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CASE REPORT

AMBIGUITY IN DIAGNOSIS: A CASE REPORT OF CENTRAL ODONTOGENIC FIBROMA

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ABSTRACT

Central odontogenic fibroma is a mesenchymal odontogenic tumor comprising less than 5% of all the odontogenic tumors. This tumor tends to occur in a wide age range with no gender predilection. Several attempts have been carried out by various authors to simplify and classify this benign tumor. The rarity and lack of specific clinic and histologic criteria have rendered the diagnosis of this tumor challenging. Also, the clinical and radiographic features mimics several other odontogenic and non-odontogenic entities. However, a biopsy is mandatory to diagnose this type of odontogenic tumors. We describe a case of central odontogenic fibroma occurring in the posterior mandible of a 13 year old male patient.

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INTRODUCTION

World health organization (WHO) describes central odontogenic fibroma (COF) as "a rare neoplasm of mature fibrous connective tissue, with variable amounts of inactive-looking odontogenic epithelium with or without evidence of calcification" (Wright *et al.,* 2017). The rarity of this mesenchymal odontogenic tumor has led to much controversy about this tumor. Most of the tumors reported are associated with coronal or radicular portion of the tooth. Therefore an origin from dental follicle or periodontal ligament has been elucidated for COF (Gardner, 1996). In the previous WHO classification (2005), COF have been categorized into epithelium rich type and epithelium poor type according to histopathology (Philipsen *et al.,* 2005). In 2017 updated WHO classification, the simple/epithelium poor type is discarded as the consensus group believe it as a poorly defined and documented entity (Wright *et al.,* 2017). Most of the COFs presents as painless swelling which are identified only upon routine radiographic analysis. Both clinical features and radiographic interpretations are not characteristic in case of an odontogenic fibroma. They usually present in the posterior region of mandible with no sex predilection and a wide age range from 11 to 80 years. In radiographs, they can either appear as small unilocular radiolucencies or large multilocular ones (Philipsen *et al.,* 2005). Therefore, a confirmed diagnosis of COF can be given only with biopsy.

CASE REPORT

A 13 year old male patient reported to the dentist for orthodontic correction of his teeth. On clinical examination, a deciduous second molar with preshedding mobility was noted. Radiographic examination revealed a deciduous second molar preventing the eruption of permanent second premolar on the right side of the mandibular arch. A small unilocular radiolucency of 1cm diameter was noted enclosing the crown of the unerupted premolar. The well-defined border of the radiolucency was seen attached to the CEJ of the tooth mimicking a dentigerous cyst. An excisional biopsy with extraction of both deciduous second molar and permanent second premolar was done. Histopathological examination revealed a fibrous connective tissue stroma containing proliferating odontogenic epithelium. The odontogenic epithelium consisted of cuboidal cells with clear to pale eosinophilic cytoplasm and hyperchromatic nuclei arranged in nests, cords and strands (Figure 1 and 2). Plump stellate like cells in a loosely arranged matrix resembling primitive mesenchyme were also evident in areas. Diffuse areas of hyalinization (Figure 3) and numerous foci of calcification were also noticed. A final diagnosis of odontogenic fibroma was given based on the histopathological evidence. The post-surgical period was incidence free with the patient undergoing regular follow up for the last one year without any signs of recurrence.

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Table 1. Different classifications of COF

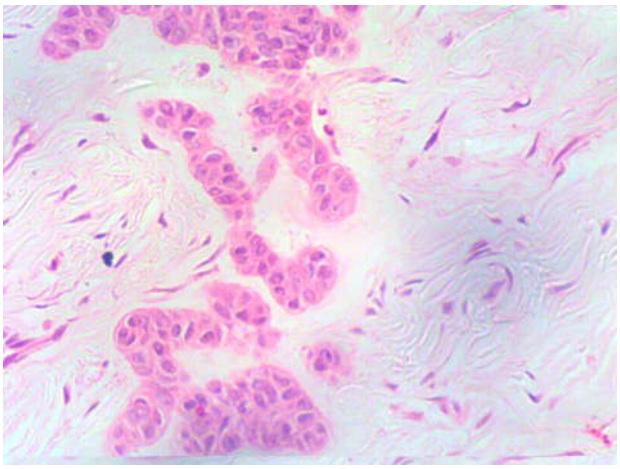
Classifications	Histologic types		
Gardner, 1980	Hyperplastic dental follicle	Simple type	WHO type
WHO, 1992		Simple	Complex
WHO, 2005		Simple type/Epithelium poor	WHO type/Epithelium rich
WHO, 2017	Epithelium poor type is discarded *COF is defined as a rare neoplasm of mature fibrous connective tissue, with variable amounts of inactive-looking odontogenic epithelium, with or without evidence of calcification*		

Table 2. Differential diagnosis of COF

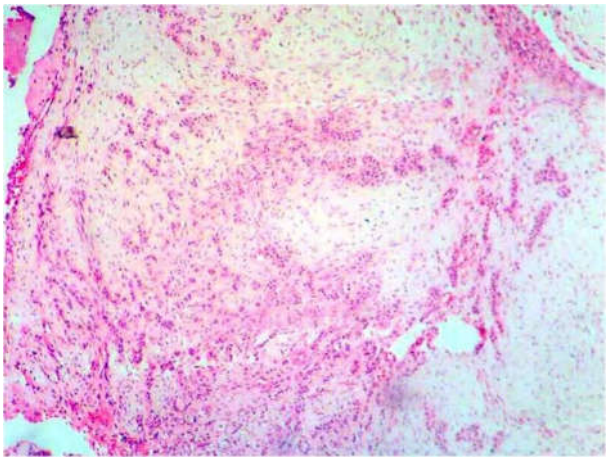
Lesion	Clinical features	Distinguishing features
Central odontogenic fibroma	2nd – 3rd decade Slight female predilection Posterior mandible and anterior maxilla	Nests and strands of inactive-looking odontogenic epithelium in a collagenous stroma calcifications can be found
Demoplastic fibroma	2nd decade Female predilection Posterior mandible	Interlaced bundles and whorled aggregates of densely collagenous tissue containing uniform spindled and elongated fibroblasts/myofibroblasts No epithelial rests or calcifications
Odontogenic myxoma	4th decade Female predilection Molar - premolar region of mandible	Spindle shaped cells in a rich myxoid stroma with few collagen fibrils. Presence of calcifications
Ameloblastic fibroma	1st-2nd decade Slight male predilection Posterior mandible	Branching and anastomosing proliferative odontogenic epithelium with peripheral rim of columnar cells in a primitive connective tissue stroma No hard tissue formation

Table 3. Differentiating features between a dental follicle and COF

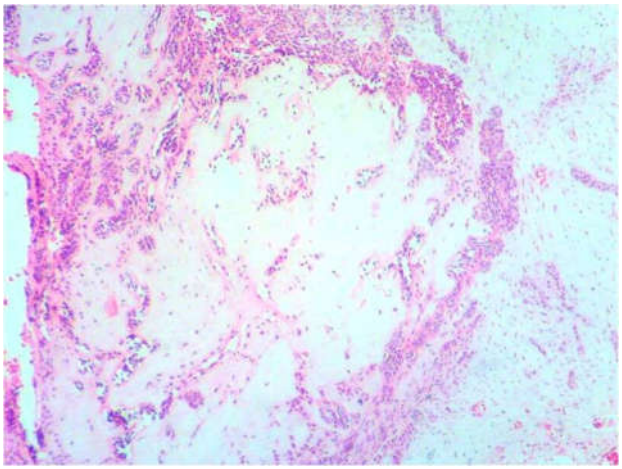
Lesion	Radiographic features	Histopathologic features
Enlarging dental follicle	Semicircular pericoronal radiolucencies <4mm More symmetric	Scattered odontogenic epithelial remnants Reduced enamel epithelial lining in the inner aspect Myxoid stroma
Central odontogenic fibroma	Pericoronal radiolucency >4mm Less symmetric Occasional findings: Root resorption and tooth displacement	Strands and cords of epithelial islands Absence of reduced enamel epithelium Fibrous stroma



Picture 1. H&E section shows odontogenic epithelial cells arranged in nests, cords and strands. (10X magnification)



Picture 2. H&E section shows odontogenic epithelial cells arranged strands in a mesenchymal stroma (40X magnification)



Picture 3. H&E shows hyalinized areas within the tumor mass (10X magnification)

DISCUSSION

COF is explicated by WHO as a rare, benign odontogenictumor of mesenchymal origin. The term central is used since an extraosseous counterpart exists for this odontogenictumor; designated as peripheral odontogenic fibroma. However, both the variants have similar histopathological features. COF is thought to arise from dental papillae or periodontal membrane tissue as they are frequently associated with crown or root portion of a tooth. However, Gardner in 1980 described three entities which comes under the same spectrum of related lesions - a hyperplastic dental follicle; a simple type with varying collagenous fibrous connective tissue containing nests of odontogenic epithelium; and a WHO type with varying amounts of odontogenic epithelium, dysplastic dentine or cementum-like tissue.[2] The 2005 WHO classification recognizes two variants of odontogenic fibroma; epithelium rich type (WHO type) and epithelium poor type (simple type) (Philipsen *et al.,* 2005). In the simple type, more of collagenous stroma, sometimes myxoid, is present with little or no odontogenic epithelium. Many authors have suggested the elimination of simple type and describing COF as a single neoplasm as it is rarely and ambiguously reported (Wright *et al.,* 2005; Adalberto *et al.,* 2011; Handlers *et al.,* 1991). However, the recent 2017 updated WHO classification discards the epithelium poor/simple type of COF (Table 1). The extreme rare nature and the ill-defined histopathologic features of simple type has prompted the consensus group to discard this variant (Wright and Vered, 2017). COFs usually occur as asymptomatic swellings or as accidental findings in routine radiography. Many authors have proposed an equal predilection among males and females; however few also suggest a female proclivity (Balaji *et al.,* 2015). It occurs in a wide age range but second to fourth decades are considered as peak ages. Both jaws are equally affected, however anterior maxilla and posterior mandible are seen to be the common sites. In our case, patient was a male in second decade of life and it occurred as an asymptomatic lesion diagnosed on a routine OPG. Radiographically these lesions do not have any diagnostic features; they can appear as unilocular or multilocular radiolucencies or as mixed lesions associated with the crown of an unerupted tooth. Root resorption and tooth displacement are occasional findings in some radiographs. Radiopaque flecks can suggest either calcifications or dentinoid materials or both.

In the radiograph, our case revealed a unilocular radiolucency associated with the crown of an unerupted second premolar tooth. The existence of ambiguity in differentiating a dentigerous cyst from COF, radiographically, was reported in a number of cases in the past. The most likely differential diagnosis includes dentigerous cyst, keratocystic odontogenic tumor (odontogenic keratocyst), unicystic ameloblastoma, ameloblastic fibroma and enlarged follicular space. Generally it is difficult to differentiate between an enlarging follicular space and many of these entities. A clue to diagnosis in such cases can be derived from radiographs. Only when the diameter of a pericoronal space is smaller than 2.5mm on an intraoral radiograph and smaller than 3mm on a panoramic radiograph, a diagnosis of follicular space can be considered.[8] According to Adalberto *et al.* pericoronal radiolucencies of dental follicles seldom exceed 4mm in diameter and are more symmetric than COF's (Adalberto *et al.,* 2011). COF can present with rare histologic components like ossification, amyloid like protein deposits, giant cell granuloma like components, pleomorphic fibroblasts etc. About 50 cases of a hybrid variant of COF and central giant cell granuloma have been described in the literature. Several theories have been proposed for the bizarre occurrence of this hybrid lesion like (a) collision tumor, (b) induction of odontogenic component by the production of cytokines and chemokines in a giant cell granuloma (c) giant cells are formed in response to trauma or other stimulus in the primary COF tumor. However, more studies need to be conducted at molecular level to detect the pathogenesis of this hybrid tumor (Upadhyaya *et al.,* 2018). Another variant, odontogenic fibroma-like hamartoma/enamel hypoplasia syndrome characterized by multiple unerupted posterior teeth with enlarged pericoronal radiolucent areas and generalized enamel defects has also been described (Eversole, 2011). In our case, though there was an unerupted tooth, no generalized enamel defect was found clinically. Another interesting finding reported by Chandreshekar C et al was the appearance of numerous mast cells in the connective tissue stroma of COF. A possible pathogenesis of C-kit overexpression by the stromal fibroblasts and resultant release of chemotactic factors causing an accumulation of mast cells were elucidated by the authors in this case report (Chandrashekar *et al.,* 2018).

Shimada *et al* reported a rare case of COF in a 14 year old patient with nevoid basal cell carcinoma syndrome (Shimada *et al.,* 2018). Presence of unusual histopathologic components like pleomorphic fibroblasts, giant cells, mast cells etc, and association with other lesions like giant cell granuloma renders COF an exceptional odontogenic tumor. Though, the recent WHO classification has made the histopathologic diagnosis of COF much simpler this rare odontogenic tumor needs to be differentiated from other histologically similar odontogenic tumors like ameloblastic fibroma, odontogenic myxoma and desmoplastic fibroma (Table 2). Also, it is essential to differentiate between an enlarging dental follicle and COF histologically, as many cases of former entity have been misdiagnosed as COF (Table 3). In our case, numerous odontogenic islands and calcifications in a fibrous stroma prompted us to reach the final diagnosis of central odontogenic fibroma. Adalberto *et al.* has done a panel of immunohistochemical markers on 14 cases of COF and found that the epithelial islands were positive for cytokeratin AE1/AE3, CK5, CK 14 and CK19 while the connective tissue stroma is positive for vimentin in all cases (Adalberto *et al.,* 2011). Enucleation with vigorous curettage is the suggested treatment for COFs. Although, the recurrence rates are very low, complete removal of the lesion should be ensured in all cases. It is reported that maxillary lesions with multilocular aspects and cortical bone perforation tend to show a higher recurrence rate (Pontes *et al.,* 2018).

Conclusion

This rare and contentious mesenchymal odontogenic tumor needs to be explained in detail for better diagnosis and understanding of its true nature. Cytogenetic and biomolecular studies are the need of the hour to shed light on the behaviour and pathogenesis of these types of less known odontogenic tumors.

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