[Downloaded free from http://www.jomfp.in on Tuesday, November 15, 2011, IP: 202.56.124.129] || Click here to download free Android application for this journal

CASE REPORT

Maxillary keratocystic odontogenic tumor with calcifications: A review and case report

Farzan Naveen, Shoaib R Tippu[1], Girish KL, Manpreet Kalra, Vela Desai[2]
Departments of Oral Pathology and Microbiology, [1]Oral and Maxillofacial Surgery, and [2]Oral Medicine and Radiology, Jaipur Dental College and Hospital, Jaipur, India

Address for correspondence:
Dr. Farzan Naveen,
516, Unique Sanghi Apartments,
Mahaveer Nagar, Tonk Road,
Jaipur – 302 015, Rajasthan, India.
E-mail: drfarza@hotmail.com

ABSTRACT

Calcifications or hard tissue deposits in the wall of keratocystic odontogenic tumor (KCOT) are rare and uncommon. Three types of calcifications have been reported in the literature: dystrophic, cartilagenous, dentinoid. A case of maxillary KCOT with calcification is reported with a review on prevalence and pathogenesis of these calcifications. The calcifications associated with the connective tissue of keratocystic odontogenic tumor were studied using special stains. These calcifications were positive for collagen stains, Van Gieson, Masson trichrome, and negative for congo red and mallory suggesting that these irregularly shaped, diffuse calcifications which form on a collagenous matrix are dysplastic dentin.

Key words: Calcifications, keratocystic odontogenic tumor, maxillary

INTRODUCTION

The so-called odontogenic keratocyst (OKC) has been recognized as a separate entity of benign cystic lesions of the jaws for decades. This entity was also referred to as cholesteatoma, epidermoid cyst, sebaceous cyst, or primordial cyst of the jaw. In 1971, the World Health Organization simplified the classification of jaw cysts and made the terms "primordial cyst" and "keratocyst" synonymous.[1] This lesion has been in controversy because of its aggressive nature and finally in 2005 in the revised World Health Organization classification, the OKC was recognized as a benign odontogenic tumors and the new terminology is "keratocystic odontogenic tumor" (KCOT).[2]

Hard tissue deposits, namely dystrophic calcifications, cartilage, dentinoid are uncommon in the connective tissue wall of the primary KCOT.[3] Brown reported a prevalence 16.9% of dystrophic calcifications in primary OKC and 33.3% in syndromic OKC (multiple jaw cysts).[4] Exceedingly rare are the findings of chondroid and dentinoid metaplasia in the wall of KCOT.

Access this article online

Quick Response Code:

Website: www.jomfp.in

DOI: 10.4103/0973-029X.86692

Here we report a case of KCOT with calcifications, in the maxillary-posterior region with a review of different calcifications seen in KCOT and insight into probable pathogenesis of these calcifications.

CASE REPORT

A 23-year-old female presented with a chief complaint of swelling from last 5 years and pain since 4 days. The present history included swelling associated with pain which was throbbing, intermittent in nature, radiating to the ear and head region, aggravated on pressing over the region. Extraoral examination revealed tenderness on palpation over the left zygoma region. Intraoral examination showed swelling in relation to upper left first molar extending posteriorly. The swelling was soft in consistency and was obliterating the vestibule [Figure 1].

Panaromic images showed, well-defined oval-mixed radiolucent lesion with thick sclerotic border present in the left maxillary third molar region at the apicodistal aspect of upper left second molar circumscribing the horizontally impacted third molar extending superiorly and medially [Figure 2]. Aspiration yielded thick blood tinged yellowish material. The cyst was enucleated under general anesthesia and the specimen was submitted to Department of Oral Pathology for histopathologic examination with a provisional diagnosis of dentigerous cyst.

Microscopic examination of aspirate which was yellowish white in color, thick in consistency, and blood tinged was

[Downloaded free from http://www.jomfp.in on Tuesday, November 15, 2011, IP: 202.56.124.129] || Click here to download free Android application for this journal

suggestive of infected cyst [Figure 3]. Histopathological examination revealed cystic lumen lined by corrugated parakeratinized stratified squamous epithelium which was 10-12 layers thick; basal cells were hyperchromatic and polarized [Figure 4a]. Epithelium was found to be separated from the connective tissue at places and the capsule comprised of dense parallel bundles of collagen and was infiltrated by acute and chronic inflammatory cells at places. Connective tissue capsule revealed numerous calcifications [Figure 4b]. Calcifications were eosinophillic with few basophillic calcospherulites [Figures 4c and 4d]. These masses stained positive for collagen (Masson's trichrome and Van Gieson) [Figures 5a-d] but were negative for amyloid (congo red) and keratin (Mallory's stain) and showed weakly positive PAS. The overall microscopic features were suggestive of kerocystic odontogenic tumor with calcifications.

DISCUSSION

Review of the English language literature disclosed that hard tissue formation within the wall of the KCOT is an uncommon finding and that it usually takes the form of dystrophic calcifications, cartilaginous tissue, or dentinoid. The most common calcification in solitary KCOT is dystrophic calcifications, reported to be 4.5-16.8%.[5,6,3] These are usually caused by degeneration and degeneration can be the result of necrobiosis or a foreign-body reaction. Additionally, injured tissue of any kind is predisposed to dystrophic calcification. High incidence of crystalline calcium phosphates, hydroxyapatite, and whitlockite, and inorganic phosphates were found in the aspirated fluid of the KCOT. This may be responsible for the higher frequency calcium deposits in the walls of these lesions. The presence of chondromatous tissues in the adjoining connective tissue of KCOT was reported in only eight cases. They were in the form of well-developed cartilage, chondroid, cartilaginous metaplasia.[7-10] There are several possible explanations for the presence of cartilage in the cyst wall which included the presence of a chondroma; persistence and displacement of vestigial remains of cartilage; a possible metaplastic change of the fibrous connective tissue in response to chronic irritation; a possible induction of the cyst wall by the epithelial lining; and/or trapped glycosaminoglycans.

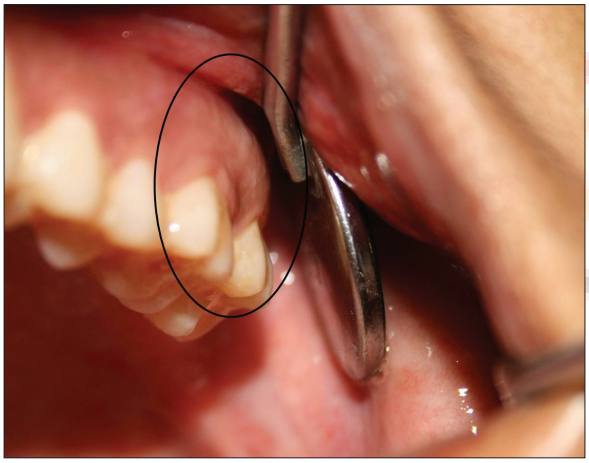

Figure 1: Swelling 2 × 2 cm in size, obliterating the vestibule in relation to upper left-posterior region extending posteriorly

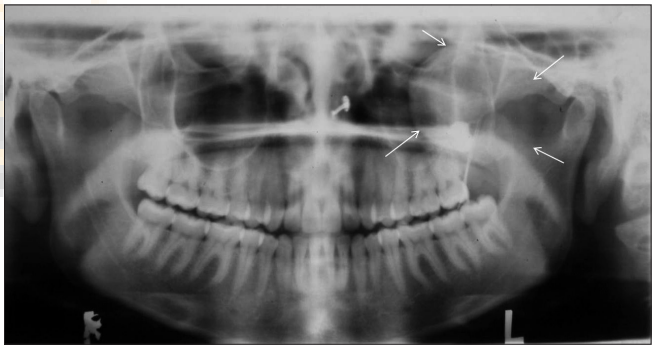

Figure 2: Panaromic radiograph reveals well-defined oval unilocular radiolucent lesion with haziness (arrows) at the apicodistal aspect of 27 circumscribing the horizontally impacted 28

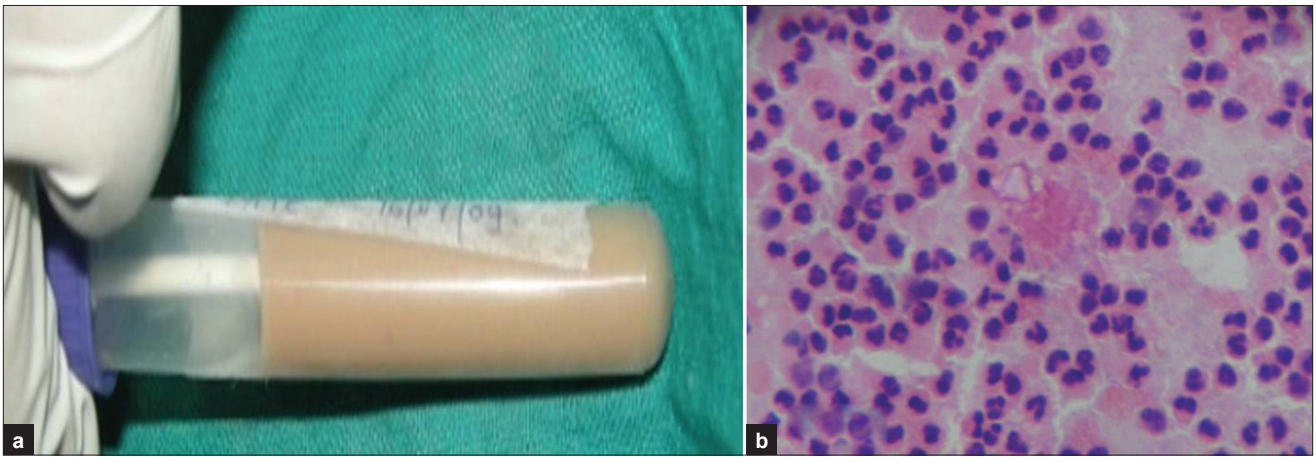

Figure 3: (a) Photograph of thick blood tinged yellowish aspirate from the cystic cavity, (b) photomicrograph of smear showing fibrinopurulent background with acute and chronic inflammatory cells

[Downloaded free from http://www.jomfp.in on Tuesday, November 15, 2011, IP: 202.56.124.129] || Click here to download free Android application for this journal

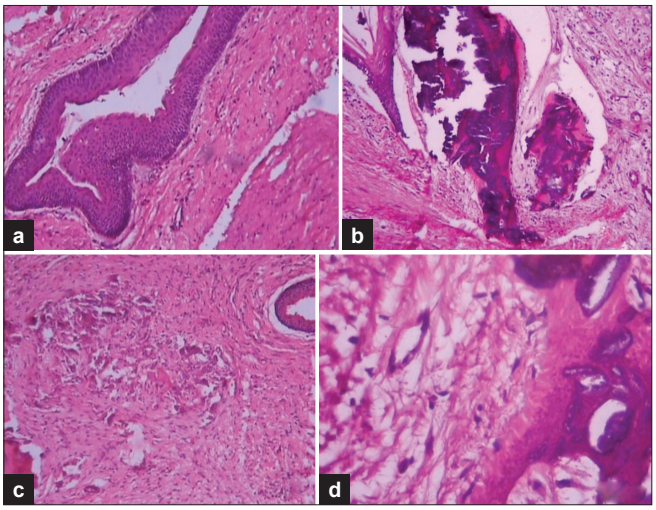

Figure 4: (a) KCOT with typical parakeratinized stratified squamous epithelial lining with corrugated surface (H and E, x20), (b) photomicrograph showing an irregular eosinophillic mass and calcospherulite type of mineralization in the connective tissue capsule (H and E, x20), (c) calcification in association with the daughter cyst (H and E, x20), (d) calcospherulite – type of mineralization at higher magnification (H and E, original magnification ×40)

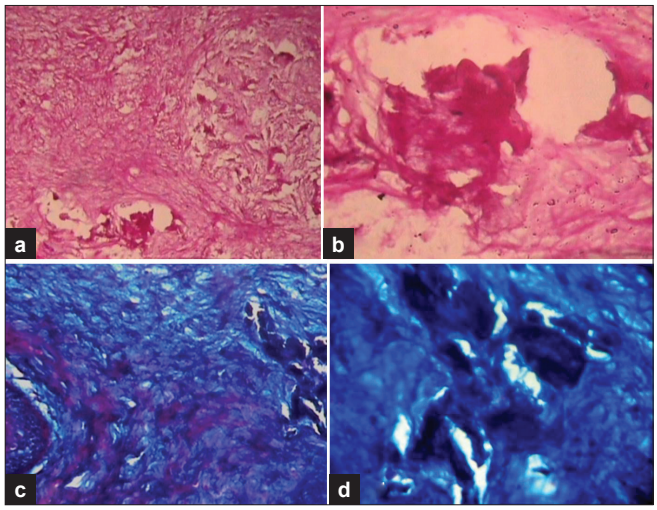

Figure 5: (a-b) Photomicrograph presenting the calcifications (arrowheads) in the connective tissue capsule (Van Gieson, ×10, ×40), (c-d) photomicrograph revealing the calcifications (arrows) in the connective tissue capsule seen adjacent to the daughter cyst epithelium (Masson's trichome, ×20, ×40)

Exceedingly rare is the presence of dentinoid which can occur as irregular eosinophilic masses with tubule formation or calcospherite-like mineralization. The possible pathogenesis could be the inductive changes which mesenchymal cells, can undergo, leading to calcium deposits. Bone sialoprotein (BSP) is synthesized and secreted by bone, dentine and cementum-forming cells and has been implicated in *de novo* bone formation and mineralization.[11] Metaplasia can be regarded as another probable explanation for the presence of these calcifications. Having considered the previous reports, the histological findings of the present lesion resembled strongly the case reported by Ng and Siar[3] who reported dentinoid calcification in the form of calcospherite-like mineralization which was Van Gieson and Masson trichome positive and negative for congo red. Gardener and Sapp[12] in their ultrastructural study of the calcifications reported diffuse calcifications which form in the collagenous matrix to be dentinoid. Kawakami *et al.*[13] reported that the calcified deposits formed in the site of fibrinoid degeneration were located partly on collagen fibers, but mainly on cell debris in foci of degeneration; therefore dystrophic calcifications are usually devoid of collagen or constitutes of only wispy collagen. So Van Gieson and Masson trichrome stains would be negative or slightly positive in cases of dystrophic calcifications. Therefore, these collagen-positive, amyloid-negative irregular eosinophilic masses noted in the connective tissue wall of the KCOT of this case may be identified as dysplastic dentin. As the calcified masses in the current case were associated with odontogenic epithelium islands, we believed that they probably arose by inductive changes. The globular appearance of this calcification may be caused due to incomplete fusion of calcospherites during the process of mineralization.

A case of OKC with dentinoid calcification in the form of calcospherites is presented, and the literature for different calcifications occurring in KCOT is reviewed. Whether hard tissues in these lesions should be regarded as a metaplastic process, or represent a true inductive effect is still to be clarified.

REFERENCES

1. Hodgkinson DJ, Woods JE, Dahlin DC, Tolman DE. Keratocysts of the jaw. Clinicopathologic study of 79 patients. Cancer 1978;41:803-13.
2. Barnes L, Eveson JW, Reichart P, Sidransky D. Pathology and genetics of head and neck tumours. WHO classification of tumours series. Lyon: IARC Press; 2005.
3. Ng KH, Siar CH. Odontogenic keratocyst with dentinoid formation. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2003;95:601-6.
4. Browne RM. The odontogenic keratocyst. Histological features and their correlation with clinical behavior. Br Dent J 1971;131:249-59.
5. Haring JI, Van Dis ML. Odontogenic keratocysts: A clinical, radiographic and histopathologic study. Oral Surg Oral Med Oral Pathol 1988;66:145-53.
6. Kakarantza-Angelopoulou E, Nicolatou O. Odontogenic keratocysts: Clinicopathologic study of 87 cases. J Oral Maxillofac Surg 1990;48;593-600.
7. Kratochvil FJ, Brannon RB. Cartilage in the walls of the odontogenic keratocysts. J Oral Pathol Med 1993;22:282-5.
8. Mosqueda-Taylor A, de la Piedra-Garza JM, Troncozo-Vazquez F. Odontogenic keratocyst with chondroid fibrous wall. A case report. Int J Oral Maxillofac Surg 1998;27: 58-60.
9. Fornatora ML, Reich RF, Chotkowski G, Freedman PD. Odontogenic keratocyst with mural cartilaginous metaplasia: A case report and a review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2001;92:430-4.

[Downloaded free from http://www.jomfp.in on Tuesday, November 15, 2011, IP: 202.56.124.129] || Click here to download free Android application for this journal

10. Arwill T, Kahnberg KE. Odontogenic keratocyst associated with an intramandibular chondroma. J Oral Surg 1977;35:64-7.
11. Chen J, Aufdemorte TB, Jiang H, Liu AR, Zhang W, Thomas HF. Neoplastic odontogenic epithelial cells express bone sialoprotein. Histochem J 1998;30:1-6.
12. Sapp JP, Gardener DG. An ultrastructural study of the calcifications in calcifying odontogenic cysts and odontomas. Oral Surg Oral Med Oral Pathol 1977;44:754-66.
13. Kawakami T, Nakamura C, Hasegawa H, Eda S, Akahane S, Yamazaki T, *et al*. Ultrastructural study of calcinosis universalis with dermatomyositis. J Cutan Pathol 1986;13:135-43.

How to cite this article: Naveen F, Tippu SR, Girish KL, Kalra M, Desai V. Maxillary keratocystic odontogenic tumor with calcifications: A review and case report. J Oral Maxillofac Pathol 2011;15:295-8.

Source of Support: Nil. **Conflict of Interest:** None declared.

Author Help: Reference checking facility

The manuscript system (www.journalonweb.com) allows the authors to check and verify the accuracy and style of references. The tool checks the references with PubMed as per a predefined style. Authors are encouraged to use this facility, before submitting articles to the journal.

- The style as well as bibliographic elements should be 100% accurate, to help get the references verified from the system. Even a single spelling error or addition of issue number/month of publication will lead to an error when verifying the reference.
- Example of a correct style
  Sheahan P, O'leary G, Lee G, Fitzgibbon J. Cystic cervical metastases: Incidence and diagnosis using fine needle aspiration biopsy. Otolaryngol Head Neck Surg 2002;127:294-8.
- Only the references from journals indexed in PubMed will be checked.
- Enter each reference in new line, without a serial number.
- Add up to a maximum of 15 references at a time.
- If the reference is correct for its bibliographic elements and punctuations, it will be shown as CORRECT and a link to the correct article in PubMed will be given.
- If any of the bibliographic elements are missing, incorrect or extra (such as issue number), it will be shown as INCORRECT and link to possible articles in PubMed will be given.