

## CASE REPORT

## Central odontogenic fibroma of the maxilla

V Veeravarmal, R Nirmal Madhavan, M Mohamed Nassar, R Amsaveni

Division of Oral and Maxillofacial Pathology, Rajah Muthaih Dental College and Hospital, Annamalai University, Tamil Nadu, India

**Address for correspondence:**

Dr. Veeran Veeravarmal,  
Division of Oral and Maxillofacial Pathology,  
Rajah Muthaih Dental College and Hospital,  
Annamalai University, Annamalai Nagar,  
Tamil Nadu - 608 002, India.  
E-mail: varmal.opath@yahoo.com

**ABSTRACT**

The central odontogenic fibroma (COF) is a rare benign odontogenic mesenchymal tumor of jaw bones. The World Health Organization (WHO) recognizes two variants of COF namely: 1) Epithelial-rich type (WHO) and 2) epithelial-poor type (simple type). Rare variants like ossifying COF, COF associated with giant cell lesions, and amyloid have been documented. This article presents a case of an epithelial-rich variant of COF in a 24-year-old female. It presented as a bony swelling of the maxilla and appeared as a mixed lesion in radiographs. Histopathology showed a highly cellular fibrous connective tissue stroma with plump fibroblasts and long strands of odontogenic epithelium exhibiting mild eosinophilic to clear cytoplasm. Numerous cementum-like hematoxyphilic calcifications of various sizes akin to dentin or acellular cementum were observed. We believe that clinical and radiographic features of this case may add valuable knowledge to the already existing literature.

**Key words:** Amyloid, central odontogenic fibroma, jaw bones

**INTRODUCTION**

Central odontogenic fibroma (COF) is a relatively rare benign odontogenic neoplasm of jawbones.<sup>[1]</sup> It has a diverse histopathology and a certain degree of controversy still exists regarding the diagnostic criteria. History indicates that a variety of conditions had been reported as odontogenic fibroma.<sup>[2]</sup> The recent World Health Organization (WHO, 2005) classification recognizes two subtypes of COF: 1) Epithelial-poor type (simple type) and 2) epithelial-rich type (WHO type).<sup>[3]</sup> There is no clear-cut margin between the two types at the microscopic level. The main aim of this report is to present a case of COF (epithelial-rich variant) and to compare its clinical, radiographic and histopathological features with the existing literature; and thus, significantly contribute to the latter.

**CASE REPORT**

A 24-year-old female patient came with the complaint of a swelling over the right cheek region for the past 1 year. There was a slow growing swelling in the middle one-third of the

face on the right side. It measured about 3 × 5 cm in size and obliterated the nasolabial fold. It was hard in consistency and asymptomatic. Intraoral examination revealed a well-defined, bony swelling measuring, about 3 × 5 cm in size in the right side of the maxilla, extending from canine to second molar in the anteroposterior direction. There was buccal cortical expansion and grade I mobility of 14 and 15 [Figure 1a].

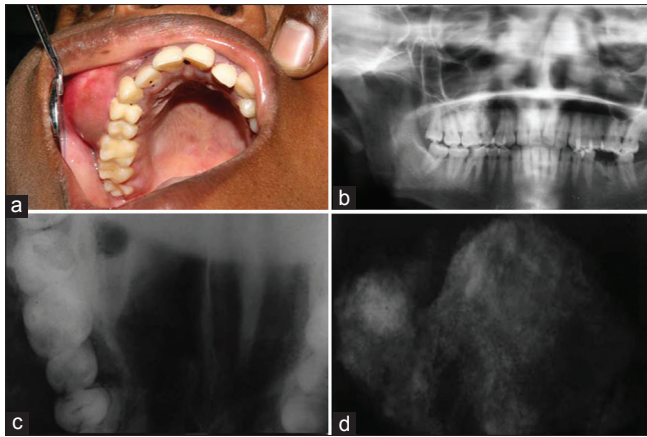
Orthopantomogram (OPG) showed an ill-defined, mixed radiolucent, and radio-opaque area; extending from the distal surface of canine to the distal aspect of second molar [Figure 1b]. Occlusal radiograph revealed a well-defined radiolucent lesion in the hard palate extending from the incisor to the second molar. It also showed buccal cortical expansion with radio-opaque flakes on the buccal aspect of 14, 15, and 16 [Figure 1c]. From the above findings, a provisional diagnosis of a fibro-osseous lesion was arrived at the clinical level.

The incisional biopsy showed a highly cellular, fibrous connective tissue stroma with plump fibroblasts and long strands, islands and nests of odontogenic epithelium seen scattered throughout the lesion and it exhibited mild eosinophilic to clear cytoplasm [Figure 2a-b, and d]. Numerous cementum-like hematoxyphilic calcifications of various sizes were seen [Figure 2]. These calcified structures demonstrated basophilic lamellated spherules to acellular eosinophilic materials akin to dentin or acellular cementum. In focal areas, loose fibrous connective tissue with fine collagen fibrils and stellate shape fibroblast were observed. The diagnosis of COF (epithelial-rich variant) was made because

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**Figure 1:** (a) A bony swelling in the right side maxilla with cortical expansion, (b, c) Orthopantomogram and occlusal view of maxilla showing a mixed lesion and cortical expansion, (d) X-ray of the excised specimen shows 'ground glass' appearance

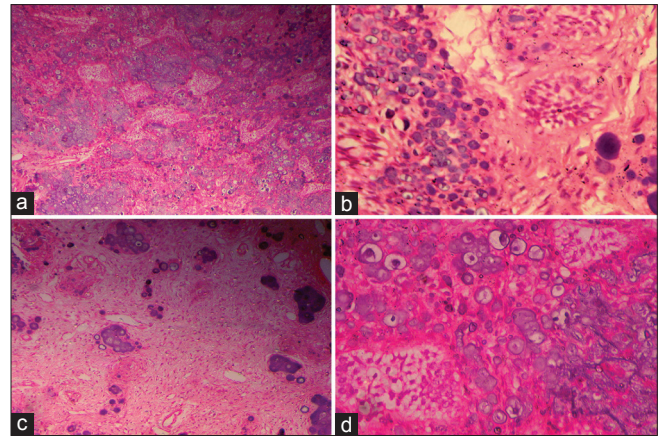
it was the predominant feature, although areas of the simple type were also seen focally.

The lesion was surgically excised and was not encapsulated. The cut surface appeared grayish-white and gritty. The X-ray finding of the excised specimen was suggestive of a fibro-osseous lesion [Figure 1d]. The histopathology of the postoperative tissue was consistent with the incisional biopsy diagnosis.

## DISCUSSION

COF is defined as a fibroblastic neoplasm containing varying amounts of apparently inactive odontogenic epithelium.<sup>[3]</sup> It is considered to be derived from mesenchymal tissue of dental origin, periodontal ligament, dental papilla, or dental follicle.<sup>[4]</sup> COF is a benign odontogenic neoplasm which remains incompletely understood.<sup>[5]</sup> Revised WHO histological typing of odontogenic tumors by Kramer (1992) included this entity under "odontogenic ectomesenchyme with or without included odontogenic epithelium".<sup>[6]</sup> The WHO panel decided to consider the simple type of odontogenic fibroma under the heading of myxoma. It is the most collagenous variant of the histologic spectrum of odontogenic myxomas, myxofibromas, and odontogenic fibromas.<sup>[5]</sup> It is suggested that the terminology "odontogenic fibroma WHO type" can be renamed as "odontogenic fibroma complex type" or "fibroblastic odontogenic fibroma" which could be considered as a more appropriate name.<sup>[6]</sup>

The literature review showed that COF is a very rare odontogenic neoplasm accounting for 0-5.5% of odontogenic tumors.<sup>[3]</sup> It is reported in wide age groups and frequently diagnosed in patients between the 2<sup>nd</sup> and 4<sup>th</sup> decades of life and in the current case it occurred in the 3<sup>rd</sup> decade. Female predilection is observed in many reports, similar like the present case. Perhaps equal distribution between males and females has also been reported by several authors.<sup>[2-4]</sup> Both



**Figure 2:** Hematoxylin and eosin (H&E) stained sections show an admixture of odontogenic epithelium and numerous basophilic and aggregates of cementum-like calcifications (a,  $\times 100$ , b,  $\times 400$ ). Cementum-like calcification, stellate-shaped fibroblasts in loose fibrous stroma with fine collagen fibres, and odontogenic epithelium (c,  $\times 100$ , d,  $\times 400$ )

the jaws have been affected equally. In the maxillary arch, it involves anterior segment and mandibular lesions affect the premolar and molar areas.

However, the present case manifested as a slow growing lesion in the posterior region of maxilla. COF causes bony expansion and displacement of the adjacent teeth.<sup>[7]</sup> Radiographically, the lesions are associated with the crown of an unerupted molar, premolar, or incisor tooth. COF usually appears as a unilocular radiolucency with well-defined borders, but may also exhibit a multilocular appearance with scalloped margins.<sup>[3-4]</sup> The presence of calcifications in the form of flecks was thus interpreted as a mixed lesion with a characteristic "ground glass" appearance. The lesion and the surrounding normal bone interface may be well-demarcated with sclerotic borders. The appearance may suggest encapsulation, but presence of a capsule has not been reported. In spite of that, many cases appeared to be infiltrative.<sup>[2,4,6,7]</sup> Presence of calcifications in the molar region in the radiographs was instrumental in making the surgeons considering it as a fibro-osseous lesion in the present case.

A review on 19 cases of odontogenic fibroma pertaining to the pathological features by Handlers, *et al.*, showed infiltrative margins. The tumor exhibited less to moderate cellularity in most of the cases. In addition, hyalinization and myxoid areas were observed in some cases. Pushing borders without encapsulation has been mentioned in few cases.<sup>[2]</sup>

Gardner made an attempt to classify COF into two variants namely: 1) Simple type and 2) WHO type. The basic difference between the simple and WHO type is that the stroma of the simple fibroma mimics that of a dental follicle from which it is probably derived. Histologically, the simple type exhibits relatively acellular delicate fibers which are interspersed with considerable amount of ground substance. On the other

hand, the stroma of the WHO type exhibits high cellularity. It occurs as fibroblastic strands which may be interwoven with less cellular areas. The epithelial rests are dispersed sparsely in the simple type. In contrast, the odontogenic epithelium is an integral component of the WHO type. The other difference between these two variants is the presence of foci of calcifications of the collagenous materials in the WHO type which are described as cementoid, osteoid, and dysplastic dentin by several authors.<sup>[1,5]</sup> The probable reason for dissimilar histological presentation of odontogenic fibromas is attributed to the tissue of origin.<sup>[5]</sup> Adalberto *et al.*, suggested that COF should be treated as a single entity that may be demonstrating two different histological patterns.<sup>[3]</sup>

The current classification of odontogenic fibroma by WHO (2005) is: 1) The WHO variant, and 2) the non-WHO variant. The WHO variant is considered as a mesenchymal odontogenic tumor and is comprised of two distinct cell types, a fibrous element, and an epithelial component that resembles dental lamina or its remnants. In contrast, the non-WHO variant lacks an epithelial component and is said to be a monomorphic fibroblastic tumor, purported to be of odontogenic mesenchymal origin and ostensibly derived from pulpal or follicular fibroblasts.<sup>[7]</sup>

In the present case, the predominant basophilic cementum-like calcifications and remnants of odontogenic epithelium in a highly cellular fibroblastic connective tissue background differentiated this epithelial rich variant of COF from the other type and also other odontogenic and nonodontogenic entities.

Many odontogenic fibromas in conjunction with the giant cell granuloma-like component have been reported over the past decades. In literature, it is suggested that this kinship between these two lesions represent a “collision” tumor as a result of synchronous occurrence of WHO type in the area of giant cell granuloma. Whether this is a hybrid or a biphasic tumor, remains unclarified.<sup>[6,8]</sup> The simple odontogenic fibroma in addition with pleomorphic fibroblasts and numerous calcifications has been reported. This histological variant was similar to the giant cell fibroma of the oral cavity.<sup>[6,9]</sup>

Rare variants like COF with ossification and with amyloid-like protein deposits have also been mentioned in the literature. Mostly the radiographic presentations of COF with ossification were interpreted as a mixed lesion. This variant showed intimately admixed odontogenic epithelial islands with tumor trabeculae. The fibrous connective tissue was described as acellular with monomorphic fibroblast nuclei and the ossifications were either irregular or ovoid in nature.<sup>[7]</sup> The current case demonstrated radiographically a mixed lesion in the posterior segment of maxilla and histologically it exhibited numerous calcifications. No evidence of ossification within the lesions was noted, which is necessary for the diagnosis.

The amyloid/dendritic cell which were associated with odontogenic fibroma histologically appeared with COF in addition to amyloid deposits. These deposits are Congo red positive and are characterized as ovoid or globular acellular hyalinised structure in the fibrous stroma and observed in the periepithelial regions. A variant called as odontogenic fibroma-like hamartoma/enamel hypoplasia syndrome is characterized by multiple unerupted posterior teeth with enlarged pericoronal radiolucent areas in association with generalized enamel defects. Under the microscope, these lesions exhibit similar presentations like COF. The histopathology demonstrated two types of calcification: Type A which presented as small calcifications or coalesced into large bodies which resembled the calcifications observed in our case and Type B which exhibited polarized calcifications with fibrillar tufts in the periphery. The current case was not in association with impacted teeth.<sup>[7,10]</sup>

The intraosseous fibrogenic tumors like myxoma and desmoplastic fibroma are considered as differential diagnoses. Gardner has emphasized that the clinical behavior of odontogenic myxoma is different from simple odontogenic fibroma. The simple type behaves like an expansile lesion, whereas the odontogenic myxoma infiltrates into the surrounding bone. The presence of epithelial islands is a prerequisite for the diagnosis of COF, whereas it is not a frequent finding in odontogenic myxoma.<sup>[5]</sup> In intraosseous fibrous tumors, the absence of odontogenic epithelium in addition to delicate collagen fibers include the diagnosis of epithelial-poor type COF.<sup>[3]</sup> The current case exhibited such an area focally. However, predominance of odontogenic epithelium and numerous cementum-like calcifications convinced us to give a confirmatory diagnosis as an epithelial-rich variant. The ossifying fibroma is ruled out by the absence of ossifications and with the presence of odontogenic islands and sheets.<sup>[3]</sup>

The cellular and collagenous stroma is the characteristic feature of desmoplastic fibroma. Other histopathological features like absence of odontogenic rests and lack of bone forming potential help to differentiate this intrabony neoplasm from COF.<sup>[3,5]</sup> On application of a panel of immunohistochemical markers, five cases of COF have been reclassified as intraosseous fibrogenic tumors like myofibroma, solitary fibrous tumor, myxoma, and nerve sheath tumor.<sup>[7]</sup>

Adalberto analyzed 14 cases of COF by subjecting it to various markers. Stellate and spindle shaped connective tissue cells in all the COF were immunoreactive for vimentin and there was focal positivity for  $\alpha$ -smooth muscle actin in some cases. The epithelial islands were positive for CK, AE1/AE3, CK5, CK 14, CK19, and 34BE12. Langerhan's cells were demonstrated within the epithelial islands which were immunoreactive for S-100 protein and CD1a. The giant cells of hybrid tumors showed clear positivity for CD68, and occasionally mononuclear stromal cells in some cases. The anti type IV collagen was positive in areas of eosinophilic globules

which were seen within the odontogenic islands.<sup>[3]</sup> COF possess limited growth potential, therefore, the recommended treatment is enucleation or curettage.<sup>[2,6]</sup>

## CONCLUSION

The epithelial-rich type COF exhibiting calcifications radiographically has to be considered as a differential diagnosis of mixed lesions of jaw bones. Despite of its low recurrence rate, a postoperative follow-up is needed. The clinical and histopathological findings of the present case widen the horizons of the existing literary mass, while recording precious knowledge of a rare condition to posterity.

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