CASE REPORT

Calcifying ghost cell odontogenic cyst: A review on terminologies and classifications

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ABSTRACT

Calcifying ghost cell odontogenic cyst (CGCOC) is a relatively uncommon odontogenic lesion characterized by varied clinical, radiographical features and biological behavior. CGCOC can exhibit either as a cystic or a solid lesion. Since its first description by Gorlin *et al*, in 1962, it has been known by different names and classified and sub-classified into various types. In this article we present a case of CGCOC and discuss the related literature regarding the terminology, classification and biological behavior of CGCOC.

Key words: Classification, cyst, odontogenic, terminology, tumor

INTRODUCTION

Calcifying ghost cell odontogenic cyst (CGCOC) is a heterogeneous group of lesion existing either as cystic or solid variant.[1-4] The cystic lesion comprises majority of CGCOC accounting for 85% of cases.[5] Clinically, CGCOC may present either as central (85%) or peripheral lesion (15%). It shows bimodal age of occurrence commonly presenting in second and seventh decade of life.[1,3,6] CGCOC shows no predilection towards any gender and occurs in equal frequency in either of the jaw bones, anterior to the first molar in the incisor-canine region.[2,3,5,6] Asymptomatic bony expansion is the most common presentation of the central lesions, while sessile or pedunculated smooth surfaced mass are features of peripheral lesions.[1-3,6] Radiographically, the central lesion appear as unilocular or sometime multilocular radiolucency with or without calcified structures.[1,2,7] Size and opacity of the calcified structure varies, sometime occupying the entire lesional area.[2] CGCOC may be associated with an odontoma (24-35%) or an impacted tooth, most commonly the canine (10-32%).[2,3,5,7]

Despite the varied clinical and radiographical presentation, microscopic features of CGCOC are characteristic.[3] It consists of cystic cavity lined by 4-10 cells thickness of odontogenic epithelium and the fibrous wall.[8] Basal layer of the epithelial lining is composed of cuboidal or columnar-shaped ameloblast-like cells, overlying the basal layer, there are loosely arranged cells appearing similar to stellate reticulum of enamel organ.[1] Anucleated epithelial cells with retention of cellular outline are present either in the epithelial lining or connective tissue is a characteristic finding and are called 'ghost cells'. Individual ghost cells may fuse together to form large sheets of amorphous eosinophilic structure on which calcification may occur. Irregular masses of calcified structure suggestive of dysplastic dentin are present in association with basal layer denoting the inductive nature of the odontogenic epithelium.[3,4,8] Conservative enucleation or local resection is the commonly practiced mode of treatment. Recurrence is uncommon.[4,6,9]

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CASE REPORT

A 89-year-old female reported to our Dental clinic with the chief complaint of swelling in the right maxillary arch for past 1 month. History revealed that the patient underwent uneventful extraction of tooth no (FDI) 14 3 months back in a private Dental clinic and was the last tooth to be extracted before her complete edentulousness. One and half months after extraction, the patient was given maxillary and mandibular complete dentures. For the past 1 month the patient was unable to wear the maxillary denture due to mild discomfort in the region of 14 and 15 for which she was referred to our clinic. The past medical history was not contributory. On intraoral examination we observed completely edentulous maxillary and mandibular arch with mild swelling in the region of 14 and 15, measuring 3 × 3 cms. Overlying mucosa was sore in few areas possibly due to irritation by the denture [Figure 1]. On palpation the swelling was bony hard with mild tenderness. Orthopantamograph (OPG) revealed a radiolucent lesion in 14

Figure 1: Intraoral photograph showing completely edentulous arch with swelling in the 14 and 15 region

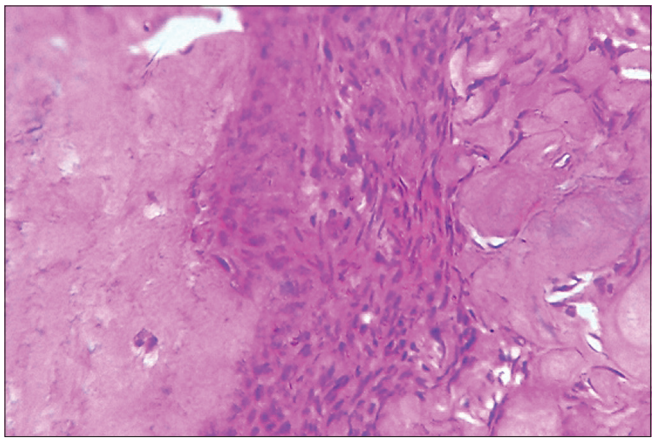

Figure 3: Low-power photomicrograph showing odontogenic epithelium and globules of eosinophilic structure suggestive of ghost cells (H and E stain, 10×)

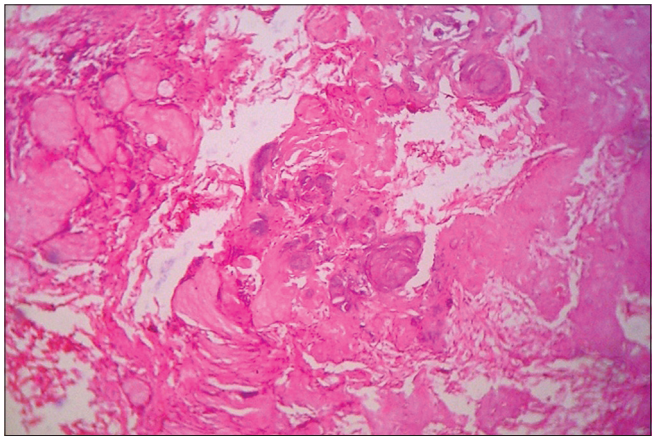

Figure 5: Early stages of calcification seen as basophilic granular areas. (H and E, ×10)

and 15 region. [Figure 2]. The history of extraction and clinical features made us to arrive at a diagnosis of residual cyst. The lesion was enucleated under local anesthesia and subjected to histopathological study. Histopathologically, the lesion

Figure 2: Ortho pantamograph showing radiolucent lesion in 14 and 15 region

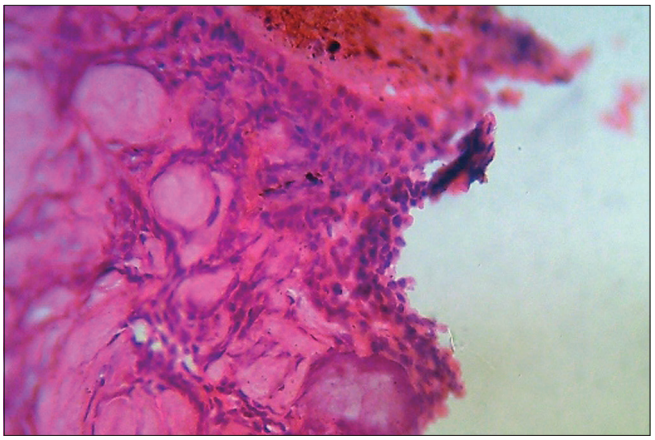

Figure 4: High power photomicrograph showing ghost cells (H and E stain, 40×)

showed a fragmented cystic odontogenic epithelial lining with associated fibrovascular connective tissue capsule. The epithelial lining was 3-4 cells thickness exhibiting globules of eosinophilic structure suggestive of ghost cells [Figures 3 and 4]. Some of the ghost cells appear to undergo dystrophic calcification [Figure 5]. With this we made a final diagnosis of calcifying ghost cell odontogenic cyst. The patient was followed-up for 1 year with no recurrence of lesion.

DISCUSSION

In 1962 Gorlin *et al*, was the first person to describe calcifying ghost cell odontogenic cyst (CGCOC) under the term calcifying odontogenic cyst (COC).[1,2,10] As the cystic variant comprises majority of CGCOC (85%), term COC was commonly used and still prevails in some literature.[3,7] From the year of description, disagreements exist regarding the nature, terminology and classification of CGCOC. These controversies and confusion about the lesion are due to existence of two variants of the lesion: cystic and the neoplastic forms. Some authors prefer to consider CGCOC as lesion existing in two forms either cyst or neoplasm - dualistic concept; others like to regard the lesion as a tumor with a marked tendency toward cystic architecture - monistic concept.[4,10]

In 1981, Praetorius *et al,* framed a classification based on dualistic concept in which they divided COC (as it was called then) into two entities: A cyst and a neoplasm and proposed the term dentinogenic ghost cell tumor (DGCT) for the neoplastic variant.[4,10] In 1991, Buchner classified COC majorly on clinical grounds – peripheral COC and central COC, further subclassifying each of them into cystic or neoplastic variants and included rare malignant variant of COC in the classification.[10] Similarly, in 1991, Hong *et al.* followed the dualistic concept and divided COC into cystic and neoplastic types. The cystic type is further subdivided into proliferative, nonproliferative, ameloblastomatous and odontoma associated. They used a term epithelial odontogenic ghost cell tumor (EOGCT) for the solid variant, the term that was originally proposed by Ellis and Shmooker (1986).[4,10]

Table 1: Classification proposed by Toida (1998)

Cyst: Calcifying ghost cell odontogenic cyst (CGCOC)
Neoplasm:
Beningn: Calcifying ghost cell odontogenic tumor (CGCOT)
Cystic variant: Cystic CGCOT
Solid variant: Solid CGCOT
Malignant: Malignant CGCOT
Combined lesion: Each of the categories described above (CGCOC, CGCOT, malignant (CGCOT) associated with the following lesions:
a. Odontoma
b. Ameloblastoma
c. Other odontogenic lesions

Table 2: Commonly used terminologies for CGCOC[3,4,10]

Terminology	Author proposed
Calcifying odontogenic cyst (COC)	Gorlin *et al*. (1962)
Keratinizing Calcifying odontogenic cyst (KCOC)	Gold *et al*. (1963)
Keratinizing ameloblastoma (KA)	Bhaskar (1965)
Calcifying ghost cell odontogenic tumor (CGOT)	Fejerskov and Krogh (1972)
Cystic calcifying odontogenic tumor (CCOT)	Freedman *et al.* (1975)
Dentinogenic ghost cell tumor (DGCT)	Praetorius *et al*. (1981)
Epithelial odontogenic ghost cell tumor (EOGCT)	Ellis and Shmooker (1986)
Calcifying ghost cell odontogenic cyst (CGCOC)	Toida (1998)
Odontogenic ghost cell tumor (OGCT)	Colmenero *et al*. (1990)
Odontogenic ghost cell ameloblastoma (OGCA)	Shear (1994)
Odontocalcifying Odontogenic tumor (OOT)	Wirshberg *et al*. (1994)
Calcifying cystic odontogenic tumor (CCOT)	WHO classification (2005)

In 1998, Toida indicated the demerits of above classification systems and clarified that the lesion should not be grouped into "cystic" or "neoplastic" variant. Toida stated that in above classifications the term "cystic" is used synonymous for "non-neoplastic" which was incorrect because the former term describes the morphological pattern while the latter term defines the biological behavior of the lesion. He further mentioned that there may be lesion with cystic architecture with extensive proliferating capacity.[10] Toida proposed a classification in which he called CGCOC for the cystic variant and used the term calcifying ghost cell odontogenic tumor (CGCOT) for the neoplastic variant, the latter term was originally proposed by Fejerskov and Krogh (1972). He further subdivided the neoplastic group into –cystic CGCOT and solid CGCOT, to include neoplasm showing cystic architecture and neoplasm with solid pattern, respectively.[10] [Table 1].

From the above described classifications it is plausible that the dualistic concept was mostly followed, considering CGCOC as a lesion existing in two forms, cyst and neoplasm. However, the approach of WHO toward CGCOC was different and mostly followed monistic concept. In 1971, WHO described the lesion as non-neoplastic cystic lesion and preferred to use the term COC.[6,11] In 1992, WHO classified this lesion under odontogenic tumor but continued to use the term calcifying odontogenic cyst. As the terminology was misleading and did not explain the complete behavior of the lesion, in 2005, WHO again renamed the lesion as calcifying cystic odontogenic tumor.[6]

From the year of description of CGCOC in 1961 till date different terminologies and classifications have been proposed and practiced in the literature [Table 2]. In these terminologies, some characteristic features of the lesion are considered and named accordingly, like origin of the lesion (odontogenic epithelium- COC, CCOT, EOGCT), histopathological features (calcifying structure, dysplastic dentin, ghost cell- DGCT, CGCOC, OGCT), and architectural pattern (cystic CGCOT, solid CGCOT, COC). In spite of various terminologies and classifications, discrepancies prevail over the usage of terminology and still some authors which prefer to use the older terminologies.

Inadvertent use of the term COC (Gorlin 1962) for these lesions carries the possibility of masking the real biological behavior of the solid neoplastic variant and neoplastic with cystic architecture, which has high proliferating index,[12] On the other hand use of the term CCOT (WHO 2005) for the lesion may result in unwanted extensive surgical procedure for the cystic subtypes.The authors would like to conclude by suggesting that, use of nomenclature should emphasize on biological behavior of the lesion rather than familiar or older terms, so that lesion can be approached and treated accordingly. For example, nomenclature carrying a phrase "cystic" is generally approached relatively less vigorously (enucleation or marsupialization), than nomenclature carrying a phrase "tumor", which are treated more

aggressively (*en bloc* resection) and followed-up precautiously for longer period.[6,9,13] Terminology and classification proposed by Toida as cystic (CGCOC), neoplastic (CGCOT) variant with subclassifying the neoplastic types into cystic CGCOT and solid CGCOT not only avoids confusion but also helps in planning the treatment accordingly.

CONCLUSIONS

Presentation of lesions with controversial historical description, terminologies and clinical behavior should be encouraged so that it provides opportunities to understand the actual incidence, biological behavior, treatment and recurrence. Nomenclature and classification of those particular lesions should be re viewed periodically and should be followed universally.

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