

Research Journal of Pharmaceutical, Biological and Chemical Sciences

Importance of Histopathology in Diagnosis of Central Odontogenic Fibroma – A Case Report.

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ABSTRACT

Central odontogenic fibroma is an unusual and controversial odontogenic tumour. We present a case of central odontogenic fibroma involving premolar- molar region of mandibular left quadrant. The lesion was asymptomatic with well defined borders, without resorption and displacement of the adjacent teeth. The case was initially diagnosed as lateral periodontal cyst based on clinical and radiographic findings, but on histopathological diagnosis it turned out to be central odontogenic fibroma. Hence, this case report gives readers a bird's eye view of the significance of role of histopathology in diagnosis.

Key words: mandible, odontogenic cysts, odontogenic tumours.

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INTRODUCTION

Odontogenic fibroma is a rare, benign, slowly growing, noninfiltrating odontogenic tumour which is mainly composed of proliferating fibrous tissue with varying amounts of inactive appearing odontogenic epithelium [1]. There are two types of odontogenic fibroma: an intraosseous or central odontogenic fibroma (COF) and an extra osseous or peripheral odontogenic fibroma, later being more common compared to COF by a ratio of 1.4:1 [2].

According to 1992 WHO publication, COF is odontogenic tumor of “odontogenic ectomesenchyme with or without included odontogenic epithelium” [3]. It is derived from mesenchymal tissue of dental origin like periodontal ligament, dental papilla or dental follicle.² In 1980, Gardner classified this entity into two histologic subtypes; Simple and WHO type, which is being used by most of the authors [4].

COF is one of the most ill defined lesions, as very few cases have been reported in the literature. Regezi and associates found no cases of odontogenic fibroma among 706 odontogenic tumours retrieved from 54,534 oral biopsy specimens [5]. Buchner and associates found 1,088 cases of COF (1.2%) among 91,178 cases accessed [6]. Mosqueda and associates found five cases of COF (1.4%) out of 349 cases of odontogenic tumours. Multiple COF lesions were also reported in a rare syndrome associated with enamel dysplasia [1].

Clinically COF could appear as an asymptomatic expansion of buccal or lingual cortical plates. Radiographically it appears as unilocular to multilocular radiolucency, surrounded by well-defined border most of the times [7]. Histopathologically they vary from hypercellular fibrous tumours to one with moderate cellularity and varying amount of inactive odontogenic epithelium seen as strands and cords.¹ The concept of COF till date remains a matter of argument [4].

We present a case of COF- simple type that was clinically asymptomatic and radiographically mimicked lateral periodontal cyst unlike reported cases.

Case Report

A 51 year old male patient reported to the O.P.D of Sri Siddhartha Dental College, Tumkur with a chief complaint of deposits on his teeth. Intra oral examination revealed generalized periodontal pockets. Orthopantomograph was advised for periodontal reasons, which revealed a well defined, corticated unilocular radiolucency measuring about 1.5cm in diameter (fig-1), extending from mesial root of 36 to apex of 35. Resorption and displacement of teeth were not evident. There was no history of pain or swelling and his medical history was non contributory. Periodontal pocket of 7mm depth was present in relation to 36 and grade I mobility with respect to 35. Both 35 and 36 were vital. A provisional diagnosis of lateral periodontal cyst in relation to 36 was considered.

Subperiosteal flap was raised and surgical window was prepared between 35 and 36 to enucleate the lesion. 10ml venous blood was drawn from patient, it was centrifuged to obtain platelet rich fibrin which was mixed with osseo-graft and was filled in the defect.

The enucleated single soft tissue specimen measured 3x1x1cm, whitish tan in colour, firm and didn't reveal any cystic area on grossing. Microscopic examination showed collagenous fibrillar stroma with prominent fibroblasts which were plump, spindle shaped having prominent nuclei (fig-2). Many inactive odontogenic epithelial islands were found in the form of strands and nests (fig-3 and fig-4). No foci of dystrophic calcifications were observed. After co-relating clinical, radiographic and histopathologic findings the case was diagnosed as Central odontogenic fibroma (simple type).

DISCUSSION

COF is a central tumour of jaw which is most ill-defined and least understood among odontogenic tumours [8]. According to WHO classification 2005 the tumour has been divided into two types - epithelium poor and epithelium rich [1]. Epithelium poor type is suggested to be derived from dental follicle, while epithelium rich type is considered to arise from periodontal ligament.² COF occurs in age range of 11 to 66 years,³ and shows female predilection of 2.8:1 [9]. Zimmerman and Dahlin found that 67% of the cases

reported were between 10 to 29 years of age [8]. In our case the tumour occurred in a male patient at his sixth decade.

Clinically, COF occurs mostly in anterior region of maxilla or in premolar and molar region of mandible [8]. Most frequently observed clinical signs are swelling, displacement of adjacent teeth leading to slowly growing diastema [7]. However, the case reported here didn't show any swelling, nor displacement of teeth and neither cortical expansion.

The radiological appearance has not been clearly described and exhibits great variation (table-1). It could range from unilocular to multilocular radiolucency with well-defined borders varying from <1cm to >5cm at longest diameter [1]. Unilocular tumours are mostly <3cm, while multilocular lesions are more than 3cm [1]. Sometimes COF may exhibit a mixed radiolucent/radiopaque appearance with poorly defined or diffused borders. Root divergence and root resorption of the associated teeth are common [8]. In our case, the lesion was unilocular with cortication showing no evidence of root resorption and displacement of adjacent teeth. The relation of radiolucency to the adjacent teeth gave an impression of lateral periodontal cyst.

The etiology of COF is not clear. It is considered to arise from odontogenic ectomesenchyme - from periodontal ligament when tumour develops around the roots and from dental sac when it is associated with crown of an unerupted tooth [1]. In 1980, Gardner attempted to clarify the criteria for diagnosis of COF [3]. He differentiated between three central lesions: Hyperplastic dental follicle, Simple COF and WHO type of COF.

He described Hyperplastic dental follicle as "well circumscribed mass of fibrous tissue that surrounds crown of an unerupted tooth and has a radiographic appearance similar to small dentigerous cyst, and this lesion should not be considered as COF" [3].

Simple type of COF is defined as "An expansile, noninfiltrating connective tissue lesion resembling dental follicle. It is relatively acellular, the fibres being quite delicate, and there is a considerable amount of ground substance yielding a fibromyxoid quality. It may exhibit inactive looking rests of odontogenic epithelium but they are seldom numerous. Occasionally, nondescript calcifications are found" [3].

WHO type of COF is defined as "A benign neoplasm composed of cellular connective tissue. It often occurs in fibroblastic strands that are interwoven with less cellular areas in which numerous small blood vessels are present. Foci of calcified collagenous matrix, resembling dysplastic cementum, osteoid or atubular dysplastic dentin often occur. Islands or strands of inactive looking odontogenic epithelium are integral component of this type of COF, they are usually conspicuous. A clearly defined capsule is not encountered" [3].

Our case was diagnosed as central odontogenic fibroma of simple type. Lateral periodontal cyst was considered in provisional diagnosis as associated teeth were vital and unilocular radiolucency was present lateral to root of 36, but there was no cystic lesion evident on histological examination. Differential diagnosis for the case can be categorized into two groups - tumours which contain odontogenic epithelium and tumours which don't contain odontogenic epithelium [1]. (table-2)

Since our case showed presence of odontogenic epithelium, only lesions which contain odontogenic epithelium were considered. Hyperplastic dental follicle shows presence of reduced enamel epithelium and odontogenic epithelium similar to COF [1], but the size, location and radiographic findings should exclude hyperplastic dental follicle. Odontogenic myxoma consists of stellate and spindle shaped cells seen in a myxoid background. Clinically it is more aggressive and invasive, thus it was excluded [1,9]. Ameloblastic fibroma shows odontogenic epithelium in the form of strands and islands, but consists of peripherally placed cuboidal cells and centrally placed stellate reticulum like cells which differs noticeably from the inactive looking cell rests found in COF [1,3]. AOT consist of duct like structures along with eosinophilic amorphous material, is associated with an unerupted tooth in maxillary anterior region [9].

Treatment for COF is surgical enucleation followed by vigorous curettage. Recurrence rate is usually considered low [3]. Heimedal and associates reported a case which reoccurred nine years post-surgery [14]. Dunlop and associates reported two cases without reoccurrence with ten years follow-up.¹ No recurrence was observed in our case on three years of follow up.

Table 1: Age, symptoms, location and radiological appearance in some of the cases reported

Authors & Year	Age In yrs	Gender	Symptoms	Jaw involved	Area of jaw involved	Tooth involved	Radiographic Findings
Kyung Soo Nah (2011) [2]	17	Male	Painless swelling	Mandible	Posterior	Right 3 rd molar region	Multilocular radiolucency with ill-defined borders.
Bruno Ramos et al (2011) [12]	7	Male	Asymptomatic	Mandible.	Posterior	Unerupted right primary 2 nd molar.	Well circumscribed pericoronal radiolucency with well-defined radiopaque rim
Khadka et al (2010) [13]	13	Female	Painless swelling	Maxilla	Posterior	Left 1 st premolar and 2 nd molar .	Well circumscribed radiolucency with cortication.
Bindu et al (2010) [10]	12.5	Female	Painless swelling	Mandible	Posterior	Right 2 nd premolar	Unilocular radiolucency associated with impacted mandibular 1 st and 2 nd molar.
Marco T Brazoa et al (2010) [9]	28	Male	Painless periodontal swelling	Mandible	Anterio-posterior	Right canine and 1 st premolar.	Unilocular radiolucency with displacement of teeth.
Ioanna Daskala et al (2009) [11]	71	Male	Painless gingival swelling.	Mandible	Anterio-posterior	Left canine and premolar region.	Multilocular radiolucency.
Hang Ping chuang (2008) [15]	20	Female	Asymptomatic	Mandible	Posterior	Left 1 st and 2 nd molars.	Unilocular radiolucency with well-defined border.
Khandekar SP et al (2007) [4]	12	Female	painful Swelling	Mandible	Posterior	right 2 nd premolar.	Well defined radiolucency with cortication
Ugo Covani et al (2005) [7]	26	Female	Painless gingival swelling.	Maxilla	Anterior	Right canine.	Unilocular radiolucency with cortication.
Eui-Hwan et al (2002) [8]	52	Female	Painless hard swelling.	Maxilla	Anterio-posterior	Left Canine up to the 2 nd premolar.	Multilocular radiolucency with cortication and displacement of teeth.

Table 2: Differential diagnosis for the case.

Tumours containing odontogenic epithelium	Tumours which doesn't contain odontogenic epithelium
Hyperplastic dental follicle	Myxofibroma
Odontogenic myxoma	Desmoplastic fibroma
Ameloblastic fibroma	Cemento ossifying fibroma
Adenomatoid odontogenic tumour	Low grade histiocytic sarcoma

Figure 1: Orthopantomograph showing unilocular radiolucency measuring 1.5cm in diameter extending from apex of 35 to mesial aspect of 36.



Figure 2: photomicrograph shows collagenous fibrillar stroma with numerous epithelial islands (Hematoxylin & Eosin stain, 40x).

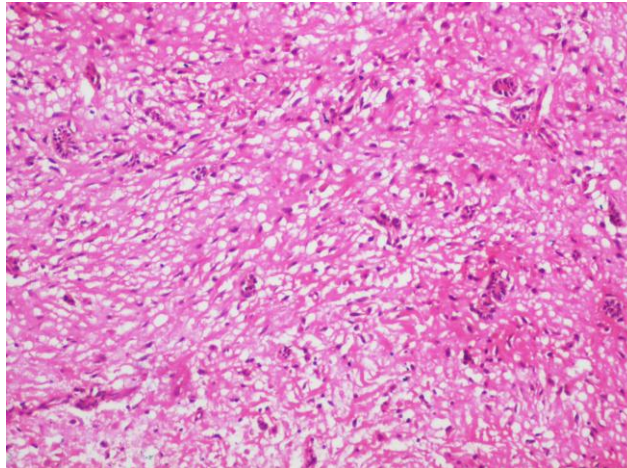


Figure 3: Photomicrograph shows fibrous connective tissue with nest and strands of inactive looking odontogenic epithelium. (Hematoxylin & Eosin stain, 200x).

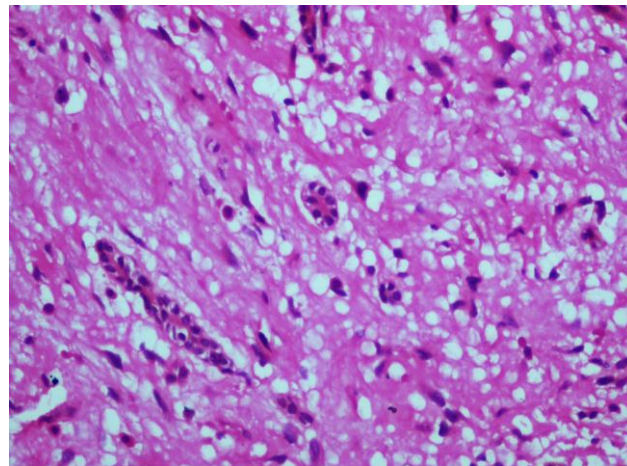
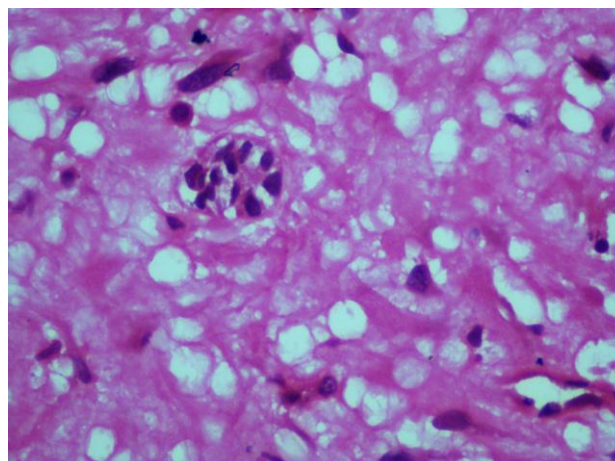


Figure 4: Photomicrograph showing odontogenic epithelial cell nest (Hematoxylin & Eosin stain, 400x).



CONCLUSION

The reported case was clinically asymptomatic, radiographically mimicked lateral periodontal cyst, but histopathologically it turned out be Central odontogenic fibroma of simple type. Due to limited number of COF

cases available in literature, it is difficult to establish diagnosis based on clinical and radiographic examination alone. Hence, the paper stresses on importance of histopathology in diagnosis. It is essential that the radiologists, maxillofacial surgeons and oral pathologists should assimilate all the pertinent information and come up with an accurate diagnosis for appropriate management.

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