

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.e-jds.com

Correspondence

Calcifying odontogenic cyst treated by marsupialization and subsequent total enucleation



KEYWORDS

Calcifying odontogenic cyst;
Marsupialization;
Anterior maxilla;
Total enucleation

Calcifying odontogenic cyst (COC) is an odontogenic cyst characterized by the presence eosinophilic or calcified ghost cells in the lining epithelium.^{1,2} In this study, we presented the clinicopathological feature of a large anterior maxillary COC which was treated by marsupialization and subsequent total enucleation.

This 19-year-old male patient came for treatment of a radiolucent and radiopaque lesion at the right anterior maxilla for more than 10 months. Approximately 10 months ago, he found a swelling at the right anterior maxilla and went to a local dental clinic for management. The panoramic radiography revealed a well-defined radiolucent and radiopaque lesion extending from the distal side of tooth 12 to tooth 16 region. The local dentist did an incisional biopsy and the histopathological report was a COC. Because the lesion was relatively large, marsupialization was performed and a resin button was placed near the apical region of tooth 13 to prevent foodstuff from falling into the bone cavity. Ten months later, he came to our dental clinic for cyst removal. The panoramic and periapical radiographies showed a well-defined radiolucent and radiopaque lesion from the distal side of tooth 12 to tooth 15 region (Fig. 1A and B). The patient was admitted for cyst enucleation. After discussing with the patient and obtaining the signed informed consent, the cystic lesion was completely enucleated under general anesthesia. The totally excised specimens exhibited a cyst partially lined by the ameloblastomatous epithelium with

the cuboidal basal cells and suprabasal stellate reticulum-like cells. The most characteristic features were the presence of eosinophilic or calcified ghost cells in the lining epithelium and in the cystic lumen (Fig. 1C, D, and E) as well as the presence of pieces of eosinophilic dentinoid material in the subepithelial fibrous connective tissues (Fig. 1F and G). At some areas of fibrous connective tissue wall with an infiltrate of lymphoplasmic cells and hemosiderin-laden macrophages, the overlying cystic lining epithelium transformed from the ameloblastomatous epithelium into the stratified squamous epithelium (Fig. 1H). All these characteristic histological features confirmed the diagnosis of a COC.

The COC is a locally aggressive odontogenic cyst. It occurs with an equal incidence in the maxilla and the mandible. Approximately 65% of COC cases are discovered in the incisor and canine areas.^{1,2} Most COCs are between 2 cm and 4 cm in greatest diameter, but if the size of a COC is relatively large, it can be treated by marsupialization and subsequent total enucleation several months later like those procedures performed for our COC case. A large mandibular COC which is treated by initial marsupialization followed by surgical removal of the lesion has been reported in a 11-year-old child. The initial marsupialization has the advantages to decrease the lesion size, induce peripheral bone formation, and reduce the risk of mandibular fracture and inferior alveolar nerve damage. The COC lesion may or may not show juxtaepithelial

<https://doi.org/10.1016/j.jds.2022.02.001>

1991-7902/© 2022 Association for Dental Sciences of the Republic of China. Publishing services by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

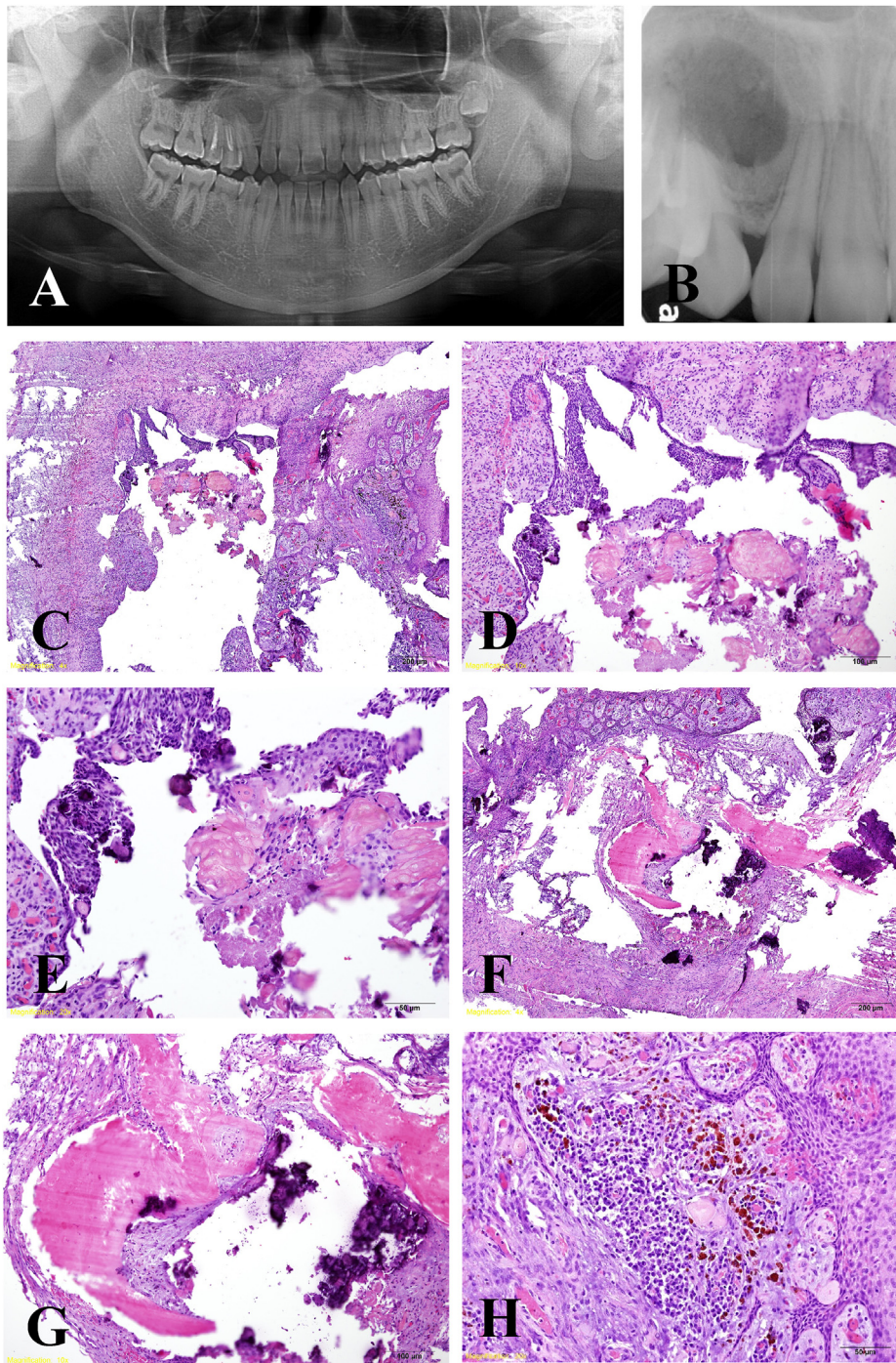


Figure 1 Radiographic and histological photographs of our case of calcifying odontogenic cyst. (A and B) The panoramic and periapical radiographies showing a well-defined radiolucent and radiopaque lesion from the distal side of tooth 12 to tooth 15 region. (C, D and E) Low-, medium-, and high-power microphotographs exhibiting a cyst partially lined by the ameloblastomatous epithelium with the cuboidal basal cells and suprabasal stellate reticulum-like cells. The most characteristic features were the presence of eosinophilic or calcified ghost cells in the lining epithelium and in the cystic lumen. (F and G) Low- and medium-power microphotographs showing the presence of pieces of eosinophilic dentinoid material in the subepithelial fibrous connective tissues. (H) High-power microphotograph revealing an infiltrate of lymphoplasmic cells and hemosiderin-laden macrophages in the fibrous connective tissue wall of the cyst and transformation of the lining epithelium from the ameloblastomatous epithelium into the stratified squamous epithelium. (Hematoxylin and eosin stain; original magnification; C and F, 4 × ; D and G, 10 × ; E and H, 20 ×).

dentinoid material. When the dentinoid material is present in the fibrous connective tissue beneath the lining epithelium, it is believed to be the result of an inductive effect by the odontogenic epithelium on the adjacent mesenchymal tissue.^{1,2} COC may occur in association with different odontogenic tumors, including odontoma, ameloblastoma, adenomatoid odontogenic tumor, odontoameloblastoma, ameloblastic fibroma, and odontogenic myxoma.^{3–5} We have reported a hybrid odontogenic tumor of COC, ameloblastic fibroma, and complex odontoma previously.⁵

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

References

1. Neville B, Damm DD, Allen C, Chi A. Odontogenic cysts and tumors. In: Neville BW, Damm DD, Allen CM, Chi AC, eds. *Oral and maxillofacial pathology*, 4th ed. St Louis: Elsevier, 2016:647–9.
2. Hwang MJ, Lang MJ, Jin YT, Chiang CP. Histopathological study of a characteristic calcifying odontogenic cyst. *J Dent Sci* 2022; 17:578–9.
3. de Moraes ATL, Soares HA, Viana Pinheiro JJ, Ribeiro AL. Marsupialization before enucleation as a treatment strategy for a large calcifying odontogenic cyst: case report. *Int J Surg Case Rep* 2020;67:239–44.
4. Mahdavi N, Khoozestani NK, Hasheminasab M, Soltani N. Hybrid odontogenic tumor of calcifying odontogenic cyst and ameloblastic fibroma: a case report and review of literature. *J Dent* 2020;21:153. 7.
5. Chen YC, Chen PR, Lee YP, Chiang CP. A hybrid odontogenic tumor of calcifying odontogenic cyst, ameloblastic fibroma, and complex odontoma. *J Dent Sci* 2022;17:595–7.

Tzu Hsien Yeh[†]

Department of Dentistry, Hualien Tzu Chi Hospital,
Buddhist Tzu Chi Medical Foundation, Hualien, Taiwan

Yen-Chang Chen[†]

Department of Anatomical Pathology, Hualien Tzu Chi
Hospital, Buddhist Tzu Chi Medical Foundation, Hualien,
Taiwan

Department of Pathology, School of Medicine, Tzu Chi
University, Hualien, Taiwan

Yi-Pang Lee**

Department of Dentistry, Hualien Tzu Chi Hospital,
Buddhist Tzu Chi Medical Foundation, Hualien, Taiwan

Chun-Pin Chiang*

Department of Dentistry, Hualien Tzu Chi Hospital,
Buddhist Tzu Chi Medical Foundation, Hualien, Taiwan

Department of Dentistry, National Taiwan University
Hospital, College of Medicine, National Taiwan University,
Taipei, Taiwan

Graduate Institute of Oral Biology, School of Dentistry,
National Taiwan University, Taipei, Taiwan

**Corresponding author. Department of Dentistry, Hualien
Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, No.
707, Section 3, Chung-Yang Road, Hualien 970, Taiwan.
E-mail address: bonbonlee20140516@gmail.com (Y.-P. Lee)

*Corresponding author. Department of Dentistry, Hualien
Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, No.
707, Section 3, Chung-Yang Road, Hualien 970, Taiwan.
E-mail address: cpchiang@ntu.edu.tw (C.-P. Chiang)

Received 29 January 2022
Available online 24 February 2022

[†] These two authors contributed equally to this work.