



Calcifying odontogenic cyst presenting as a lateral periodontal cyst — a case report

YEONG-LEI HUANG, LI-MIN LIN, CHENG-CHUNG LIN AND YAT-HANG YAN

School of Dentistry, Kaohsiung Medical College, Kaohsiung, Taiwan, ROC.

We report a case of the calcifying odontogenic cyst occurring on the left anterior mandible of a 41-year-old male with clinical and radiographic features which made it initially to be a lateral periodontal cyst. This unusual presentation of a calcifying odontogenic cyst is discussed and its significance for the clinician is reviewed. A calcifying odontogenic cyst may present some problems for the differential diagnosis both clinically and histopathologically. The differential diagnosis of a lesion occurring in a lateral periodontal location should include the calcifying odontogenic cyst.

Key words: calcifying odontogenic cyst, lateral periodontal cyst.

鈣化齒源性囊腫，側牙周囊腫。

In general, radiographic differential diagnose of lateral periodontal radiolucencies usually include lateral radicular periodontal cysts, deep periodontal pockets, radiolucent odontogenic tumors and benign mesenchymal tumors¹. Lateral periodontal cysts (LPC) are generally considered to be either of a developmental odontogenic origin² or of an infection origin³. They occur most frequently in premolar regions associated with vital teeth, and as a small, well-circumscribed radiolucencies between the roots of the teeth. They appear to arise directly from the lateral periodontal ligament of an erupted tooth⁴. However, different theories explaining their mode of development have recently been proposed⁵⁻⁸. Seventeen percent of the cases of LPC presented with a keratinizing epithelial lining in a study reported by Fantasia. The calcifying odontogenic cyst (COC) was first presented as a separated entity by Gorlin and his associates in 1962 occurred as solid tumor masses. It is defined as follows¹⁰: a nonneoplastic cystic lesion in which the epithelial lining shows a well-defined basal layer of columnar cells, an overlaying 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. The ghost epithelial cells may become calci-

fied. Dysplastic dentin may be laid down next to the basal layer of the epithelium. The purpose of this article is to present a case of COC that was diagnosed radiographically as LPC. The clinician should be alerted that COC must be included in the differential diagnosis of radiolucencies occurring on the lateral root surface of vital teeth.

CASE PRESENTATION

This 41-year-old male patient was referred by a local dentist to the dental department of the Kaohsiung Medical College for further evaluation of a painless swelling mass over the lingual portion of the left mandibular lateral incisor and canine region (Figure 1). This slow growing mass had been presented for over 5 years and had been treated locally. After treatment, the swelling subsided, but it recurred a year ago. Oral examination revealed a 2 cm in diameter, dome-shaped, firm, and elevated mass which was covered by a slight reddish oral mucous membrane and was located on the lingual side from the left mandibular canine to the central incisor region. Physical examination and electrical pulp test (EPT) for associated teeth #22-27 were unremarkable except for a weak response to EPT noted for the two lateral incisors. The periapical and occlusal radio-



Figure 1. A dome-shaped, firm, and elevated mass over the lingual portion of the left mandibular lateral incisor and canine area (arrows).

graphs revealed a well-defined cystic radiolucent lesion extending from the mesial surface of the left mandibular lateral incisor to the mesial surface of the first premolar on the same side and measuring 1.5 cm in diameter (Figure 2). The patient denied paresthesia in his lip and chin area and his medical history was noncontributory. A tentative diagnosis at this stage was LPC. The lesion was excised under local anesthesia and the recovery of the patient was satisfactory. The surgical

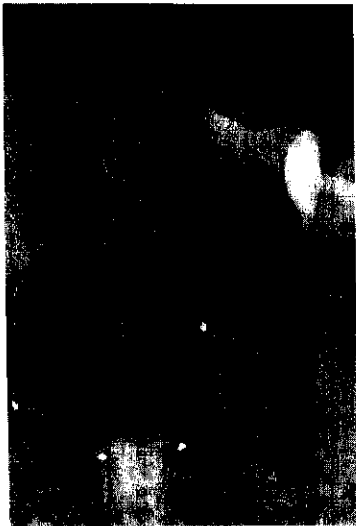


Figure 2. A well-defined cystic radiolucency extending from the mesial surface of the left mandibular lateral incisor to the mesial surface of the first premolar on the same side (arrows).

specimen submitted for pathologic investigation consisted of one tissue fragment measuring 1.4 X 0.9 X 0.8 cm in size. Grossly, it was grayish white and soft with focal hemorrhagic areas. After cutting it in half, a lumen-like structure with a fibrous capsule was found. Microscopically, the lesion was cystic with a dense fibrous connective tissue wall which was lined with a thin layer of dark-stained cuboid "basoloid" cells. Scattered among the epithelial cells were numerous eosinophilic "ghost cells" which represented keratinizing epithelial cells (Figures 3 and 4). Beneath the epithelium in the connective tissue were masses of hyalinized material that resembled dentinoid or osteoid substances (Figure 5). In some areas, the cyst was lined with stratified



Figure 3. A lumen-like structure surrounded by compact fibrous connective tissue and lined with the stratified squamous epithelium on the inner surface. (H & E, 4 X).

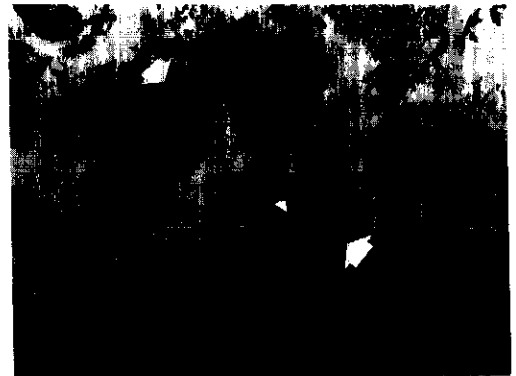


Figure 4. The cystic lining exhibiting intraepithelial keratinizing (arrows) and calcifying (arrowhead) ghost cells. (H & E, 100 X)



Figure 5. Inductive formation of dentinoid or osteoid materials (DM) in the fibrous connective tissue wall just beneath the cystic lining epithelium containing ghost cells (arrow). (H&E, 100 X)

equamous epithelium, while in other areas, epithelial proliferation into the connective tissue was found. There was also focal calcification in the collagenous tissue of the cyst wall. The histopathologic features suggested that it was a case of COC.

DISCUSSION

The histopathologic appearance of the odontogenic epithelial component in cysts and tumors varies to a large extent. The calcifying odontogenic cyst was first described in 1932 by Rywkind¹¹, who reported a lesion of the jaws which he thought was the same lesion as cholesteatoma of the ear. Therefore, he called it cholesteatoma of the jaw. Thoma and Goldman¹² described another similar lesion which they called a strange variant of an ameloblastoma. COC was first recognized as a distinct pathologic entity by Gorlin et al⁹ in 1962. Gorlin et al¹³ considered this lesion to be an analogue of the cutaneous calcifying epithelioma of Malherbe (the pilomatrixoma). Although the designation of COC was referred to as a cyst, many cases appeared as solid tumor masses¹⁰. In the International Histological Classification of odontogenic tumors issued by WHO in 1971¹⁰, COC was indicated to be associated with complex odontoma or with tissue resembling ameloblastic fibro-

odontoma. In a survey of these cystic lesions,

Praetorius¹⁴ pointed out that COC has a more complex nature, which may complicate differential diagnosis. At least four types of odontogenic tumors are known to occur in combination with COC – the ameloblastoma, the odontoameloblastoma, the ameloblastic fibroodontoma, and the complex odontoma¹⁴.

LPC is a cyst which develops on the lateral root surface. The proposed etiologies of LPC are: (a) pulpal inflammation which results in a cyst formation adjacent to an accessory lateral root canal; (b) a dentigerous cyst which assumes a lateral position in relation to an erupting tooth; (c) a result of chronic inflammation of periodontal origin; (d) a primodial cyst of a supernumerary tooth germ; and (e) idiopathic cystication of the rest of Malassez⁷. Although proof of origin is very difficult, the correlation of histology and clinical data often suggests one of the aforementioned theories. Therefore, it is necessary that the clinical, radiographic and surgical findings be reported and the excised tissue examined, in order to determine the pathogenesis of cysts which assume a lateral periodontal position. The clinical and histologic features of lateral periodontal cysts have been analyzed by several authors^{6,7,15}. The clinical course of a lateral periodontal cyst is one of insidious and slow growth^{8,16,17}. It can cause cortical bone expansion, perforation, and root displacement^{7,16,18,19}. The epithelial lining of a lateral periodontal cyst may range from a single flat layer of epithelium to a thickened, proliferating epithelium with varying degrees of exocytosis. The neoplastic potential of a lateral periodontal cyst has been discussed on the basis of its capability for aggressive growth and its common predilection to become an ameloblastoma²⁰. Also the presence clear odontogenic epithelial cells in an LPC and the propensity of these cells for neoplastic change have been correlated with the development of an ameloblastoma. In the case presented here, there was a prominent keratinized epithelium with ghost cells as well as dystrophic calcified material deposited in the ghost cells and in the fibrous connective tissue. This appeared to be a very rare case of COC presenting as LPC.

CONCLUSION

The lateral root surface of the teeth is an unusual location for a COC. A diagnosis of COC can only be made after careful evaluation of the clinical, radiographic, and histologic findings. Treatment of a COC is surgical enucleation with conservation of the surrounding tissues.

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臨床上似側牙周囊腫的鈣化齒源性囊腫

黃湧澧 林立民 林正仲 印憶恆

高雄醫學院牙醫學系

四十歲男性患者，於左側下顎側門齒與犬齒的舌側牙齦部，有一無痛性腫脹，經X光片檢查發現有一放射性透射區位在上述牙齒間區，臨床上診斷為側牙周囊腫，但手術後摘除之組織標本經病理檢查結果是鈣化齒源性囊腫，該病在文獻報告中很少有發生於門齒及犬齒區，因此臨床上在做發生於側牙根區病灶之鑑別診斷時，應將鈣化齒源性囊腫包括其中。

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Reprint requests to: Dr. Yeong-Lei Huang, School of Dentistry, Kaohsiung Medical College, No. 100, Shih-Chuan 1st Road, Kaohsiung, Taiwan 80731, ROC.

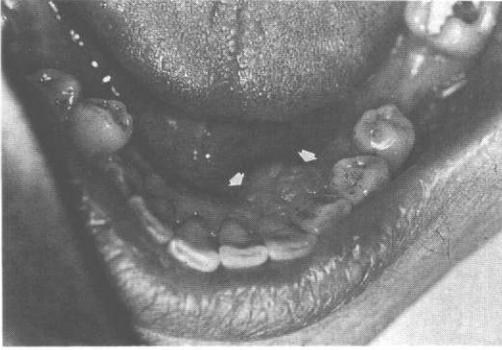


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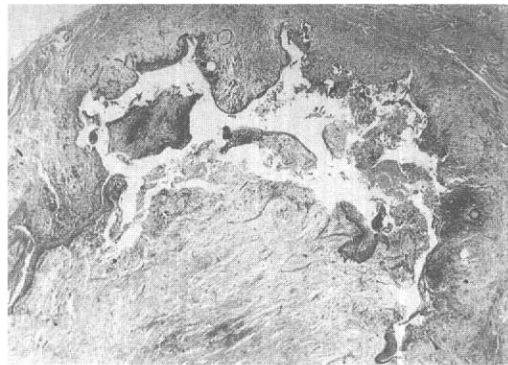


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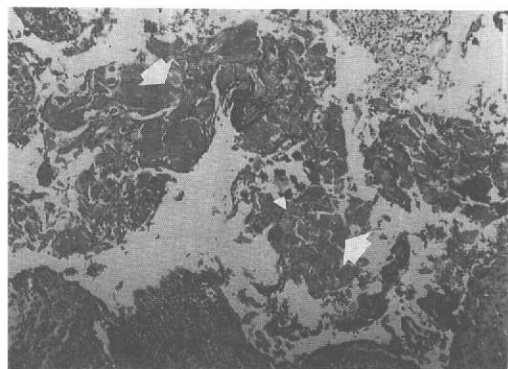


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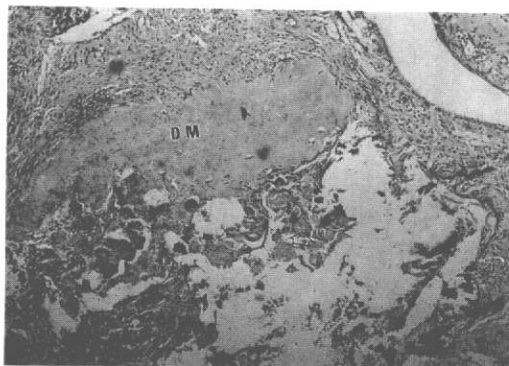


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