

CASE REPORT

CALCIFYING ODONTOGENIC CYST ASSOCIATED WITH ODONTOME- A CASE REPORT

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ABSTRACT:

Odontogenic cysts are classified according to their origin as developmental or inflammatory. The calcifying odontogenic cyst (COC) is a rare example of a developmental odontogenic cyst comprising between 0.37 - 2.1% of all odontogenic lesions. A significant source of disagreement prevails in the terminology of this complex lesion which stems from the fact that there are two different concepts or schools of thought when looking at the nature of COC; the monistic and the dualistic concept. We report a calcifying odontogenic cyst associated with odontoma in a 10 year old boy.

Key words: Calcifying odontogenic cyst, developmental cyst, ghost cells, odontome.

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INTRODUCTION

Odontogenic cysts are classified according to their origin as developmental or inflammatory. The calcifying odontogenic cyst (COC) is a rare example of a developmental odontogenic cyst comprising between 0.37 - 2.1% of all odontogenic lesions. Gorlin and colleagues (1962) were the first to describe this entity. They initially regarded it as the oral analogue of the cutaneous calcifying epithelioma of Malherbe but later labelled it as calcifying odontogenic cyst. Prior to this, Rywkind (1932) described it as a variant of the cholesteatoma, while Maitland (1947) regarded it as a type of ameloblastoma.¹

In 1971, World Health Organisation (WHO) classification described COC as a “non-neoplastic cystic lesion” In 1992 the World Health Organization (WHO) classified COC within the group of neoplasm and tumors that originate from odontogenic tissues but confirmed that most of the cases are non-neoplastic.^{1,2} A significant source of disagreement prevails in the terminology of this complex lesion which stems from the fact that there are two different concepts or schools of thought when looking at the

nature of COC; the monistic and the dualistic concept.³ Toida made a comprehensive review of the attempts at classifications presented during the last 25 years and added a new dualistic classification.

Although the 1992 WHO classification cited the terms dentinogenic ghost cell tumor (DGCT, suggested by Praetorius et al) and odontogenic ghost cell tumor (OGCT, suggested by Colmenero et al), especially for the solid lesion whose neoplastic nature is not apparent, the authors continue to use the term calcifying odontogenic cyst.^{3,4}

In 1995 Langlais and colleagues had also proposed the term calcifying odontogenic lesion (COL), which encompassed both the cystic and tumorous forms as well as combined lesions containing elements of both. In 1981, Praetorius et al recognized four different histological patterns of Calcifying Odontogenic Cyst and classified them as type 1A (simple unicystic), type 1B (odontome-producing), type 1C (ameloblastomatous proliferating) and type 2 (dentinogenic ghost cell tumour). Its frequent association has been related with other odontogenic tumours, such as the complex or compound odontoma, ameloblastoma, odontoameloblastoma,

ameloblastic fibroma, ameloblastic fibro-odontoma and the adenomatoid odontogenic tumour. In 1994, Hirsberg et al⁵ reviewed COCs associated with an odontoma i.e. type 1b to clarify the pathogenesis of this particular variant. COCs were reported to be associated with an odontoma in 22% to 47% of cases with a mean age of 16 years, primarily in the 2nd decade.^{6,7} The clinical and radiological features of COC are distinct but not pathognomic, and it is characterized by histological diversity. Radiographically it is a well defined mixed lesion and histologically consists of a large cyst. In the central area of the cyst enamel and dentin deposits can be found, irregularly distributed in areas and in other parts it takes on a well defined organoid aspect.^{1,6} Immunohistochemically, it is described using keratin proteins and involucrin. We report a calcifying odontogenic cyst associated with odontoma in a 10 year old boy.

CASE REPORT

A 10 year old male, reported with a complaint of missing upper front tooth since 1 yr. Patient had a history of exfoliation of upper front tooth 1yr ago and was asymptomatic for next 6 months after which the patient noticed a swelling in the gums of upper front region. Patient also gave history of pain in the same region for last 2 months on application of finger pressure. Extra-orally no significant finding was present.

On intra-oral examination, #11 and #12 were found missing and a swelling was present in that region. The

swelling was firm to hard in consistency and measured approximately 2×1.5cm with indistinct margins. The swelling had a smooth texture with no discharge and no tenderness associated with it.

Radiographic examination revealed a round to oval mixed radioopaque-radiolucent mass in the maxillary anterior alveolus with respect to #11, #12 and #53. Based on these findings a provisional diagnosis of Odontome was reached at.

Pathological gross findings revealed two bits of tissue about 1×1.7cm in diameter, hard white in colour, resembling a tooth.

In the present case, excisional biopsy was performed. A trapezoidal mucoperiosteal flap was raised and the hard tissue mass, 2 x 2 cm in diameter leaving a margin of 1-2 mm from the surrounding core was enucleated. Microscopic examination of the section studied showed a cystic lumen lined by 2-3 layers of cuboidal cells (Fig 1a). In certain areas the basal cells were columnar in appearance. Ghost cells were seen intercellularly in the epithelial lining. Dystrophic calcification is seen towards the lumen. Underlying connective tissue capsule was loosely cellular while at the periphery it was more fibrous.

Decalcified section of hard tissue mass showed enamel, dentin and cementum arranged in a haphazard manner (Fig 1b). Enamel was uneven in nature with fishscale appearance. Dentin was highly irregular. The ground section and clinic-radiographic findings were suggestive of composite odontome. A final diagnosis of Calcifying Odontogenic Cyst-Odontome type was confirmed.

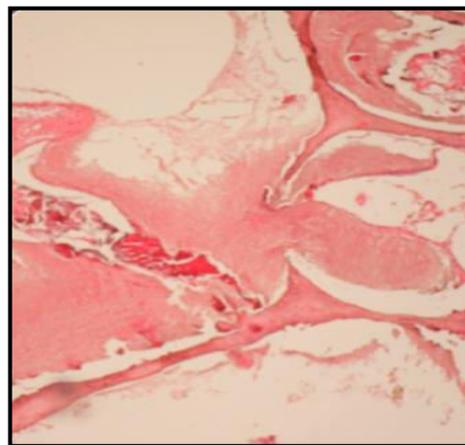
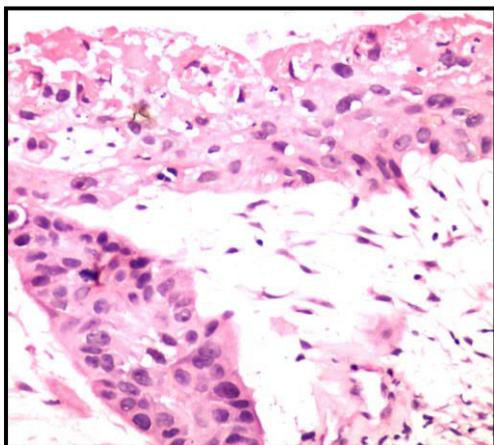


Fig 2: (a): A 2-3 layered epithelial lining with ghost cells and underlying loosely cellular connective tissue.

(b): Decalcified section showing the mineralized component; enamel, dentine and cementum arranged in haphazard manner.

DISCUSSION

COC is an unusual and unique lesion with characteristics of a solid neoplasm and of a cyst. It shows considerable amount of histopathological diversity, with variable clinical behavior such as cystic, neoplastic and infiltrating malignant behavior. Odontomes are best known as hamartomatous benign tumors rather than true neoplasms, arising from odontogenic tissues. Histologically, they are classified as compound and complex variety.^{6,8}

It was not until 1994 (Hirsberg et al⁵) that COCs associated with an odontome (COCaO) were first reviewed in an attempt to clarify the pathogenesis of this particular variant. The three classifications previously proposed are all commonly based on the dualistic concept.⁶ Toida⁹ raised the point that in the three classifications the authors seem to have used the term cystic as a synonym for non-neoplastic. Cystic is a morphologic term that does not necessarily cover the term non-neoplastic, which is a biologic one. In other words, there may well be neoplastic lesions with a cystic histoarchitecture. In the suggested classification of COCs Type 1b is characterized as a non-neoplastic (simple cystic) variant with non proliferative (or proliferative) epithelial lining associated with odontomas.⁶ Several possibilities are suggested regarding the pathogenesis of COC with odontome. One possibility is that COC and odontoma may represent coincidental juxtaposition of COC and an odontoma, because other odontogenic tumors like ameloblastoma have been reported to be associated with COC.¹⁰ Other investigators suggest that the COC develops secondarily from odontogenic epithelium that participates in the formation of the odontoma. However, it also has been suggested that the odontoma develops secondarily from lining epithelium of the COC.¹¹ Some authors think that the COCaO should be regarded as a separate entity, characterized by the clinical differences in location and in age distribution between simple COC and COCaO.² COC may occur in association with other odontogenic tumors, the most common of these is the odontoma. Buchner shows this association in 35% of his cases, Nagao et al in 22% and Shamaskin et al in 47%.⁶ COCs are reported to be associated with an odontoma in cases with a mean age of 16 years, primarily in the 2nd decade. COCaO presents a female predominance (2:1) and most frequently occurs in the maxilla (61.5%).¹¹ The present case has been reported

in a male patient of 10 years of age in the maxillary anterior alveolus. Our case also presented with typical radiographic findings associated with a COCaO.

The COCaO should be treated conservatively by surgical enucleation because recurrences are very uncommon. Finally, the malignant transformation of a preexisting benign COC could happen, but is extremely uncommon.^{12,13} COC occurring along with complex odontoma is a rare finding among the cystic lesions of the jaws. Early diagnosis and management is important to prevent its potential complications.

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