniformly present throughout the tissue	Varying mineralization, Unenc-apsulated lesion having cell-rich fibrous stroma. Psammomatoid type - rounded acellular psammoma like bodies having basophilic centres with peripheral eosinophilic osteoid rim. Trabecular type- delicate, anastomosing bony trabeculae rimmed by osteoblasts, less prominent vascularity, haemorrhage at periphery of lesion.
Treatment	Curettage.	Active treatment is not necessary but some trimming of the bone may be required for cosmetic reasons.	No active treatment is required for COD. Curettage of infected/necrotic bony fragment if secondary infection occurs.	Complete surgical removal.

to restore function and esthetics. Patient was followed up for 15 months without any report of recurrence.

Discussion: Central odontogenic fibroma (COF) has been described as an unusual, slow-growing tumor of the jaw. It appears as an asymptomatic extension of the cortical plate of the mandible or maxilla.^[4] Unlike ameloblastoma, it is not locally invasive.^[3] Shafer et al considered odontogenic fibroma a distinct neoplasm with its own histopathologic and clinical features separating it from other odontogenic tumors.^[2] COFs usually occur in adults and shows a female predilection (M:F= 1:2.8).^[6] The sex and the age of the patient we described in this paper are consistent with the literature. The most common site for the presentation of COFs is the mandibular posterior (premolar/molar) region, while in the maxilla they are mostly observed in the anterior region^[1] unlike in our case where the lesion was present in maxillary posterior region and which was rare according to its site.

Usually, COF exhibits slow growing painless swelling that results in cortical expansion. Clinical symptoms such as pain and paresthesia are uncommon^[7] but in our case, patient presented with gradually increasing asymptomatic swelling which eventually became painful. In some cases, it presents as slowly enlarging hard swelling which is sometimes recognized by presence of slow growing diastema due to dislocation of adjacent teeth. The affected patients may have tooth mobility or displacement.^[8] In present case, the bony hard swelling was associated with mobility of involved teeth (14, 15,16).

Radiological examination showed that the odontogenic fibroma is an eminently unilocular radiolucency with well-defined margins and large size lesions tend to have multilocular appearance with poorly defined or diffused borders in rare case.^[7] In few

cases, radiopacity can be seen within a radiolucent area, as in our case, the lesion showed mixed radiopaque- radiolucent areas with glass ground appearance.^[9] The great variation and overlap in radiologic appearance of the COFs with other lesions means it should be carefully differentiated from similar fibrous lesions.(Table 1)^[2,5,10,11]. Histologically, the CODF is categorized into two types: the epithelium-rich type (WHO-type) and the epithelium poor type (Simple type). The epithelium rich type shows more epithelial islands or strands distributed in fibrous tissue. It contains mineralized material that has been interpreted as osteoid, cementum-like or dysplastic dentin. The epithelium poor type lesions are less cellular and the epithelial tissue is not necessarily presented. It is generally believed that odontogenic epithelium is not always presented in the COF and the WHO type occurs much less commonly than the simple type.^[8,12] In our case, thin strands and islands of odontogenic epithelium were seen scattered within cellular fibrous stroma. Foci of calcification and few multinucleated giant cells were seen along with odontogenic epithelium. Thus the diagnosis of central odontogenic fibroma i.e epithelium-rich or WHO type was made.

Different treatment recommendations were suggested in the literature for CODF. Recurrences have been reported, but are very uncommon. One piece enucleation of the lesion is possible because it has no attachment to a tooth root and bone but, a few cases, which have grown large, may necessitate tooth extraction to have more access for total removal.^[6] In our case, enucleation followed by curettage was done and the lesion was removed. Extraction of 15 and 16 was done followed by prosthetic rehabilitation to restore function and esthetics. The present case showed no sign of recurrence after 15 months of follow-up.

We retrieved pubmed and medline literature accessed on April 3, 2014 and we obtained 20 cases of central odontogenic fibroma-WHO type.

Conclusion: It is imperative that all fibrous lesions of the jaws should also be taken into account to arrive at final diagnosis. Keeping in mind that OF can mimic fibro-osseous lesions so, we recommend that vigilant comprehensive analysis of all the pertaining clinical, radiographic and histopathological aspects should be carried out. We also emphasize on the fact OF have tendency for recurrence so it should be followed up for long term to detect any signs of recurrence.

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