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Letter to Editor

Keratocystic odontogenic tumor with mural calcification: A case report

Dear Editor,

The odontogenic keratocyst (OKC), first described by Phillipsen in 1956, has metamorphosized into a keratocystic odontogenic tumor (KCOT) as reported in the World Health Organization (WHO) classification of Head and Neck tumors in 2005.[1,2] Although uncommon, hard tissue formation, namely dystrophic calcifications or cartilage has been reported to occur in the wall of the odontogenic keratocyst. However, calcification in the form of dentinoid is extremely rare.[3]

A 31-year-old male reported to a private clinic in Mangalore with a complaint of swelling in the upper canine region since six months. The swelling was gradually increasing, and causing discomfort. Swelling was slightly painful. Clinical examination revealed swelling extending from the left lateral incisor region to the first premolar region. The overlying mucosa was normal, and all teeth were vital. Radiograph showed a single radiolucency with sclerotic margin extending from the left upper central incisor to the second premolar.

The gross specimen consisted of multiple soft tissue bits that appeared like cystic linings; the largest bit measuring 2.5 × 1.2 × 1.4 cm.

Microscopic examination revealed cyst linings composed of fibrous connective tissue lined by para-keratinized stratified squamous epithelium with surface corrugation [Figure 1]. The lining epithelium exhibited palisading of the basal columnar cells and a flat epithelial connective tissue interface. Some areas of the fibrous connective tissue wall showed extensive basophilic masses with evidence of tubule formation and calcospherite-type of mineralization [Figures 2 and 3]. These masses stained positive for collagen and minerals (picrosirius red and von kossa; Figures 4 and 5) but were negative for amyloid (Congo red). In other parts of the cyst wall, foci of dystrophic calcification and irregular, hairpin-shaped structures were also observed. The remaining areas were made up of dense fibrous tissue. On the basis of these findings, a diagnosis of keratocystic odontogenic tumor with dentinoid-like calcification was made.

The keratocystic odontogenic tumor formerly known as odontogenic keratocyst is a benign developmental odontogenic tumor with many distinguishing clinical and histologic features. WHO recommends the term keratocystic odontogenic tumor as it better reflects its neoplastic nature.[1] KCOT is believed to originate from the dental lamina, occurs most commonly in the mandible; especially in the posterior body and ramus regions. Distinctive clinical features include potential for local destruction and tendency for multiplicity. KCOTs have a high recurrence rate, reportedly between 25% and 60%.[2] A review of the literature suggests that the recurrence rate is relatively low with aggressive treatment; whereas more conservative

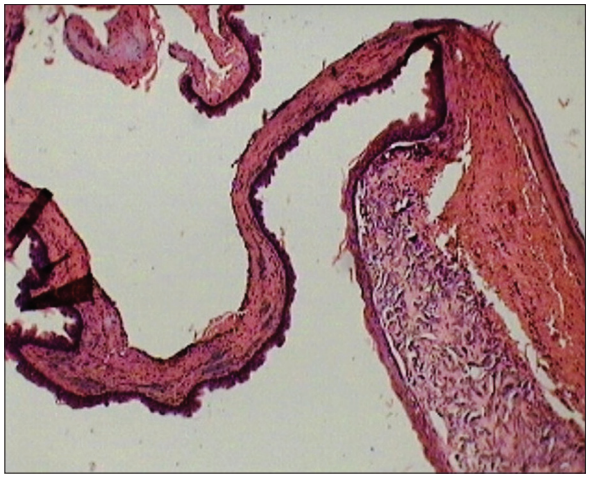

Figure 1: Para-keratinized stratified squamous epithelial lining with surface corrugation and portion of the capsule showing basophilic calcification (H and E, x40)

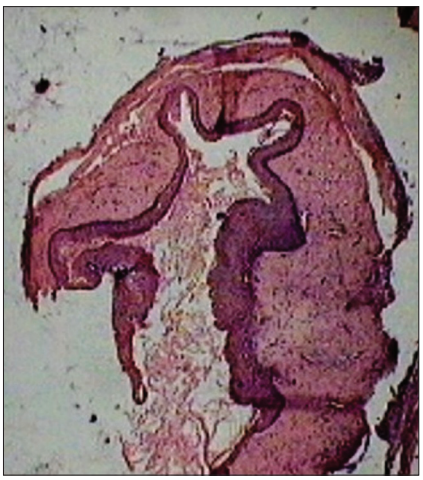

Figure 2: Para-keratinized stratified squamous epithelial lining with surface corrugation, basal cells arranged in palisading arrangement, and lumen filled with keratin (H and E, x100)

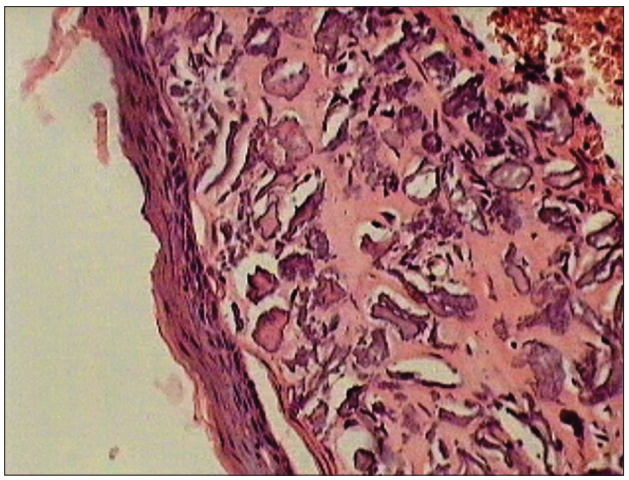

Figure 3: Dentinoid calcification juxtaposed with epithelial lining (H and E, x400)

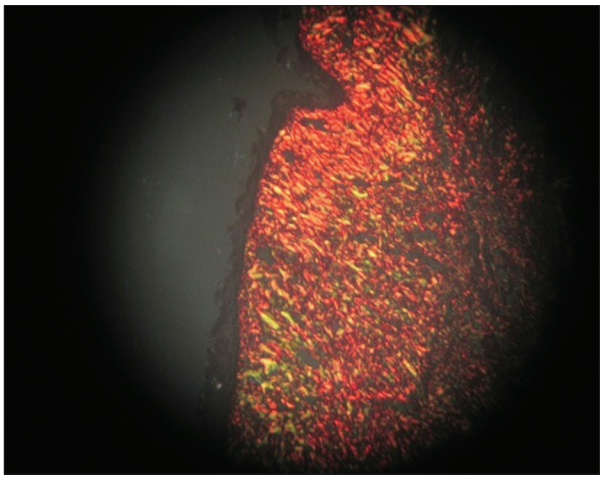

Figure 4: Dense collagen fibers showing orange red birefringence in the cyst wall (picrosirius red stain, original magnification ×100)

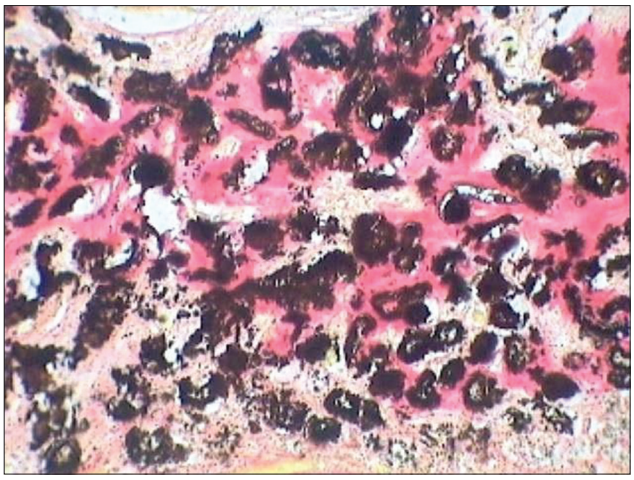

Figure 5: Mineral deposits in the wall of KCOT (von-kossa stain, original magnification ×400)

methods tend to result in more recurrences.[4] Hard tissue formation within the wall of the keratocystic odontogenic tumor is an uncommon finding and usually takes the form of dystrophic calcification. The presence of cartilage and hyaline bodies is also reported. Calcification in the form of dentinoid is extremely rare.[3]

The pathogenic mechanism of amorphous calcified deposits in parts of the connective tissue wall of this cyst is not known. The presence of calcification closer to the epithelium in the present case suggests an induction activity. Presence of short tubules at the periphery of these masses and globular appearance caused by incomplete fusion of calcospherites during the process of mineralization suggest the dentinoid nature of calcification.[3]

Dystrophic calcifications were also seen in the present case which is a comparatively unusual finding. Browne *et al.,* found a high incidence of crystalline calcium phosphates, hydroxyapatite and whitlockite, and inorganic phosphates in the aspirated fluid of the odontogenic keratocysts. This may be responsible for the increased frequency of calcific deposits in the walls of these cysts.[5] It was speculated that in other soft-tissue microenvironments, namely the dermis, dystrophic calcification occurs in a homogeneous matrix produced by pre-existing structures such as sweat glands, nevus cells, or even Rushton's hyaline bodies which are structures of putative epithelial or hematogenous origin that are typically found within the cyst epithelial linings and may occur in about 4.6-11% of odontogenic keratocysts (presently known as KCOT). These bodies may present in a variety of shapes, including linear, curved, or hairpin, and they may calcify. The hairpin-shaped deposits observed in this case belong to this category.[3] The significance of calcification in the behavior of keratocystic odontogenic tumor is not clear. A few studies have shown that primary non-recurrent keratocystic odontogenic tumor showed a slightly higher prevalence for dystrophic calcification than recurrent keratocystic odontogenic tumors.

The inductive effect of the odontogenic epithelial component is the favored explanation for the presence of dentin or dentinoid material in dentin-forming odontogenic neoplasms, and their deposits often appear juxtaposed to the odontogenic epithelium.[3,5]

Dentinoid formation is an unusual finding in the keratocystic odontogenic tumor; tumors which usually show dentinoid materials are calcifying odontogenic cyst, odontoma, ameloblastic fibro odontome, central odontogenic fibroma, and adenomatoid odontogenic tumor. In the present case, histopathological features were classical of keratocystic odontogenic tumor, however the presence of calcification is an unusual finding.[3,5,6,7]

The WHO working group recently highlighted the neoplastic nature of this lesion and reclassified it from cyst to a tumor. Because of the paucity of reported cases of KCOT with dentinoid, the behavior and prognosis of this variant of KCOT is yet to be established.

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