Calcifying odontogenic cyst with atypical features

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ABSTRACT

The calcifying odontogenic cyst (COC) was first delineated in 1962. It is a rare developmental odontogenic cyst with notable presence of histopathological features which include a cystic lining demonstrating characteristic "Ghost" epithelial cells with a propensity to calcify. In addition, the COC may be associated with other recognized odontogenic tumors. This gives rise to a spectrum of variants of COC according to clinical, histopathological, and radiological characteristics. Very few reports have actually captured the actual transformation while most reported cases are documents of co-existing lesions. This article presents one such entity, where the asymptomatic presentation misled the diagnosis and on histopathological examination revealed the COC with areas suggestive of adenomatoid odontogenic tumor.

Keywords: Adenomatoid odontogenic tumor, calcifying odontogenic cyst, cysts, epithelial–mesenchymal interaction, odontogenic neoplasm, periapical lesion

INTRODUCTION

In clinical situations, unusual presentation of uncommon lesion or early presentation of rare lesions can mimic routine pathological entities. The signs and symptoms of such lesions are often misleading and pose a diagnostic and therapeutic challenge. They may require an alternative in treatment plan which in turn could influence the outcome of treatment. Uncommonly, rare pathologies mimic the commonly encountered typical periapical lesions.

True bone cysts are often encountered in facial bones due to the presence of embryonic epithelial rests in these bones. A majority of them are remnants of odontogenic apparatus. The calcifying odontogenic cyst (COC) is a developmental odontogenic cyst and its occurrence constitute about 0.3–0.8% of all odontogenic cysts.[1] Since its description in 1930's and delineation as a unique entity in 1960s, owing to the variations it presents with respect to the histopathology, an extensive classification has been proposed for this entity with four groups and several subgroups.[2] The documented literature confirms that that COC has a spectrum of variants, ranging from that of a developmental odontogenic cyst to benign and possibly a malignant odontogenic tumor.[2] Atypical presentation of COC is not rare. It has been reported to occur in association with odontogenic entities suggestive of adenomatoid odontogenic tumor (AOT).[3]

The aim of this article is to present a case that was nonsymptomatic, which initially mislead the diagnosis. It highlighted the fact that a common clinical diagnosis could be a rare and histopathological surprise. This article also discusses the clinical and histopathological correlation of this condition and its impact on treatment/prognosis.

CASE REPORT

A 43-year-old male electrician reported to our center for prosthetic replacement of his missing lower posterior teeth. His medical history and dental history was non-contributory except that he had multiple uneventful extractions of mandibular second and third molars as well as maxillary third molars several months back. On clinical examination, no abnormalities were observed, except for an indistinct, nontender, hard bulge over the right maxillary canine-lateral incisor region on palpation and also had attrition of anterior teeth. The right maxillary canine and lateral incisor appeared to be diverged. Few posterior teeth also had dental

caries in various stages. Routine panoramic examination revealed a radiolucent lesion in the right anterior region, measuring 1.7 cm × 1.4 cm [Figure 1]. The oval, radiolucent lesion involved the distal part of the root of lateral incisor, the apex, and mesial part of the root of the right maxillary canine with diffuse margin. Closer radiological examination with periapical radiography revealed that the maxillary right canine root was in juxtaposition with the entity while the lateral incisor had a loss of lamina dura [Figure 1]. Clinically, there was a delayed response to pulp vitality test, indicating a compromise in pulpal vitality in both the teeth.

The patient had a parafunctional, occupation related habit of traumatizing his canines repeatedly. Based on the history, radiographic finding, clinical presentation, and pulpal vitality test a provisional diagnosis of periapical granuloma was made. Clinical differential diagnosis of a lateral periodontal cyst, squamous oontogenic tumor, and globulomaxillary cyst were considered. The nontender, indistinct, clinical bulge was attributed as a reactive, buccal cortical expansion of this probable long-standing condition. Uneventful, single visit root canal treatment was done in canine and lateral incisors. The curettage of the periapical granuloma was planned after root canal treatment.

On elevating a wide, semilunar flap, a well-circumscribed area of elevated, thinned cortex was identified [Figure 2]. On removal of the bone over the lesional area, an ovoid fluctuant, swelling with what clinically appeared to be a thick capsule was identified. Carefully, the margins were explored and the entire lesion was removed in total after careful blunt dissection [Figure 3]. The residual bone was clear with no evidence of cystic lining [Figure 4A]. The entire area was thoroughly curetted till healthy bone with pinpoint bleeding spots in bone appeared. Apicectomy and retrograde filling was done in lateral incisor and canine. The area was irrigated with Povidone-Iodine, saline, and primarily closed.

The macroscopic appearance was nonspecific, except for the prominent superficial vasculature [Figure 4B and C]. The cystic mass was subjected to histopathological study. The lesion exhibited cystic lumen lined by a thin layer of odontogenic epithelium (OE). The basal layer of this OE was made up of

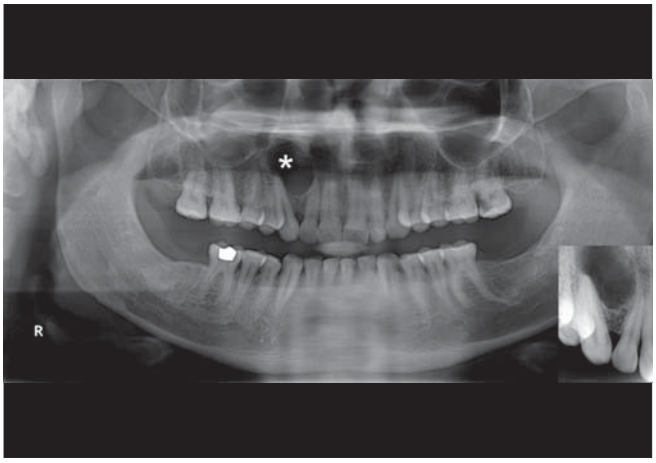

Figure 1: Orthopantomogram of the patient; asterix indicates the extent and nature of the radioluceny in between the apices of the right maxillary canine and lateral incisor. Inset shows the periapical radiograph of the area

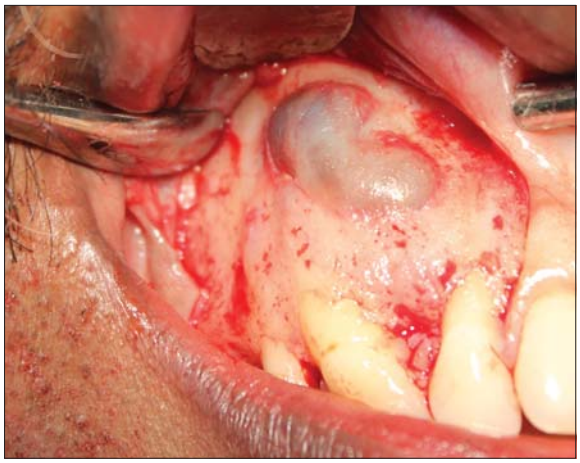

Figure 2: The lesional area after raising the semilunar flap. Note the shape of the lesion and bone overlying the cyst

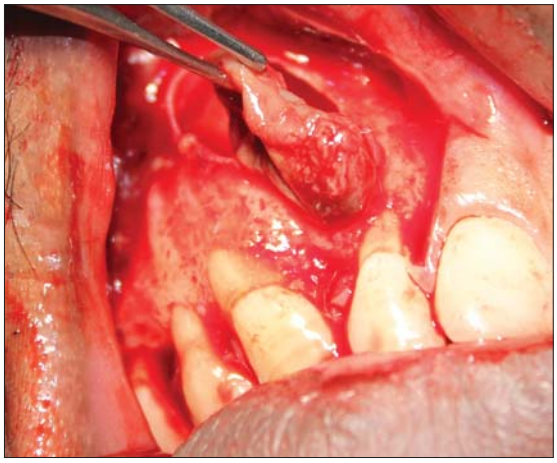

Figure 3: Total removal of the cystic content *in toto*

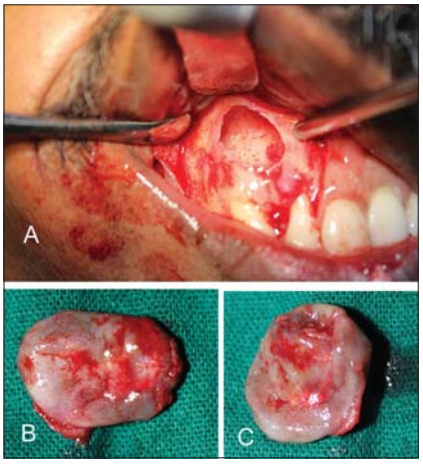

Figure 4: Composite image of A. The residual bone with healthy bleeding after removal of the cyst B. Buccal surface of the cyst C. Lingual surface of the lesion

palisaded columnar or cuboidal cells, resembling ameloblasts of varying thickness. The hyperchromatic nuclei of these cells were polarized away from the basement membrane. These cells were overlined by loosely arranged epithelial cells resembling stellate reticulum. There were a number of epithelial ghost cells which were devoid of nuclei, eosinophilic, and retaining their basic cell outline. These eosinophilic ghost cells were often, enlarged, and were of different shape with well-defined outlines. A few of them contained nuclear remnants [Figure 5].

In one area of the section, thickening of OE was seen. In such areas, islands of odontogenic cells were arranged in the form of thin anastomosing strands in a plexiform and lattice-work pattern. Toward the center of the lesion, in this area, arrangement of ameloblast-like cells in the form of ductal or rosette pattern was observed [Figure 6]. In the center of this ductal arrangement was a pale, eosiniophilic material with varying texture and pattern. Also in one particular foci, cholesterol clefts were identified [Figure 6B]. Based on the histopathological picture, a diagnosis of COC with AOT-like areas was made.

The healing was uneventful and patient was followed up for 6 months with no evidence of recurrence.

DISCUSSION

Periapical pathologies are common in oral and maxillofacial surgical practice, and periapical cysts are more than 50% of all odontogenic cysts reported.[1] Asymptomatic periapical lesions are not uncommon and routine classical description of the radiological appearance of the periapical cysts are a round or ovoid radiolucency lined by a narrow radiopaque margin that extends from the lamina dura of the involved tooth. In enlarging cysts, this margin may be absent. A periapical cyst involving proximal surface of tooth, with or without root resorption, is not uncommon and is attributed to accessory root canals.[4] Care should be taken to rule out periodontal cyst in such conditions. Electricians often engage in parafunctional oral habits such using teeth to strip wires or hold screws. Minor, repeated such insults could compromise the vitality of the pulp. In the present case, as also observed in radiographs, the cuspal tips of maxillary canines are blunt indicating the extent of parafunctional nature. Hence clinically the presentation was thought to be that of a periapical lesion. Absence of a well-defined sclerotic border in imaging indicated that the lesion did not evoke a chronic healing reaction and probably the lesion is a periapical granuloma.

The periapical curettage was planned. As the surgeon encountered a cystic swelling, after removal of bone over the lesional area, the working diagnosis was proved wrong and an enucleation was planned to remove the cyst *in toto*. The dense connective tissue capsule facilitated the *in toto* removal of the lesion. The cystic lesion was carefully removed. As there was a strong attachment of the capsule to bone, careful repeated curettage of residual bone was essential to prevent recurrence. The area was closed after careful debridement. Healing was uneventful.

The histopathological study threw more light into the composition of the lesion. The lesion predominantly was COC with certain areas suggestive of AOT. Similar cases have been reported in maxillofacial surgical literature.[3,5,6]

Odontogenic pathologies arise from odontogenic epithelium (OE). OE by itself has the potential for diverse differentiation under the influence of the ectomesenchyme. In this case, the proliferation of strands of lesional tissue resembled the inner enamel epithelium and hence possibly had the primary ectomesenchymal induction potential. It has been pointed that the combined occurrence of COC with odontogenic pathology is a possibility owing to the inherent potentiality of the OE. The cystic lesion could assume any part of the spectrum. Depending on its degree and direction of differentiation, the initial inductive stimulus, the degree of

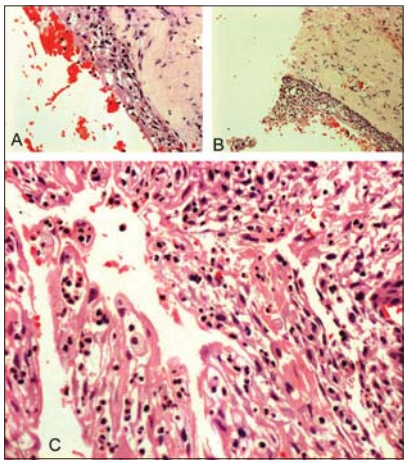

Figure 5: A) The area showing thin OE with basal cells resembling ameloblasts, ghost cells, and extravasated red blood cells. (H and E section, 20×) B) A foci showing the thickened lining with palisaded basal cells resembling ameloblasts, ghost cells, and extravasated red blood cells. (H and E section, 10×) C) A foci showing thickened lining with palisaded basal cells resembling ameloblasts, ghost cells, superficial stellate reticulum-like cells, and extravasated red blood cells. (H and E section, 40×)

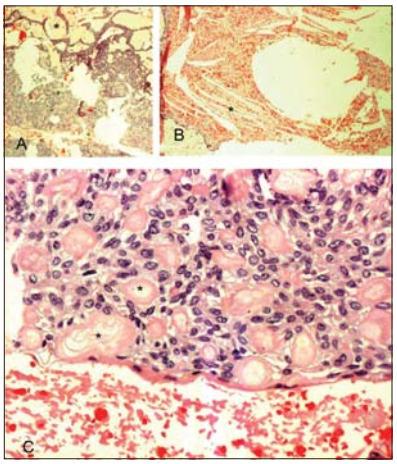

Figure 6: A) Foci showing arrangement of solid areas of odontogenic cells near cystic lumen with cords and lattice pattern, indicative of AOT features (H and E section, 4×) B) Foci showing areas of cholesterol clefts in an uninfected area. C) Foci showing features suggestive of AOT. Note the ductal or rosette-like appearance, reversal of polarity, centrally placed atypical material that has varying pattern (asterix), prominent intercellular bridges, and duplicated basal membrane in certain areas. (H and E section, 20×)

"odontogenesis" prior to the stimulus, and individual human variability, the second lesion might have been initiated.[7]

In the present case, the etipopathogenesis of COC could be from either a mutated clone of presecretory ameloblast or a postsecretory ameloblast that has acquired its mesenchymal inducting potential. It is probably the earlier one that has served as the source of the neoplastic clone. In course of the disease process, they have produced the characteristic ghost cells. Probably, if left for more time, there could have been the characteristic calcifications of the COC. The proliferating cells, at some point of time, by their induction potential have produced features similar to AOT in certain areas near the lumen. It is to be considered that in AOT, the duct-forming cells are reported to possess secretory granules and coated vesicles near the luminal pole, a feature which is highly reminiscent of preameloblasts. These structures were also reported in the non-duct forming columnar cells in various patterns and quantities.[8] Probably this case is the one of the few that serves as evidence to the hypothesis that AOT could evolve from the pluripotent cell lining of COC. If the current case had been diagnosed at a later stage, the lesion probably would have presented with both COC and AOT.

The presence of cholesterol clefts and associated macrophage is unusual in uninfected odontogenic lesions and is probably associated with the degenerating cells. The extensive presence of red blood cells in the tissue section could be due to minor areas of bleeding or due to extensive tissue manipulation during root canal treatment/surgery as mentioned in the case report. The presence of bleeding inside the COC had been reported in the literature.[9] Cholesterol clefts, in radicular cysts, have been postulated to be associated with disintegrating red blood cells in a form that readily crystallizes in the tissues.[10] To the best of our knowledge, cholesterol clefts in uninfected COC have been described for the first time. The presence of cholesterol clefts together with the absence of sclerotic border in imaging indicates that the lesion had been actively enlarging.

In abnormal and rare odontogenic pathologic processes, it is now clearly recognized that a histologic diagnosis can be quite difficult and that a definitive diagnosis is advised to be made in conjunction with clinical and radiologic correlation.[9] The specimen obtained in our present case proved to be a nonaggressive cyst, and the clinical impression/diagnosis was done for a painless, periapical lesion. Although conventional imaging did not reveal any calcification, literature cites that computed tomography had better accuracy in such situations.[9]

The present case concurs with previous reported literature regarding COC. The case which occurred in a male in fifth decade of life, remained asymptomatic, presented as hard swelling. The diagnosis was made on routine radiological examination.[11] The treatment of COC is enucleation and the response depends on any associated odontogenic tumor.[2] As AOT has good and favorable prognosis, the condition would not recur again probably in future. However, we intend and continue to follow up the patient for further monitoring and documentation.

CONCLUSION

A case that had been initially misdiagnosed as periapical pathology and was later identified as COC has been presented. Clinician has to carefully assess all the features before treatment planning. Intraoperative changes in findings have to be included for diagnosis and treatment modified in the better interest of the patient. The case probably is an apt example of a COC giving rise to an odontogenic pathology. Furthermore, this case stresses the fact that all enucleated specimen need to be examined histopathologically to confirm the working diagnosis, so as appropriate precautious treatment plan and follow-up can be instituted.

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