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Coc Of Anterior Maxilla.

- Reporting an uncommon entity

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Abstract
Calcifying odontogenic cyst (COC), or Gorlin cyst, is a rare developmental lesion constituting <1% of odontogenic cysts and characterized by ghost cells with variable calcification[1]. Its diverse biological behaviour and clinicopathologic features lead to uncertainty and ambiguity in nomenclature and classification[2]. This report presents two cases: a 12-year-old female and a 17-year-old male with pain and mild swelling in the maxillary anterior region. Radiographic and histopathological findings confirmed COC in both patients, each linked to unerupted or supernumerary teeth. Surgical enucleation resulted in uneventful healing. These cases emphasize the importance of integrating clinical, radiographic, and microscopic findings for accurate diagnosis.

Keywords: Gorlin cyst, Ghost cells, Odontome, calcifications.

Introduction

COC, also known as Gorlin cyst, is an uncommon odontogenic lesion characterized by the presence of ghost cells and varying amounts of calcification. It may present as a cystic or solid lesion and is often associated with impacted or unerupted teeth. The lesion has both developmental and neoplastic potential, showing a wide histopathological spectrum[3]. **Gorlin et.al (1962)** were the first to document COC[4]. Earlier World Health organization (WHO) termed the entity as calcifying cystic odontogenic tumor[5]. However, recent WHO Classification of Head and Neck Tumours (2017) considered the entity as a cyst. The current WHO classification uses COC for cystic lesions and dentinogenic ghost cell tumor for the neoplastic entities[6]. This ambiguity in the nomenclature and classification has arisen as the entity exists in three histomorphologic distinct forms – benign, cystic lesions, solid masses(neoplastic) and aggressive variants(malignant)[3]. Reporting such cases is essential because of their rarity, variable presentation, and the ongoing evolution in diagnostic criteria and classification.

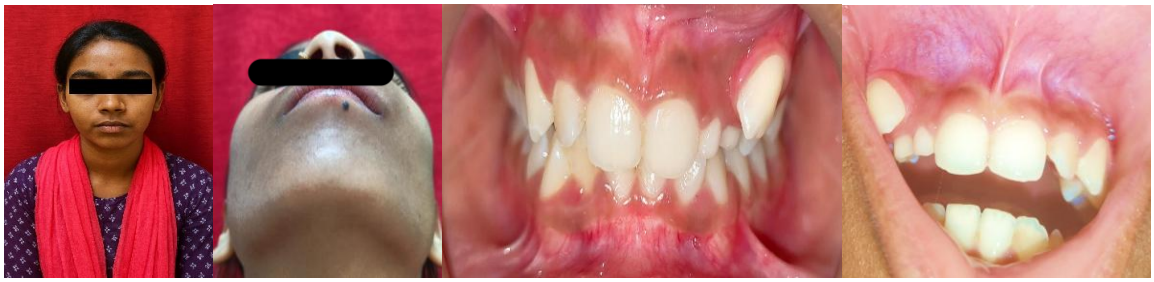
Case report - 1

A 12-year-old female patient presented to Srisai college of dental surgery to the Outpatient Department of Oral Medicine and Radiology (OMR) with a chief complaint of swelling in the left upper front tooth region that had been gradually increasing in size over the past two months. The patient was apparently normal before the onset of swelling, and there was no history of pain, pus discharge, or trauma.

Her medical, family, and social histories were non-contributory. There was no history of chronic illness or deleterious habits, and the patient had no known drug allergies. On general examination, she was conscious, cooperative, and well oriented, with no pallor, icterus, clubbing, cyanosis, or lymphadenopathy.

Extraoral examination revealed gross facial asymmetry due to a swelling on the left side of the face. The swelling extended supero-inferiorly from 1cm below the infraorbital region to 1cm above the upper lip and antero-posteriorly from the left ala of the nose to about 5cm anterior to the tragus. The overlying skin appeared normal, with no sinus opening, pigmentation, or discoloration, although the nasolabial fold was obliterated Figure 1(a).

On palpation, a solitary swelling with ill-defined borders and firm consistency was felt. There was no local rise in temperature, tenderness, pulsation, or crepitus. Mild paresthesia was elicited over the left cheek region. The temporomandibular joint was non-tender, and lymph nodes were palpable but non-tender.



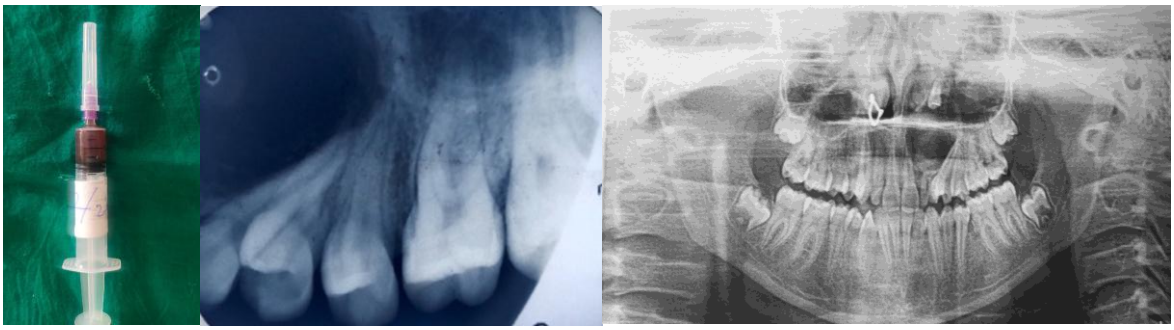
(a) (b)

Figure 1(a)Extraoral swelling in left upper front tooth region (b)Intra oral swelling.

Intraorally, there were 27 teeth present, with the permanent left lateral incisor (22) missing and retained teeth 62 and 63. A well-defined swelling was observed on the buccal aspect extending from distal of 21 to mesial of 25, causing buccal vestibular obliteration Figure-1(b). The palatal aspect appeared normal, with no sinus tract, erythema, or discharge. On palpation, the swelling was firm in consistency, with mild tenderness and buccal cortical expansion noted from 21 to 25, particularly around 62 and 63. There was no discharge or paresthesia intraorally.

Aspiration cytology revealed a protein content of 5.6mg/dl, suggesting a cystic nature Figure 2(a).

Radiographic evaluation through occlusal view showed buccal cortical expansion in the anterior maxillary region Figure 2(b).

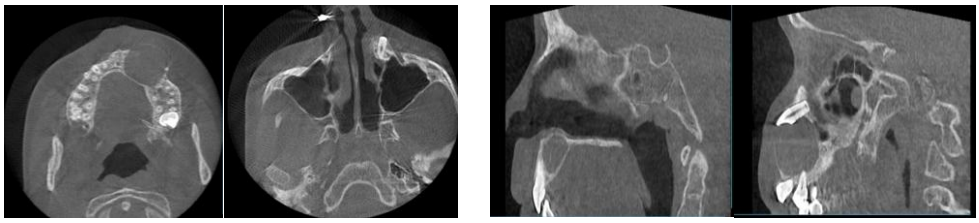


(a) (b) (c)

Figure 2(a) shows aspiration content in a syringe, (b)Buccal cortical expansion in anterior maxillary region in an occlusal view radiograph, (c) well-defined radiolucent lesion in the left anterior maxilla.

Orthopantomogram (OPG) demonstrated a well-defined radiolucent lesion in the left anterior maxilla associated with an unerupted 22 suggesting a cystic lesion with calcific specks. The lesion caused displacement of adjacent teeth and thinning of the cortical plates Figure 2(c).

Computed tomography (CT) scans in axial and sagittal views confirmed a well-circumscribed cystic lesion with cortical expansion and thinning, without evidence of invasion into surrounding structures Figure 3(a) & (b).



(a) (b)

Figure 3(a) represents Computed tomography (CT) Axial view,(b) saggital view

Based on the clinical and radiographic findings, a provisional diagnosis of an odontogenic cyst of the left anterior maxilla was made. Adenomatoid odontogenic tumor (AOT), ameloblastoma, and dentigerous cyst were given a place in the differential diagnosis.

Surgical management involved a crevicular incision extending from 21 to 25 with releasing incisions. A bony window was created in the region of 62 and 63, through which the cyst lining was identified and completely enucleated along with the impacted tooth 22. The retained deciduous teeth 62 and 63 were extracted, and primary closure was achieved, and the tissue of size measuring approximately 3cm × 4cm was sent for histological examination.

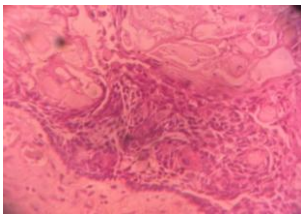


Figure 4 shows Hematoxylin and eosin (H & E) stained section with epithelium containing ameloblast like cells and numerous ghost cells.

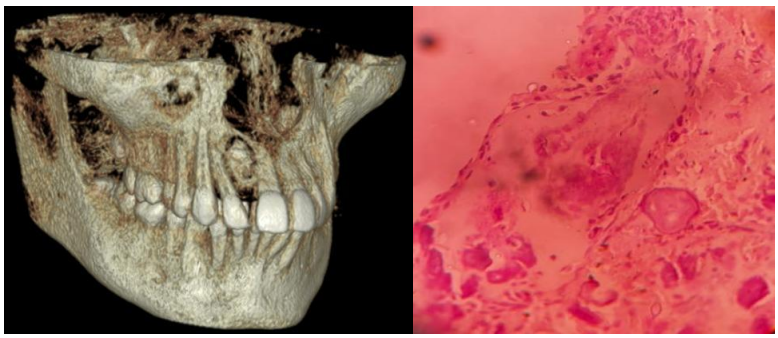
Histopathological examination of the excised specimen from the left anterior maxilla revealed cystic tissue lined by odontogenic epithelium. The epithelium in certain areas was composed of cuboidal basal cells with hyperchromatic nuclei, resembling ameloblast-like cells, and a scant stellate reticulum–like layer above them. Other areas of the epithelial lining showed a thickness of about 4–8 cell layers. Numerous ghost cells were observed within the lining epithelium; these cells appeared pale eosinophilic, enlarged, and lacked distinct nuclei, some showing evidence of calcification. The underlying connective tissue capsule consisted of delicate collagen fibers with numerous blood vessels engorged with red blood cells (RBCs). Mild inflammatory cell infiltration and areas of extravasated RBCs were also evident. These microscopic features particularly the presence of ghost cells and calcifications were consistent with the diagnosis of a COC.

Case report – 2

A 17-year-old male patient reported to the outpatient department with a complaint of pain in the upper right front region of the mouth. The discomfort had been present for some time and was localized to the buccal vestibular region corresponding to teeth 11 and 12.

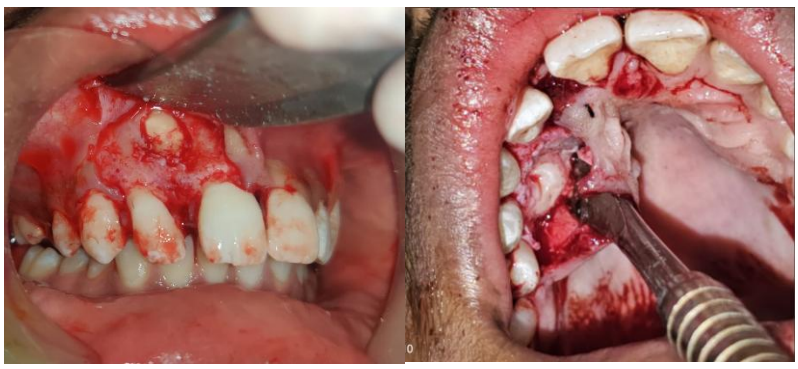
The patient was generally healthy, with no relevant medical, dental, or family history. On clinical examination, a mild diffuse swelling was observed in the buccal vestibule in relation to 11 and 12. The overlying mucosa appeared normal, with no evidence of ulceration, discharge, or sinus tract formation. On palpation, the swelling was firm in consistency, non-tender, and appeared to cause slight buccal cortical expansion.

Radiographic examination revealed an ill-defined radiolucent lesion involving the periapical area of teeth 11 and 12, extending toward 14. A supernumerary tooth was noted in the same region, which appeared impacted. The radiographic appearance suggested a cystic lesion with mild cortical thinning Figure 5(a). Based on the clinical and radiographic findings, a provisional diagnosis of odontome in relation to 11 and 12 with an impacted (supernumerary) tooth in relation to 14 was made.

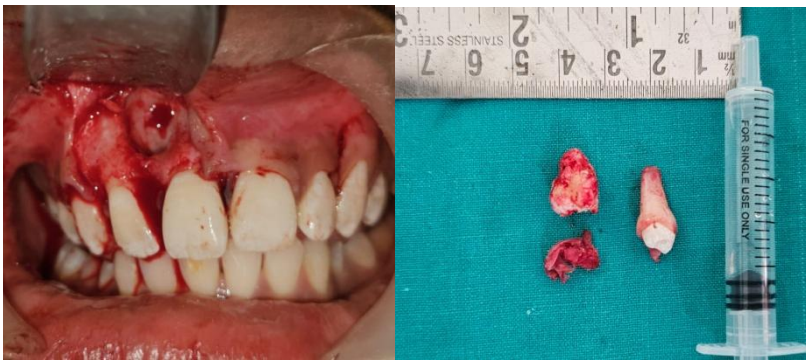


(a) (b)

Figure 5(a) shows CBCT of maxilla and mandible with maxillary anterior region shows a possible radiolucent area or cystic expansion around a developing tooth (b) Histopathological image stained with H & E showing scanty epithelium with ghost cells and calcifications.



(a) (b)



(c) (d)

Figure6 (a) Intraoperative procedure with crevicular incision showing odontome irt 11 & 12, (b) Intraoperative procedure image showing impacted supernumerary tooth irt 14 and (c)&(d) Post operative picture.

An excisional biopsy was performed, and the specimen was submitted for histopathological examination. Microscopic evaluation of H&E-stained sections revealed a cystic lumen lined by scanty odontogenic epithelium. The epithelial lining appeared thin, and in several focal areas, ghost cells which are large, eosinophilic, anucleated with indistinct outlines were evident. These ghost cells were associated with areas of calcification, a characteristic feature of COC. The underlying connective tissue capsule consisted of delicate to dense collagen fibers, odontogenic epithelial rests, and scattered fat cells. Focal areas within the connective tissue exhibited basophilic calcifications of varying sizes Figure 5(b).
The lesion was benign in nature, showing cystic characteristics with no signs of aggressive behavior. The final diagnosis was recorded as a COC in relation to 11 and 12 with an impacted supernumerary tooth in relation to 14.

Discussion

COC is classically a benign cystic entity lined by odontogenic epithelium and simulates as ameloblastoma. The lesion is categorized under ghost cell lesions as it manifests with distinctive ghost cell keratinization[7]**. Altini and Farman (1975)** reported that the entity had been initially documented in the German literature[8]. Ever since its documentation, there has been disagreement concerning its classification and nomenclature. This ambiguity in the nomenclature and classification has arisen as the entity exists in three histomorphologic distinct forms – benign, cystic lesions, solid tumor(neoplastic) masses and aggressive(malignant) variants[9].COC classification is based on two hypotheses – monistic and dualistic. The "monistic" theory considers COCs to be neoplastic, even though most lesions seem benign and cystic. The "dualistic" theory suggests that the lesion exists in two different forms – a cystic and neoplastic form. The WHO (1992) advocated the monistic theory and considered COC as an odontogenic tumor, However, the dualistic theory is recommended nowadays by most researchers[10].
In our cases the findings were suggestive of unicystic type of the COC based on the proposed classification by **Praetorius et.al (1981)** which was in coherence with findings of published literature of **Ahmad et.al (2022)**[11].
The benign or cystic form is most frequently seen (80%–98%). The solid tumor mass/neoplastic variant contributes for 11.5% of cases. COC may be seen anywhere in the oral cavity, however, most of the cases are seen in the anterior jaw region[12]. COC equally affects both maxilla and mandible, with no gender predilection.
The lesions are mostly seen in the second decade of life. Majority of COC occurs anterior to the first molar region, with more than 75% of cases occurring in the incisor-canine region or intercanine region[13].
In our present case of 12 year old female patient, swelling was in the upper front tooth region that had been gradually increasing in size over the past 2 months and in a 17 year male patient,pain was in the upper right front tooth region with mild diffuse swelling which was localized to the buccal vestibular region corresponding to 11 and 12.
Radiographically, most of the lesions exhibits a unilocular pattern with a well-demarcated sclerotic border, however, few cases are multilocular (5%–13%). COC has been documented in association with odontomas and impacted teeth (usually canines) in 24%–35% and 35% of cases, respectively. Another radiographic finding is that the lesion is frequently associated with unerupted teeth (32% of cases), thus, radiographically simulating dentigerous cyst[14].
In our cases the radiographic findings of 12 year old female revealed buccal cortical expansion in the anterior maxillary region and a well-defined radiolucent lesion in the same area associated with an unerupted 22 suggesting a cystic lesion with calcific specks and 17 year old male revealed an ill-defined radiolucent lesion involving the periapical area of teeth 11 and 12, extending toward tooth 14 suggesting a cystic lesion with mild cortical thinning associated with a impacted supernumerary tooth.
The salient microscopic features of the COC are epithelial basal lining arranged in a cuboidal/columnar fashion simulating ameloblasts. A cellular pattern mimicking the stellate reticulum of the enamel organ in the suprabasal layers is also a common feature.Ghost cells are the characteristic histopathological feature of COC, which are eosinophilic cells devoid of a nucleus. Eventually, the ghost cells may get calcified, thus, losing the cellular configuration and result in foci of calcified keratin[12].
In our cases microscopic features of a 12-year-old female patient showed odontogenic epithelium with ameloblast-like cells, and a scant stellate reticulum like layer above them and numerous ghost cells were observed within the lining epithelium and also showed evidence of calcification which were similar to findings of 17 year old male patient.
Enucleation is the preferred treatment for central cystic lesions. En bloc resection with a vigilant and prolonged follow-up is the recommended management protocol for neoplastic COC[13].
Our cases were surgically enucleated and follow up was adviced for one year post surgically.

CONCLUSION

In conclusion, these cases demonstrate the typical features of a COC occurring in the anterior maxilla of a young female and male patient. The lesion exhibited classic clinical as well as histopathological characteristics, including association of the lesion with impacted teeth, ghost cells and calcifications, which are diagnostic hallmarks. Early detection and surgical management result in an excellent prognosis, highlighting the importance of correlating clinical, radiographic, and histopathological findings for accurate diagnosis and appropriate treatment. Despite common features, the rarity of this lesion emphasizes the importance of reporting such cases to enhance understanding and clinical awareness.

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