A rare presentation of hybrid odontogenic tumor involving calcifying cystic odontogenic tumor and plexiform ameloblastoma

Snehal S. Chaubey, Sunil S. Mishra[1], Shirish S. Degwekar, Saujanya Chaubey[2]

Abstract

A hybrid odontogenic tumor comprising two distinct lesions is extremely rare. We presented a hybrid odontogenic tumor composed of a calcifying cystic odontogenic tumor (CCOT) and a plexiform ameloblastoma. This tumor was observed in the anterior area of the mandible of a 17-year-old Indian male. Masses of ghost epithelial cells with the characteristics of CCOT were seen in the lining of the cyst. The odontogenic epithelia with the features of plexiform ameloblastoma were also observed.

Keywords: Calcifying cystic odontogenic tumor, odontogenic tumor, plexiform ameloblastoma

Introduction

Calcifying odontogenic cyst (COC) was first described as a distinct clinicopathologic entity by Gorlin *et al.,* in 1962.[1] Ever since then controversy and confusion have existed regarding its nature. COC is a developmental cyst derived from the odontogenic epithelial remnants.[2] The lesion shows extreme diversity in its clinical and histopathological features as well as in its biological behavior.[3] Initially, COC was considered as a non-neoplastic cystic lesion, but solid lesions with neoplastic nature were also seen. COCs are frequently associated with odontogenic tumors, a finding which is a rare event in other types of odontogenic cysts or tumors.[4] The most common of these is odontoma, but rarely, ameloblastoma, adenomatoid odontogenic tumor (AOT), odontoameloblastoma, ameloblastic fibroma, ameloblastic fibro-odontoma and odontogenic myxofibroma have been identified.[4] In this article, we report the first case of a hybrid odontogenic tumor composed of calcifying cystic odontogenic tumor (CCOT) and plexiform ameloblastoma of the anterior mandible that occurred in a 17-year-old Indian male is described.

Departments of Oral Medicine and Radiology, Sharad Pawar Dental College and Hospital, DMIMSU, Sawangi (Meghe), Wardha, [1]CSMSS Dental College and Hospital, Kanchanwadi, Aurangabad, Maharashtra, [2]Department of Oral and Maxillofacial Surgery, Hitkarni Dental College, Jabalpur, Madhya Pradesh, India

Correspondence: Dr. Sunil S. Mishra, Department Oral Medicine and Radiology, CSMSS Dental College and Hospital, Kanchanwadi, Aurangabad, Maharashtra, India.
E-mail: aegisthesunil@gmail.com

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Case Report

A 17-year-old male patient reported with a presenting complaint of swelling in the lower part of face on the side since 1 month. The swelling was initially small in size and gradually kept increasing. The increase in the size of swelling was associated with tooth mobility in the mandibular right quadrant. The swelling was not associated with any episode of pain or paresthesia and could not be linked to any history of previous trauma or dental infection. Patient visited a private practitioner to seek treatment, but instead was referred to out-patient department.

On extraoral examination, a diffuse swelling was evident in the chin region on the right side extending to the mandibular body to some extent [Figure 1]. The swelling was painless and soft to firm on palpation. Lymph node examination revealed palpable single right submandibular lymph node which was firm and non-tender.

Intraoral examination [Figure 2] revealed diffuse swelling with 41, 42, 43 region involving both labial and lingual aspect of size around 1.5 cm × 1 cm each, which was soft to firm and non-tender on palpation. Radiographic diagnosis [Figures 3 and 4] revealed a large ill-defined, non-corticated interradicular radiolucent lesion with 43 and 42 region causing pathologic drifting and external root resorption of 42, 41, 31. An important finding was evidence of a small focus of calcification near the periapical region of 41.

Under the clinic-radiographic diagnosis of calcifying epithelial odontogenic cyst, patient was advised for aspirational biopsy, which yielded clear fluid. The lesion was enucleated along with the involved teeth, viz. 43, 42, 41, 31. The histopathological examination of the specimen

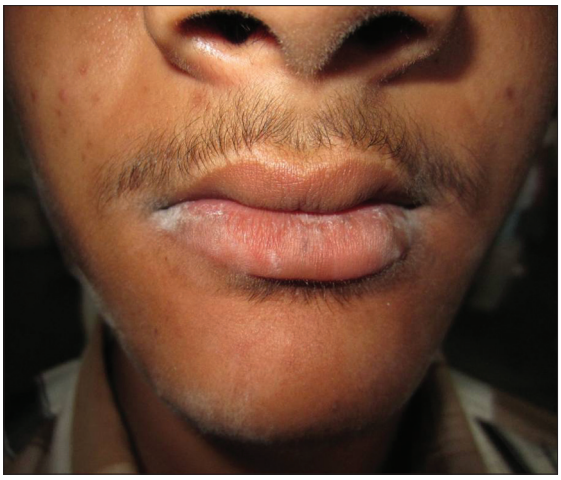

Figure 1: Extraoral view of the patient showing diffuse swelling at the chin region

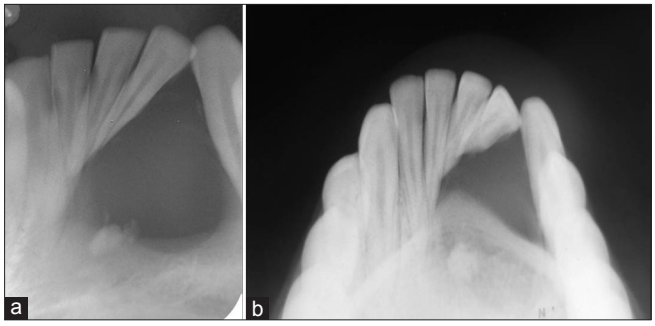

Figure 3: Intraoral radiographic investigation; (a) Anterior Islands organic producers association and (b) Anterior mandibular occlusal topography; showing a bony defect between 42 and 43 with displacement of teeth, root resorption of 42, 43 and a focus of calcification

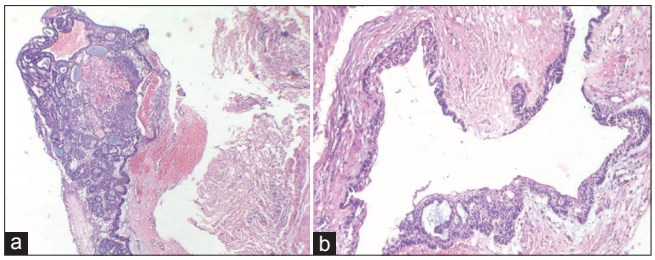

Figure 5: (a) Under scanner view (×4); odontogenic epithelium and connective tissue can be seen, (b) Under low power view (×10); cystic cavity lined by tall columnar odontogenic cells are seen

showed a cystic cavity lined with tall, columnar odontogenic cells along with the presence of ghost cell keratinization and basophilic material with cystic cavity lined by tall columnar odontogenic cells, which is suggestive of CCOT associated with plexiform ameloblastoma [Figure 5]. The patient has been followed-up for 6th month until date and shows no signs of recurrence.

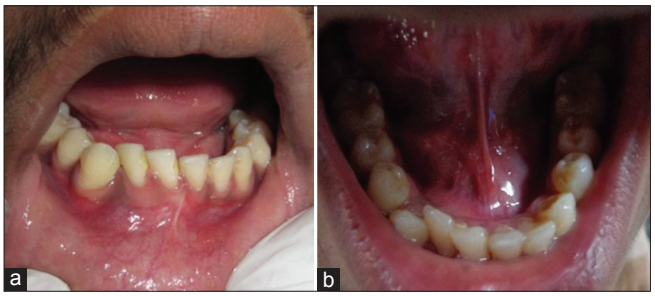

Figure 2: Intraoral view; (a) Labial vestibule showing diffuse swelling; (b) Lingual extension of the swelling

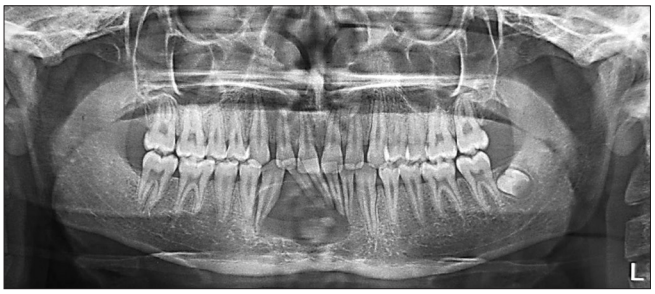

Figure 4: OPG view showing the extension of an ill-defined radiolucent bony lesion with root resorption of the involved teeth

Discussion

COCs are rare odontogenic cystic tumors constituting 0.37-2.1% of all odontogenic tumors. Due to their low prevalence, reports since the first description by Gorlin *et al*., have by necessity involved only a small number of cases.[1,5] The WHO classified COC as CCOT under "benign neoplasm related to odontogenic apparatus" and defined it as "a cystic lesion in which the epithelial lining shows a well-defined basal layer of columnar cells, an overlying layer that is often many cells thick and that may resemble stellate reticulum and masses of ghost epithelial cells that may be in the epithelial cyst lining or in the fibrous capsule."[6]

Classification by Praetorius *et al*. 2006

The latest classification by Praetorius *et al*., 2006[7] categorizes the lesion into four categories and considers each one of them to be distinct in its architectural presentation and biological behavior, as follows

- Grade 1
  - Simple cyst
  - COC.
- Group 2
  - Cysts associated with odontogenic hamartomas or benign neoplasms: CCOT. The following combinations have been published
  - CCOT associated with an odontome
  - CCOT associated with AOT
  - CCOT associated with ameloblastoma
  - CCOT associated with ameloblastic fibroma
  - CCOT associated with ameloblastic fibro-odontoma

- CCOT associated with odontoameloblastoma
- CCOT associated with odontogenic myxofibroma.

- Group 3
  - Solid benign odontogenic neoplasms with similar cell morphology to that in the COC and with dentinoid formation
  - Dentinogenic ghost cell tumor.
- Group 4
  - Malignant odontogenic neoplasms with features similar to those of the dentinogenic ghost cell tumor
  - Ghost cell odontogenic carcinoma.

The CCOT may occur in association with other odontogenic tumors, frequently odontoma. Other lesions showing combined histopathological features with CCOT include orthokeratinized odontogenic cyst, ameloblastoma, dontogenic myxofibroma, ameloblastic fibroma, ameloblastic fibro-odontoma and both ameloblastic fibro-odontoma and AOT. The CCOT simultaneously occurring with another lesion at different locations is rare. However.,[8] in this article, this is the first report of a CCOT lesions associated with plexiform ameloblastoma in anterior mandible. The mechanism is unknown of such unique mixture but it is widely considered that the development of CCOT, which possesses the features of other odontogenic tumors results in the development of these tumors secondarily, rather than that these tumors are themselves secondary phenomena of a preexisting odontogenic tumor.

These lesions are observed in association with an unerupted tooth in 10-32% of the cases. Radiographically, it appears as either a unilocular or multilocular radiolucent area with either well-circumscribed or poorly defined margin. Differing amounts of radiopaque materials are observed.[9] Some reports have indicated that computed tomography (CT) may be more successful than plain film radiography in depicting such calcification because such calcifications may be obscured in plain radiographs by superimposition of anatomic details. CT findings characteristic of COC include calcification along the outline of the bony cavity and have been reported previously.[9] In this article, the radiographic findings were in accordance to previous reports.

The microscopic feature of CCOT is characterized by the lining of a well-defined ameloblastomatous epithelium with variable amounts of ghost cells. Several arguments have been proposed regarding the nature of ghost cells. Some studies suggested that the ghost cells might present the product of abortive enamel matrixin odontogenic epithelium because enamel proteins were immunohistochemically expressed in the ghost cells. However, other studies indicated that the ghost cells represented different stages of normal and aberrant keratin formation or the metaplastic transformation and coagulative necrosis of the odontogenic epithelium.[10]

Surgical enucleations had been performed in all cases. In this case having adjacent teeth with resorbed root, these teeth were removed simultaneously with enucleation of the lesion. Although, enucleation and excision appeared to cure the hybrid lesion, long-term follow-up data and additional cases are still needed to clarify the clinical significance of these lesions.

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