

## Peripheral Calcifying Odontogenic Cyst of Mandible: A Case Report and Discussion

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**Abstract:** The calcifying odontogenic cyst (COC) was described by Gorlin et al. in 1962 as a distinct entity from other odontogenic lesions. Calcifying odontogenic cyst accounts for less than 2% of all odontogenic cysts and tumors. This paper presents a case report of 40 year male patient present with history of non-tender swelling of left mandibular vestibule area. Periapical and panoramic radiograph showed No bone changes. Excisional biopsy was performed and Microscopic examination revealed odontogenic cystic lesion lined by ameloblastoma like epithelium, numerous ghost cells and irregular calcified tissue. Patient was diagnosed with Peripheral variety of COC. PCOC usually appears in the age group of 40-45 years with no signs and symptoms except non symptomatic swelling, surgical excision is the treatment of choice and presents with rare recurrence.

**Key Words:** peripheral calcifying odontogenic cyst, extraosseous variety, mandible posterior

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### I. Introduction

The calcifying odontogenic cyst (COC) is an uncommon odontogenic lesion. COC presents both in intraosseous (central) and extraosseous (peripheral) locations. The intraosseous COC is a unilocular or multilocular radiolucent lesion with destructive nature that may contain irregular calcifications and ghost cells. The peripheral COC (PCOC) represents less than 25% of the all COC. Carson et al. 1998 suggested that COC represent a non-neoplastic lesion, but it has a potential for continuous growth. A lot of confusion and disagreement is present in the terminology and classification of COC. Some investigators have considered COC as a tumor with a tendency for marked cyst formation. This concept, called "monistic" by Toida (1998), has lead some researchers to substitute the terms "calcifying ghost cell odontogenic tumor" or "cystic calcifying odontogenic tumor" for that of COC. In addition, a "dualistic" approach (Toida 1998) has been suggested, in that COC can contain 2 entities: a cyst and a neoplasm. Indeed, Praetorius et al. (1981) divided COC in 2 groups, cystic (1) and neoplastic (2), recognizing different histologic patterns in them: (1a) simple unicystic; (1b) odontome producing; (1c) ameloblastomatous proliferating; (2) dentinogenic ghost cell tumor. The World Health Organization-2005 (2005 WHO) [2] renamed these lesions as calcifying cystic odontogenic tumors.

### II. Case Report

A 41-year-old male patient with a non-contributory medical history reported to the Department of Oral and Maxillofacial Surgery, Government Dental College, Thiruvananthapuram, Kerala with a chief complain of soft tissue swelling inside mouth since past 2 month. No history of past debilitating medical condition was mentioned. The patient denied the history of smoking or any other harmful habits, however occasional consumption of alcohol was mentioned.

On detailed examination no extra oral abnormalities were noted. Intra oral examination revealed a well-defined firm to semi firm swelling of around 2×2×1.5cm near vestibule of 36, 37 tooth region. The lesion clinically appeared to be a non-reactive. No surface changes evaluated on clinical examination. No missing tooth, caries or periodontal problems identified on clinical examination. No lymphadenopathy noted.

A Orthopantomogram and a periapical radiograph showed that there was no erosion of the underlying bone or the presence of radiolucency. Clinical differential diagnosis of lateral periodontal cyst, minor salivary gland neoplasm, soft tissue neoplasm was expected. Lesion was planned for excisional biopsy under local anaesthesia, the lesion was completely removed. Intra operative lesion appeared well encapsulated cystic

swelling arising from vestibule region of 36, 37 tooth region. Bony cupping was there with smooth margins. Primary closure was done.

Microscopic examination reveals serial sections of H&E stained tissue showing a moderately collagenous and moderately cellular connective tissue capsule lined by a lining of odontogenic epithelium. The basal cells of epithelium were columnar and similar to ameloblasts showing reversal of polarity and palisading pattern. The overlying layer of loosely arranged epithelium resembled stellate reticulum. Epithelium and connective tissue showed a variable number of ghost cells with some of the ghost cells fused to an amorphous eosinophilic matrix. Eosinophilic matrix resembling dentinoid was seen adjacent to epithelium. Stroma showed a diffuse collection of chronic inflammatory cells, mainly lymphocytes and plasma cells. Vascularity was moderate. Periphery showed some osteoid tissue. The definite microscopic diagnosis was Peripheral calcifying odontogenic cyst. No recurrence was present at a three-month follow-up.

### **III. Discussion**

COC is usually asymptomatic and usually found on routine radiographs. Seventy-four percent of the maxillary lesions affected the anterior region while 56% of the lesions were located in the mandible. Radiographically, the lesions appear as unilocular or multilocular well-defined radiolucencies, and may be associated with unerupted teeth. PCOC has been reported to represent less than 25% of all cases. There is also the possibility that PCOC is somewhat more common, and that some cases of PCOC have been probably classified as other lesions, such as peripheral ameloblastoma.

They are usually located on the gingiva or edentulous ridge. Their clinical appearance is that of a painless, circumscribed, pink or red, and sometimes papillary nodule. The swelling is mostly smooth, with a firm or soft cystic texture, and is about 0.5-1 cm in diameter. The incisor-canine or premolar regions of the mandible are most frequently involved. About 55% of PCOC have been found between the canines. Literature suggests about 25% of cases associated with bony erosions. About 30% of PCOC are solid rather than cystic (against 2% of CCOC), which can be related to their small size. PCOC has a less aggressive behavior than the intraosseous counterpart (CCOC) and a simple excision is curative.

Microscopically, it is possible to find the presence of thick-walled cysts that have a smooth outer surface and a semisolid content. Usually there is a single cystic cavity or multiple smaller cavities. The cysts are lined by an irregular epithelium, variable in thickness, and is composed of a columnar or cuboidal layer of preameloblast-like basal cells with reversed polarity of their nuclei. The epithelial lining of COC sometimes has the capacity to induce the formation of dental tissue in the connective tissue wall, mainly in the form of dentinoid type material or odontomas. Indeed, COC can be found in association with odontogenic tumors such as odontoameloblastoma, adenomatoid odontogenic tumor, calcifying epithelial odontogenic tumor, and ameloblastoma. The ghost cells forming the suprabasal layers are large and lightly eosinophilic, with a cytoplasm containing diffuse tonofilaments, but not ortho- or para-keratin, and showing a faint outline of the cellular and nuclear membrane. These ghost cells may form small foci within the epithelial lining or fuse into large masses, forming extensive sheets of an amorphous, acellular eosinophilic material, extending or even filling the cyst lumen. Mineralization of the ghost cells is not uncommon. They may also invade the connective tissue, causing a foreign body reaction. The presence of ghost cells in COC is not pathognomonic, having been described in ameloblastoma, ameloblastic fibroma, ameloblastic fibro-odontoma, and odontomas. The microscopic differential diagnosis must be made with peripheral ameloblastoma.

Like PCOC, peripheral ameloblastoma also occurs at a significantly older age than its counterpart. Both lesions share the presence of prominent elongated basal cells and stellate reticulum zone, but the presence of ghost cells is rare in ameloblastoma. It is still very difficult to determine whether an individual lesion having a cystic architecture is truly cystic or neoplastic. Further studies, including immunohistochemical investigation on cell proliferation activity, may help in resolving the question. Proliferating cell nuclear antigen labeling index is a possible parameter for differentiating benign from malignant COC, and the proliferative features in the lining seem to be the main factor influencing the proliferating activity of COC.<sup>18</sup>

The presented case, similar to the other reports in the literature, had an asymptomatic localized gingival swelling. Clinical presentation of the PCOC is often described as variable or nonspecific. In the reported case, we believe the neoplastic epithelium arose from the odontogenic remnants of the overlying mucosa due to the lesion's intimacy with the oral surface and absence of tooth or bone involvement. But whether the cyst develops as a central or peripheral lesion probably depends on the location of odontogenic epithelium, which constitutes the source of the lesion. Nevertheless, the location does not seem to have any relation to either behavior or histologic features of the cyst. Treatment of the peripheral COC is surgical excision, and recurrences are rare.

Generally, cystic COCs have a good prognosis, but the neoplastic cases are uncertain. When a COC is associated with other odontogenic tumors, treatment and prognosis must be based on the associated lesion.

**PRE OPERATIVE:**

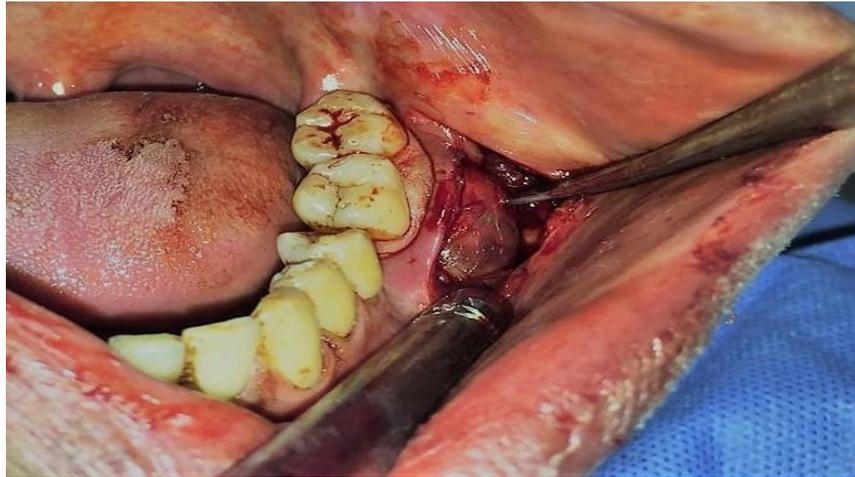


**RADIOGRAPH:**



**INTRA OPERATIVE:**





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