

# 1 Calcifying Cystic Odontogenic Tumor -A Case Report and 2 Review on Diverse Presentation of the Tumor

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## 7 **Abstract**

8 The calcifying odontogenic cyst (COC) accounts for about 1

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10 **Index terms**— calcifying odontogenic cyst; diverse clinical, radiological and histological presentation;  
11 odontogenic tumors; calcifying cystic odontogenic tumor.

## 12 **1 Introduction**

13 calcifying odontogenic cyst (COC) was first described as a distinct clinicopathologic entity by Gorlin et al., in  
14 1962. Ever since then controversy and confusion have existed regarding its nature 2 . According to the WHO  
15 classification in 2005, COC has now been reclassified as calcifying cystic odontogenic tumor (CCOT). The lesion  
16 shows extreme diversity in its clinical and histopathological features as well as in its biological behavior. CCOTs  
17 are frequently associated with odontogenic tumors, a finding which is a rare event in other types of odontogenic  
18 cysts or tumors ??, 4, 5, 6, 7, 8. Central and peripheral forms of calcifying odontogenic cyst occur equally in  
19 the upper and lower jaws. Johnson et al reported the occurrence of 60% of the tumors in mandible, 30% in the  
20 form of peripheral calcifying odontogenic cysts and anterior part of the jaw was involved in 53% of cases9. On  
21 the basis of 52 examples of calcifying odontogenic cysts associated with an odontoma Hirschberg et al concluded  
22 that the upper jaw was affected in 61.5% and the anterior region of the jaw in 75% of the reported cases 10 .

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

24 Cases have also been recorded of patients in their eighties 11 . However, in the majority of cases it occurs in  
25 the second decade of life 12,13 .

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

29 A hybrid odontogenic tumour composed of a calcifying cystic odontogenic tumour (CCOT), a solid multicystic  
30 ameloblastoma (A-S/M) and an Adenomatoid odontogenic tumour (AOT) was reported in the anterior part of  
31 the mandible of a 64-year-old Chinese woman 5 . This further confirms the diverse histopathologic presentation  
32 of the tumor.

## 33 **2 II.**

## 34 **3 Case Report**

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

42 On local extraoral examination, a solitary ill defined oval swelling measuring about 4 X 5 centimeters is present  
43 over the left body and angle of mandible. Superiorly the swelling extends from 1 cm below the zygomatic arch

## 5 DISCUSSION

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44 to 1 cm beyond the lower border of the mandible. Anteriorly, the swelling extends from 1cm distal to the angle  
45 of mouth to the ramus of mandible posteriorly. The skin over the swelling is stretched with obliteration of the  
46 Nasolabial fold. There is no evidence of pus, blood or watery discharge and no secondary changes were noted.  
47 The surrounding area appears normal.

48 Intraoral hard tissue examination revealed Grade I mobility i.r.t ??5 and Grade III mobility i.r.t ??6, ??7 .  
49 On Soft tissue examination vestibular obliteration and tenderness present i.r.t ??4, ??5, ??6, ??7 . Color of the  
50 mucosa appeared normal with no sinus opening noted over the vestibule. No evidence of ulcer or growth noted  
51 in the soft tissue. Buccal and lingual cortical plate expansion with perforation of the buccal cortical plate was  
52 present.

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

58 Based on history, clinical features and radiographic appearance, a differential diagnosis of Ameloblastoma,  
59 Keratocystic Odontogenic Tumor, Calcifying Cystic Odontogenic Tumor and Odontogenic myxoma was consid-  
60 ered.

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

### 64 4 III.

### 65 5 Discussion

66 CCOT represents 2% of all the odontogenic pathological changes of the jaws, although it can be found in isolation,  
67 it is usually associated with other odontogenic tumours, most frequently with odontoma in 24% of the cases 15 .

68 According to few studies CCOT is more common in females and in maxilla where as there are reports of CCOT  
69 occurring more in males and in mandible 16 .Cases have also been reported where Calcifying cystic odontogenic  
70 tumor is provisionally diagnosed as a residual cyst 15 as well as a periapical pathology 17 .

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

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

76 However, this association is a challenge for diagnosis using only conventional images, due to the presence of  
77 numerous over lapped images of anatomical structures of the maxillofacial region. Root resorption and divergence  
78 of roots of the associated teeth are common radiographic findings, and an association with an impacted tooth  
79 occurs in approximately one-third of cases 18 . This further suggests the diverse radiographic presentation of  
80 CCOT.

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

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

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

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

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

91 Although rare, because of its variable presentation calcifying cystic odontogenic tumor should be included in the  
92 differential diagnosis of jaw lesions. involving calcifying cystic odontogenic tumor and plexiform Ameloblastoma.  
93 Contemp Clin Dent. 2013 Jul-Sep; 4(3):406-408. 5. Zhang W et al. A case report of a hybrid odontogenic  
94 tumour: Ameloblastoma and adenomatoid odontogenic tumour in calcifying CCOT was first described in 1932  
95 by Rywkind who reported a lesion of the jaw which resembled cholesteatoma of the ear and thereafter called it as  
96 cholesteatoma of the jaw. In 1946, Thoma and Goldman described a lesion which they called a strange variant  
97 of ameloblastoma. It was in 1962 that Gorlin first described it ??, 3, 14 .

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