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CALCIFYING ODONTOGENIC CYST IN EDENTULOUS MAXILLA: AN UNUSUAL PRESENTATION

Medicine

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ABSTRACT

The calcifying odontogenic cyst (COC) or Gorlin Gold cyst is an uncommon benign cyst of odontogenic origin.COC shows mixed radiolucency and radiopacity showing cystic and solid neoplastic characteristics. This cyst often shows characteristic features of ameloblastoma and may contain ghost cells, tooth like material and calcified deposits. There are variants of COC according to clinical, histopathological, and radiological characteristics. Therefore a proper categorization of the cases is needed for better understanding of the pathogenesis of each variant. Here, we report a case of calcifying odontogenic cyst in upper anterior edentulous area in old female patient. In this case cone beam computed tomography (CBCT) could accurately reveal the extent and the internal structure of the lesion for the diagnosis of the lesion as COC.

KEYWORDS

Calcifying Odontogenic Cyst, Gorlin Gold cyst, Cone Beam Computed Tomography.

INTRODUCTION

Calcifying odontogenic cysts (COC) is a very common odontogenic cyst. It is also called as Calcifying cystic odontogenic tumor (CCOT) which is a mixed radiolucent-radiopaque lesion of the jaws with features of both a cyst and a solid neoplasm.[1] *Gorlin* initially called this cyst as Gorlin's cyst on 1962 and in1963 *Gold* termed it as Keratinising and Calcifying odontogenic cysts. *Gorlin* reported that COC may be the oral analogue of dermal calcifying epithelioma of Malherbe. The first author to publish COC was *Rywkind* and later on he termed it as cholesteatoma of jaw.[1]

The WHO Classification (2005) of odontogenic tumors re-named this entity as Cystic calcifying odontogenic tumor (CCOT) and Dentinogenic ghost cell tumor (DGCT) representing the cystic and neoplastic variants respectively contains 'ghost cells' and spherical calcification.[2]Definition of COC according to WHO as "a cystic lesion in which the epithelial lining shows a well-defined basal layer of columnar cells, an overlying layer that is often many cells thick and that may resemble stellate reticulum, and masses of ghost epithelial cells that may be in the epithelial cyst lining or in the fibrous capsule".[3] The histological variation of CGCOC has led to different terminologies such as CGCOT (Fejerskov and Krogh,1972), dentinogenic ghost cell tumour, epithelial odontogenic ghost cell tumour (Ellis and Shmookler, 1986),and odontogenic ghost cell tumour (Colmenero et al,1990).The epithelial lining shows unifocal or multifocal intraluminal proliferative activity that resembles ameloblastoma, although it also contains isolated or clustered ghost cells and calcifications.[4]

We hereby describe a case report of an old patient with COC in edentulous maxilla focusing on the CBCT findings of the lesions.

CASE REPORT

A 84-year-old female patient reported to the department with the chief complaint pain and swelling in the upper front tooth region of the jaw since 2 months. Pain was insidious in onset, continuous, mild in nature and localized associated with swelling.Swelling gradually increased in its size.

Figure 1: (a) and (b): Clinical photographs showing solitary swelling in the left anterior maxillary region (c): Post-Operative Intraoral View.

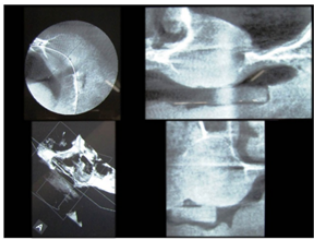

Figure 2: CBCT shows axial,coronal and sagittal section demonstrates the lesion extending into the maxillary sinus.

On intraoral examination we observed completely edentulous maxillary and mandibular arch with a solitary swelling in the region of 21 and 25, measuring 3 × 3 cms. On palpation the swelling was bony hard with mild tenderness. Buccal and palatal cortical expansion seen .The mucosa over the lesion was intact (Fig 1 (a and b).On Fine needle aspiration straw coloured fluid seen.

Intraoral periapical radiograph showed a well defined radiolucency seen. Panoramic radiograph disclosed a unilocular wellcircumscribed round radiolucency extending from 21 to 25 regions. The lesion was surrounded by a sclerotic margin.CBCT images revealed a large, well-defined unilocular radiolucent expansile lesion with thinned cortical outline involving anterior maxilla.(Fig 2).

Based on the clinical and radiographical features, differential diagnosis of residual cyst, radicular cyst and calcifying odontogenic cyst were assigned.

Surgical enucleation under local anesthesia was performed. The H&E staining demonstrated thin odontogenic epithelium with areas of loosely arranged and flattened superficial cells. Numerous isolated and conglomerates of ghost cells present .Numerous small cystic space enclosed by epithelium and ghost cells are evident within the stroma. Few hematoxyphic calcifications are evident within the epithelium and among ghost cells in the stroma. Homogenous dentinoid like material present subepithelially. The histopathological features were suggestive of calcifying odontogenic cyst. Postoperatively wound healing was satisfactory. The patient was followed-up for 1 year with no recurrence of lesion. (Fig 1c).

DISCUSSION

The COC is a rare example of a developmental odontogenic cyst, its occurrence constituting about 0.37% to 2.1% of all odontogenic tumors.[5] Higher incidence of these cysts seen in younger age group, 70% occurred in the second and third decades. The most common site

of occurrence seen in the anterior part of the jaws.In the mandible, several cases have crossed the midline, but this is less usual in the maxilla .[6,7]

It occurs almost equally in both sexes with slight male predilection. The most common sites of occurrence are anterior maxilla 41.2%, posterior mandible 35.3%, anterior mandible (17.6%), and posterior maxilla (5.9%).[8]

Several classifications of CGOC subtypes have been proposed, but most of them have limitations in separating cystic and neoplastic variant.

Classification of COC by Reichart

1. Non neoplastic (simple cystic) variant (CGCOC):

- with non-proliferative epithelial lining,
- with non-proliferative (or proliferative) epithelial lining associated with odontomas
- with proliferative epithelial lining,
- with unicystic, plexiform ameloblastomatous proliferation of epithelial lining.

2.Neoplastic variant:

(A) benign type (CGCOT):

(a) cystic subtype (cystic CGCOT)

- SMA ex epithelial cyst lining,

(b) solid subtype (solid CGCOT)

Peripheral ameloblastoma-like

SMA-like,

(B) malignant type (malignant CGCOT or OGCC):

(a) cystic subtype

(b) solid subtype.

(a.) Calcifying ghost cell odontogenic cyst.(b.) compound (or complex) cystic ghost cell odontomas.(c.)Does not completely fulfill the histopathologic criteria of early ameloblastoma as suggested by Vickers and Gorlin. (d.)Calcifying ghost cell odontogenic tumor. (e.)With histopathologic features of early ameloblastoma as suggested by Vickers and Gorlin. (f.)Resembling a peripheral amelobastoma, hence denoted as peripheral epithelial odontogenic ghost cells tumor. (g.) Often called central epithelial odontogenic ghost cell tumor. (h.)Odontogenic ghost cell carcinoma).[3]

Radiographic feature of the COC is usually a mixed lesion, with radiolucent area, uni or multilocular appearance, containing different amounts of radiopaque material. [9]*Marx et al*. in 2003 discussed three patterns of radiopacity with this tumor; first, salt and pepper pattern of flecks, second, fluffy cloudlike pattern throughout, and third, a crescent shaped pattern on one side of the radiolucency .[10]

McGowan and Browne in 1982, found that the presence of mineralization was approximately twice as frequent in microscopic examination compared to radiographic analysis.[11]

Very limited data is available in the literature about the value of CBCT in COC diagnosis. CBCT of a limited area is also very effective in achieving high spatial resolution in comparison with conventional CT .[12]CBCT examination was useful for complete evaluation of the lesions and related structures involving the maxillofacial complex.

Differential diagnosis can be Dentigerous cyst, Odontogeic Keratocyst, and Ameloblastoma as they have little or no mineralization and therefore may present as radiolucencies.

The final diagnosis of COC can be made more appropriately only based on histological finding due to the variable clinical, radiological and biological features.

Histopathological features shows Epithelial lining showing a palisaded layer of columnar basal cells, presence keratinization, dentinoid, and calcification.Ghost cells, tooth like material and calcifications are also seen scattered in the epithelium. ghost cells are not exclusively seen in COC [1].Some theories reveal that it may represent coagulative necrosis or a form of normal or aberrant keratinization of odontogenic epithelium or an underlying ischemic process, which may result in squamous metaplasia and later tends to calcify .[12]

In our case report, 84 years old female patient shows and cystic lesion on left side of maxilla. Radiographically well-circumscribed unilocular radiolucency seen and no detectable calcified bodies/flecks in the lesion.Correlating with the clinical features and radiographic findings, residual cyst, dentigerous cyst, and COC was assigned. The treatment of cystic lesion involves enucleation with long-term followup. Recurrence depends on completeness of cyst removal. Recurrence rate of calcifying odontogenic cyst is very rare, there are some case reports of development of ghost cell odontogenic carcinomas from calcifying odontogenic cyst. Our follow-up radiograph did not show any signs of recurrence.

CONCLUSIONS

COC is a unique lesion which possessing both cystic and neoplastic potential and shows number of variants clinically, radiographically, and histopathologically. CBCT was useful advanced imaging modality in determining the extent and relationship of the COC to surrounding structures.

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