Concurrent central odontogenic fibroma and dentigerous cyst in the maxilla: A rare case report

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Abstract
Central odontogenic fibroma (COF) represents a rare group of benign odontogenic tumor, while dentigerous cyst (DC), on the other hand, is the odontogenic cyst that encloses the crown of an unerupted tooth. A combination of COF and DC is rare and not reported in literature till date. The aim of this paper is to report a case of COF occurring concurrently with DC. The simultaneous occurrence of COF and DC raises the question of whether it is a collision tumor or their simultaneous occurrence is just a coincidence. In our case, the probable cause of simultaneous occurrence might be that COF would have displaced 23 resulting in the formation of DC.

Keywords: Central odontogenic fibroma, dentigerous cyst, simultaneous

INTRODUCTION
Central odontogenic fibroma (COF) is an elusive benign tumor because it accounts for <1.5% of all odontogenic tumors. It is believed to be derived from ectomesenchymal tissue of dental origin.\(^1\) Since it is a rare entity, the location, age and sex cannot be clearly determined. In general, it manifests as a painless swelling with slow and progressive growth pattern often with cortical plate expansion.\(^2\) Root resorption and displacement of adjacent teeth have also been reported in more severe cases.\(^3\) It is more commonly seen among patients in the third and fourth decades of life with female predilection with mandible being most commonly affected.\(^4,5\) Radiographically, COF appears as a unilocular or multilocular radiolucent area with a well-defined often sclerotic border.\(^2\) Histologically, it is defined as fibroblastic neoplasia that ranges from poorly cellular and myxoid without significant odontogenic epithelial component to cellular with abundant odontogenic epithelia and occasional foci of calcifications. The terms simple (epithelium poor) type and complex or the WHO (epithelium rich) type have been designated to those lesions.\(^6\)

Dentigerous cyst (DC) is defined as cyst that encloses the crown of an unerupted tooth by expansion of its follicle and is attached to its neck.\(^7\) It is the second most common cyst in the oral cavity and it is most commonly associated with impacted mandibular third molar.\(^7\) Clinically, the smaller cysts are usually asymptomatic and detected during routine radiographic examination. Larger cysts (>2 cm) may present with swelling, tooth displacement, mobility and sensitivity.\(^8\) Radiographically, DC presents as unilocular radiolucency with well-defined sclerotic border, surrounding the crown of an unerupted tooth.\(^7\)
CASE REPORT

A 14-year-old male presented with a complaint of an asymptomatic swelling in the left cheek for 6 months which gradually increased to the present size. His medical and family histories were noncontributory and there was no history of trauma. On extraoral examination, a diffuse swelling was present on the left middle third of the face causing facial asymmetry [Figure 1]. On palpation, the swelling was nontender and hard in consistency. Intraoral examination showed 3 cm × 5 cm swelling extending from the left upper permanent canine to the second permanent premolar which obliterated the vestibule [Figure 2]. Panoramic radiograph and computed tomography (CT) imaging were carried out. A panoramic radiograph [Figure 3] was obtained, which revealed the presence of a multilocular radiolucent lesion near the teeth 21, 22, 63, 24 and 25. Incidentally, another radiographic finding of an impacted tooth was also seen in the floor of maxillary sinus. CT image showed an expansile lytic lesion arising from the left maxilla approximately measuring 2.2 cm × 2.1 cm × 2.4 cm with few areas of calcifications. Thinning of adjacent bone was seen. It also revealed another lesion which appeared to be associated with impacted tooth present in the floor of maxillary sinus [Figure 4]. Differential diagnosis of adenomatoid odontogenic tumor associated with DC, keratocystic odontogenic tumor, ameloblastoma and odontogenic myxoma was made. A bony window was made and an incisional biopsy was taken, after which a diagnosis of COF was made. The lesion was resected under general anesthesia, together with the unerupted canine displaced to the floor of maxillary sinus and was sent for confirmatory diagnosis. The excised mass was white with irregular surface [Figure 5]. The impacted canine along with cyst lining was found adjacent which was separated by a bony wall [Figure 6]. Microscopic examination of mass from the body of maxilla revealed mature collagen fibers interspersed with plump fibroblasts and nests of odontogenic epithelium [Figure 7]. Focal areas of calcification resembling cemento-ossous material were
evident, and a section attached to the neck of the canine showed cystic wall composed of fibrous tissue and lined by stratified squamous, nonkeratinized epithelium [Figure 8]. Based on the histopathological examination, confirmatory diagnosis of coexistence of COF with a DC in the maxilla was made.

**DISCUSSION**

COF represents a rare group of benign odontogenic tumor, while DC, on the other hand, is the odontogenic cyst that encloses the crown of an unerupted tooth. A combination of COF and DC is rare and not reported in literature till date.

Shafer et al. in 1983 described odontogenic fibroma as a different entity based on its histopathological and clinical features. The odontogenic origin of this rare tumor is confirmed by its occurrence only in jaws and by the presence of epithelial rests.\(^3\) The lesion may evolve from a mesenchymal dental tissue such as dental papilla, dental follicle or from the periodontal ligament, and therefore is invariably related to the coronal or radicular portion of teeth.\(^8\) This tumor is seen in individuals ranging from 5 to 80 years of age; it is more frequently seen in the third and fourth decades of life in mandible with female predilection.\(^4,5\) However, in this case, tumor was seen in a 14-year-old male in the maxillary anterior region. It manifests as a painless swelling with slow and progressive growth pattern, often with cortical plate expansion.\(^2\)

COF may demonstrate great variability in radiographic appearance. It can present as an unilocular or multilocular radiolucent area often with a well-defined margin. Multilocular radiolucencies are considered to be more aggressive than that of unilocular because they are associated with complications such as severe reabsorption of the roots of adjacent teeth, displacement of adjacent or associated teeth and expansion of the cortical bones.\(^9,10\)

*Figure 5:* Photograph showing the excised mass with irregular surface and a bony window

*Figure 6:* Photograph showing impacted canine along with cyst lining

*Figure 7:* Photomicrograph showing mature collagen fibers interspersed with pump fibroblasts and nests of odontogenic epithelium ×10

*Figure 8:* Photomicrograph showing cystic wall composed of fibrous tissue and lined by stratified squamous, nonkeratinized epithelium ×10
In this case, radiographic features were similar to that described by Daniel and Bucher et al., pertaining to multilocular appearance of lesion and displacement of 23. Histopathologically, in COF, one or more of the following features can be identified (a) fibrous or myxoid stroma, (b) odontogenic epithelium and (c) calcifications with scattered fibroblasts within a collagen-rich background. The COF is classified histologically into two types: the epithelium-rich type (formerly termed the WHO type) and the epithelium-poor type (formerly termed the simple type). The epithelium-rich type lesions are often characterized by epithelial islands or strands and the epithelium-poor type lesions are less cellular and the epithelial tissue is not constantly present. When present, epithelial nests or epithelial cords are probably distributed throughout the tumor.[1] This case was compatible with epithelium-rich type of COF. Histological features of COF may imitate a wide range of pathosis from benign lesions such as ameloblastic fibroma, odontogenic myxoma to more aggressive lesions such as desmoplastic fibroma, juvenile aggressive fibromatosis or fibrosarcoma to avoid diagnostic pitfalls.[3] Enucleation or vigorous curettage remains the treatment of choice of the COF. Although the recurrences of COF are not common, few cases have been reported by Daniels;[9] hence, postoperative tracking should be carried out on the patient after surgical intervention.[11]

DC is an epithelial-lined developmental cavity that encloses the crown of an unerupted tooth at the cementoenamel junction. Thus, it is also called tooth-containing cyst,[1] which accounts for approximately 20%–24% of all epithelial-lined jaw cysts. The exact histogenesis of the DC is not known. It is stated that the DC develops around the crown of an unerupted tooth by accumulation of fluid either between the reduced enamel epithelium and enamel or in between the layers of the enamel organ.[12] The cyst is typically seen in the second or third decade of life in the maxillary or mandibular third molar or maxillary canine regions.[13] It is common among the male gender and classically presents as an asymptomatic slow-growing swelling and initially associated with the crown of an impacted, embedded or unerupted tooth.[7,14] In this case, the cyst was seen in association with impacted 23. DC radiographically shows a unilocular radiolucency with well-defined sclerotic margins in association with the crown of an unerupted tooth.[15] Histopathologically, it is generally composed of thin connective tissue wall with a thin layer of stratified squamous epithelium lining. The cyst wall is composed of a very loose fibrous connective tissue or sparsely collagenized myxomatous tissue and may also show varying numbers of inactive odontogenic islands.[14] In the present case, there were 2–3 layers of nonkeratinized stratified squamous epithelium with mature fibrous connective tissue wall with few odontogenic rests. The recommended treatment for DCs depends on the size of the cyst, small cysts can be enucleated along with the cyst contents and associated tooth. For extremely large lesions or in cases when the involved tooth is desired to be retained in the arch, marsupialization may be done.[7]

COF and DC are two different odontogenic lesions that run entirely diverse clinical courses with varying treatment options and differing prognosis. Two distinctively different lesions occurring in the maxilla of a patient are exceptionally rare and not reported in literature. The concurrent occurrence of these two lesions raises the question of whether it is a collision tumor or their simultaneous occurrence is just a coincidence. In this case, the probable cause of simultaneous occurrence might be that COF could have displaced 23 resulting in the formation of DC.

Given the fact that COF is a rare tumor, accurate diagnosis is imperative combining clinical, radiological and histopathological features to prevent over or under treatment.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

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