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By Nishath Khanum, Naresh Lingaraju, Srisha Basappa & Mahesh M.S
Farooqia Dental College and Hospital, India

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Calcifying Cystic Odontogenic Tumor – A Case Report and Review on Diverse Presentation of the Tumor

Nishath Khanum ^α, Naresh Lingaraju ^σ, Srisha Basappa ^ρ & Mahesh M.S ^ω

Abstract- The calcifying odontogenic cyst (COC) accounts for about 1% of all the jaw cysts, found most commonly within the bone. The lesion shows extreme diversity in its clinical, radiological and histopathological features as well as in its biological behavior. In addition, the calcifying odontogenic cyst may be associated with other odontogenic tumors, most commonly odontoma. Calcifying odontogenic cyst associated with Ameloblastoma and Adenomatoid Odontogenic Tumor has also been reported¹. Here we present a case of a 29 year old male patient with a large COC associated with an impacted 38 and involving the left body and angle of mandible with diverse clinical and radiological appearance.

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I. INTRODUCTION

Calcifying odontogenic cyst (COC) was first described as a distinct clinicopathologic entity by Gorlin et al., in 1962. Ever since then controversy and confusion have existed regarding its nature². According to the WHO classification in 2005, COC has now been reclassified as calcifying cystic odontogenic tumor (CCOT). The lesion shows extreme diversity in its clinical and histopathological features as well as in its biological behavior. CCOTs are frequently associated with odontogenic tumors, a finding which is a rare event in other types of odontogenic cysts or tumors^{3, 4, 5, 6, 7, 8}.

Central and peripheral forms of calcifying odontogenic cyst occur equally in the upper and lower jaws. Johnson et al reported the occurrence of 60% of the tumors in mandible, 30% in the form of peripheral calcifying odontogenic cysts and anterior part of the jaw was involved in 53% of cases⁹. On the basis of 52 examples of calcifying odontogenic cysts associated with an odontoma Hirschberg et al concluded that the upper jaw was affected in 61.5% and the anterior region of the jaw in 75% of the reported cases¹⁰.

Calcifying cystic odontogenic tumor can occur in very young patients, even in the first year of life.

Cases have also been recorded of patients in their eighties¹¹. However, in the majority of cases it occurs in the second decade of life^{12, 13}.

*Author α σ ρ ω: Department of Oral Medicine and Radiology Farooqia Dental College and Hospital, Mysore, Karnataka, India.
e-mail: maheshmvs@rediffmail.com*

CCOT may clinically be diagnosed as Ameloblastoma, Calcifying epithelial odontogenic tumor, Adenomatoid odontogenic tumor, Ameloblastic fibroodontoma, complex or compound odontoma and dentigerous cyst or as other types of odontogenic cysts.

A hybrid odontogenic tumour composed of a calcifying cystic odontogenic tumour (CCOT), a solid multicystic ameloblastoma (A-S/M) and an Adenomatoid odontogenic tumour (AOT) was reported in the anterior part of the mandible of a 64-year-old Chinese woman⁵. This further confirms the diverse histopathologic presentation of the tumor.

II. CASE REPORT

A 29-year-old male patient visited the department of oral medicine and radiology of Farooqia Dental College and Hospital, with a chief complaint of swelling in the left side of face since 7 months. Swelling which started gradually increased to attain the present size. No history of pus, blood or watery discharge, color change or parasthesia noted over the swelling. No history of difficulty in opening the mouth, speaking and swallowing food. History of difficulty in chewing the food from affected side. No history of pain and any other associated symptoms like fever, loss of appetite, loss of weight, diarrhea or fatigue. Patient gave history of noticing mobility of teeth in the region of complaint since 4 months.

On local extraoral examination, a solitary ill defined oval swelling measuring about 4 X 5 centimeters is present over the left body and angle of mandible. Superiorly the swelling extends from 1 cm below the zygomatic arch to 1 cm beyond the lower border of the mandible. Anteriorly, the swelling extends from 1cm distal to the angle of mouth to the ramus of mandible posteriorly. The skin over the swelling is stretched with obliteration of the Nasolabial fold. There is no evidence of pus, blood or watery discharge and no secondary changes were noted. The surrounding area appears normal.

On palpation the swelling was tender, firm - hard in consistency, with smooth surface and well defined borders. The swelling was non fluctuant, non compressible, non depressible, immobile and no discharge was elicited.

Intraoral hard tissue examination revealed Grade I mobility i.r.t.³⁵ and Grade III mobility i.r.t.^{36, 37}. On Soft tissue examination vestibular obliteration and tenderness present i.r.t.^{34, 35, 36, 37}. Color of the mucosa appeared normal with no sinus opening noted over the vestibule. No evidence of ulcer or growth noted in the soft tissue. Buccal and lingual cortical plate expansion with perforation of the buccal cortical plate was present.

The orthopantomograph revealed a multilocular radiolucency on the left side of the mandible extending from the periapical region of first premolar up to the angle of mandible. The lesion contained the unerupted third molar displaced distally near the angle of mandible. Resorption of the apical 1/3rd of the root of 35 and apical and middle 1/3rd of the mesial and distal roots of 36 and 37 was evident. There was break in the continuity of lower border of mandible without any pathological fracture.

Based on history, clinical features and radiographic appearance, a differential diagnosis of Ameloblastoma, Keratocystic Odontogenic Tumor, Calcifying Cystic Odontogenic Tumor and Odontogenic myxoma was considered.

An incisional biopsy was obtained from the lesion to establish the final diagnosis. Histopathologic examination revealed epithelial lining consisting of tall columnar basal and superficial stellate reticulum like cells along with areas of eosinophilic ghost cells suggestive of Calcifying cystic odontogenic tumor.

III. DISCUSSION

CCOT was first described in 1932 by Rywkind who reported a lesion of the jaw which resembled cholesteatoma of the ear and thereafter called it as cholesteatoma of the jaw. In 1946, Thoma and Goldman described a lesion which they called a strange variant of ameloblastoma. It was in 1962 that Gorlin first described it^{2, 3, 14}.

CCOT represents 2% of all the odontogenic pathological changes of the jaws, although it can be found in isolation, it is usually associated with other odontogenic tumours, most frequently with odontoma in 24% of the cases¹⁵.

According to few studies CCOT is more common in females and in maxilla where as there are reports of CCOT occurring more in males and in mandible¹⁶. Cases have also been reported where Calcifying cystic odontogenic tumor is provisionally diagnosed as a residual cyst¹⁵ as well as a periapical pathology¹⁷.

Radiographically, they are clearly delineated and appear as unilocular or multilocular radiolucencies. Scattered irregular sized calcifications producing a mixed radiopaque radiolucent lesion may also be encountered, which may coalesce later and give an appearance of tooth like densities within the lesion^{2, 13}.

COC can occur alone or in association with other odontogenic tumors such as odontomas (20%), adenomatoid odontogenic tumors and ameloblastomas.

However, this association is a challenge for diagnosis using only conventional images, due to the presence of numerous overlapped images of anatomical structures of the maxillofacial region. Root resorption and divergence of roots of the associated teeth are common radiographic findings, and an association with an impacted tooth occurs in approximately one-third of cases¹⁸. This further suggests the diverse radiographic presentation of CCOT.

The present case occurred in a 29 year old male and is in the mandibular left posterior teeth region associated with an impacted tooth.

It demonstrated a gradual increase in size of the swelling with associated mobility of teeth and was found to be much larger measuring 3x7 cm which is in contrary to what has been reported.

Expansion of the labial or buccal cortical plate invariably occurs usually sparing the lingual cortical plate.

The reported case here is unusual to what has been published in literature since lingual cortical expansion was noted along with perforation of the buccal cortical plate and resorption of lower border of mandible without any evidence of pathological fracture.

In conclusion, the diverse clinical and radiological presentation of calcifying odontogenic cyst makes it difficult to diagnose clinically. Calcifying cystic odontogenic tumor (CCOT) is an uncommon odontogenic tumor.

Although rare, because of its variable presentation calcifying cystic odontogenic tumor should be included in the differential diagnosis of jaw lesions.

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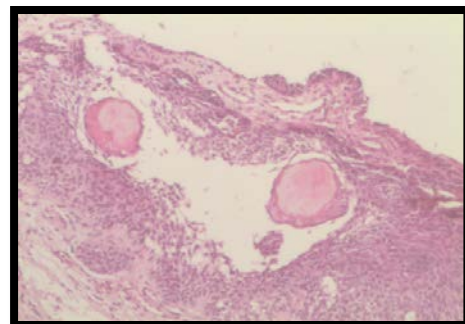
Extra Oral Photograph



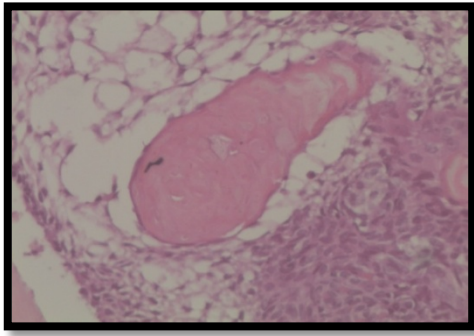
Opg Showing The Multilocular Radiolucency



Cropped Image Of The Opg



Histopathologic Section – 20x Magnification



Histopathologic Section Showing The Ghost Cell –
40x Magnification