Introduction

Calcifying odontogenic cyst (COC) was first categorized as distinct entity by Gorlin et al., in 1962 and was named after him since then. According to Shear it account for 1% of the jaw cysts. Once it was thought to be a cutaneous counter part of benign calcifying epithelioma of malherb described in 1980. The COC is an unusual and unique lesion with characteristics of solid neoplasm and of a cyst. It shows considerable amount of histopathological diversity with variable clinical behavior, such as cystic, neoplastic and infiltrating malignant behavior. There may be variants of COC according to clinical, histopathological and radiological characteristics. There proper characterization of cases is needed for better understanding of pathogenesis of each variant.

The central giant cell granuloma (CGCG) of the jaws account for approximately 7% of all benign tumors of the jaws. These lesions mainly occur in young adults with a predilection for females. The lesions must be differentiated from a variety of the jaw lesions such as cysts, odontogenic tumors, fibroosseus lesions, vascular malformations and even malignancies. Radiographically it is a unilocular or multilocular radiolucency. As this is a slow growing lesion, borders are usually well defined. Teeth displacement, root and lamina dura resorption of the teeth may also be observed.

Case Report

A 30 year old female reported to Out Patient Department with chief complaint of an asymptomatic swelling in the lower jaw. The swelling was present since three months and was slowly enlarging to the present size of 6 x 3 cm. On extraoral evaluation an asymmetry involving lower right and left face region extending from right corner of mouth to two centimeter behind left corner of mouth was present. Borders of swelling were indistinct and variable in consistency on palpation. Consistency was firm on periphery and fluctuant at the center. Paraesthesia was present in the region of swelling. Intraoral examination revealed buccal cortical expansion extending from tooth #43 to #36 region crossing the midline. Mucosa over the swelling was intact. On palpation swelling was non tender and cystic. Mobility was present with tooth #42, #41, #31, #32, #33, #34 and #35. Displacement of involved teeth was also noted.

Radiographic examination showed a well-defined unilocular radiolucency extending anteroposteriorly from distal of tooth #42 to mesial of #36. Superio-inferiorly the lesion extends from periapical region of involved teeth to lower border of mandible. There was
external root resorption of seven teeth from tooth #42 till the tooth #35 in apical 1/3rd area (fig 1c). Based on the clinical presentation and radiographic appearance differential diagnosis of central giant cell granuloma, unicystic ameloblastoma and aneurysmal bone cyst was considered.

Before going for surgery 16 ml of cystic aspirate was drained with wide board needle using the thinnest part present on the buccal cortical plate. Cytological smear of the cystic aspirate was prepared and subjected to microscopic examination. It showed numerous pus cells, RBC’s and few cholesterol crystals which were non-conclusive. Full thickness envelop flap when raised showed loss of buccal cortical plate at the center of the lesion and thinning at the periphery. A well-defined cystic lesion was enucleated in toto and well separated from the surrounding bone. Inferior alveolar nerve was sacrificed, and then the whole specimen was subjected to histopathological examination.

On histopathological examination the microscopic features include a fibrous capsule with a stratified lining of odontogenic epithelium. The basal layer was made up of ameloblast-like columnar or cuboidal polarized cells of four to ten cell thickness over-lined by a loosely arranged spindle or stellate shaped odontogenic epithelial cells bearing similarity to stellate reticulum of the enamel organ. There exists varying number of epithelial cells devoid of nuclei which are eosinophilic and retain their basic cell outline suggestive of ghost cells (fig 1d). These ghost cells show calcification at some places and lose their cellular outline to firm sheet like area of calcified keratin. Calcification and dentinoid material was found. Ghost cells and dentinoid were very well demonstrated by the special stains like Mallory and Van Geison Stain. Accentuated ghost cells appear orange colored in Mallory stain and yellow colored in Van Geison stain (fig 1e & f).

Discussion
The age ranges from 1 to 82 years with peak incidence in the 2nd decade. In an observation of 215 lesions Bruchner et al., have drawn attention to bimodal age distribution in support of their contention that two different entities may be involved with second peak in sixth / seventh decade.9,10 The lesion has no sex predilection and is equally distributed between maxilla and mandible.11 Radiographically majority of the lesions present in an unicocular form with well-defined margin,9,12 while in 5 - 13% of the cases they are multicellular. They have scattered irregular sized calcification producing a variable range of opacities [salt and pepper type of patterns].13 They may be associated with tooth like densities in 50% of the cases and one-third of the cases show association with unerupted tooth, most often a canine.13

The central COC presents as an asymptomatic hard swelling of the jaws that produces expansion rather than erosion of bone. Pain indicates secondary infection.14-16 Early lesions are usually detected following routine radiographic examination and they are often associated with an unerupted tooth.1,7 The peripheral COC presents on the gingiva as a non-specific well-circumscribed sessile or pedunculated mass with a smooth surface. They resemble other epulis, such as gingival fibromas or peripheral giant cell granulomas.15 The definitive diagnosis of COC is made histologically, due to the lesion’s lack of characteristic clinical and radiological features, as well as its variable biological behaviour.17

According to Buchner’s classification16 this lesion would be classified as a central multicystic COC (type B 1a). The classification of Praetorius does not recognize multicystic lesions as a separate entity. The presence of radiopacities could suggest a diagnosis of an adenomatoid odontogenic tumour, a calcifying epithelial odontogenic tumour or an ameloblastic fibro-odontoma.18 Nagao and colleagues19 reported in a series of 23 cases only three cases of multicellular lesions, which converts to a ratio of 7.6:1. In 33% of cases COCs are associated with unerupted teeth.

Classification of calcifying odontogenic cysts as proposed by Praetorius et al.20
1 a: Simple unicystic type with little or no dentinoid
1 b: Odontoma producing type
1 c: Ameloblasomatous proliferating type
2: Neoplastic variant (dentinogenic ‘ghost’ cell tumor)

Classification of calcifying odontogenic cysts as proposed by Buchner16
A. Peripheral (extraosseous) COC
1. Cystic variant
2. Neoplastic (solid) variant
B. **Central (intraosseous) COC**
   1. Cystic variant
      i. Simple (unicystic or multicystic)
      ii. Associated with an odontoma
      iii. Associated with odontogenic tumours (other than odontoma)
      iv. Other variants (such as clear cell variant, pigmented variant)
   2. Neoplastic (solid) variant
   3. Malignant COC

Clinically the lesion was present in lower anterior region crossing the midline as an asymptomatic swelling; age of presentation was 3rd decade in female. The lesion was unilocular with well-defined borders without any radiopacities in the cystic lumen. All these findings were in favor of central giant cell granuloma, unicystic ameloblastoma both of which are aggressive lesions and show definite degree of recurrence. After histopathological examination it came out to be calcifying odontogenic cyst which is a benign neoplasm and do not show recurrence.

![Image of a clinical case](image)

**Figure 1:** Extraoral swelling in lower left chin region (a) with Intraoral buccal expansion (b) and drifting of teeth in the region of swelling and orthopantomograph showing radiolucency crossing the midline (c). The photomicrograph under low power view showing Ghost Cells in hematoxilin & eosin stain (d), Mallory Stain (e) and Van Geison Stain (f).

The clinical significance of this case is that clinical and radiographical observations appeared to be simple, unambiguous and obvious. However, the histopathological observations showed that it was a rare odontogenic lesion. Hence, this case report emphasizes the role of biopsy in the diagnosis of such deceptive clinical presentations. This also proves the importance of microscopic examination of
each and every case before going for the treatment which may change the final diagnosis of the patient and hence the treatment plan and the final outcome of the treatment.

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References

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