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DENTISTRY- CASE REPORT

Calcifying Cystic Odontogenic Tumour Mimicking As A Residual Cyst

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ABSTRACT

BACKGROUND: Calcifying cystic odontogenic tumour (CCOT), first described by Gorlin in 1962, accounts for less than 2% of all odontogenic tumours. This rare cystic odontogenic lesion, sometimes referred to as the 'keratinizing and calcifying odontogenic cyst', is characterized by the presence of 'ghost' epithelial cells and by its resemblance to the pilomatrixoma of the skin. **CASE:** In this report, we present a rare case of calcifying odontogenic cyst mimicking as a residual cyst in the maxillary anterior region, in a 60-year-old female patient. The lesion was surgically removed. After enucleation, no recurrence has been recorded. **CONCLUSION:** Correlation of clinical and radiological information with histological features is important in the diagnosis of odontogenic tumours and cysts.

Key Words: calcifying odontogenic cyst, residual cyst, calcifying cystic odontogenic tumour, Gorlin cyst, odontogenic tumour

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Introduction

The calcifying odontogenic cyst (COC) was first described as a distinct entity by Gorlin et al [1] in 1962. However, Kramer et al. [2] classified it as a benign odontogenic tumour, with the SNOMED code 9301/0, in the World Health Organization's (WHO) publication, Histological Typing of Odontogenic Tumours. Prætorius et al [3] proposed that calcifying odontogenic cyst comprises of two entities: a cyst and a neoplasm. In the WHO publication on odontogenic tumours which was published in 2005 [4], it was renamed as a calcifying cystic odontogenic tumour (CCOT).

Several classifications have been proposed because of a variety of clinical, radiological and histological

appearances of these lesions. Li and Yu [5] divided the lesions into three groups – cysts, benign tumours and malignant tumours – and suggested that the term COC should be used specifically to designate the unicystic lesions, with or without an associated odontoma. In the more recent edition of 'Cysts of the oral and maxillofacial regions' which was edited by Shear [6], a new classification as suggested by Praetorius [Table/Fig 1], was included.

A. Group 1 'Simple' cysts 1. Calcifying odontogenic cyst (COC)
B. Group 2 Cysts associated with odontogenic hamartomas or benign neoplasms: calcifying cystic odontogenic *tumours* (CCOT). The following combinations have been published: 1. CCOT associated with an odontome 2. CCOT associated with adenomatoid odontogenic tumor 3. CCOT associated with ameloblastoma 4. CCOT associated with ameloblastic fibroma 5. CCOT associated with ameloblastic fibro-odontoma 6. CCOT associated with odonto-ameloblastoma 7. CCOT associated with odontogenic myxofibroma
C. Group 3 Solid benign odontogenic neoplasms with similar cell morphology to that in the COC, and with dentinoid formation 1. Dentinogenic ghost cell tumour
D. Group 4 Malignant odontogenic neoplasms with features similar to those of the dentinogenic ghost cell tumour 1. Ghost cell odontogenic carcinoma

[Table/Fig 1] Suggested classification of the odontogenic ghost cell lesions. (Prætorius, 2006)[6]

CCOT is an uncommon lesion and represents about 0.03% of the biopsy lesions and less than 2% of all odontogenic cysts and tumours. [7, 8] The lesion occurs over a wide age range. The youngest recorded patient was 1 year old, the oldest was 82 years old, and there was an impressively high peak in the second decade, a feature which was confirmed in the reports by many groups. There seems to be an equal gender distribution of both the intraosseous and the extraosseous cases of CCOT. Similarly, the mandible and the maxilla seem to be affected with equal frequency. [6] The extraosseous variety is more likely to occur in the sixth decade of life or in older people, as observed in a review of 29 extra-osseous cases. [9]

The COC normally appears as a painless, slow-growing tumour, affecting equally the maxilla and the mandible, with predilection to the anterior segment (incisor/canine area). It generally affects young adults in the third to fourth decades of life, without gender predilection. Patients with CCOT usually present with a slowly growing, asymptomatic swelling, with hard bony expansion in case of intraosseous lesions. Lingual expansion, displacement of teeth and perforation of cortical plates are occasionally observed. The extraosseous lesions usually appear as well-circumscribed, pink to red, elevated masses. [6-10]

Radiographically, CCOT is usually a mixed lesion which is unilocular or multilocular, with radiolucent areas, that contains varying amounts of radiopaque material. Its association with impacted teeth is described in 10-32% of the cases. [6, 10, 11] Its association with unerupted/impacted teeth have been reported in some cases. Displacement of the teeth and resorption of the roots of the adjacent teeth are frequent findings. [12]

Histologically, COC is usually composed of a cystic cavity with a fibrous capsule, lined by an odontogenic epithelium. [13] The typical microscopic characteristic of this lesion is the presence of variable amounts of aberrant epithelial cells, without nuclei, which are named as ghost cells. In addition, dysplastic dentine can be found and occasionally, the cyst is associated with an area of dental hard tissue formation which resembles an odontoma. [6]

CCOT lesions can also be found in association with other odontogenic tumours, like ameloblastomas, ameloblastic fibroodontomas, odontoameloblastic tumours, calcifying epithelial odontogenic tumours and adenomatoid odontogenic tumours. [14, 15]

Case Report

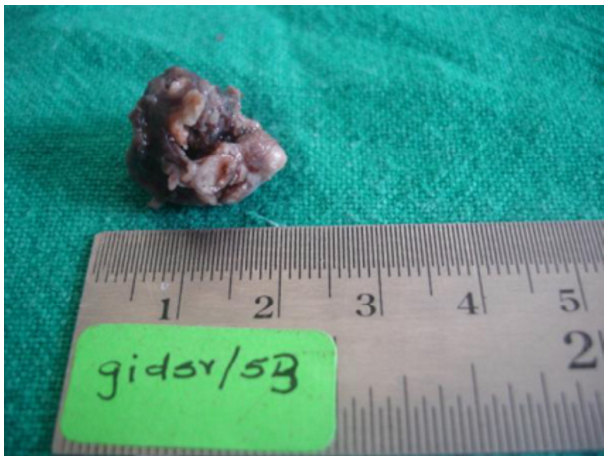
A 60 year old female patient reported to the Department of Oral Pathology and Microbiology, Genesis Institute of Dental Sciences and Research, Ferozepur, with the chief complaint of painless swelling of the front area of her right upper jaw. She

gave a history of full mouth extractions in the preparation for complete dentures. She had persistent irritation after extractions in the maxillary right side central and lateral incisor region. The swelling had been present for two months and had slowly enlarged to the present size of approximately 1 x 1.5 cm.

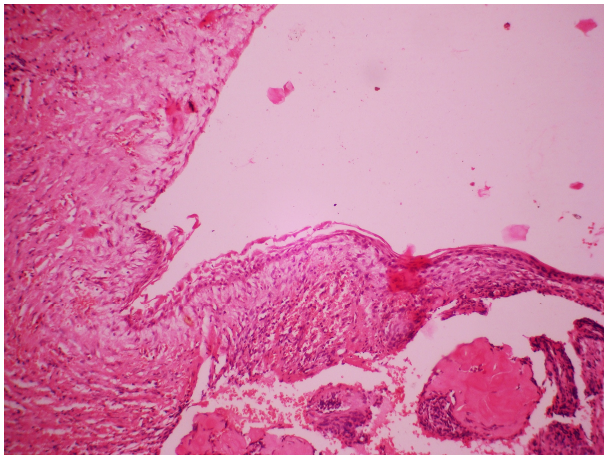
Intraoral examination revealed a labial cortical expansion of the residual ridge in the region of 11 and 12, which had been extracted earlier. The mucosa over the lesion was intact. Palpation revealed a non-tender hard bony expansion of the right maxilla in the area of 11 and 12. Radiographical examination showed a well defined, unilocular radiolucency at the site of the previously extracted teeth 11 and 12 .A differential diagnosis of residual cyst was considered, based on clinical and radiological findings. Excisional biopsy was performed and the sample was sent to the Department of Oral Pathology for histopathological examination. The gross specimen was roughly ovoid in shape, measuring 1.5 x 2.0 cm in dimension, reddish brown in color and firm in consistency [Table/Fig 2].

Microscopically, a cystic lumen lined with stratified squamous epithelium was observed. The basal layer of the lining epithelium was composed of a columnar layer of ameloblast-like cells, with nuclei which was polarized away from the basement membrane [Table/Fig 4]. The suprabasal layers were composed of cells which resembled stellate reticulum. Few areas of the cystic lining showed an irregular arcading pattern of stratified squamous epithelium which resembled a radicular cyst [Table/Fig 5].

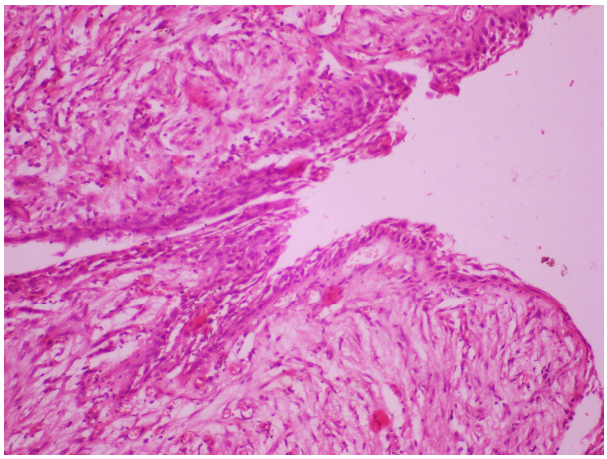
The most prominent and unique features were the presence of keratinized ghost cells within the loose spinous cell layer and lining epithelium-induced dentinoid matrix, which was found in the connective tissue wall. In some areas, there was fusion of ghost cells which formed extensive sheets of an amorphous, acellular eosinophilic material, filling the lumen of the cyst [Table/Fig 6]. Irregular masses of hyalinized acellular calcified material (dentinoid) were also observed in the connective tissue, in relation to both the epithelial lining and the masses of ghost cells [Table/Fig 6]. Based on these features, the lesion was diagnosed as intraosseous calcifying cystic odontogenic tumour (CCOT).



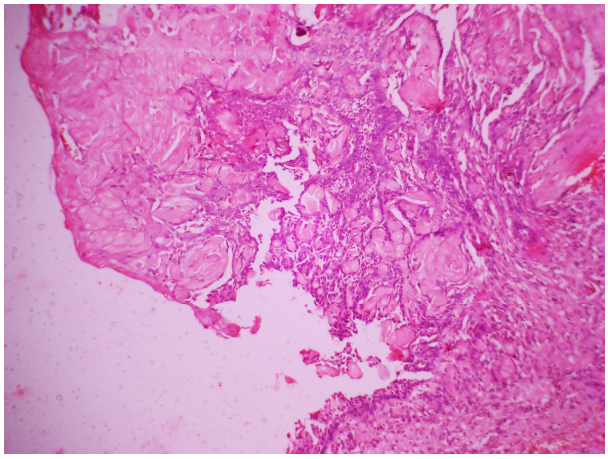
[Table/Fig 2]: Gross specimen of cyst, measuring 1.5cm x 2.0cm in dimension.



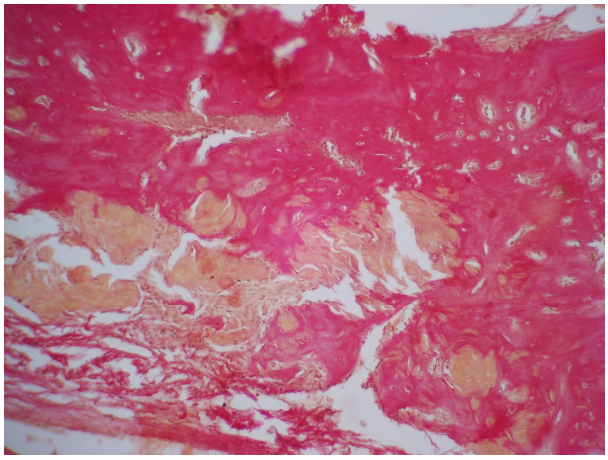
[Table/Fig 3]: Photomicrograph showing lumen, thin epithelial lining, and an area of lining with ameloblast-like cells and stellate-reticulum-like cells, and an area of dense collagen within the connective tissue stroma. (H & E, 100x).



[Table/Fig 4]: Photomicrograph showing arcading type of epithelial lining, resembling the lining of a radicular /residual cyst. (H & E, 100x).



[Table/Fig 5]: Photomicrograph showing clusters of ghost cells within the epithelial lining. (H & E, 100x).



[Table/Fig 6]: Photomicrograph showing red-staining dentinoid and yellow-staining ghost cells. (Van Gieson, x 100).

Discussion

The calcifying odontogenic cyst (COC) is an odontogenic lesion that has been characterized as a discrete entity since 1962. [1] Though it is a well-recognized lesion, COC is not very commonly encountered. In 1971, COC was described as a non-neoplastic cystic lesion, but in 1992, the WHO classified this lesion with the odontogenic tumours because of its histological complexity and diversity. [2] According to the new WHO classification in 2005, COC has now been reclassified as calcifying cystic odontogenic tumour (CCOT). [5] Although there is no consensus regarding the classification and the terminology of COC, this benign lesion is categorized as either a cyst or neoplasm (solid). [4, 6, 16] In the cyst variant, three different types may be found: the simple unicystic type, the unicystic odontoma-associated type, and the unicystic ameloblastomatous proliferating type.

CCOT represents 2% of all the odontogenic pathological changes of the jaws, [6, 17] although it can be found together with other odontogenic tumours, most frequently with odontoma in 24% of the cases. [10, 14, 15]

Praetorius et al. [3] and Buchner et al. [10] believe that the COC cystic epithelium originates from the reduced enamel organ, from islands of odontogenic epithelium within the tooth follicle or from the remnants of odontogenic epithelium in the bone or gingival tissues. In the reported case, we presume that the neoplastic epithelium must have arisen from the remnants of odontogenic epithelium which was present within the bone.

The histological features of a classical calcifying odontogenic cyst are characteristic and present a few diagnostic problems. The clinical appearance and radiological features of the present case gave an impression that it was a residual cyst. However, its histopathological features proved otherwise.

A case of the calcifying odontogenic cyst in the mandible of a 41-year-old male with clinical and radiographical features, which suspected it initially to be a lateral periodontal cyst, has been reported by Huang et al. in 1990. [18] In a retrospective study of non-endodontic periapical lesions in Chile, only 26 cases (0.65%) out of 32,423 biopsy specimens had a histopathological diagnosis of non-endodontic pathology. Out these 26 cases, there was only one case of CCOT. [19] There have been case reports where persistent periapical periodontitis with unsuccessful attempts at root canal treatment (RCT) turned out to be CCOT on histopathological examination of the surgically enucleated periapical tissue. [20] The authors opined that CCOT can mimic apical periodontitis and therefore, should be considered in the differential diagnosis of large

lesions associated with the apex of the root of the tooth.

The presence of two distinct epithelial odontogenic patterns in a single lesion is probably not a collision of a COC and a residual cyst, but instead an expression of the histodifferentiation potential of the odontogenic epithelium from which these lesions are derived. This assumption is supported by the finding of both histological types in the epithelial lining of the same cystic cavity.

Conclusion

The clinical significance of this case is that radiographical observations appeared to be simple, unambiguous and obvious. However, the histopathological observations showed that it was a rare odontogenic lesion. Hence, this case report emphasizes the role of biopsy in the diagnosis of such deceptive clinical presentations. This also proves the importance of microscopic examination of the entire specimen when giving a diagnosis based on histopathological observations.

References

[1] Gorlin RJ, Pindborg JJ, Clausen FP, Vickers RA. The calcifying odontogenic cyst. A possible analogue of the cutaneous calcifying epithelioma of Malherbe. Oral Surg Oral Med Oral Pathol 1962; 15:1235-43.

[2] Kramer I.R.H., Pindborg J.J. and Shear M. Histological Typing of Odontogenic Tumours Berlin: Springer Verlag: 1992

[3] Praetorius F, Hjorting-Hansen E, Gorlin RJ, Vickers RA. Calcifying odontogenic cyst. Acta Odont Scand 1981; 39:227-40

[4] Prætorius F. and Ledesma-Montes C. Calcifying cystic odontogenic tumour. In: Barnes L., Eveson J.W., Reichart P., Sidransky D., eds. Pathology and Genetics. Head and Neck Tumours. Lyon: World Health Organization Classification of Tumours, IARC Press. 2005. p. 313.

[5] Li T.J. and Yu S.F. (2003) Clinicopathologic spectrum of the so-called calcifying odontogenic cysts: a study of 21 intraosseous cases with reconsideration of the terminology and classification. American Journal of Surgical Pathology 2003; 27: 372–384.

[6] Shear M. and Speight P. Cysts of the Oral and Maxillofacial Regions 4th edition Blackwell-Munksgaard 2007 p. 100-107

[7] Swan RH, Houston GD, Moore SP. Peripheral calcifying odontogenic cyst (Gorlin cyst). J Periodontal 1985; 56:340-3.

[8] Iglesias S, Aguado A, Seoane JM. Quiste odontogénico calcificante asociado con odontoma. Caso clínico. Revisión de la literatura. Rev Esp Cirug Oral yMaxilofac 1997; 19:140-4.

[9] Kaugars C.C., Kaugars G.E. and DeBiasi G.F. Extraosseous calcifying odontogenic cyst: report of case and review of literature. Journal of the American Dental Association 1989; 119: 715–718.

[10] Buchner A. The central (intraosseous) calcifying odontogenic cyst: an analysis of 215 cases. J Oral Maxillofac Surg 1991; 49:330-9.

[11] Freedman PD, Lumerman H, Gee JK. Calcifying odontogenic cyst: a review and analysis of 70 cases. Oral Surg Oral Med Oral Pathol 1975; 40:93-106.

[12] Tanimoto K., Tomita S., Aoyama M., et al. Radiographic characteristics of the calcifying odontogenic cyst. International Journal of Oral and Maxillofacial Surgery 1988; 17, 29–32.

[13] Arthur J, Mark F, Lionel G. Calcifying odontogenic cyst: A clinicopathologic study of 57 cases with immunohistochemical evaluation for cytokeratin. J Oral Surg 1997; 55:108-11.

[14] Altini M, Farman AG. The calcifying odontogenic cyst. Oral Surg Oral Med Oral Pathol 1986; 40:751-9.

[15] Lello GE, Maken M. Calcifying odontogenic cyst. Int J Oral Maxillofac Surg 1986; 15:637-44.

[16] Ledesma-Montes C, Gorlin RJ, Shear M, Prae Torius F, Mosqueda-Taylor A, Altini M, Unni K, Paes de Almeida O, Carlos-Bregni R, Romero de León E, Phillips V, Delgado-Azañero W, Meneses-García A. International collaborative study on ghost cell odontogenic tumours: calcifying cystic odontogenic tumour, dentinogenic ghost cell tumour and ghost cell odontogenic carcinoma. J Oral Pathol Med 2008; 37(5): 302-8.

[17] Altini M, Farman AG. The calcifying odontogenic cyst. Eight new cases and a review of the literature. Oral Surg Oral Med Oral pathol. 1975; 40:751-759.

[18] Huang YL, Lin LM, Lin CC, Yan YH. Calcifying odontogenic cyst presenting as a lateral periodontal cyst--a case report. Zhonghua Ya Yi Xue Hui Za Zhi. 1990 Mar; 9(1):42-5.

[19] Ortega A, Fariña V, Gallardo A, Espinoza I, Acosta S. Nonendodontic periapical lesions: a retrospective study in Chile. Int Endod J. 2007 Aug; 40(8):661.

[20] Estrela, C ; Decurcio, D A ; Silva, J A ; Mendonça, E F ; Estrela, C R A. Persistent apical periodontitis associated with a calcifying odontogenic cyst. Int Endod J 2009, Jun, 01; 42(6):539-45.