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CENTRAL ODONTOGENIC FIBROMA: A CASE REPORT WITH IMMUNOHISTOCHEMICAL ANALYSIS

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ABSTRACT Odontogenic fibroma (OF) is a rare odontogenic tumor with a variable incidence that the World Health Organization (WHO) defines as a benign odontogenic neoplasm that arises from mesenchymal odontogenic tissue, comprising less than 5% of all the odontogenic tumors. It has been reported in patients aged 11 to 80 years old, with a mean age of 34 years and shows no gender predilection. It presents a diagnostic dilemma because its clinical and radiological features resemble other odontogenic and/or non-odontogenic tumours, and the differential diagnosis is based on histological examination. In this report, a 65 years old male patient is presented, who was diagnosed with central odontogenic fibroma of simple type from clinical and histopathological findings.

KEYWORDS : CD99; Immunohistochemistry; Ki-67; Mesenchyme; Odontogenic tumor

INTRODUCTION

Odontogenic fibroma (OF) is a rare tumor of odontogenic origin with variable percentages of incidence, regarded by the World Health Organization (WHO) as a benign odontogenic neoplasm derived from mesenchymal odontogenic tissue.[1] Two variants can be distinguished: an intraosseous or central type (COF) and an extraosseous or peripheral type (POF). Although initial reports indicated a slight predilection for the mandible, recent studies have reported equal proportions of cases in maxilla and mandible. In the maxilla, the lesion appears frequently to involve the anterior region, whereas in the mandible, the lesion tends to be in the posterior area. It has been reported in patients ranging in age from 11 to 80 years with a mean age of 34 years.[2]

COF is an uncommon benign neoplasm composed of varying amounts of inactive looking odontogenic epithelium embedded in a neoplastic mature and fibrous stroma.[3] Central odontogenic fibroma has been subdivided into simple type and the WHO/complex type. COF is a rare tumor with uncertain histogenesis.[4] The WHO variant is considered a mesenchymal odontogenic tumor and comprises two distinct cell types: a fibrous element, and an epithelial component that resembles dental lamina or its remnants. On the other hand, the non-WHO variant lacks an epithelial component and is a monomorphic fibroblastic tumor.[5]

The classical histological picture of COF comprises of mature collagenous tissue with variable amount of myxomatous stroma containing proliferating fibroblasts. Nests of odontogenic epithelium and calcified material may be seen dispersed in the connective tissue. Apart from these classical features, the occurrence of granular cells, giant cells and pleomorphic fibroblasts has also been reported.[6] Accounting for about 0.1% of all odontogenic tumours and 6.1% of all central odontogenic tumours, COFs present a diagnostic dilemma to the clinician and the pathologist.[7] Usually, the lesion is asymptomatic except swelling of the jaw.[8]

COF usually appears radiographically as a unilocular radiolucency with well-defined borders but may also exhibit a multilocular appearance with scalloped margins.[9] It sometimes presents as a radiolucent expansile multilocular tumor resembling ameloblastoma. Radiolucent lesions often associated with the peri-radicular area of erupted teeth. Around 12% of the COF may show radiopaque flecks.[10] Due to its rarity and radiolucent nature, it often gives clinical appearance resembling as an odontogenic cyst or tumor such as an odontogenic keratocyst, ameloblastoma, odontogenic myxoma and ameloblastic fibroma. Histopathological examination therefore is necessary to elucidate the exact nature of the lesion.[11]

The aim of this case report is to analyse clinical, radiographic, and histopathological features of central odontogenic fibroma (COF) with the present literature that enabled the correct differential diagnosis.

CASE REPORT

A 65 years old male reported with a chief complaint of pain and swelling in his left back tooth region since 6 months. Past family, dental, medical and habit history were non-significant and the lymph nodes were non-palpable.

Extra oral examination revealed facial asymmetry with swelling in the left zygoma region. Intraoral examination revealed swelling w.r.t. lateral wall of maxillary antrum. It was pale in color and the margins were indistinct. On palpation, the lesion was tender, and no discharge was seen on compression. The clinical differential diagnosis include odontogenic keratocyst, ameloblastoma, odontogenic myxoma, ameloblastic fibroma, and central giant cell tumor.

The radiographic examination revealed destruction and erosion of lateral maxillary antrum's anterior and lateral wall and part of zygomatic arch. Excisional biopsy was done and sent for the histopathological examination.

Gross examination of biopsy specimen showed multiple soft tissue bits collectively measuring 8.5x7.5x2mm, greyish-brown in color, irregular in shape and firm in consistency. The soft-tissue specimens were sectioned, and all the bits were processed following routine tissue processing protocols.

Microscopic examination revealed epithelial nests, strands and cords composed of polyhedral cells with central hyperchromatic nuclei suggestive of odontogenic epithelial cell rests in a dense mature fibrous connective tissue stroma. Focal areas showed squamous metaplasia within the odontogenic islands. Epithelial cells are isomorphic with vesiculated nuclei and few cells shows altered nuclear-cytoplasmic ratio. Plump stellate like cells seen in a loosely arranged matrix resembling primitive mesenchyme were also evident. Connective tissue stroma shows collagen fibres which were associated with plump fibroblast and mild chronic inflammatory cell infiltration such as lymphocytes and plasma cells. Blood vessels were lined by cuboidal endothelial cells.

Immunohistochemistry was performed to investigate the immunohistochemical profile of odontogenic epithelium of the tumor

and found to be positive for CD99 and Ki-67. The microscopic characteristics together with clinical findings confirmed the tumor to be central odontogenic fibroma (COF).

Table 1: Classifications Of Central Odontogenic Fibroma

Classifications	Histological Type		
Gardner (1980)	**Hyperplastic dental follicle**	**Simple Type**	**WHO Type**
WHO (1992)	Simple	Complex	
WHO (2005)	Simple/ Epithelium Poor	WHO/ Epithelium rich	
WHO (2017)	Intraosseous/ Central	Extraosseous/ Peripheral	

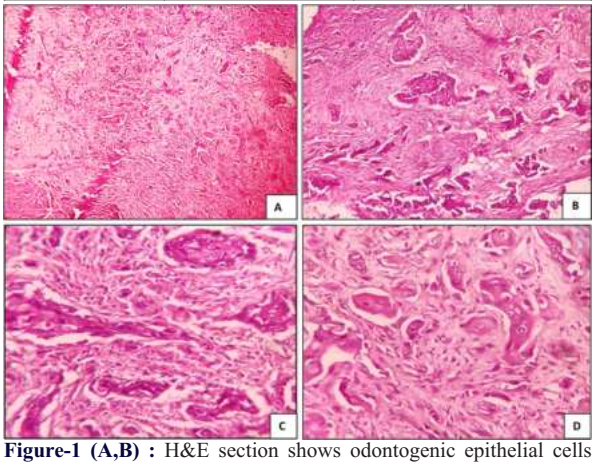

Figure-1 (A,B) : H&E section shows odontogenic epithelial cells arranged in nests, cords and strands in a mature fibrous connective tissue stroma. (A- 4x, B-10x magnification) **(C,D) :** H&E section shows epithelial cells with vesiculated nuclei and altered N:C ratio. (40x magnification)

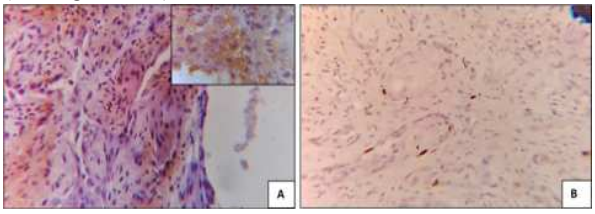

Figure-2: (A) The odontogenic epithelium and fibroblast-like cells in the stroma were positive for CD99. (40x magnification, IHC) **(B)** Few cells of odontogenic epithelium showed positivity for Ki-67. (40x magnification, IHC)

DISCUSSION

Odontogenic tumors constitute an unusually diverse group of lesions ranging from hamartomatous ones to benign and malignant neoplasms that arise from the odontogenic apparatus and their remnants. Histological characteristics and typing of these tumors is important for prediction of their biological behavior. Odontogenic tumors are unique due to their exclusive origin from tooth forming apparatus/ tooth germ even after cessation of odontogenic activity in the jaw bone. The classification of these tumors has been continuously evolving over the years.

Central odontogenic fibroma constitutes 0.1% of all odontogenic tumors. 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.[12] The 2005 WHO classification recognizes two variants of odontogenic fibroma; epithelium rich type (WHO type) and epithelium poor type (simple type). In the simple type, more of collagenous stroma, sometimes myxoid, is present with little or no odontogenic epithelium.[13] Many authors have suggested the elimination of simple type and describing COF as a single neoplasm as it is rarely and ambiguously reported.[14] However, the recent 2017 updated WHO classification discards the epithelium poor/simple type of COF. The extreme rare nature and the ill-defined histopathologic features of simple type has prompted the consensus group to discard this variant.[15](Table-1).

COF shows slight female predilection with peak incidence in the 2nd to 4th decades. The most common locations are the anterior region of the maxilla (71%) and posterior region of the mandible (73%). Clinically, COF tends to manifest as an asymptomatic swelling, although it can appear in a more aggressive way, resulting in displacement of teeth. The tumor occurs more frequently in the anterior maxilla and may result in a distinctive palatal cleft or depression, which is considered characteristic of this tumor.[5] Clinical differential diagnosis includes odontogenic cysts, ameloblastoma, adenomatoid odontogenic tumor, and ameloblastic fibroma. The current patient is male, lesion was central in the jaws (posterior maxilla) and exhibited a symptomatic lesion with slow persistent growth. Radiographically it is mostly unilocular, larger lesions show multilocular radiolucency with well-defined borders. They usually show more aggressive behavior with complications of resorption of teeth, radicular displacement of adjacent teeth, and expansion of cortical bones. The radiographic features of the COF are similar to other radiolucent lesions, such as traumatic bone cyst, ameloblastoma, odontogenic cysts, and central giant cell granulomas.[16]

Histologically, the epithelium-poor type is characterized by a fibromyxoid stroma with scattered odontogenic epithelial islands and cords. The epithelium-rich type, which is more frequently observed, consists mainly of cellular, fibrous connective tissue stroma where islands of odontogenic epithelium are an integral component. Occasional foci of calcifications that consist of osteoid, dentinoid or cementum-like materials may occur. The histological differential diagnosis of COF includes ameloblastic fibroma, desmoplastic fibroma, and myxoma.[17]

The treatment of COF involves conservative surgery through enucleation of the lesion as the lesion doesn't show tendency to undergo malignant transformation. The prognosis of the lesion is usually good with very few cases of recurrence reported in the literature. The cause of recurrence is thought to be incomplete removal of the lesion rather than the type of COF.[5]

CONCLUSION

Central odontogenic fibroma is a rare benign tumor of the tooth-forming apparatus. Due to its rare occurrence, these lesions may often be misdiagnosed with a periapical or a malignant lesion. COFs have peculiar histopathologic features. Because the presence of odontogenic epithelium is a requisite to confirm its diagnosis, immunohistochemistry can be helpful in epithelium-poor cases. Hence, assessment of clinical, radiological and histopathological findings help establish a correct diagnosis and help in therapeutic

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