



Calcifying Epithelial Odontogenic Cyst- A Case Report With 7 Years Follow Up

KEYWORDS

Calcifying epithelial odontogenic Cyst; Cyst; ghost epithelial cells.

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ABSTRACT

The calcifying epithelial odontogenic cyst (CEOC) was first delineated in 1962. Calcifying epithelial odontogenic cyst (CEOC), is a rare benign odontogenic cyst of locally aggressive behavior. It is more common in the posterior part of the mandible, typically in the fourth to fifth decades. It usually starts as a painless swelling and is often concurrent with an impacted tooth with notable presence of histopathological features which include a cystic lining demonstrating characteristic "Ghost" epithelial cells with a propensity to calcify. In addition, the COC may be associated with other recognized odontogenic tumors. This gives rise to a spectrum of variants of COC according to clinical, histopathological, and radiological characteristics. This article presents one such entity of case report of CEOC with good prognosis and no recurrence.

Introduction:

Epithelial-lined cysts seldom occur in skeletal bones, because embryonic epithelial rests are normally not found in them. They do, however, occur in the jaws where the majorities are lined by epithelium derived from remnants of the odontogenic apparatus. These odontogenic cysts are classified as either of developmental or inflammatory origin. The calcifying ghost cell odontogenic cyst (CGCOC) is a rare example of a developmental odontogenic cyst, its occurrence constituting about 0.37% to 2.1% of all odontogenic tumors [1]. Periapical pathologies are common, and periapical cysts are more than 50% of all odontogenic cysts reported [2]. Asymptomatic periapical lesions are not uncommon and routine classical description of the radiological appearance of the periapical cysts are a round or ovoid radiolucency lined by a narrow radiopaque margin that extends from the lamina dura of the involved tooth. In enlarging cysts, this margin may be absent. A periapical cyst involving proximal surface of tooth, with or without root resorption, is not uncommon and is attributed to accessory root canals [3]. The calcifying epithelial odontogenic cyst is a rare lesion of the jaws. Gorlin et al [4] first described the condition and pointed out its close resemblance to the calcifying epithelioma of Malherbe. Gold [5] recommends the more descriptive term of calcifying and keratinizing odontogenic cyst. The condition has been referred to as keratinizing ameloblastoma [6] or melanotic ameloblastic odontoma. One of the histological features of the condition is the presence of ghost cells. Fejeskov and Krough [7] are of the opinion that the lesion initially presents as a solid tumor, consisting mainly of ghost cells, and that the cyst development is a secondary phenomenon. They suggested a new descriptive term of ghost cell odontogenic tumor for the lesion.

This article discusses the clinical and histopathological correlation of the condition and its treatment and prognosis.

Report of a Case:

A 6-year-old male patient reported to our outpatient clinic with the complaint of swelling in lower left side of the jaw that had been present for approximately 1 month. On evaluation, there was an asymmetry involving the left lower

face region. Swelling was approximately 4 cm × 3 cm in size. Palpation revealed nontender hard bony expansion of the left mandible. It was presented with the lymph node enlargement in the submandibular region.

Intraoral examination revealed buccal as well as lingual cortical expansion extending anteroposteriorly from 72 to 75 regions, superiorly obliterating the mandibular vestibule. The mucosa over the lesion was intact (Figures 1). Radiographic examination disclosed a unilocular well circumscribed round radiolucency in the 74 region involving the unerupted 33. It involves the body of the mandible to lower border of mandible (Figures 2).



Figure 1: Intraoral view



Figure 2: Panoramic view

The operation was performed under local anesthesia by enucleation of the lesion (Figure 3), in agreement with the principle of clinical method for treating small cystic lesions of jaws. The enucleated specimen was cystic approximately 5mm to 4mm in diameter, entire specimen was sent for histopathological evaluation, and it was revealed as calcifying ghost cell odontogenic cyst (Figure 4).



Figure:3 Enucleation

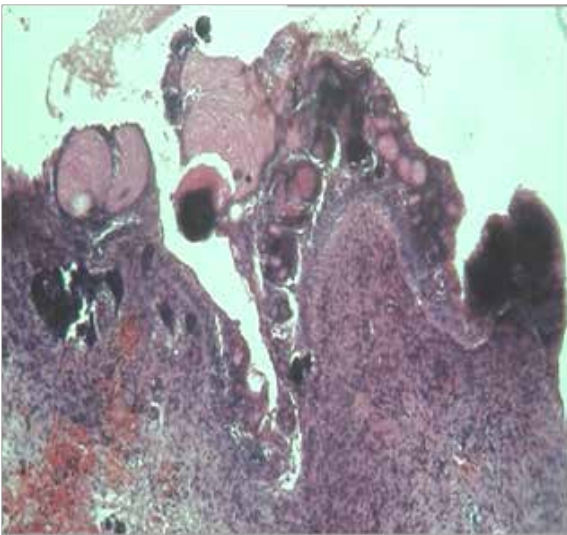


Figure 4: Histological View

The patient was reviewed every 6 months for past 7 years. The prognosis is good with no recurrence of the cyst (Figure 5, 6).



Figure 5:post operative view



Figure 6 post operative radiograph

Discussion:

In 1971, the WHO described CGOC as a "non-neoplastic" cystic lesion; nevertheless, it decided that the lesion should be classified as a benign odontogenic tumor. In 1992, the World Health Organization (WHO) classified CGOC as a neoplasm rather than a cyst but confirmed most of the cases are nonneoplastic. In view of this duality, many different terminologies have been applied to cystic and solid CGOC variants, but calcifying odontogenic cyst is the preferred term [8]. There was an almost even gen-

der distribution. In Asians, it showed a higher incidence in younger age group; almost 70% occurred in the second and third decades, whereas in whites, only about 53% occurred in the respective decades. Moreover, in the Asians, the lesions showed a predilection for the maxilla (65%), whereas in whites, the predilection was for the mandible (62%) [8]

The C.E.O.C. is known to involve mandible and maxilla with equal frequency [9]. 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. The cyst is usually asymptomatic unless secondarily infected.

In a case report of calcifying cystic odontogenic tumor mimicking as a residual cyst Manveen et al [10] concluded that a correlation of clinical and radiographic information with histological features is important in the diagnosis of odontogenic tumor and cyst. There are variety of lesions besides the typical granulomas and cysts that can appear at the apices of teeth. These other lesions must receive consideration in the diagnosis of periapical disease because of their potential impact on patient treatment and outcome. Unless the clinician is thinking in broad rather than narrow terms, serious conditions may go undiagnosed and untreated for an inappropriate period of time. [11,12]

Our case represents the classical features of calcifying odontogenic cyst, according to Praetorius et al. It comes under category of Type 1(a) simple unicystic type, and according to Reichart, it comes under the category of calcifying ghost cell odontogenic cyst (CGCOC) nonneoplastic (simple cystic) variant with proliferative epithelial lining. In the present case, the etiopathogenesis of COC could be from either a mutated clone of presecretory ameloblast or a postsecretory ameloblast that has acquired its mesenchymal inducing potential. It is probably the earlier one that has served as the source of the neoplastic clone. In course of the disease process, they have produced the characteristic ghost cells. The treatment of COC is enucleation and the response depends on any associated odontogenic tumor[13]. It has good and favorable prognosis, the condition would not recur again probably in future. However, we intend and continue to follow up the patient for further monitoring and documentation.

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