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Calcifying Odontogenic Cyst – a case report.

Dental Science

KEYWORDS: Calcifying Odontogenic Cyst , Peripheral Calcifying Odontogenic Cyst, Ghost Cells.

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ABSTRACT

Calcifying odontogenic cyst is an uncommon lesion that demonstrates considerable histopathological diversity. It has recently been classified under the group of odontogenic neoplasms and designated as dentigenic ghost cell tumor. Predominantly, it occurs in the anterior region of the mouth and in the second and third decades of life. Odontogenic tumors such as ameloblastoma have been reported to be associated with CCOT.

1. INTRODUCTION -TThe calcifying odontogenic cyst (COC) is a rare odontogenic lesion which was first described as a distinct entity by Gorlin et al[1] in 1962. COC is a cystic lesion in which the epithelial lining shows a well-defined basal layer, an overlying layer resembling stellate reticulum, and masses of ghost epithelial cells located in the epithelial cyst lining or in the fibrous capsule. COC represents about 1% of jaw cysts, most of which are located anterior to the first permanent molars. Central COC (CCOC) is found most commonly in the second decade of life (more than 40% of cases); while peripheral COC (PCOC) affects individuals most commonly in their sixth decade of life[2].

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[3] has led 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 has been suggested, that COC can contain two entities: 1) cyst 2) neoplasm (benign: calcifying ghost cell odontogenic tumor; or malignant: malignant calcifying ghost cell odontogenic tumor); and combined lesion: each of the categories described above associated with odontoma, ameloblastoma, or other odontogenic lesions.

Indeed, Praetorius et al.4 divided COC to two groups, 1) cystic and 2) neoplastic, considering the different histologic patterns in them. The cystic form can be (1a) simple unicystic, (1b) odontoma producing, and (1c) ameloblastomatous proliferating. The neoplastic form comprises dentinogenic ghost cell tumor.[4] The latter tumor that may have an infiltrative growth pattern is a predominantly solid lesion with features of ameloblastoma, ghost cells, and dentinoid.[3] Other investigators proposed for this neoplastic counterpart the term "epithelial odontogenic ghost cell tumor" and "odontogenic ghost cell tumor".[5]

Subsequently, WHO, which first recognized and defined the COC as a non-neoplastic cystic lesion, classified the entity and its variants as an odontogenic tumor rather than an odontogenic cyst. The neoplastic lesions were subdivided to three subgroups based on location (intraosseous and extraosseous) and histologic features.[6]

2.CASE REPORT

A female patient of 40 years age reported the out patient department with a complain of pain and swelling in left upper back region of jaw. The lesion had an abrupt onset which gradually increased in size.

On evaluation, there was an asymmetry involving the left midface region. Swelling was approximately 3cm x 2cm in size, extending superioinferiorly from 1cm below infraorbital rim to angle of the mouth and anteroposteriorly from left ala of the nose to about 3 cm in front of the tragus (Fig 1a). Palpation revealed non tender hard bony expansion of the left maxilla.

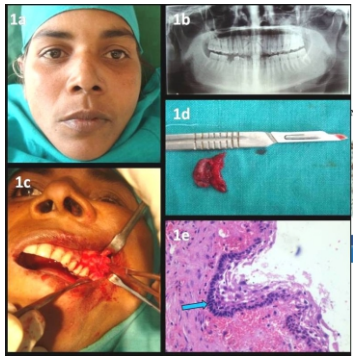
On inspection Intraoral examination revealed distinct boundaries extending anteroposteriroly from 25 to 28 region and the color of the mucosa is red. The mucosa overlying the lesion was intact. On palpation the borders of the lesion are well defined, consistency was soft and the lesion is mobile.

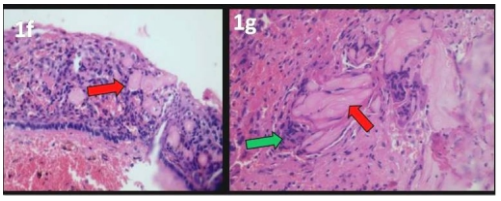
Radiographic expression in the form of OPG revealed unilocular lesion with loss of lamina dura involving 25-28 region (Fig1b).

Based on clinical and radiological findings, differential diagnosis of 1. Cystic Odontome, 2. Odontogenic Keratocyst, 3. Calcifying Odontogenic Cyst, 4. Eruption Cyst, 5. Dentigerous Cyst was made.

FNAC was done but it was not conclusive.

The following treatment was then carried out. Enucleation of the cyst was done (Fig 1c) and the tissue biopsy (Fig1d) was sent to the diagnostic laboratory for investigations. Macroscopic features shows the tissues were brownish black in color, firm in consistency and together measured 2.8 x 2.2 x 2.0 cm. The larger tissue was cut into two. One tissue part was kept for routine processing. Microscopic features reveals a cystic lumen surrounded by a fibrous connective tissue capsule with a small fragment of lining epithelium lying separated from capsule. The epithelium shows basal layer made up of ameloblast like tall columnar cells and a supra basal layer of stellate reticulum like cells (Fig 1e). Small foci of large round to polygonal cells with abundant eosinophillic cytoplasm with some have pyknotic nucleus and other devoid of nucleus suggestive of ghost cells, are seen within the epithelium(Fig 1f). The connective tissue capsule is moderately collagenous with foci of chronic inflammatory infiltrate and few islands of odontogenic epithelium with giant cells and reactive bone formation at the periphery. Small fragments of overlying gingival epithelium and lamina propria are also evident(Fig 1g)





3. DISCUSSION

Gorlin and colleagues (1962)[1] were the first to describe this entity. They initially regarded it as the oral analogue of the cutaneous calcifying epithelioma of Malherbe but later labelled it the calcifying odontogenic cyst. Prior to this, Rywkind (1932)[7] described it as a variant of the cholesteatoma, while Maitland (1947)[8] regarded it as a type of ameloblastoma. The COC has also been reported under a variety of other designations including: dentinogenic ghost cell

odontogenic tumour[4] epithelial odontogenic ghost cell tumour,[9] ghost cell cyst,[10] calcifying ghost cell odontogenic tumour,[11] and dentino-ameloblastoma.[12] The WHO presently describe the calcifying odonto- genic cyst as a non-neoplastic cyst, but classify it as a benign tumour.[6].Langlais and colleagues (1995)[13] proposed the term calcifying odontogenic lesion (COL), which encompasses both the cystic and tumourous forms as well as combined lesions containing elements of both.

A dentigerous cyst is one which encloses the crown of an unerrupted tooth by expansion of its follicle and is occurs in first decade of life and more frequent in males. Radiographically it appears as a unilocular radiolucent area associated with the crown of unerupted teeth having well defined sclerotic margins. Eruption cyst occurs when a tooth is impeded in its eruption within the soft tissues overlying the bone and is found in children of different ages and occasionally in adults. It produces a smooth swelling over the erupting tooth which may be of normal gingival colour or blue. It is painless. On the radiograph the cyst may throw a soft tissue shadow but there is no bone involvement. Cystic Odontome is a rare condition where a cyst develops in relation to the complex composite or compound composite odontome. They usually develop in the lower molar or upper incisor region. The radiographic appearances are those of an area of bone destruction which appears as a dark shadow having well defined margins lined by a thin white layer of cortical bone. The odontogenic keratocyst may occur at any age but the peak incidence is in the 2nd and 3rd decades of life. The mandible is more frequently affected than the maxilla and has the characteristic “egg shell crackling” on palpation. The lesion appears as either a unilocular or multilocular radiolucency with a thin sclerotic border which may be smooth or scalloped but is generally sharply demarcated. Calcifying odontogenic cyst is n extremely rare lesion with peaks in 2nd and 6th decades and the anterior part of the jaw is the commonest site of occurrence.

Radiographically, they appear as radiolucent areas often associated with complex odontone leading to the appearance of dense opacities in the cyst. Resorption of roots of adjacent teeth is a frequent finding and a very important x-ray feature.

Hence, after correlating he clinical and radiographic picture a provisional diagnosis of 'Calcifying Odontogenic Cyst (COC)' was made.

4.CONCLUSION

The diagnosis of pathological alteration of odontogenic etiology appears very simple. But an excellent knowledge of the structure is necessary for correct diagnosis. An optimal solution would be the existence of a clinical pathologist, who would arrive at the final diagnosis in co-operation with an oral surgeon. For better determination of histopathologic & biologic nature of COC more investigations based on histomorphological and immunohistochemical studies are needed.

Legends for figures:

Fig 1a: Photograph showing asymetry of face on the left side.

Fig 1b: Photograph showing unilocular lesion with loss of lamina dura involving 25-28 region.

Fig 1c: Surgical view of cystic lesion

Fig 1d: photograph showing cystic lesion

Fig 1e: The epithelium shows basal layer made up of ameloblast like tall columnar cells and a supra basal layer of stellate reticulum like cells.

Fig 1f: Small foci of large round to polygonal cells with abundant eosinophilic cytoplasm with some have pyknotic nucleus and other devoid of nucleus suggestive of ghost cells are seen with in the epithelium.

Fig 1g: Focal areas showed calcification of ghost cell mass(red) ,herniation of ghost cells(green) into the lumen as well as the fibrous capsule.

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