

Odonto calcifying cyst

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Abstract

The calcifying odontogenic cyst (COC) is reported to be associated with odontoma in 24% of cases. Separation of the cases of calcifying odontogenic cyst associated with odontoma (COCaO) may lead to a better understanding of the pathogenesis of this lesion. The literature revealed 52 cases of COCaO. The male to female ratio was 1:1.9, with a mean age of 16 years. Most common location was the maxilla (61.5%). The radiographic appearance of most cases (80.5%) was a well-defined, mixed radiolucent-radiopaque lesion. Histologically, the lesions consisted of a single large cyst with tooth-like structures as an integral part, giving the impression of a single lesion. In addition to the unique histologic features, differences in gender and distribution were found between the cases of COCaO and those of simple COC. COCaO may be regarded as a separate entity and classified as a benign, mixed odontogenic tumor. The term odontocalcifying odontogenic cyst is suggested.

Keywords: Odontogenic cyst, calcification, odontome

Introduction

The calcifying odontogenic cyst (COC) was first defined as a separate entity by Gorlin *et al.*, in 1962.^[1] The COC represents about 1% of jaw cysts, and although it may occur in soft tissue, it is most commonly found within bone.^[2,3] Both the intraosseous and extra osseous forms occur with about equal frequency in the maxilla and mandible, mainly in the incisor and canine areas.^[4-7] Although, the peripheral COC affects individuals in the sixth decade of life, the central variant is observed most commonly in the second decade and does not show gender predilection.

COC is usually asymptomatic and may be an incidental radiographic finding.^[2] Radiographically, the lesion appears as a unilocular or multilocular well-defined radiolucency that may contain small irregular calcified bodies of varying sizes, and it may be associated with an odontoma or an unerupted tooth.^[2,8-10]

Ghost cells are a characteristic feature of COC, although they also occur (less frequently) in other lesions such as the odontoma. In addition, dentin like material and enamel may be present in the COC. The World Health Organization (WHO)'s International Classification of Odontogenic Tumors and Allied Lesions^[11] define it as “a non-neoplastic lesion” and mention the possible association with other odontogenic lesions, such as odontoma. Nevertheless, few cases of true odontoma found concurrently with COC have been reported.^[12-14]

Case Report

A 13-year-old boy reported with the chief complaints of space in the maxillary front tooth region since 1 month [Figure 1].

History reveals that the patient visited a dentist for improper arrangement of the maxillary front tooth region and had undergone uneventful extraction. On clinical examination, extra orally there was no obvious facial asymmetry.

Intra oral examination showed an ill-defined swelling of the buccal gingiva in the region of right maxillary central and lateral incisors measuring 1.5 cm × 1 cm in size. The swelling was slightly obliterating the labial vestibule; surface over the swelling was smooth, normal in color and texture. On palpation, the swelling was bony hard in consistency and non-tender [Figure 2].

On correlating the patient's complaint, with the history and clinical examination a provisional diagnosis of benign tumor of maxilla was made.

On radiographic examination, IOPA Intra Oral Periapical Radiograph showed radiopaque mass with multiple teeth like structures present in relation to 11, 12, 13 measuring 0.5 cm in size. Occlusal radiograph showed expansion and thinning of the labial cortical plate [Figure 3]. OPG Ortho Pan Tomography shows a tooth like radio opaque mass in

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relation to right central and lateral incisor surrounded by a well-defined radiolucent rim towards the inferio-left lateral aspect of the mass [Figure 4].

The radiographic differential diagnosis that were considered are odontome, cystic odontoma, multiple supernumerary teeth, COC, Adenomatoid odontogenic tumour, ameloblastic fibro-odontoma.

The patient was admitted, and enucleation of the lesion was done under local anesthesia. The lesion was easily detached from the surrounding bone and was removed together with hard tissue. The excised specimen consisted of a cystic sac approximately 2 cm in diameter; the odontoma-like hard tissue was attached to the cystic wall.

Microscopically, the histopathological section of the specimen showed odontogenic lining epithelium which was single layered in one area and two to three layers in the remaining areas lining the cystic cavity [Figure 5]. Above the odontogenic epithelium stellate reticulum like cells were arranged. Eosinophilic masses suggestive of ghost cells were seen. A fibrous connective tissue wall was also present [Figure 6].



Figure 1: A 13-year-old boy complains of space in the maxillary front tooth region



Figure 3: Occlusal radiograph showed radiopaque mass with multiple teeth like structures present in relation to 11, 12, 13 with expansion and thinning of the labial cortical plate

The decalcified section of the hard tissue showed enamel and dentin like material arranged in disorganized fashion [Figure 7]. The histopathologic features were suggestive of COC associated with complex composite odontoma.

Discussion

Since 1971, COC has been described by the WHO as a non-neoplastic cystic lesion in which the epithelial lining shows a well-defined basal layer that is often many cells thick and that may resemble the stellate reticulum of an enamel organ, and masses of ghost epithelial cells that may be in the epithelial cyst lining or in the fibrous capsule.^[11] The ghost cells may become calcified and dysplastic dentin may be laid down next to the basal cell layer of the epithelium. It may be associated with complex odontoma or with tissue resembling an ameloblastic fibro-odontoma.

Praetorius^[8] proposed a sub-classification for the heterogeneous group of COC's, in which the cystic lesions



Figure 2: Intra oral examination showing an ill-defined swelling of the buccal gingiva



Figure 4: OPG Ortho Pan Tomograph showing a lesion in relation to the right central and lateral incisor



Figure 5: Odontogenic epithelium lining the cystic cavity

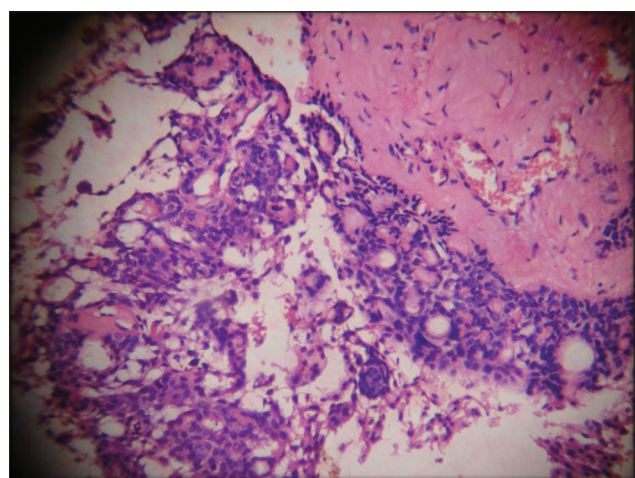


Figure 6: Stellate reticulum like cells arranged above the odontogenic epithelium, the eosinophilic masses suggestive of ghost cells

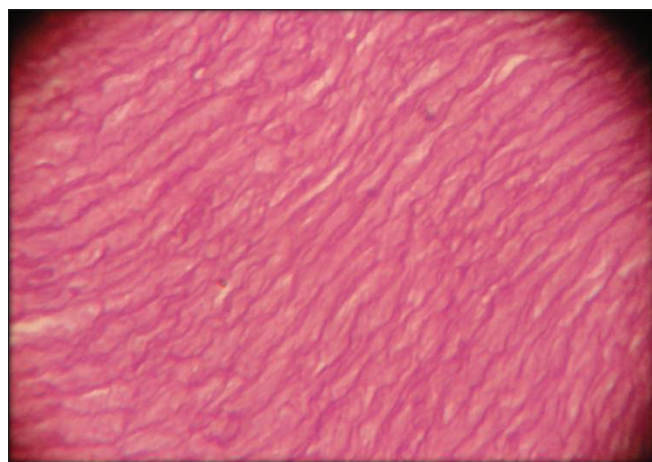


Figure 7: Decalcified section showing hard tissue with enamel and dentin like material arranged in disorganized fashion

were separated from the neoplasms (solid lesions). These researchers further divided the cystic entity into three types,

the simple unicystic type, the unicystic odontoma-producing type, and the ameloblastomatous proliferating type. Other authors have reported only 13-21% of the cysts to be peripheral (extraosseous) lesions.^[6,7] The area affected in this case has been considered the most commonly affected site: About 65% of the reported cases were found in the incisor-canine area.^[8] The radiographic findings of this case (unilocular radiolucency with a radiopaque mass and well-circumscribed borders) are encountered in the majority of odontoma-producing intraosseous COC.

The radiopaque mass observed in the panoramic radiograph of this case, later diagnosed as odontoma components, are considered to be present in 24-50% of the reported cases.^[7] These cases were classified as a subtype of COC under the term odontoma-producing type, or as a cystic variant associated with odontoma.^[11] Hirschberg *et al.*,^[15] proposed that this variant to be classified as a separate entity as odontocalcifying odontogenic cyst. They observed that this variant was more prevalent in females, with a mean age at discovery of 16 years (compared with 34.4 years in the simple COC group) and that most cases were located in the maxilla. The findings of our case were mostly in accordance with the above mentioned observations. This variant may be more common in younger adults, but it may also be discovered in older persons. The possibility of occurrence of an odontoma associated with COC may result from factors present at a certain age, but it seems that the same odontogenic epithelium is present in COC and in this particular variant. Furthermore, it seems that the same characteristic epithelium is involved in the various clinical presentations of this lesion.

Finally, most authorities regard the COC as a lesion with little, if any, tendency to recur, and enucleation is the most commonly reported method of treatment. Several cases of recurrent COC have been reported. Wright *et al.*,^[16] also have reported that recurrence has developed 5 years or more following initial therapy. McGowan and Browne^[17] stated that follow-up periods of up to 10 years are advisable. Therefore, further follow-up examination will also be conducted in the present case, considering the neoplastic nature of the lesion.

Summary

A COC associated with a complex odontoma is described. The presence of reduced enamel epithelium showing extensive proliferation accompanied by dentinoid and enameloid formation, and the morphologic connection of the lining epithelium of the cyst with the enamel epithelium, indicate that the possible origin of the cyst is the enamel epithelium of the odontoma.

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