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Complex Cystic Ghost Cell Odontoma Mimicking As a Residual Cyst: A Case Report.

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ABSTRACT

Odontogenic lesions result from aberrant events during the development of a tooth or at a later phase. These odontogenic lesions have the potential to produce abnormal matrices such as enameloid, dentinoid, cementum like material, abnormal calcifications and also altered epithelial cells. Such altered epithelial cells appear swollen, pale eosinophilic with shadowy appearance and are termed as ghost cells. These ghost cells may be seen in a spectrum of odontogenic pathologies ranging from hamartomas to tumorous proliferations. Ghost cells are commonly encountered in Calcifying odontogenic cyst (COC) and COC may exhibit varied manifestation clinically, radiographically and histopathologically. A 62 year old female patient reported with a chief complaint of painless swelling in the lower front tooth region of mouth. Upon clinical examination, a provisional diagnosis of residual cyst was given. Macro and microscopic examination of the lesion came up with a diagnosis of calcifying odontogenic cyst associated with complex odontoma (complex cystic ghost cell odontoma). The present paper emphasizes the significance of clinicopathologic correlation and also highlights the varied manifestation, and histopathological types of COC.

Keywords: calcifying odontogenic cyst, ghost cell, odontoma

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INTRODUCTION

Odontogenesis is a well orchestrated phenomenon destined to produce enamel, dentin and other components owing to the active epithelial mesenchymal interactions. When there is aberrancy in the normal production of any component of a tooth, a hamartoma, a cyst or a tumor develops. These odontogenic lesions may exhibit enameloid, dentinoid, cementum like material, areas of calcification and altered epithelial cells. Ghost cells are such altered epithelial cells which are swollen pale eosinophilic masses, exhibiting shadowy appearance and hence the name. These aberrant epithelial cells may be encountered in a variety of pathologies ranging from hamartomas to malignant neoplasms. The ghost cells are seen in calcifying odontogenic cyst, odontomas, ameloblastomas, craniopharyngiomas, pilomatricoma, ghost cell odontogenic tumors and so on [1-3].

Calcifying odontogenic cyst (COC) was first described by Gorlin et al in1962 [4]. Since then, even though several cases have been reported in the English literature [5], COC appears to be relatively rare when compared to other odontogenic cysts. COC exhibits variable clinical behaviour and histopathologic diversity.

The present paper describes a case report of COC associated with complex odontoma (complex cystic ghost cell odontoma) which was thought to be a residual cyst and also emphasizes the importance of correlation of biopsy findings with clinical and radiographic features. In addition, this paper highlights the histopathologic diversity of COC.

Case Report

A 62 year old female reported to the outpatient department of DAPM R V Dental College and Hospital, with a chief complaint of painless swelling in the lower front tooth region. Upon clinical examination, a well defined, firm, non tender swelling measuring about 1.0 cm X 1.0cm was seen on the lower lingual anterior gingival region (Fig 1). There was history of extraction of 31 and 41. Intra oral periapical radiograph showed unilocular radiolucency with foci of radiopacity in the apical region of previously extracted 31 and 41 along with root resorption of 32 (Fig 2). A provisional diagnosis of residual cyst was given. Excisional biopsy of the lesion was carried out in the department of Oral and Maxillofacial Surgery and the specimen was submitted to the department of Oral and Maxillofacial Pathology for evaluation.

Macroscopy of the biopsy specimen revealed a soft tissue that measured 2.0cm x 1.0cm x 1.4cm, reddish brown, soft to firm in consistency. During grossing, a small hard tissue (0.75cm in maximum dimension) suggestive of odontoma was noticed (Fig 3A). The soft tissue bits were taken for processing and odontoma for decalcification.

Light microscopic examination of haematoxylin and eosin stained sections of soft tissue specimen revealed cystic lesion lined by odontogenic epithelium of variable thickness and connective tissue capsule. The lining epithelium showed basal cuboidal to tall columnar cells resembling ameloblasts and over this was found loosely arranged cells suggestive of stellate reticulum. Small to large areas of acellular eosinophilic material lacking nuclear details, suggestive of ghost cells were observed in the epithelial lining. Areas of dentinoid and dystrophic calcification were also seen (Fig 3B). The connective tissue was fibrovascular with islands and rests of odontogenic epithelium.

Haematoxylin and eosin stained decalcified hard tissue sections revealed haphazardly arranged areas of dentinoid, foci of cementum like masses and pulp like tissue. Areas of ghost cells (Fig 3C) and odontogenic epithelium were observed close to the dentinoid.

Considering the observations of soft tissue and hard tissue specimens, a histopathological diagnosis of Calcifying Odontogenic Cyst With Complex Odontoma (Complex Cystic Ghost Cell Odontoma) was given. The patient is kept under follow up.

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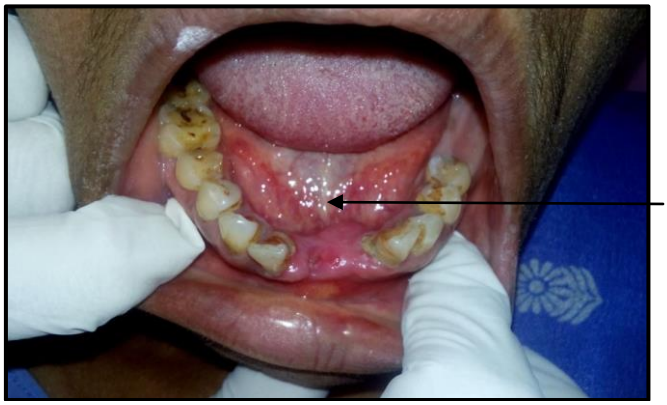

Figure 1: Clinical Photograph of the lesion (Arrow)

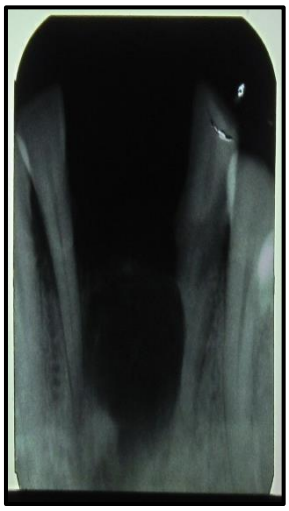

Figure 2: Intra oral Periapical radiograph showing unilocular radiolucency with foci of radiopacity

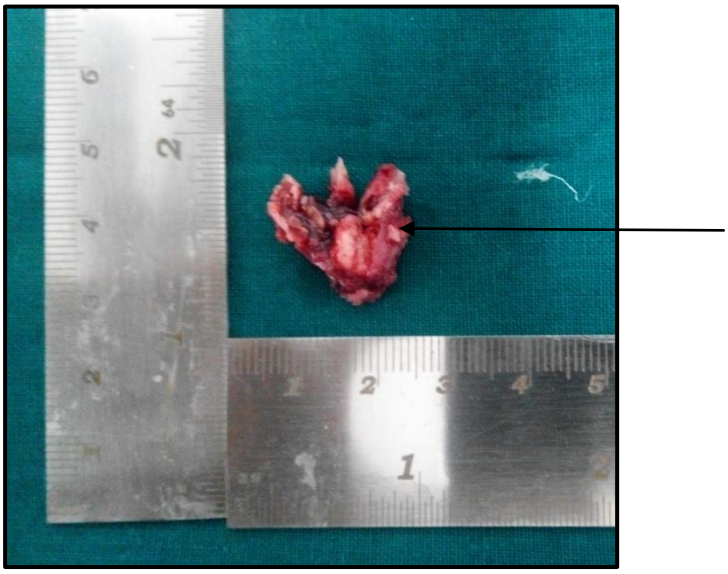

Figure 3A: Macroscopy of the cystic lesion with an odontoma (arrow)

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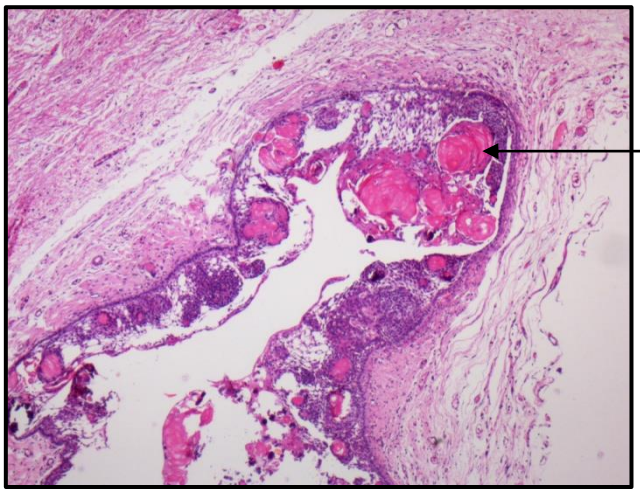

Figure 3B: Microscopy of the cystic lesion showing lining epithelium with ghost cells (arrow) and connective tissue wall (Haematoxylin and eosin, X 100)

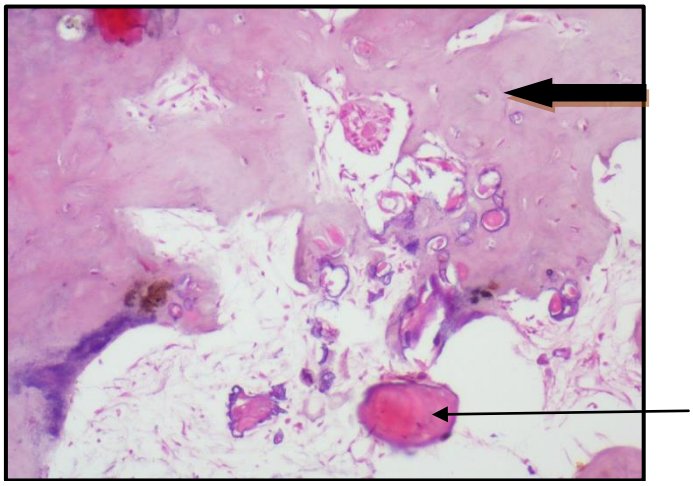

Figure 3C: Microscopy of the odontoma exhibiting ghost cells (arrow) and dentinoid (block arrow)

DISCUSSION

Periapical (radicular) cyst is a common entity associated with a non vital tooth. Periapical inflammatory tissue which is not removed after tooth extraction may give rise to a residual periapical cyst. This residual cyst presents as a round to ovoid radiolucency of differing size in the alveolar ridge at the site of a previous tooth extraction. In an aging residual cyst, luminal radiopacity may be seen due to the presence of dystrophic calcification resulting from degeneration of the cellular contents within the lumen [6].

Calcifying odontogenic cyst is a relatively rare entity which is predominantly an intraosseous lesion. Most cases are diagnosed in the second and third decades of life (Mean age 33 years). However, COCs which are found to be associated with odontoma tend to occur in younger individuals (mean age, 17 years) [7].

Central COC usually presents as a well defined unilocular radiolucency with foci of radiopacities [6].

The cystic epithelium of COC may arise from reduced enamel epithelium, and odontogenic epithelial remnants found in the bone or gingiva [8, 9].

The pathology of COC has diversity (Table 1) [3] and can be broadly classified as cystic (non-neoplastic) or solid (neoplastic) types. Most commonly encountered is a well defined cystic lesion composed of fibrous capsule and an odontogenic epithelial lining of 4 – 10 cell thickness. The basal cells of the epithelial

lining appear cuboidal to columnar, similar to ameloblasts. The overlying layer is composed of loosely arranged epithelial cells resembling stellate reticulum. The most characteristic feature of COC is the presence of altered epithelial cells termed as ghost cells which appear as pale eosinophilic masses with loss of nuclei. Some investigators believe that this change represents accumulation of enamel proteins or coagulative necrosis and others contend that it could be a form of normal or aberrant keratinisation of odontogenic epithelium.

Table 1: Suggested classification of COCs [3]

1. Non-neoplastic (simple cystic) variants (CGCOC)
    a. With nonproliferative epithelial lining
    b. With nonproliferaive (or proliferative) epithelial lining associated with Odontomas (compound or **complex cystic ghost cell odontoma**)
    c. With proliferaive epithelial lining
    d. With unicystic, plexiform ameloblastomatous proliferation of epithelial lining
2. Neoplastic variants
    A. Benign type (CGCOT)
        a. Cystic subtype (cystic CGCOT)
        α.SMA ex epithelial cyst lining
        b. Solid subtype (solid CGCOT)
        α.Peripheral ameloblastoma-like
        β.SMA-like
    B. Malignant type (malignant CGCOT or OGCC)
        a. Cystic subtype
        b. Solid subtype

CGCOC- Calcifying ghost cell odontogenic cyst
CGCOT-Calcifying ghost cell odontogenic tumor
OGCC- Odontogenic ghost cell carcinoma

About 20% cases of cystic COCs are associated with odontomas. This variant presents as a unicystic lesion showing features of COC together with those of a compound or complex odontoma [6].

Neoplastic (solid) COCs are uncommon (2% - 16% of all COCs) and may occur intra or extraosseously. The solid variant of COC exhibits islands and nests of proliferating odontogenic epithelium similar to ameloblastoma. Very few cases of malignant epithelial ghost cell tumors (odontogenic ghost cell carcinomas) have been reported, which exhibit cellular pleomorphism, mitoses and invasion into the surrounding tissues.

Odontoma is the most common odontogenic tumor composed of enamel, and dentin with variable amounts of pulp and cementum; these components may be organized in a pattern similar to a tooth (compound type) or may show haphazard arrangement of tooth components (complex type) [6]. Complex odontoma is frequently seen in the posterior parts of the jaw bone [3]. About 20% of complex odontomas exhibit small islands of ghost cells, which may mark the remnants of odontogenic epithelium that would have undergone aberrant keratinisation and cell death due to local anoxia [6].

The present case showed few contrasting features and some similarities when compared to the reported cases of COC in English literature. The contrasting features are age of the patient, location and presentation of the lesion.

The present case revealed the features of cystic COC associated with complex odontoma. The suggested pathogenesis for this association is that COC with odontoma may be regarded as an odontoma in various stages of development, in which the epithelial component initiates the development of a complex (or compound) odontoma and also at a certain stage forms an epithelial cyst lining, that eventually envelopes the odontoma. As the odontoma and the cyst component are continuous with each other, COC with odontoma should be regarded as a separate entity. The lesion is not a cyst but an odontoma in which there is secondarily formed epithelial cyst. The cyst lining may occasionally show some proliferative activity. Thus it should be called as compound / complex cystic ghost cell odontoma [3]. It is of worth mention that even odontomas (compound and complex types) may exhibit ghost cells in 11% to 18% of the examined cases [10]. The feature that distinguishes odontoma containing ghost cells from the COC associated with an odontoma is the definite formation of a cyst lined by odontogenic epithelium in the latter.

Lesion in the present case would have arised from the remnants of odontogenic epithelium found in the jaw bone.

One case of COC mimicking as a residual cyst has been reported in the English literature [11]. The present case showed COC with odontoma, the observation of which was confirmed only after pathologic examination of the excised lesion. Then, clinical and radiographic findings were reevaluated. COC may be considered under differential diagnoses of periapical radiolucencies with foci of radiopacity, and histopathologic evaluation is mandatory for confirmatory diagnosis.

CONCLUSION

The present paper reported a case of intraosseous calcifying odontogenic cyst associated with complex odontoma which was thought to be a residual cyst. COC with odontoma has been designated as complex cystic ghost cell odontoma. Significance of clinicopathologic correlation is emphasized. In addition, histopathologic diversity of COC has been highlighted.

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REFERENCES

[1] Main JH, Waheed MA. J Natl Cancer Inst 1971;47:711-726
[2] Lucchese A, Scivetti M, Pilolli GP, Favia G. Oral Surg Oral Med Oral Pathol Oral Radiol Endo. 2007;104:391-394
[3] Reichart PA, Philipsen HP. Odontogenic tumors and allied lesions. Quintessence Publishing Co, Ltd. 2004: P- 155- 168
[4] Gorlin RJ, Pindborg JJ, Odont, Clausen FP, Vickers RA. Oral Surg Oral Med Oral Pathol. 1962;15:1235-1243
[5] Freedman PD, Lumerman H, Gee JK. Oral Surg Ora Med Oral Pathol. 1975;40:93-106
[6] Neville B, Damm DD, Allen CM, Bouquot J. Oral and Maxillofacial Pathology. Philadelphia:WB Saunders; 2009. P-695-697
[7] Li TJ, Yu SF. Am J Surg Pathol 2003;27:372-384
[8] Praetorius F, Hjorting-Hansen E, Gorlin RJ, Vickers RA. Acta Odont Scand 1981;39:227-240
[9] Buchner A. J Oral Maxillofac Surg 1991;49:330-339
[10] Philipsen HP, Reichart PA, Praetorius F. Oral Oncol 1997;33:86-99
[11] Manveen JK, Subramanyam RV, Simmerpreet SV, Ramandeep NS. J Clin Diag Res 2010;4:2979-2983