Case Report

Calcifying odontogenic cyst with luminal and mural component (Type 1c)

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Abstract

Calcifying odontogenic cyst (COC) was first described and classified by Gorlin *et al*. It is defined as a cystic lesion in which the epithelial lining shows a well defined basal layer of columnar cells, an overlying layer that often resemble stellate reticulum and masses of ghost cells that may be in the epithelial cystic lining or in the fibrous capsule. The lesion generally occurs in the region anterior to maxillary and mandibular molars and either intraosseous or extraosseus. This entity might present as a cystic or solid lesion. Praetorius *et al*. classified COC into 2 main entities namely a cyst (Type 1) and a neoplasm (Type 2). The present case report exhibit a cystic lesion with both luminal and mural component.

Key words: Calcifying odontogenic cyst, cystic lesion, luminal, mural

INTRODUCTION

Calcifying odontogenic cyst (COC) was first described and classified by Gorlin *et al*.[1] Recently, the WHO changed the name of COC to the calcifying cystic odontogenic tumor to emphasize the neoplastic nature of the lesion which was previously categorized as an odontogenic cyst,[2,3] and have 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 that they may resemble stellate reticulum and masses of ghost cells that may be in the epithelial cystic lining or in the fibrous capsule.[4] Ghost cells are anucleate cells with homogenous pale eosinophilic cytoplasm and very pale to clear areas instead of a basophilic nucleus,[5] and they have the propensity to calcify and are occasionally associated with certain odontogenic tumors.

The epithelial lining of the COC appears to have the ability to induce the formation of dental tissues in the adjacent connective tissue, and its association with the odontoma is relatively common. Praetorius *et al*. classified COC into two main entities, namely, a cyst (Type 1) and a neoplasm (Type 2). The Type 1 variety was subdivided into three groups simple and unicystic (Type 1a), odontoma producing (Type 1b), and ameloblastomatous proliferation (Type 1c).[6]

The other odontogenic tumors such as ameloblastoma, adenomatoid odontogenic tumor, ameloblastic fibroma, and ameloblastic fibro-odontoma may also sometimes be associated with the COC.[5]

CASE REPORT

A 22-year-old male patient reported to the Christian Dental College with the complaint of a swelling in the lower front region for the past 2 months and pain associated with the swelling for the past 25 days. The swelling was progressively increasing. The pain which was associated with the lower

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front teeth was gradual in its onset, continuous, severe, aggravated on mastication, and relieved on medication. The patient's medical history was not contributory. Extraoral examination revealed an ill-defined swelling seen on the lower jaw in the chin region, which was tender on palpation [Figure 1]. The right submandibular lymph node was palpable; single, firm, and nontender. The skin over the swelling was of normal color. Intraorally, a well-defined swelling was observed in the labial vestibule. The teeth associated with the swelling, 31, 33, 41, and 83 were mobile. 43 and 32 were missing. Obliteration of the lower labial vestibule and lingual expansion of the swelling was seen [Figure 2]. On palpation, the lesion was hard in consistency and tender. An orthopantomograph was taken which revealed a large radiolucency extending from 43 to 33 region. 43 and 32 were impacted within the lesion [Figure 3]. Flecks of radioopacities were also seen. Aspiration revealed a straw colored fluid. Computed tomography scan of the area was advised which showed a large bony expansile lesion with perforation of the labial cortical plate [Figure 4].

Based on the above findings, provisionally, it was diagnosed as an adenomatoid odontogenic tumor, with a differential diagnosis of the dentigerous cyst, COC, and calcifying epithelial odontogenic tumor.

Excisional biopsy was performed, following which the specimen was sent for histopathological examination [Figure 5]. Microscopically, one section showed a cystic lining composed of 2–3 cell layers thickness, resembling the reduced enamel epithelium, with an underlying fibrous connective tissue capsule [Figure 6]. The other sections showed a proliferating lining epithelium, in areas opening up to form stellate reticulum like cells. Most of these areas showed numerous ghost cells and calcifications, some of these calcifications were ovoid with a lamellated appearance [Figure 7]. The underlying connective tissue capsule showed amelobalstomatous proliferation with the presence of odontogenic follicles [Figures 8 and 9]. Based on the clinicopathologic findings, a final diagnosis of COC is given.

DISCUSSION

COC represents approximately 5–7% of all odontogenic tumors,[7] 1% of all cysts of the jaws, and <10% of all COCs are odontogenic ghost cell tumor.[8] It is found in the incisor and canine area of the maxilla and mandible with approximately equal frequency,[9] seen in the third to fourth decades and presents as an asymptomatic swelling.

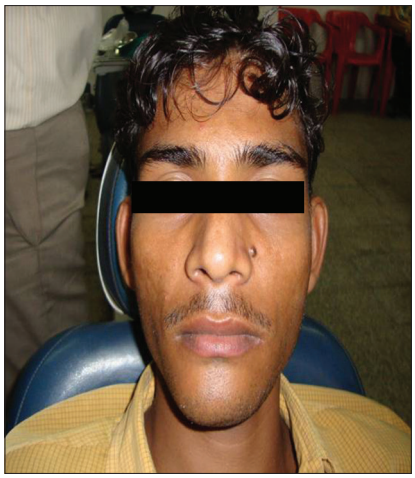

Figure 1: Extraoral photograph showing swelling on the lower jaw in the chin region

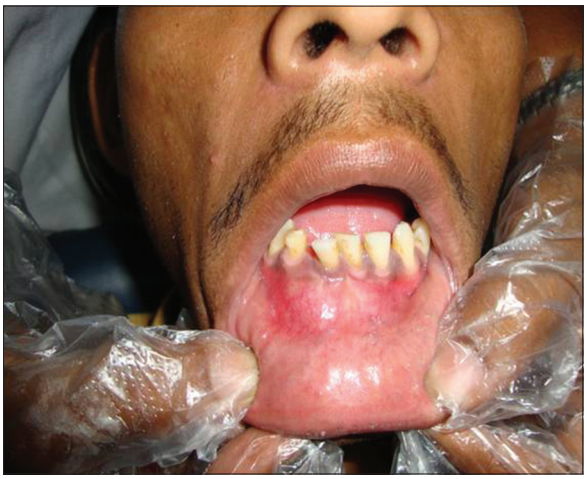

Figure 2: Intraoral photograph showing obliteration of lower labial vestibule

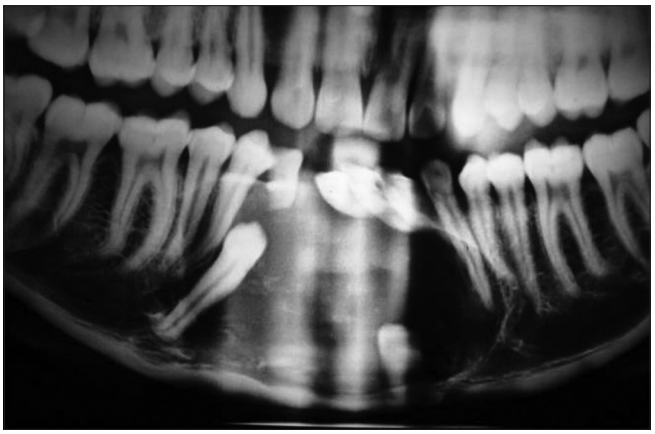

Figure 3: Orthopantomograph revealed a large radiolucency extending from 43 to 33 region, 43 and 32 were impacted within the lesion

The cyst occurs as three variants:

- A simple unilocular cyst with moderate mural proliferations of epithelium and no, or sparse

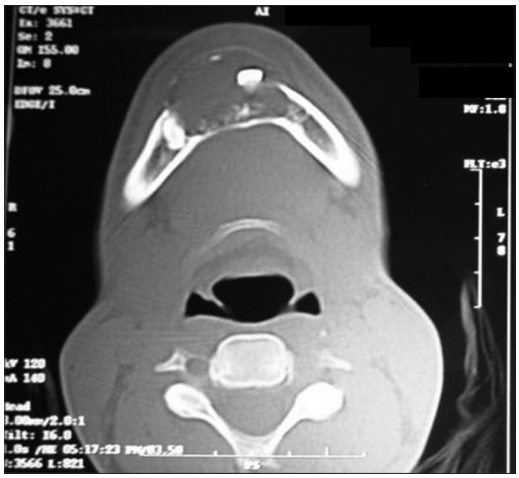

Figure 4: Computed tomography scan showed a large bony expansile lesion with perforation of labial cortical plate

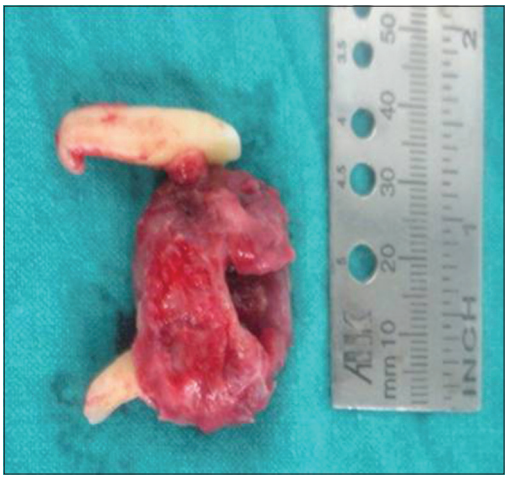

Figure 5: Excisional biopsy specimen with impacted teeth

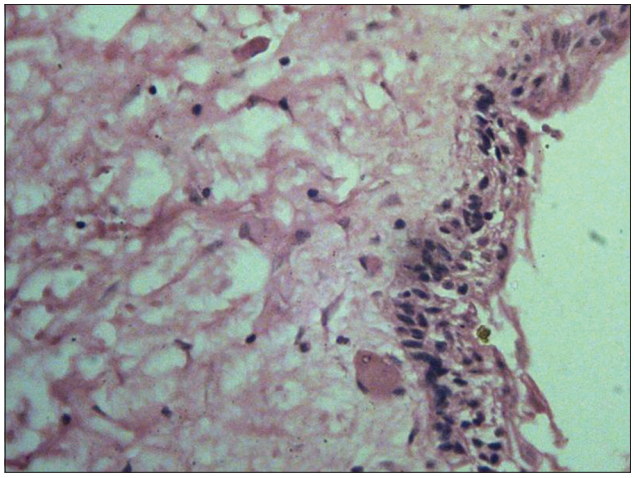

Figure 6: Photomicrograph showing cystic lining composed of 2–3 cell layers thickness with an underlying fibrous connective tissue capsule (×400 view)

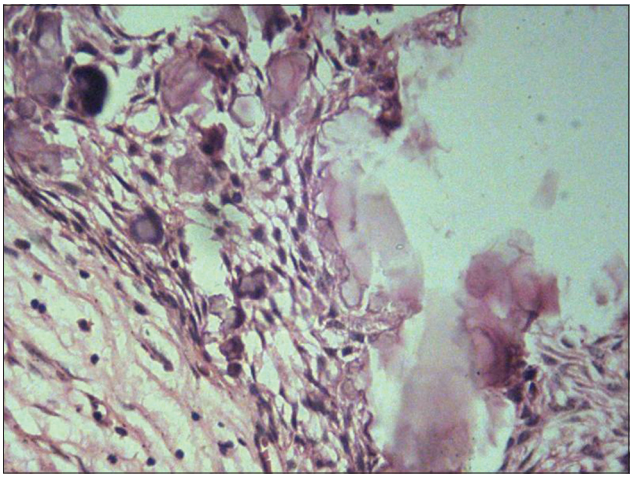

Figure 7: Photomicrograph showing ghost cells and calcifications (×400 view)

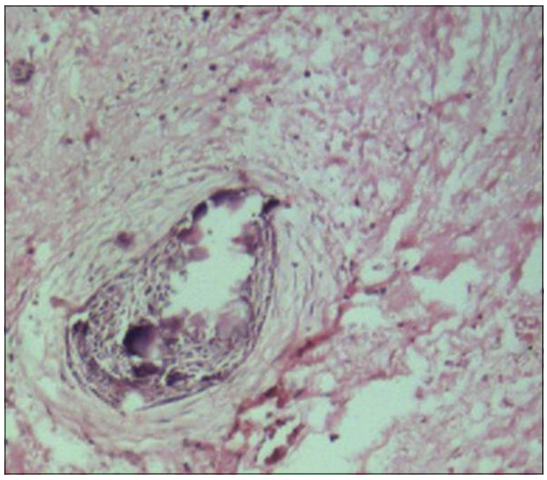

Figure 8: Photomicrograph showing amelobalstomatous proliferation with the presence of odontogenic epithelial islands (×100 view)

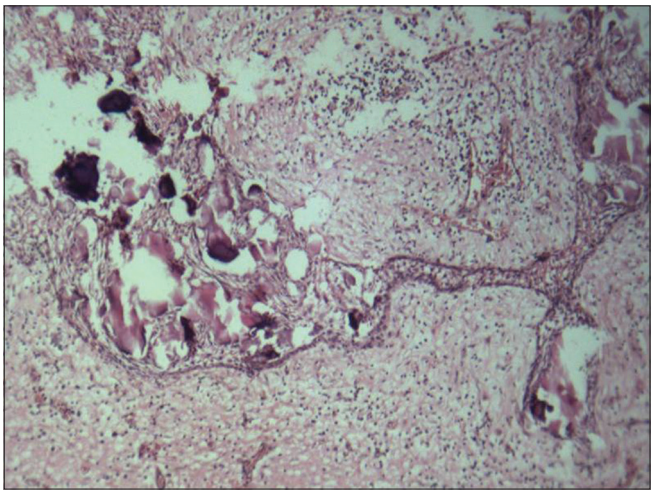

Figure 9: Photomicrograph showing amelobalstomatous proliferation with the presence of odontogenic follicles (×400 view)

amounts of dentinoid (dysplastic dentin); it seems to occur during man's entire life span

- A unilocular cyst which produces compound or complex odontomas in its luminal part, more rarely it may instead produce an intramurally

growing ameloblastic fibroma, which may call for more radical surgery. It occurs mainly in patients between 10 and 29 years of age
- A unilocular cyst with extensive luminal as well as mural ameloblastoma-like proliferations of epithelium.[6]

The present case reflects the third variant having both luminal as well as the mural proliferation of odontogenic epithelium, and therefore categorized as COC (Type 1c). Radiologically, it appears as a unilocular or multilocular (5%) radiolucency with well-defined margins and a variable amount of irregularly shaped radioopacities representing dentinoid or dystrophic calcifications or may be associated with unerupted teeth.[10,11] Calcifications are detected in about half cases,[12] and appear as varying amounts of radioopacities ranging from tiny flecks to large masses.[13] Association with unerupted teeth is also a common radiographic feature of COC, the frequency of enveloped tooth approximately 32% which may simulate the dentigerous cyst when viewed on a radiograph.[12,13] Multiple impacted teeth are a well-known feature of COC and Buchner indicated that the COC tends to cause the second and third tooth to be displaced preventing eruption and occurred in the canine region.[14] COC shows a higher frequency for the mandible.[14] The site does not seem to have any relation to the behavior or histological features of the cyst.

Our case showed a majority of the features mentioned in the literature. This case report highlights the importance that a differential diagnosis of COC should be considered in the list of cystic radiolucencies in the mandibular anterior region.

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.

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Conflicts of interest

There are no conflicts of interest.

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