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Case Report

Calcifying Epithelial Odontogenic Cyst - A Case Report

V. Swabna[1], B.Sekar[2], Maya Ramesh[3], B.S. Saravanan[4]

[1]Post Graduate Student, [2]Professor and HOD, [3]Reader, Department of Oral Pathology, Vinayaka Mission's Sankarachariyar Dental College, Salem.4 Oral and Maxillofacial Cosmetic Surgeon, Dentofacial Multispeciality Dental Clinic.

***ABSTRACT*:**
Calcifying odontogenic cyst or Gorlin cyst, was first described by Gorlin. It is an unusual and unique lesion, which may show characteristics of both a solid neoplasm and a cyst.This lesion occurs intraosseously as well as extraosseously, with about equal frequency in the mandible and maxilla (1:1). It has also been said that both the "calcifying odontogenic cyst" and the calcifying epithelial odontogenic tumor share a process of epithelial proliferation with subsequent calcification of some portions. Histologically calcifying cystic odontogenic cyst consists of ameloblastoma -like odontogenic epithelium with reverse polarization and calcifying ghost cells overlying a mature connective tissue with odontogenic rest. In this case report a middle aged patients lesion in the mandible is been discussed.

Key words: Calcifying epithelial odontogenic tumor, calcifying epithelial odontogenic cyst, ghost cells, Gorlin cyst.

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Corresponding Author: Dr. V. Swabna, Post Graduate Student, Department of Oral Pathology, Vinayaka Mission's Sankarachariyar Dental College, Salem, India

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INTRODUCTION:

The Calcifying epithelial odontogenic cyst (CEOC) was first categorised as a distinct entity by Gorlin in 1962 (1). It is also called as Gorlin's cyst or calcifying odontogenic cyst (COC). In the second decadeof life this cyst is commonly witnessed (2). In terms of its clinical presentation, histopathological features and biological behaviour, this cyst is extensively diverse. Many of the cases were present with cystic characteristics, while a few were of the solid type (15%), and rare occurrence of malignant transformation (3). The common site in which CEOC arises is the gingiva or alveolar process of the maxilla or mandible. And so it has been confused with the calcifying epithelial odontogenic tumor(4). In most of the cases examined, varying degree, shape, and size of opacity was found. In this cyst it was found that root resorption and tooth displacement was not rare. Other odontogenic lesions mainly odontomas and amelobastomas is been accomplished by CEOC(5).

CASE REPORT:

A 30 -year-old male patient visited our Vinayaka Mission's Sankarachariyar dental college at Salem with a chief complaint of a hard swelling in mandible for more than a year. The swelling was then examined intraorally which was seen to be extending from 35 to 42. The radiograph was obtained which showed root resorption present from 35 to 42.

The provisional diagnosis was given as calcifying epithelial odontogenic tumor.

Microscopically the given histopathologic section of soft tissue specimen shows hyperchromatic non keratinized lining epithelium and fibrous connective tissue. The lining epithelium showing proliferation into the lumen and is composed of ghost cells with eosinophilic cytoplasm. The connective tissue is more fibrous. Numerous normal bony trabeculae are also seen. Blood vessels and inflammatory cells are also present.

The final diagnosis was given as Calcifying Epithelial Odontogenic Cyst.

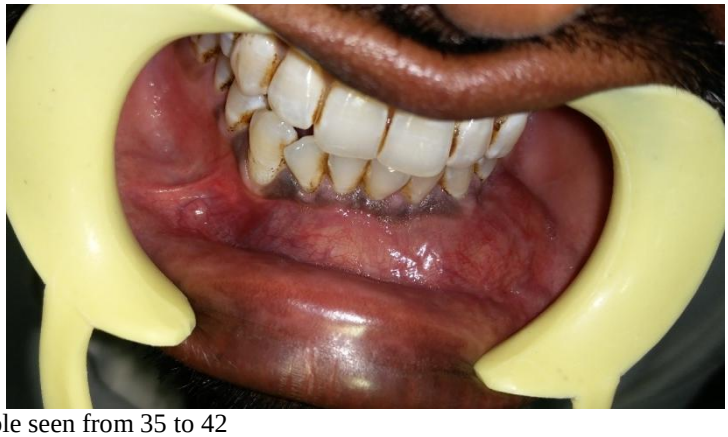

Fig 1: Swelling of mandible seen from 35 to 42

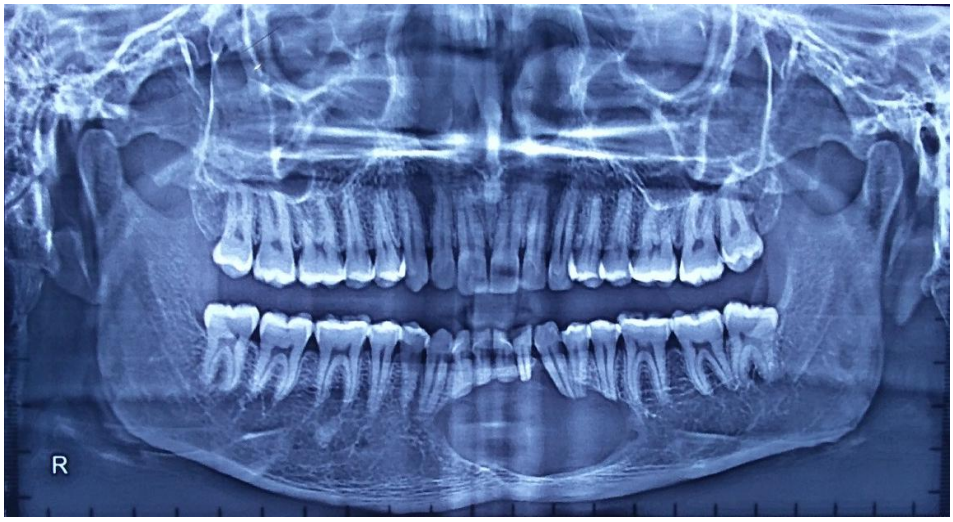

Fig 2: The OPG shows clear swelling involving the mandible from 35 to 42. The root apices of the corresponding teeth are also resorbed.

DISCUSSION:

It is seen that 1% of all the cysts of the jaw bones is been represented as calcifying epithelial odontogenic cyst. First time in the year 1932 this lesion was described by Rywkind, Gorlin, who was often credited as he defined it as an entity, histologically distinct from the calcifying epithelial odontogenic tumor(6). The WHO in 1971 described CEOC as a non-neoplastic cystic lesion by choosing it to be classified as a benign odontogenic tumor(3). In 1992, WHO classified CEOC as a neoplasm rather than a cyst but confirmed most of the cases are non-neoplastic. Because of this duality, many different terminologies have been applied to cystic and solid CEOC variants, but calcifying epithelial odontogenic cyst is the preferred term (3).

In 2005 the WHO reclassified COC into three subgroups:

- Calcifying Cystic Odontogenic tumor (CCOT),
- Dentinogenic ghost cell tumor (DGCT),
- Ghost cell odontogenic carcinoma (GCOC)(7).
-

In the mandible anteriorto the first molar region this cyst is most commonly witnessed. In which 75% of cases are in the incisorcanineregion or inter-canine region, usually crossing the midlinein the mandible. This cyst is however rarely seen in the maxilla (8). This is very much similar to our presented case where the lesion is in the mandible crossing the mid line.

In our case report the provisional diagnosis was given as calcifying epithelial odontogenic tumor and the final diagnosis was calcifying odontogenic cyst. The odontogenic tumors and as well as the cysts have diverse histological appearances which would be originated from the epithelium or the mesenchyme or both. This diversity causes difficulties on consensus about theclassification of these lesions since 1960's (5). The definitive diagnosis of CEOC can be made more appropriately only histologically, due to the lesion's lack of characteristic clinical and radiological features, as well as its variable biological behaviour (3). Since CEOC hasdistinct histopathological finding it was classified in heterogeneous group of entities that included solid tumour. In spite of the low frequency of this lesion, most cases are surgically removed and heal uneventfully(1).

It is seen that CEOC if it is associated with apices of teeth, there is a high incidence of root resorption(1) this was similar to our case where there was root resorption seen in the apices.

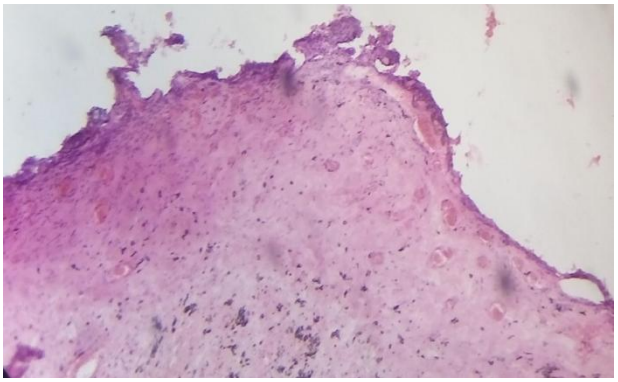

Fig 1: Photomicrographic view under Low power showing cystic lumen, cystic epithelium and underlying connective tissue. Odontogenic epithelial islands are seen in connective tissue.

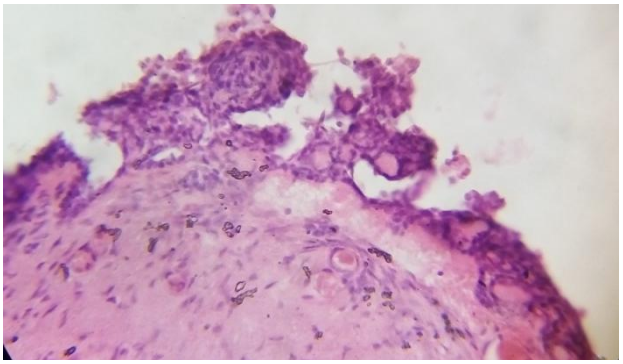

Fig 2: Photomicrographic view under High power showing stratified squamous epithelium showing ghost cells. Underlying connective tissue is loose and fibrillar.

In the histopathological examination Robert .J. Gorlind et al told that the shadow cells, like those of the "calcifying odontogenic cyst," have lost all nuclear and most cytoplasmic detail. They are eosinophilic and presumably consist of keratin(4). This feature is similar to the ghost cell feature seen in our case.

Microscopically, it is possible to find the presence of thick-walled cysts that have a smooth outer surface and a semisolid content. The cysts are lined by an irregular epithelium, variable in thickness, and is composed of a columnar or cuboidal layer of pre- ameloblast-like basal cells with reversal of polarity of their nuclei (9). This feature is present in our case where the lining epithelium showed proliferation into the lumen and is of variable thickness which was been composed of ghost cells with eosinophilic cytoplasm. The connective tissue was more fibrous.

The ability to induce dental hard tissue formation appears to be a property of epithelial cell lining of the CEOC. The malignant transformation of a pre-existing benign CEOC could occur, yet is extremely uncommon (3).

Generally, cystic CEOCs have good prognosis, but the neoplastic cases are uncertain. When a CEOC is associated with other odontogenic tumors, treatment and prognosis must be based on the associated lesion (9).

CONCLUSION:

CEOC is always seen as a unique lesion which is rare in its occurrence. It has both cystic and neoplastic potential and showing considerable number of variants clinically, radiographically, and histopathologically(3). In addition it is proved that a specific knowledge in oral histopathology is required to differentiate all other odontogenic lesions from CEOC (2).The CEOC may also be associated with other odontogenic tumors such as adenomatoid odontogenic tumor, ameloblastoma, ameloblastic fibro-odontoma and ameloblastic fibroma, where wider excision may be required. The recurrence and malignant transformation of CEOC is uncommon (8).

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