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Calcifying Odontogenic Cyst of Anterior Maxilla With Multiple Impacted Teeth : Case Report

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Abstract

Epithelial-lined cysts seldom occur in skeletal bones, because embryonic epithelial rests are normally not found in them.However true bone cysts were frequently detectable in facial bones due to the existence of embryonic epithelial residues in all of these bones. Plenty of them are ruins of the odontogenic apparatus.A majority of them are remnants of odontogenic apparatus.(1-2)This is a well-circumscribed, firm or cystic lesion originating from odontogenic epithelium that emerges from reduced enamel epithelium or residue of odontogenic epithelium in follicles, gingival tissues or bones , but contains "ghost cells"and spherical calcifications.COC is sometimes alluded to as symptomless slow-growing swelling of the jaws. This is a well-circumscribed, firm or cystic lesion originating from odontogenic epithelium that emerges from reduced enamel epithelium or residue of odontogenic epithelium in follicles, gingival tissues or bones , but contains "ghost cells"and spherical calcifications.(2)This constitutes 2% of all odontogenic pathological changes in the jaw. The COC usually occurs intraosseously, but it can also occur extraosseously in the mandible and maxilla (1:1) at about the same incidence. The age of the patients may range from 5 to 92 years, with peak incidence in the second and sixth decade of life.(6,7)

Introduction

Epithelial-lined cysts seldom occur in skeletal bones, because embryonic epithelial rests are normally not found in them.However true bone cysts were frequently detectable in facial bones due to the existence of embryonic epithelial residues in all of these bones. Plenty of them are ruins of the odontogenic apparatus.A

majority of them are remnants of odontogenic apparatus.(1-2)

Gorlin and colleagues first identified the calcifying odontogenic cyst (COC) in 1962, describing a cyst lined by an ameloblastoma-like epithelium containing large quantities of ghost cells and calcifications.(3) Around that time, it was hypothesized that this cyst might well reflect the oral portion of the dermal calcifying epithelioma of Malherbe.(1) From 1971, the World Health Organization (WHO) characterized the lesion as a non-neoplastic cystic lesion and named it as COC. In 1992, WHO classified the lesion as odontogenic tumor, but did not change its name.It was dubbed calcifying cystic odontogenic tumor (CCOT) in 2005. The consensus group classifies the cyst as calcifying odontogenic cyst and the neoplasm as dentinogenic ghost cell tumor in the latest 4th edition of the WHO classification in 2017. (4)

This condition is often referenced to as Gorlin's cyst, keratinizing ameloblastoma, or melanotic ameloblastoma. COC is sometimes alluded to as symptomless slow-growing swelling of the jaws. This is a well-circumscribed, firm or cystic lesion originating from odontogenic epithelium that emerges from reduced enamel epithelium or residue of odontogenic epithelium in follicles, gingival tissues or bones , but contains “ghost cells”and spherical calcifications.(2)

The pathogenesis is, however, still controversial. While it is agreed that the epithelial lining that induce dentinoid or odontoma formation, the question remains unanswered whether COCs with features of other odontogenic tumors produce them secondarily or whether the COCs themselves are a secondary phenomenon in pre-existing odontogenic tumors.The former theory is widely accepted, but the latter is still advocated by several investigators.The calcifying odontogenic cyst (COC) is a rare example of an odontogenic developmental cyst, comprising around 0.37% to 2.1% of all odontogenic tumors.(1,5) This constitutes 2% of all odontogenic pathological changes in the jaw. The COC usually occurs intraosseously, but it can also occur extraosseously in the mandible and maxilla (1:1) at about the same incidence. The age of the patients may range from 5 to 92 years, with peak incidence in the second and sixth decade of life.(6,7)

Case Report

A 20 year old male patient, reported to the department of oral and maxillofacial surgery, K.D. Dental College & Hospital with a chief complaint of swelling in upper left front teeth region on both buccal & palatal aspect on same side. Pt got aware of this swelling 1 month back ,after that patient visited multiple dental clinics before coming to our department.The clinical exam revealed a firm and expansile lesion involving the buccal & palatal cortical plate.The lesion extended from tooth #21 to tooth # 25.Retained deciduous tooth #62 & #63 also present.(Fig.1) No clinical evidence of caries or periodontal disease was detected and the patient denied history of trauma to the area.The mucosa around the lesion area had normal color, and bone hardness was felt at the area. (Fig.2) The patient did not have any symptoms such as pain or paresthesia or discharge associated with it.The radiographic examination done using CBCT , shows the solitary large expansile well-defined corticated hypodense Osteolytic lesion is seen in maxilla extending:

1. Mesiodistally: crossing the midline from impacted 22 to mesial of 26.
2. Superioinferiorly: from the alveolar crest to the crown of impacted 23 in periorbital region. Breaching the wall of nasal cavity on left side.

3. Buccopalatally: axial section shows buccopalatal expansion, breaching the labial/ buccal and palatal cortical plates. There is thinning and breach in floor of the maxillary sinus is also seen.(Fig.4)

The Sagittal and Axial section shows: 22# is horizontally impacted on the palatal aspect. The Crown is directed towards the root of 11# and 62#. The root of 22# is directed towards the floor of nasal cavity.(Fig.5) 23# is vertically impacted in left periorbital region. The crown is directed towards the left maxillary sinus and root is directed towards the nasal cavity, involving nasal bone. Which gave radiographic diagnosis of benign Odontogenic Cyst i.r.t Impacted tooth 22 and 23.Fine needle aspiration cytology of the lesion was performed,(Fig.3) which yielded straw colored fluid and a provisional diagnosis of dentigerous cyst was made.Patient was allocated for surgical evaluation under general anesthesia and thus the lesion was enucleated employing intraoral vestibular approach and perhaps the entire lesion was excised , along with the submerged and deciduous teeth.(Fig. 6 ,7)The cavity was closed primarily after aggressive curettage. The excised tissue was sent for histopathological examination.(Fig.8) Histopathological analysis of the section showed cystic lumen filled with odontogenic epithelium backed by a connective tissue capsule.The lining epithelium shows ameloblastoma like cells with basal cuboidal & stellate reticulam like cells. The lining epithelium also shows varaible number of ghost cells.Masses of calcified structures are seen within the epithelial lining & in ghost cells.the stroma shows islands of odontogenic epithelial follicles ,areas of hyalinization , presence of eosinophilic dentinoid like material & calcified masses suggestive of calcifying odontogenic cyst.

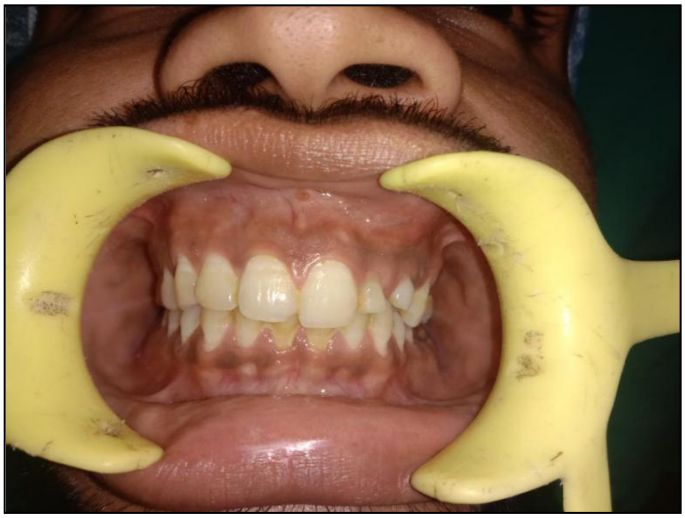

Fig.1: Pre operative occlusion profile showing retained deciduous teeth 62,63.

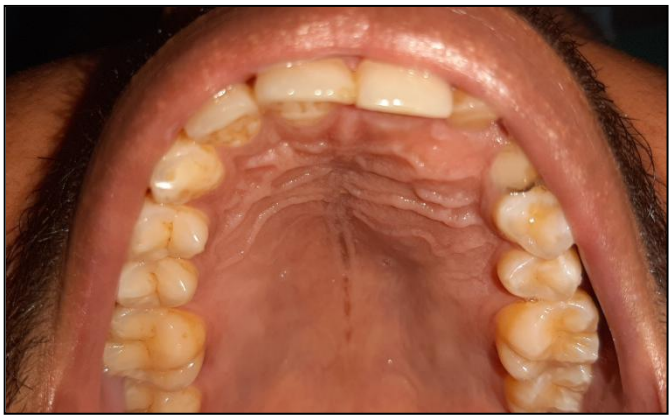

Fig. 2: Pre operative palatal profile showing swelling over left anterior palate.

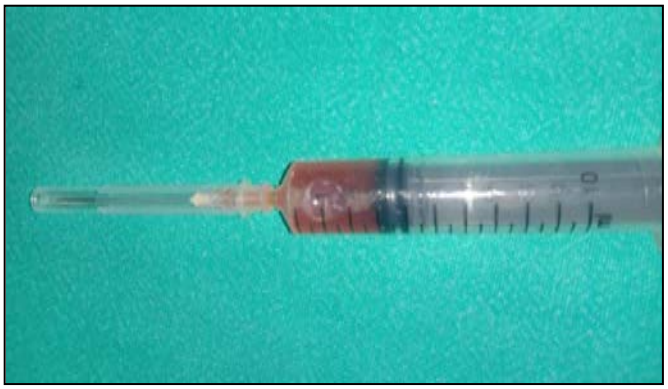

Fig. 3: Fine needle aspiration showed “Straw Colored Aspirate”.

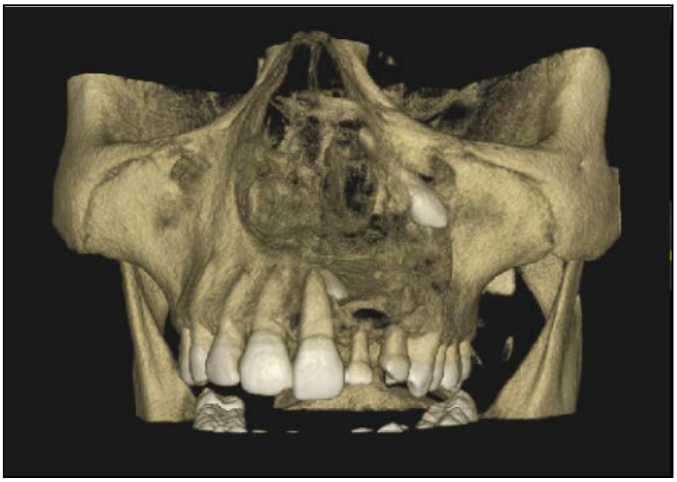

Fig.4: CBCT showing impacted teeth breaching the wall of nasal cavity on left side & extension of cyst.

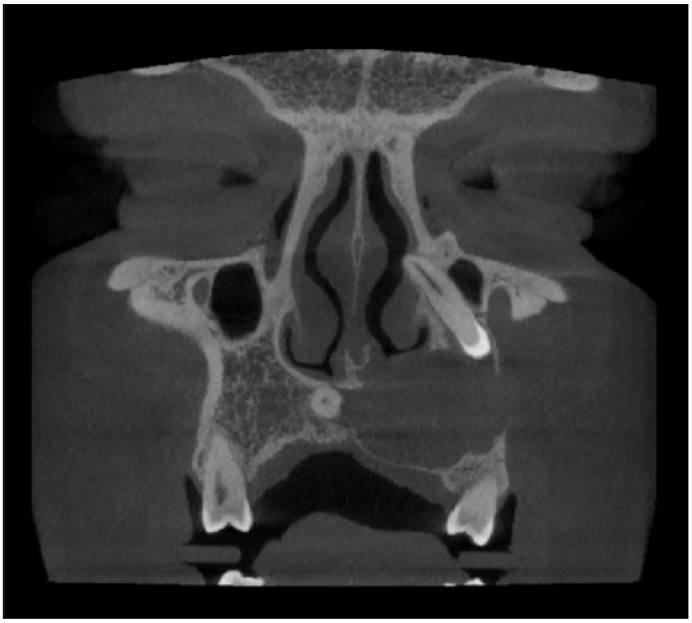

Fig. 5: Coronal view , of CBCT showing the solitary large expansile well-defined corticated hypodense Osteolytic lesion .

Discussion

The calcifying odontogenic cyst (COC) is a rare example of a developmental odontogenic cyst.its occurrence constitutes about 0.3-0.8% of all odontogenic cysts.(9)The COC has also been reported under a variety of other designations including keratinizing cyst, keratinizing cyst and calcifying odontogenic cyst (KCOC), calcifying ghost cell odontogenic tumor, dentinogenic ghost cell odontogenic tumour, epithelial odontogenic ghost cell tumour, ghost cell cyst, calcifying ghost cell odontogenic tumour, and dentino-ameloblastoma , Calcifying epithelial odontogenic cyst (CEOC) ,by various authors.(8)

The COC is considered to equally frequently include mandible and maxilla.The calcifying odontogenic cyst can occur in any location of the oral cavity and approximately 65–67.5% of cases occur in the anterior jaws .The age of occurrence of the cyst has been reported to vary from 3 years to 80 years with definite peaking in the second decade.(2 ,10) In our case lesion was situated in maxilla & the age of patient was 20 years , which is in accordance with the available literature.

Radiographically, it manifests as a unilocular or, at sometimes, multilocular radiolucency which may include calcified radiopacities or tooth-like densities. Thus, unilocular lesions are consistent with dentigerous, radicular or residual cysts, whereas multilocular lesions are consistent with multilocular lesions may mimic ameloblastomas or keratocysts.(5) In our case the lesion was manifests as a unilocular radiolucency hence initial diagnosis of odontogenic cyst were made. Calcifying epithelial odontogenic cysts are believed to be a unicystic process resulting from reduced enamel epithelium or vestiges of odontogenic epithelium in follicles, gingival tissues or bones.(9) There is still no unique indication of lesion whether clinically or radiographically.Differential diagnoses include dentigerous cyst, central giant cell granuloma, keratocystic odontogenic tumor and ameloblastoma, which have benign radiolucent appearance.(10) In our case as there were impacted teeth , fine needle aspirate was straw coloured fluid differential diagnosis of dentigerous cyst was made.

Mortazavi et al . reported that the least common pathology associated with impacted teeth is calcifying odontogenic cyst.It has also been reported that calcifying odontogenic cyst is often associated with anterior impacted teeth.(10,11) ,COC may be associated with other odontogenic

lesions, especially to odontomas (ranging from 22% to 47%), ameloblastomas, adenomatoid odontogenic tumors, and ameloblastic fibromas.(3) so in our case calcifying odontogenic cyst association with multiple impacted teeth makes it rare phenomenon.Its location is also as per descriptions available in literature.There was no association of odontome in our study.

Histopathological analysis of the section showed cystic lumen filled with odontogenic epithelium backed by a connective tissue capsule.The lining epithelium shows ameloblastoma like cells with basal cuboidal & stellate reticulam like cells. The lining epithelium also shows varaible number of ghost cells.Masses of calcified structures are seen within the epithelial lining & in ghost cells.The overlying connective tissue capsule consists of dense collagen fibers , endothelial lined blood vessels & areas of inflammatory cell infiltrate chiefly consists of lymphocytes & plasma cells. the stroma shows islands of odontogenic epithelial follicles ,areas of hyalinization , presence of eosinophilic dentinoid like material & calcified masses suggestive of calcifying odontogenic cyst.

Since its discovery various authors have tried to give classification of these lesion & organize in to groups.Praetorius (1981) envisioned to classify CEOC as Type I (cystic type) & Type II (neoplastic type [dentinogenic ghost cell tumor]).The cystic variant (Type I) was further subdivided into three different types:

a) Simple unicystic type,

b) Odontome-producing type, and

c) Ameloblastomatous proliferating type.(9)

Toida's latest dualistic classification of so-called COC (1998), in which he referred to the cystic variant as calcifying ghost cell odontogenic cyst (CGCOC) and used the term calcifying ghost cell odontogenic tumor (CGCOT) for the neoplastic variant

1). Cyst: CGCOC

2). Neoplasm:

A. Benign – CGCOT

a. Cystic variant — Cystic CGCOT

b. Solid variant — Solid CGCOT

B. Malignant - Malignant CGCOT

3).Combined lesion: the following lesions are associated with each of the above categories: a. Odontoma.

b. Ameloblastoma.

c. Other odontogenic lesions.(7)

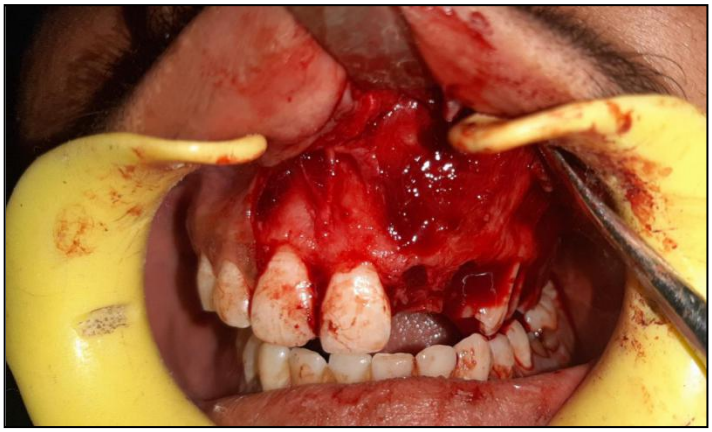

Fig 6: Intraoperative photograph showing the exposed cystic lining using the vestibular incision.

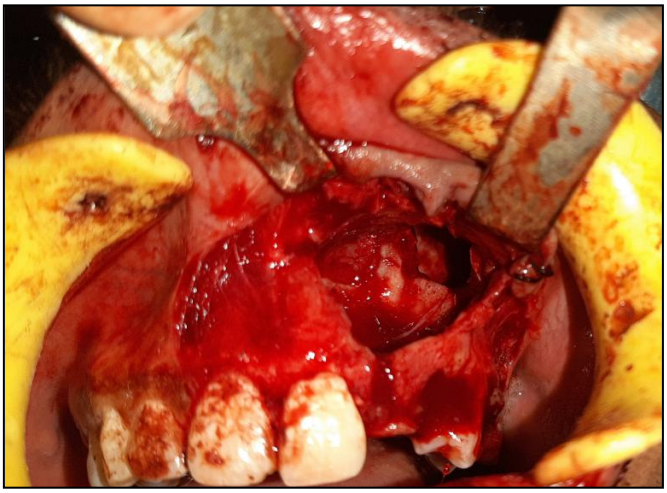

Fig. 7: Defect After Enucleation of Cyst

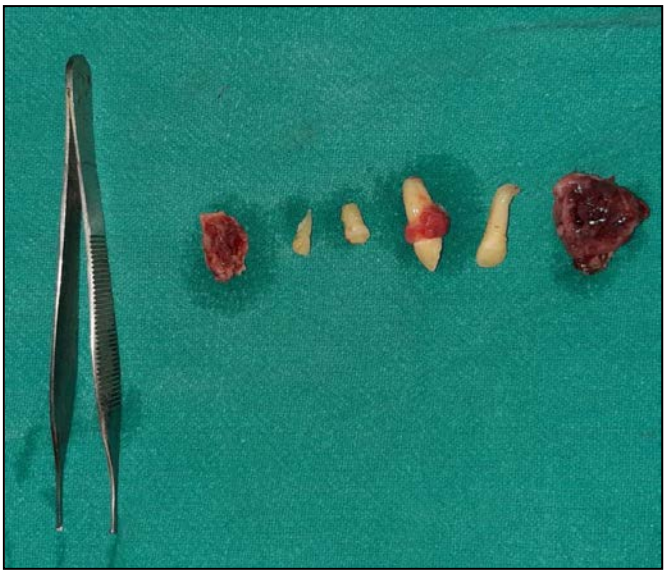

Fig. 8: Excised surgical specimen showing the lesion along with the associated impacted & deciduous teeth.

Enucleation is the treatment of choice with few recurrences reported in the literature for the most intraosseous COC.Only nine cases have been reported and all recurred within 8 years. A follow-up period of at least 10 years is, therefore, recommended.The extraosseous form is treated with surgical excision and recurrences for this type have not been reported.(5,10)Cases with a long track record as well as those who develop reoccurrence is at risk of malignant transformation. Li et al .reported a case of COC with recurrence that showed transformation to giant cell odontogenic carcinoma.(4)

Conclusion

COC is a rare developmental odontogenic cyst with numerous variations ranging from cyst ,tumour to neoplasmic variant .It lacks pathognomic features , no significant radiographic finding ,diagnosis purely based on histological examination.Although its association with impacted teeth is rare ,when radiolucency involving anterior jaw along with impacted teeth or odontome having aggressive nature differential diagnosis of calcifying odontogenic cyst should be considered.complications seen in this cases includes involvement of the nasal floor and maxillary sinus thus ,Early diagnosis and management is important.There are also chances of recurrence as well as malignant transformation so long term follow up is mandatory in this cases to avoid further complications.

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