

## CALCIFYING ODONTOGENIC CYST OF MANDIBLE – A CASE REPORT

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### ABSTRACT

Calcifying odontogenic cyst is the rare odontogenic lesion of jaw and was first described by Gorlin.*et.al* in 1962 and hence commonly known as Gorlin's cyst. WHO defined it as "a cystic lesion in which the epithelial lining shows a well-defined basal layer of columnar cells, an overlying layer that is often many cells thick and that may resemble stellate reticulum and masses of ghost epithelial cells that may be in the epithelial cyst lining or in the fibrous capsule". It constitutes about 0.37% to 2.1% of all odontogenic tumours. Clinically, it presents as a painless slow growing swelling of mandible without any sex predilection. Histopathologically, it shows numerous sheets of ghost cells along with odontogenic epithelial lining. The treatment is by surgical enucleation. The case presented here is a Calcifying Odontogenic cyst that occurred in a 17yr old male patient which is very rare.

**KEYWORDS:** Cyst, Calcifying Odontogenic Cyst, Ghost Cells, Mandible

### INTRODUCTION

A cyst is defined as a pathological cavity which may or may not have an epithelial lining and which has a fluid, semi-fluid, or gaseous content and is not formed by the accumulation of pus. Calcifying odontogenic cysts (COC) is first identified by Gorlin.*et.al* in 1962 as a specific odontogenic lesion and the characteristic feature of this lesion is presence of "Ghost cells".<sup>(1)</sup> It is commonly known as Gorlin's cyst. It was earlier thought to be an oral presentation of dermal calcifying epithelioma of Malherbe. In 1971, WHO classification described the COC as "non-neoplastic cystic lesion". Various suggested names are dentinogenic ghost cell tumour by Praetorius.*et.al*, odontogenic ghost cell tumour by Colmenero.*et.al*, Calcifying ghostcell odontogenic tumour by Fejerskov and Krogh. Recently, the World Health Organization (WHO) in 2005 updated its classification of odontogenic tumours, the name of the COC revised universally to the calcifying cystic odontogenic tumour (CCOT) to emphasize the neoplastic nature of a lesion previously categorized as an odontogenic cyst.<sup>(3)</sup>

### CASE REPORT

A 17 year old male patient came to department of oral surgery with a chief complaint of swelling in the lower left front tooth region since 4months. No relevant previous history was present. Extra-orally a diffuse swelling is seen from lower front side of face extending from para symphysis area to symphysis region (fig-1). No palpable lymphnodes and no tender on percussion and rise in temperature were seen.

Intraorally, a diffuse swelling of size 3x2 cm of size is seen in the lower left alveolar mucosa at the mucobuccal fold from central incisor to pre-molar region on left side (fig-2). On palpation, the swelling is non-tender, firm to hard in consistency. Radiographically, OPG revealed unilocular radiolucency of size 25x15mm surrounded by sclerotic boarder (fig-3). Thinning of cortical bone is seen. Root resorption is noted in lower central & lateral incisor region. Aspiration showed serosanguinous fluid. Provisional diagnosis of odontogenic cyst was given & incisional biopsy was done under local anaesthesia.

The incised specimen is 1.8x1.4cm in size, whitish grey in colour, irregular in shape, firm in consistency and the whole tissue is kept for processing. Histopathologically the section showed numerous sheets of ghost cells with odontogenic epithelial lining and fibrous connective tissue. Ghost cells are fused at some areas & few were calcified. Few bony spicules were also evident

## DISCUSSION

Calcifying ghost cell odontogenic cyst (CGCOC) is a heterogeneous group of lesion existing either as cystic or solid variant. In 1981, Praetorius. *et.al*, framed a classification based on dualistic concept in which they divided COC (as it was called) into two entities: A cyst and a neoplasm and proposed the term dentinogenic ghost cell tumour (DGCT) for the neoplastic variant<sup>(2)</sup>. Malignant transformation is rare in occurrence. It is a developmental odontogenic cyst constituting about 0.37% to 2.1% of all odontogenic tumours & is comparatively rare in occurrence. Calcifying epithelial odontogenic cyst (COC) represents approximately 5–7% of all odontogenic tumours, 1% of all cysts of the jaws. About <10% of all COCs are odontogenic ghost cell tumour.

The COC appears clinically as a painless, slow growing tumour, which affects the maxilla and mandible, showing a strong predilection for the anterior segment (incisor/canine area). It generally affects the young adults in the third to fourth decade of life. The age of these patients may range from 5 to 92 years, with a peak incidence in the second decade of life. Other authors however state a bimodal age distribution, with a second peak in the 6th-7th decade of life. The lesion has no sex predilection and is equally distributed between the maxilla and mandible<sup>(2)</sup>. The present case is reported in a 17year old male patient with a complaint of swelling in lower front tooth region and is asymptomatic.

**Different Terminologies for COC were given. These were as follows<sup>(3)</sup>**

- Gorlin.*et.al*, 1962 - Calcifying odontogenic cyst
- Gold, 1963 - Keratinizing calcifying odontogenic cyst (KCOC)
- Fejerskov and Krogh, 1972 - Calcifying ghost cell odontogenic tumour (CGCOT)
- Freedman.*et.al*, 1975- Cystic calcifying odontogenic tumour (COCT)
- Praetorius.*et.al*,1981 - Dentinogenic ghost cell tumour (DGCT)
- Ellis and Shmookler, 1986 - Epithelial odontogenic ghost cell tumour (EOGCT)
- Colmenero.*et.al*, 1990 - Odontogenic ghost cell tumour (OGCT)

Various authors proposed different classifications and following were the some of the classifications used in the literature.

**Praetorius (1981) proposed a classification for grouping CEOC as <sup>(3)</sup> Type I (cystic type)**

- Simple unicystic type,
- Odontome-producing type, and
- Ameloblastomatous proliferating type

**Type II (neoplastic type [dentinogenic ghost cell tumour]) Toida (1998)**

proposed a classification, called the cystic variant as calcifying ghost cell odontogenic cyst (CGCOC) and calcifying ghost cell odontogenic tumour (CGCOT) for the neoplastic variant, as below <sup>(5)</sup>:

- Cyst: CGCOC
- Neoplasm: A. Benign – CGCOT
- Cystic variant — Cystic CGCOT
- Solid variant — Solid CGCOT
- Combined lesion: associated with Odontoma, Ameloblastoma, Other odontogenic lesions.

**According to Reikart.*et.al*, Non–neoplastic (simple cystic) variants**

- With non proliferative epithelial lining
- With non proliferative epithelial lining associated with odontomas
- With proliferative epithelial lining
- With unicystic, plexiform, ameloblastomatous proliferation of epithelial lining

**Neoplastic Variants**

- Benign type (CGCOT)
- Cystic sub type (cystic CGCOT)
- SMA ex epithelial cyst lining
- Solid subtype (solid CGCOT)
- Peripheral ameloblastoma-like
- SMA-like

**Malignant type (malignant CGCOT or OGCC)**

- Cystic subtype
- Solid subtype

Radiographically, the COC is usually a mixed lesion, with radiolucent area, present with a unilocular/multilocular appearance. In our case well defined, unilocular, radiolucent area with sclerotic boarder is seen. Additionally, root resorption and displacement was also noted (fig-3).

Histological features of a classic calcifying odontogenic cyst (COC) include a fibrous capsule with a lining of odontogenic epithelium. The basal layer is made up of columnar or cuboidal cells of 4-10 cell thickness lined by a loosely arranged epithelial cells having similarity to stellate reticulum of the enamel organ. It also characterised by number of epithelial cells devoid of any nuclei, which are eosinophilic with their basic cell outline retained known as ghost cells. Sometimes these ghost cells may undergo calcification and lose their cellular outline.

Current case report showed histologically 4-6 layers of odontogenic epithelium with basal cells low columnar in shape & supra basal cells resembling stellate reticulum (fig-5). Sheet of numerous ghost cells were seen overlying the epithelium and at some areas they are invading into epithelium. Ghost cells are eosinophilic without any nucleus but in some faint nuclear outline is evident (fig-6). These were fused at some areas and various shapes of ghost cells like fusiform, circular and oval were also found (fig-7). These ghost cells were also calcified at few focal areas. The connective tissue showed loosely arranged collagen fibres with few inflammatory cells and final diagnosis of calcifying odontogenic cyst was given based on all the above findings.

The COC is treated conservatively by surgical enucleation and recurrences are very uncommon (recurrences depend on the completeness of cyst removal). The malignant transformation of a pre-existing benign COC can occur but is extremely uncommon. The COC may also be associated with other odontogenic tumours such as adenomatoid odontogenic tumour, ameloblastoma, ameloblastic fibro-odontoma and ameloblastic fibroma where wider excision may be required<sup>(5)</sup>.

## CONCLUSION

COC is a rare lesion & it is difficult to diagnose clinically. Differential diagnosis includes adenomatoid odontogenic tumour, unicystic ameloblastoma, odontogenic cysts, odontoma. Its clinical and radiographic features may mimic other odontogenic cysts/tumours, and a definitive diagnosis can only be made histologically. So, histopathological features along with radiographic features were important for its diagnosis. Conservative treatment by surgical enucleation is best treatment for these cases.

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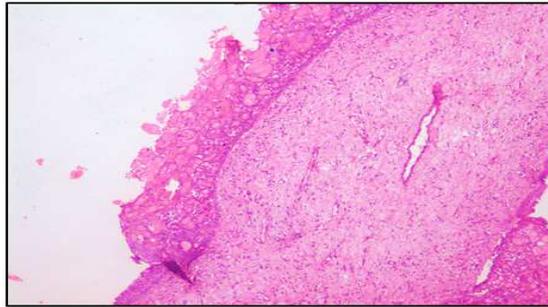
Figure 1: Swelling in the left side of Mandible



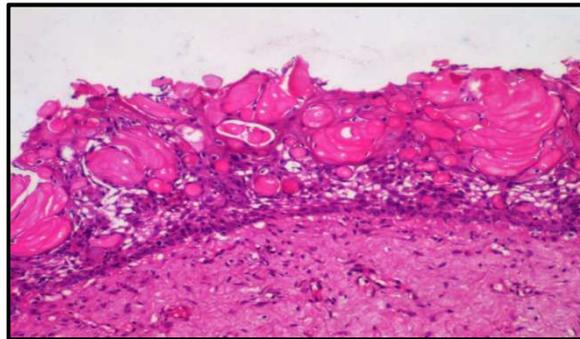
Figure 2: Diffused Swelling in the Lower Anterior Teeth



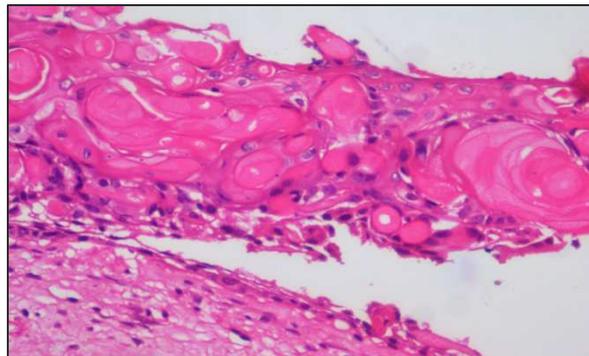
Figure 3: OPG showing well defined Radiolucency with Sclerotic Boarder



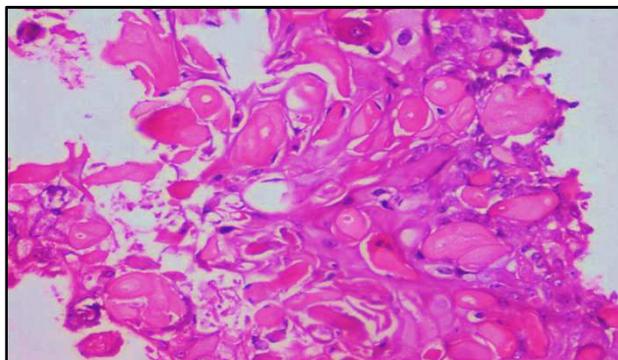
**Figure 4: Cystic Odontogenic Epithelial Lining with Overlying Sheets of Ghost Cells**



**Figure 5: under high Power 4-6 Layers of Odontogenic Epithelium with numerous Ghost Cells at Epithelial Surface**



**Figure 6: Sheets of ghost cells with faint nuclear outline**



**Figure 7: Numerous Ghost Cells with Various Shapes**