

CASE REPORT

Calcifying Epithelial Odontogenic Cyst: A Case Report

N.Anisa¹, G.R.Karthikeyan², M.Kandasamy³, K.R.Shakila⁴**ABSTRACT**

Introduction: Calcifying odontogenic cyst (COC) shows an extreme diversity in clinical features and histopathologic features as well as in biologic behaviour, because of this diversity there has been disagreement in the concerning the terminology as calcifying odontogenic cyst, Gorlin's cyst, keratinizing and calcifying odontogenic cyst, epithelial odontogenic ghost cell tumor and odontogenic cyst. Hence, its necessary to know the lesions pathogenesis, clinical behaviour and histomorphologic differentiation for better outcome of the treatment.

Case Report: We report a case of an intra ossous calcifying odontogenic cyst (ICOC) in a 27 year old male is presented with its clinical, histological, radiographic feature s and review of literature.

Conclusion: Despite the fact, that (COC) shows a unique characteristics for both cystic and neoplastic behaviour, calcifying epithelial odontogenic cyst can be diagnosed with its clinical feature, radiographic appearance and histopathologic presentation.

Keywords: Calcifying odontogenic cyst, Calcifying odontogenic tumor.

How to cite this article: N.Anisa, G.R.Karthikeyan, M.Kandasamy, K.R.Shakila. Calcifying Epithelial Odontogenic Cyst: A Case Report. International Journal of Contemporary Medical Research. 2015;2(2): 315-317

¹Senior lecturer, Department of Oral Medicine and Radiology, ²Senior lecturer, Department of Oral and Maxillo Facial Surgery, Karpaga Vinayaga Institute of Dental Sciences, Kanchipuram, ³Senior lecturer, Department of Oral Medicine and Radiology, Rajas Dental College and Hospital, Kavalkinaru, ⁴Reader, Department of Oral Medicine and Radiology, Karpaga Vinayaga Institute of Dental Sciences, Kanchipuram, India.

Corresponding author: Dr. N.Anisa Senior lecturer, Department of Oral Medicine and Radiology, Karpaga Vinayaga Institute of Dental Sciences, Kanchipuram, India

Source of Support: Nil

Conflict of Interest: None

INTRODUCTION

Calcifying odontogenic cyst (COC) is a benign cystic neoplasm of odontogenic origin that represents 1% of jaw cyst.¹ They are developmental odontogenic lesions. A solid variant known as odontogenic ghost cell tumor is believed to potentially exhibit more aggressive clinical behaviour.¹ Hoffman et al, reported 78.5% of COC arise centrally in bone and 21.5% are observed in the gingiva.² COC are believed to be derived from odontogenic epithelial remnants within the gingiva or within the mandible or maxilla. "Ghost cell keratinization," the characteristic microscopic feature of this cyst, is also a defining feature of the cutaneous lesion known as calcifying epithelioma of Malherbe or pilomatrixoma. In the jaws, ghost cells may also be seen in other odontogenic tumors, including odontomas, ameloblastomas, adenomatoid odontogenic tumors, ameloblastic fibroodontomas, and ameloblastic fibromas.¹ As a result of its neoplastic behaviour, CCOT is characterised by an ameloblastoma like epithelium with ghost cells that may calcify¹.

CASE REPORT

A 27 old patient came to the department of Oral Medicine and Radiology, Karpagavinayaga Institute of dental sciences, with the chief complaint of swelling in the right upper back tooth region since 2 months. Swelling is insidious onset for past 2 months, occasionally noticed the swelling increasing in size. No history of trauma, pus discharge and paraesthesia in that region. His medical and surgical histories were non – contributory. He has no deleterious habits. On extra oral examination, Face is asymmetrical towards right mid part of the face. There was a single diffuse ill-defined swelling seen on the right mid part of the face, measuring 2 * 2 cm approximately, extending superiorly from the 0.5 mm short of infra orbital margin to inferiorly to

the line joining the corner of the mouth to the tragus of the ear. The swelling extended medially from the ala of nose to laterally 3 cm short of tragus of ear. The skin over the swelling was normal to adjacent surrounding area with no visible pulsation. No other secondary changes were evident. On palpation, borders are diffuse, surface over the swelling was warm and firm in consistency. It was tender on palpation. No paraesthesia was evident. On intra oral examination, there was a single ovoid swelling seen at the upper right 14, 15 region, measuring 2.5*1 cm approximately, extends anteriorly from mesial of 13 region obliterating the buccal vestibule extends posteriorly till distal of 15 region. Superoinferiorly, extended from mucobuccal fold till attached gingiva in relation to 13,14, 15 region. No sinus or pus discharge were evident. On hard tissue examination, 14 is non – vital, 13 and 15 are vital. No periodontal pocket present in relation to 13, 14, 15. On clinical history, clinical examination we arrive at a diagnosis of radicular cyst in relation to 13, 14, 15.

INVESTIGATIONS

OPG: reveals missing 38, with a well-defined radiolucency measuring 1*1 cm seen in relation between 15 and 16 with radiopacity in centre of it suggestive of odontome with cyst or dentigerous cyst.

FNAC (Fine Needle Aspiration Cytology): - on FNAC - Yellow color clear fluid is aspirated and protein analysis done.

Protein Analysis: 5.3g/dl.

Patient was advised complete blood hemogram and all parameters are found to be within normal limits. Cytological smear of the Aspirate revealed given H & E stained smear shows a few anucleated polygonal epithelial cells along with a few inflammatory cells comprising of lymphocytes and polymorphous nuclear cells, suggestive of a cyst. Computed tomography (CT): reveals moderate sized well defined lobulated expansile heterodense cystic lesion involving alveolar ridge of right maxilla, extending anteriorly causing premaxillary swelling in the medial aspect with projection superficially into adjacent buccal space and mild projection superiorly into adjacent

maxillary sinus. Minimal erosion of adjacent buccal margin. Features of deformed tooth within the cystic lesion, suggestive of odontogenic cystic mass lesion. Biopsy report reveal that the given H & E staining soft tissue section shows a cystic lesion lined by odontogenic epithelium with a fibrovascular connective tissue. The epithelium is composed of tall columnar cells with reversal of polarity resembling ameloblasts. Stellate reticulum like cells seen in the suprabasal layer, eosinophilic, round or oval structure suggestive of ghost cells are seen within the epithelium. The underlying connective tissue consists of dense irregularly arranged collagen fibers with spindle shaped fibroblasts. Vascularity is moderate. Suggestive of Calcifying epithelial odontogenic cyst.

DISCUSSION

The calcifying odontogenic cyst is rare and the solid form is even less common. It is a benign neoplasm but has a malignant variant.³ All centrally located (COC) are likely to originate from reduced enamel epithelium or remnants of odontogenic epithelium, both the variants of COC should be considered regarding its histogenesis of the peripheral and neoplastic variant. The CEOC is known to involve mandible and maxilla with equal frequency.² 74% of the maxillary lesions affected the anterior region as opposed to 56% of the lesion located on the mandible (Cawson et al 1998).⁴ The age occurrence of the cyst has been reported to vary from three years to eight years with definite peaking in the 2nd decade. The cyst is usually asymptomatic unless secondarily affected⁵. The COC occurs more commonly as an intra-osseous lesions with a frequency of extraosseous presentation ranging from 16% - 22%. However, the COC predominantly occurs as an intra-osseous cystic lesion in which varying degrees of radiopacity is recorded as in our case.⁶ The radiographic appearances of COC have been reported by many authors. Unilocular or multilocular radiolucencies with discrete, well-demarcated margins. Within the radiolucency there may be scattered, irregularly sized calcifications. Such opacities may produce a salt-and-pepper type of pattern, with an equal and diffuse distribution. In some cases mineralization may develop to such

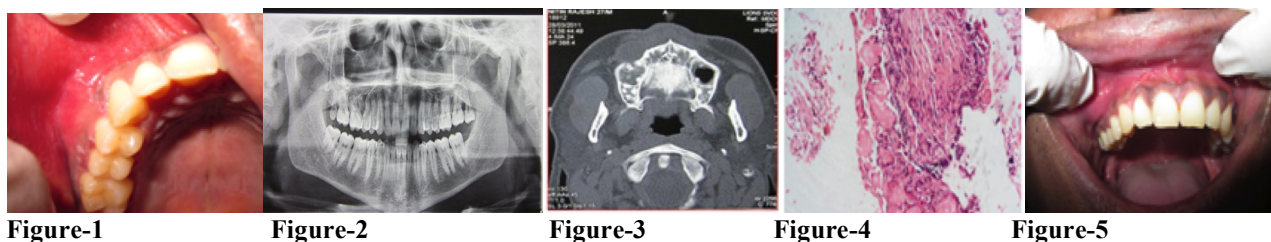


Figure-1: Intra Oral View showing obliteration in relation to 15 & 16; **Figure-2:** Orthopantomogram view Shows mixed Radiopaque lesion surrounded by radiolucent in relation to 15 and 16 region; **Figure-3:** Axial View of CT hyperdense mass surrounded by hypodense in relation to 15 and 16; **Figure-4:** Photomicrography showing Ghost cells; **Figure-5:** Post-Operative patient's Intra Oral view

an extent that the radiographic margins of the lesion are difficult to determine.⁵ As an extra-osseous lesion, COC exhibits no or minimal radiographic changes. If present, it can appear as radiolucent area with scattered radiopacity without involvement of adjacent structures.

The ghost cells typically calcify in patchy fashion, and where keratin-like material comes into contact with connective tissue it excites a foreign body reaction.³ Approximately 10% of calcifying odontogenic cysts is associated with odontomas or other odontogenic tumours.

Differential Diagnosis can be Calcifying epithelial odontogenic tumor, odonto ameloblastoma, ameloblastic fibroodontoma and complex odontoma. Enucleation of the cyst has provided total eradication of the lesion.⁵ In the present case, no signs of recurrence was detected one year after the surgery.

CONCLUSION

In conclusion, preliminary radiographic examination and computed tomography scan played an important role to arrive at proper differential diagnosis and in making appropriate surgical planning. Definitive diagnosis is required by histopathological examination. Long term follow up is necessary when adjacent structures are involved

REFERENCES

1. Joseph A. Regezi, James J. Sciubba, Richard C.K.Jordan. Oral pathology: clinical pathologic correlations. (2008) 5th ed, Saunders, St Louis, 261-270.
2. Peter. A. Reichart, Hans P. Philipsen. Odontogenic tumors and allied lesions, (2004), Quintessence pulication. 155- 168.
3. Cawson RA, Binne WH, Barrett AW, Wright JM. (2001) Oral disease: clinical and pathological correlations. 3rd ed, Mosby, London, 127.
4. Archana Sonone, V. S. Sabane and Rajeev Desai. Calcifying Ghost Cell Odontogenic Cyst: Report of a Case and Review of Literature Case Reports in Dentistry 2011;2011:328743
5. T. Saini, H.A. El-Abdin, N.O. Nartey et al, Calcifying epithelial odontogenic Cyst of the maxillary sinus. The Saudi Dental Journal 1990;2:2-5
6. V.E.Rushton, K.Hornor et al Calcifying Odontogenic Cyst – a characteristic finding. British Journal of Oral and Maxillofacial Surgery 1997;35: 196-198.