Synchronization of Calcifying Odontogenic Cyst and Aneurysmal Bone Cyst: A Case Report

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Abstract

Although aneurysmal bone cysts and calcifying odontogenic cysts accompanied with other lesions are reported in the literature, the simultaneous occurrence of these two distinct lesions has not been reported. To the best of our knowledge, this is the first report describing co-occurrence of these two lesions located in the left mandibular ramus in a 36-year-old woman.

Key words: Aneurysmal, Bone Cysts, Calcifying, Neoplasms, Odontogenic Cyst.

Introduction

Calcifying odontogenic cyst (COC) or calcifying cystic odontogenic tumor (CCOT) is one of the ghost cell odontogenic lesions that mostly grows as a cystic lesion. Based on the current WHO classification system, these lesions can also have a solid or malignant nature (1).

COCs mainly occur in the anterior areas of both jaws and generally account for about 2% of all the jaw cysts (2).

Although these painless, slow-growing lesions affect people across a wide age range, most cases are diagnosed in the second decade of life and do not demonstrate a gender predilection (3). Histopathologically, COCs have a fibrous capsule and lined with odontogenic epithelium.

The main characteristic of these lesions is the presence of a ghost cells within the epithelial lining (1).

Radiographically, COCs show a wide variety of internal structures. It may appears completely radiolucent or demonstrate calcified materials in the radiolucent area. Multilocular appearance is rare (4).

Aneurysmal bone cyst (ABC) is an intraosseous pseudocyst that can be created de novo or as a secondary lesion arising in association with other bone lesions (1).

ABCs essentially occur in long bones and vertebrae. Only 1.9% of all ABCs arise in the jaws.

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The body and ramus of the mandible are the most affected areas by these lesions (5).

ABC mostly manifests itself in the first two decades of life with a slight inclination towards females (4). Microscopically, the particular feature of ABC is blood-filled cavities without endothelial or epithelial lining (1). The typical radiographic appearance is that of a multilocular with wispy, ill-defined bony septa. A desire to make an extreme expansion of the outer cortical plates is characteristic of this lesion (4).

Here, we describe the first case of COC in association with ABC in a 36-year-old woman.

Case Report

A 36-year-old woman presented with the chief complaint of tenderness on the lingual surface of the left for four months. Overlying mucosa was normal and there were no signs of inflammation, bleeding, pus discharge, expansion or facial asymmetry (Fig.1). The patient's medical history was unremarkable.

The orthopantomograph and cone beam computed tomographic (CBCT) images showed a multilocular radiolucent lesion located in the left mandibular ramus (Fig.2, 3).

An excisional biopsy was done. Macroscopically, the specimen consisted of three pieces of soft tissue ranging in size from 14 x 4 x 4 mm to 15 x 10 x 5 mm with cream to brown color and elastic consistency and a piece of hard tissue with brown color which measured 5 x 4 x 2 mm.

Histopathologic examination showed a cystic cavity lined with three to multiple layers of columnar basal cells that resembled ameloblastoma and superficial stellate reticulum like cells.

Based on clinical, radiographic and histopathological findings of the lesion, unicystic ameloblastoma and calcifying odontogenic cyst type I (ameloblastic type) were included as differential diagnosis (Fig. 4A-D).

The lesion was then completely resected in a specimen measuring 70 x 45 x 18 mm that extended from the mesial edge of the mandibular left second molar to the left mandibular ramus and coronoid process (Fig. 5). The condylar head and neck were preserved.

Microscopically, several slices from a tissue sample revealed two distinct lesions. Some slices of the specimen demonstrated a pathologic cavity lined by multiple layers of odontogenic epithelial cells in company whit ghost cells, areas of calcification and juxta-epithelial hyalinization. These features were consistent with a calcifying odontogenic cyst (Fig. 4A-D). Other slices showed loose connective tissue, chronic inflammatory cells, foreign body giant cells and vascular spaces filled with red blood cells. These characteristics were clearly demonstrated the aneurysmal bone cyst (Fig. 6A-D).

Finally, the histopathological diagnosis of calcifying odontogenic cyst in association with aneurysmal bone cyst was made. There was no recurrence after the one year follow-up and no significant complications were noted in patient (Fig. 7).

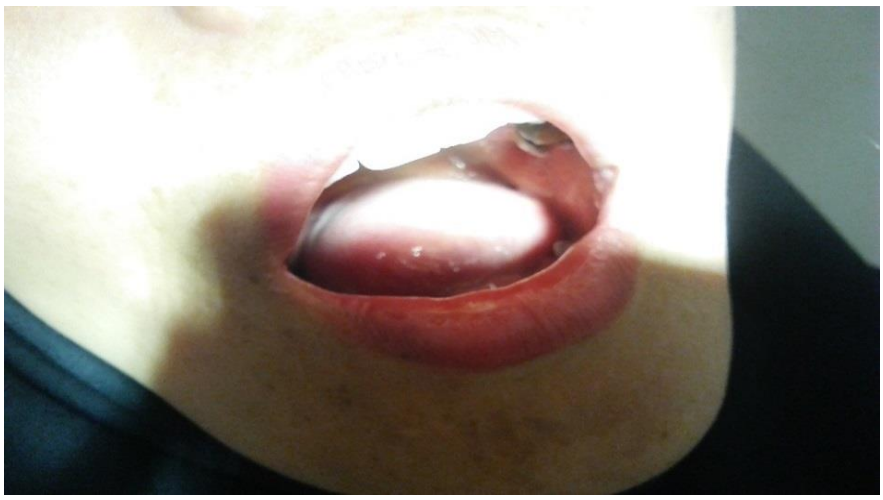

Figure 1. Intra oral photograph.

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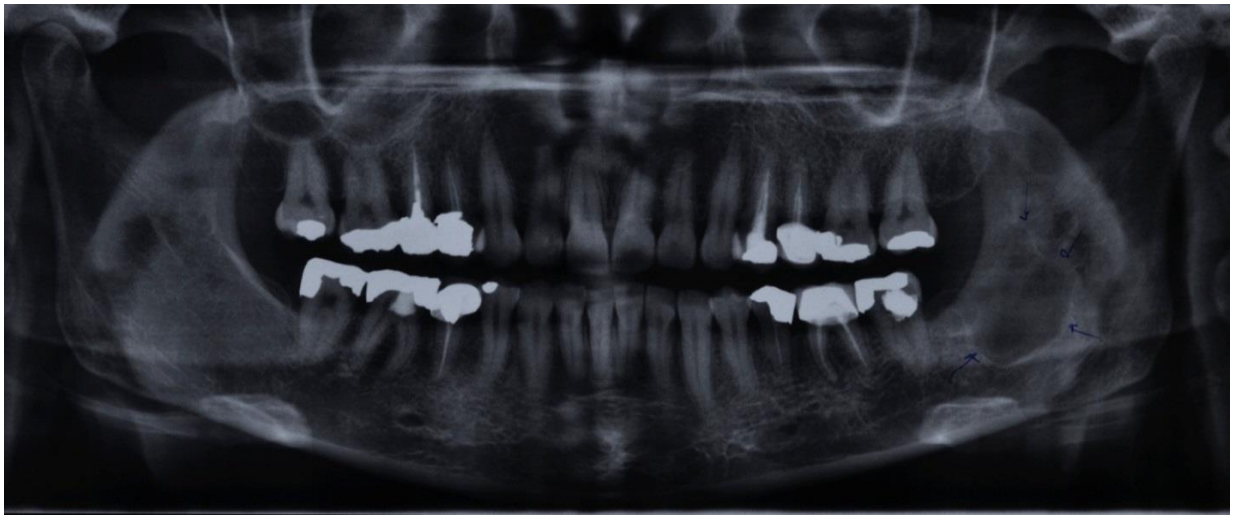

Figure 2. Panoramic image of multilacunar radiolucency occupying the left mandibular ramus.

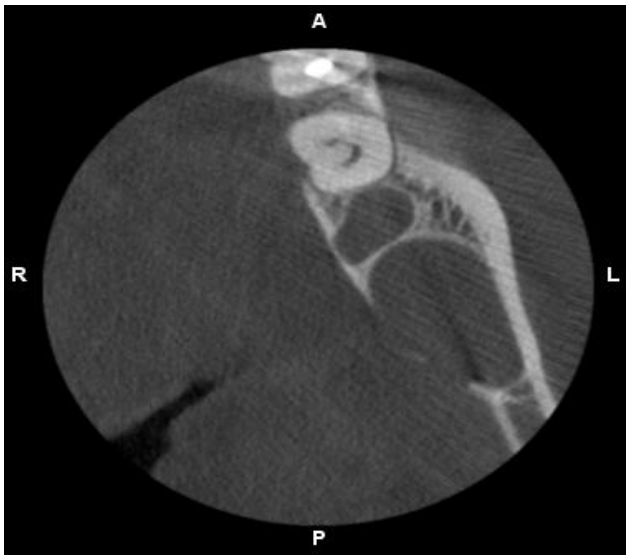

Figure 3. Axial CBCT showing a multilocular lesion at the left ramus of the mandible with thinning of the lingual cortex

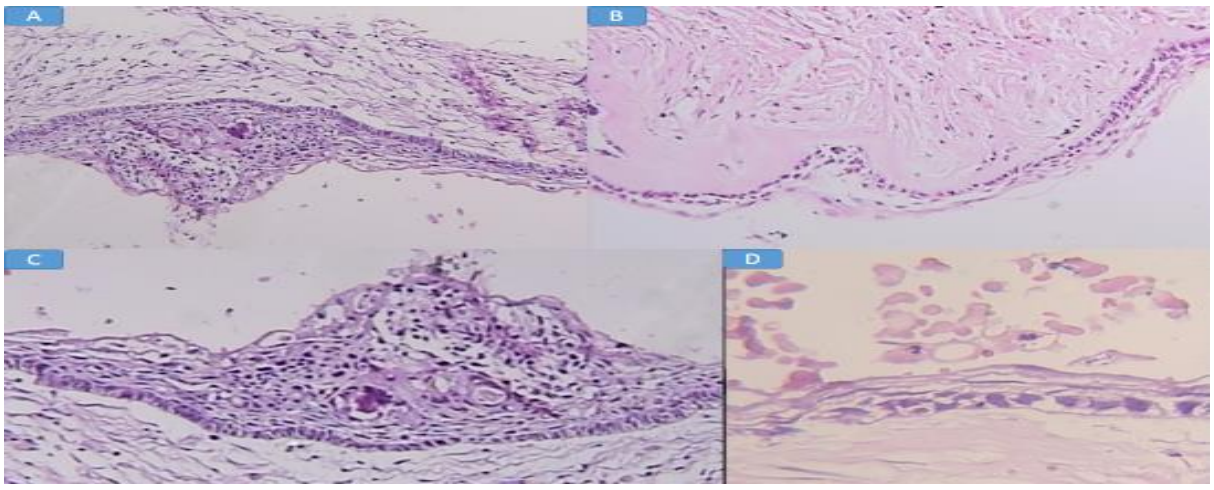

Figure 4. Macroscopic view of the surgical resection.

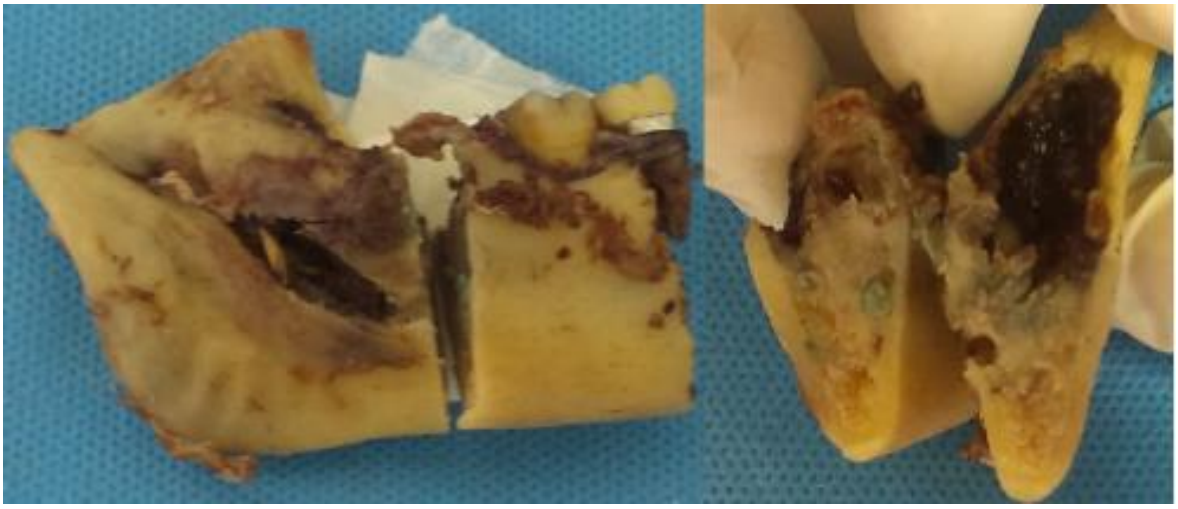

Figure 5. (A) The epithelial lining of the calcifying cystic odontogenic tumor displays palisading of the basal layer, proliferation of epithelial cells towards the cyst lumen and Ghost cells in the cystic epithelium. (B) Juxtaepithelial hyalinization. (C) Numerous calcified particles and (D) ghost cells are present adjacent to the epithelial lining of COC.

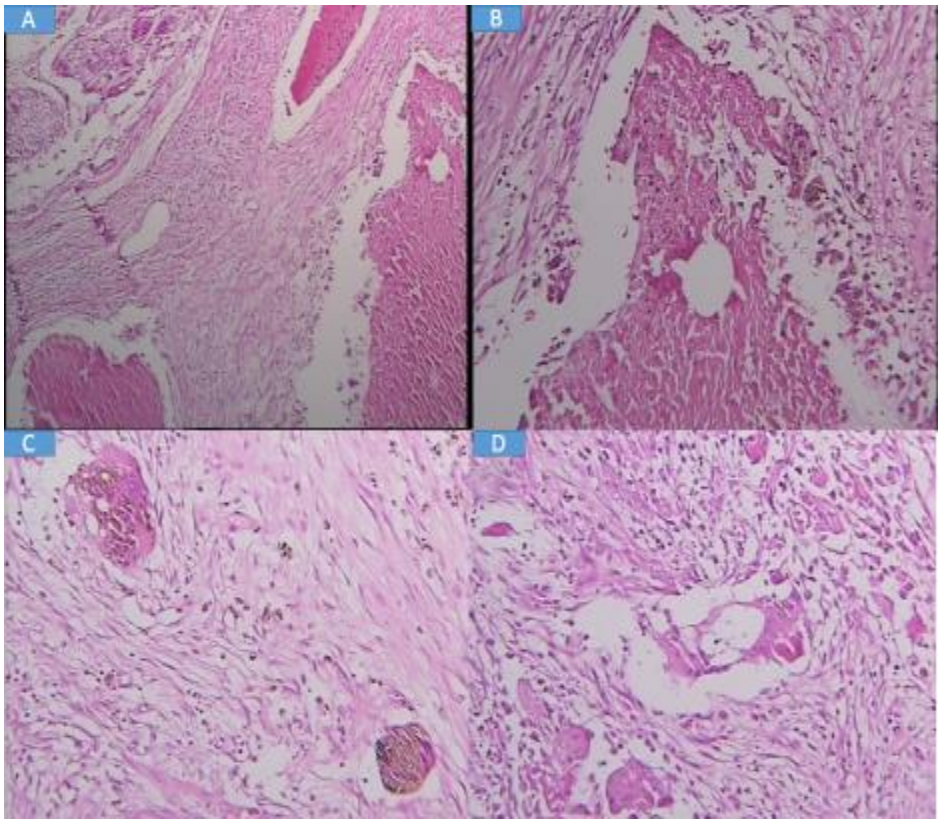

Figure 6. Microscopic examination of the ABC showing cystic spaces filled with red blood cells (A,B), giant cells, loose connective tissue(C), and hemosiderophages(D).

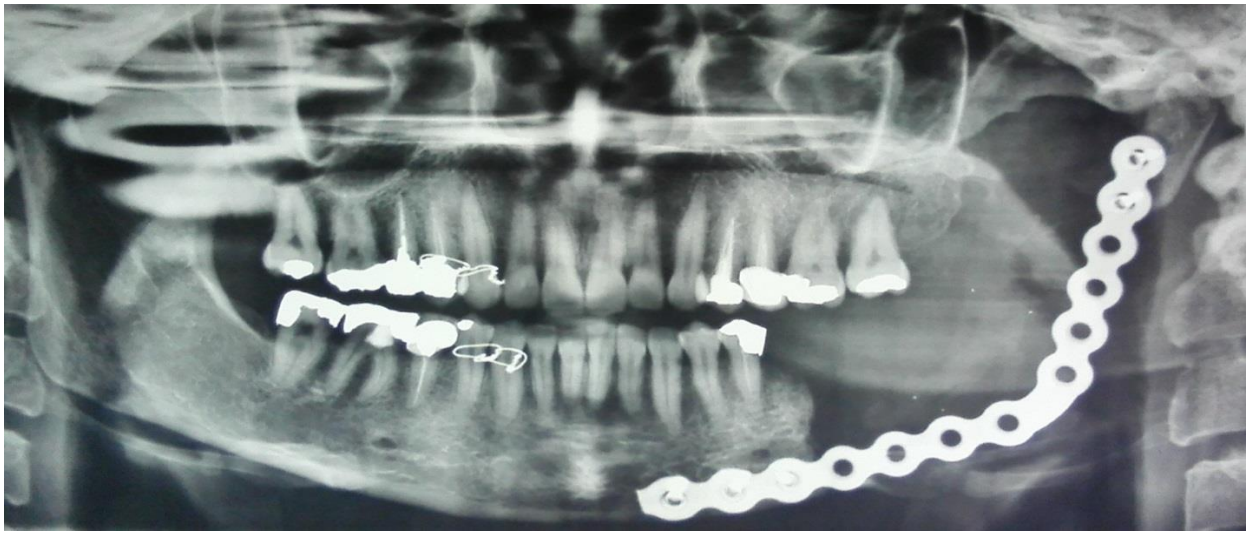

Figure 7. Postoperative panoramic radiographs after one year follow up.

Discussion

After introducing calcifying odontogenic cyst as a distinct lesion by Gorlin *et al*. in 1962, discord and dissension have prevailed regarding its nature (6, 7). This lesion has been considered as a developmental odontogenic cyst or a solid lesion with neoplastic nature (8).

The concomitance of COC and other odontogenic tumors is frequent. COCs in association with odontoma, ameloblastoma, adenomatoid odontogenic tumor, odontoameloblastoma, ameloblastic fibroma, ameloblastic fibro-odontoma, odontogenic myxofibroma and orthokeratinized odontogenic cyst have been reported (9).

Aneurysmal bone cyst was first defined by Jaffe and Lichtenstein in 1942 and has been regarded as a reactive process. Although primary ABCs are mesenchymal neoplasm, secondary ABCs which are associated with other bone lesions such as cement-ossifying fibroma, ossifying fibroma, giant cell tumor, chondroblastoma, osteoblastoma, fibrous dysplasia and dentigerous cyst, aren't considered as a neoplastic process (10-12).

The exact etiology of secondary ABC is unknown, although there are studies suggest that vascular disrupting due to the primary lesion growth and degeneration of pre-existing lesions could lead to creation of ABCs (13, 14).

This paper presents an incomparable combination of calcifying cystic odontogenic tumor and aneurysmal bone cyst, so we have no previous data to make comparisons across the same entities. With regard to age and gender of COC and ABC, patients' age and gender were compatible with both lesions.

Concerning the current location of the COCs, the occurrence of a lesion in the posterior mandibular area was unusual. However, ramus is a common area for ABCs. Expansion, which is the main symptom of ABC, didn't exist in this case. ABCs may occasionally accompanied by tenderness, same the present case. Multilacunar appearance can be rare in COCs, but common in ABCs.

COCs are cured by simple enucleation of the lesion and when they are present associated with other lesions; the treatment is based on the associated lesion (15).

In the present case, the ABC was located behind the COC and not included in the biopsy, so both the pathologist and the surgeon weren't aware of this lesion. The surgeon compelled to do en bloc resection because suddenly confronted with a hemorrhagic lesion. As ABCs are associated with a higher risk of recurrent, resection is safer than simple curettage for treatment (4).

Only a small percentage of secondary ABCs had the radiological appearance of aneurysmal bone cyst; in the other cases the associated lesion dominated the radiographic view (16).

In addition, biopsy specimens are often taken from part of a lesion, so they may show the features of only one lesion and a concomitant lesion may be missed. Therefore, An accurate diagnosis and proper treatment planning requires a close collaboration between the radiologist, the surgeon and the pathologist.

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