Case Report

Calcifying Epithelial Odontogenic Cyst - A Case Report

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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 patient's 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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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 decade of 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 Sankaracharyar 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 anterior to the first molar region this cyst is most commonly witnessed. In which 75% of cases are in the incisor-canineregion or inter-canine region, usually crossing the midline in 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 the classification 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 has distinct 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.
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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-
odonta and ameloblastic fibroma, where wider
excision may be required. The recurrence and malignant
transformation of CEOC is uncommon (8).

REFERENCES:
1. Priyank Rai et al Calciying Odontogenic Cyst of
Mandible: A Case Report. Indian Journal of Mednodent
and Allied Sciences Vol. 5, No. 2, June 2017, pp- 129-
133.
2. GoranKne`evi et al Calciying Odontogenic Cyst –
3. AshutoshVatsyayan et al Calciying Odontogenic Cyst:
Case Report and Review of Literature. International
Journal Dental Medical Research [ NOV - DEC 2014 ]
VOL 1 | ISSUE 4
4. ROBERJT. GORLIND,.D .s., M.s., et al The Calciying
Odontogenic Cyst - A New Entity and Possible Analogue of
the Cutaneous Calciying Epithelioma of Malherbe.
CANCER June 1964 Vol. 17. 723- 729.
5. BurceuSengiven et al Mandibular Calciying Odontogenic
Cyst: A Case Series. Journal of Dentistry and Oral
Biology. 2017; 2(17); article 1100.
6. ManikkathAparna et al Calciying Odontogenic Cyst: A
Rare Report of a Nonneoplastic Variant associated with
Cholesterol Granuloma. The Journal of Contemporary
Dental Practice, November-December 2013;14(6):1178-
1182.
7. Sang Yoon Park et al Ghost cell odontogenic carcinoma
on right mandible and its respective surgical
reconstruction: a case report. Journal of Korean
8. Nigel R. Figueiredo et al Calciying odontogenic cyst of
mandible. Journal of oral research and
9. Dr. Manoj S et al Peripheral Calciying Odontogenic Cyst
of Mandible: A Case Report and Discussion. IOSR
Journal of Dental and Medical Sciences (IOSR-JDMS).

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