Journal of the Formosan Medical Association (2015) 114, 781–782

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.jfma-online.com

CORRESPONDENCE

Noncalcifying variant of calcifying epithelial odontogenic tumor with Langerhans cells

Chih-Huang Tseng [a], Yi-Ping Wang [a,b,c], Jang-Jaer Lee [a,b], Julia Yu Fong Chang [a,b,c,*]

[a] *Graduate Institute of Clinical Dentistry, School of Dentistry, National Taiwan University, Taipei, Taiwan*
[b] *Department of Dentistry, National Taiwan University Hospital, College of Medicine, National Taiwan University, Taipei, Taiwan*
[c] *Graduate Institute of Oral Biology, School of Dentistry, National Taiwan University, Taipei, Taiwan*

Received 2 April 2015; accepted 16 April 2015

Calcifying epithelial odontogenic tumor (CEOT) is a rare benign epithelial odontogenic tumor. It accounts for approximately 1% of all odontogenic tumors. Most CEOTs are intraosseous and the mandible is affected twice as often as the maxilla with a predilection for the molar-ramus region. Local resection or enucleation is the treatment of choice for CEOTs, which have a recurrence rate of approximately 14%.[1]

We herein report a rare noncalcifying variant of CEOT at the left maxilla in a 24-year-old male patient. The patient had biting pain at the left upper canine and premolars for 1 month. Intraoral examination revealed prominent exostoses but no obvious swelling at the left upper canine and premolar areas (Fig. 1A). Percussion pain and increased mobility of the left upper canine and premolars were noted. Electronic pulp test revealed that the left upper canine and the first and second premolars are vital. Periapical and panoramic radiographs revealed a relatively well-defined unilocular radiolucency in the left upper canine and premolar regions and prominent root resorption of the left upper canine and first and second premolars (Fig. 1B and C). Incisional biopsy of the lesion confirmed the histopathological diagnosis of a CEOT. The tumor was totally excised with extraction of the three root-resorbed teeth under general anesthesia.

Histologically, the surgical specimen showed a poorly demarcated odontogenic epithelial tumor perforating the cortical plate of the maxilla with involvement of the lamina propria of the gingiva. The tumor was composed of scattered small epithelial nests and amorphous eosinophilic materials in the fibrous stroma (Fig. 1D). The tumor cells were oval to polygonal in shape and arranged in small nests or strands. Mild cellular and nuclear pleomorphism was noted but no increased number of mitosis was found. Prominent amorphous eosinophilic amyloid-like materials were distributed among the epithelial nests, but no calcification was identified. Most of the amyloid-like materials were negative in Congo red staining with only scanty apple-green birefringence. Anti-S-100 protein and CD1a immunostaining revealed dendritic Langerhans cells in the epithelial islands and fibrous stroma of the tumor (Fig. 1E and F).

There were some differences between this CEOT and the conventional CEOT. A conventional CEOT is often associated with an impacted tooth in the posterior mandible of the middle-aged or elderly patients. Radiographically, most conventional CEOTs present as a mixed radiolucent and

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

* Corresponding author. Department of Dentistry, National Taiwan University Hospital, Number 1, Chang-Te Street, Taipei 10048, Taiwan.
E-mail address: jyfchang@ntu.edu.tw (J.Y.F. Chang).

http://dx.doi.org/10.1016/j.jfma.2015.04.004

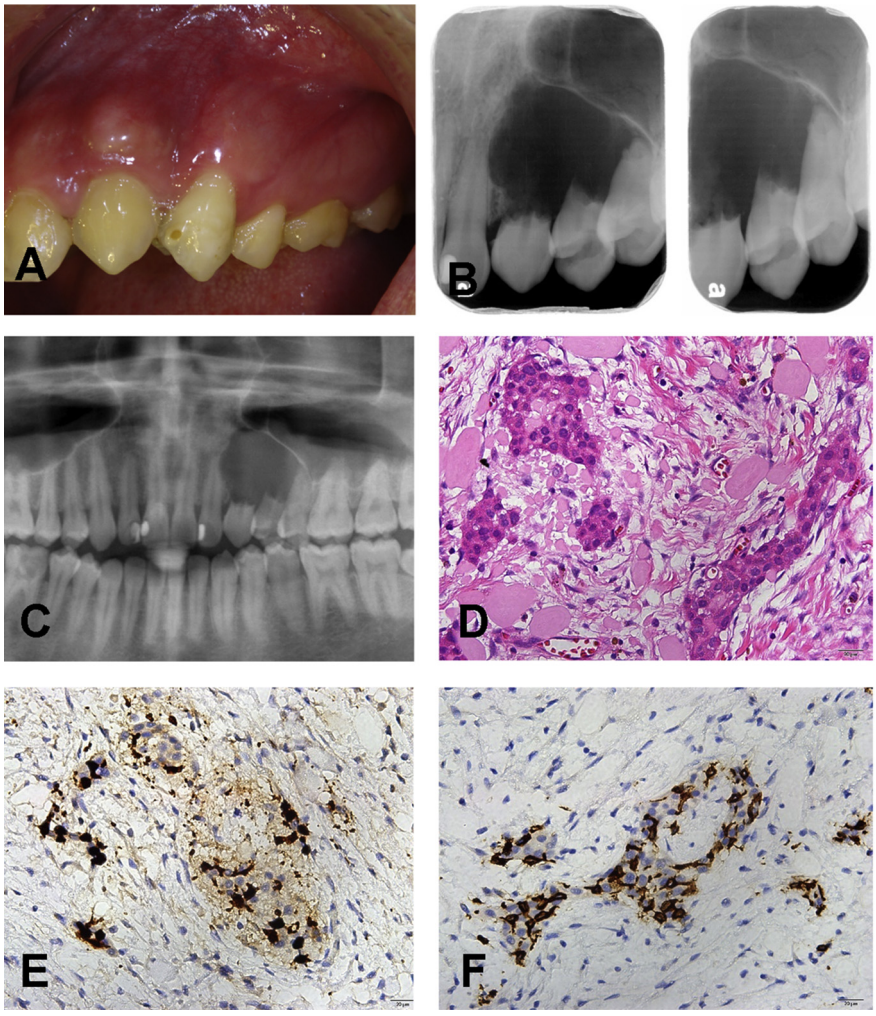

Figure 1 Clinical, radiographical, and histopathological photographs of a noncalcifying variant of calcifying epithelial odontogenic tumor. (A) Clinical photograph of the lesion showing prominent exostoses but no obvious swelling at the left upper canine and premolar areas. (B and C) Periapical and panoramic radiographs of the lesion revealing a relatively well-defined unilocular radiolucency in the left upper canine and premolar regions and prominent root resorption of the left upper canine and first and second premolars. (D) Histopathological microphotograph of the lesion exhibiting scattered small odontogenic epithelial nests and amorphous eosinophilic materials in the fibrous stroma. No calcification was identified (hematoxylin and eosin stain, original magnification, 20×). (E) Immunohistochemical staining showing S-100 protein-positive and (F) CD1a-positive dendritic Langerhans cells in the epithelial islands and fibrous stroma of the tumor (original magnification, 20×).

radiopaque lesion. Histologically, they consist of sheets or large aggregates of polygonal epithelial cells and amyloid and calcified materials in the fibrous stroma.[1] By contrast, the noncalcifying variant of CEOT reported here occurred in the canine and premolar regions of the maxilla of a young adult. It showed a well-defined unilocular radiolucency without foci of calcification. Microscopically, it contained scattered small epithelial nests and cords and some amyloid-like materials without foci of calcification. Immunohistochemically, S-100 protein- and CD1a-positive Langerhans cells were observed in the epithelial islands and fibrous stroma. Actually, the anti-S-100 protein or anti-CD1a immunostaining can also be used to identify the Langerhans cells in the central granular cell odontogenic tumors,[2,3] odontogenic epithelia of odontogenic fibromas,[4] and lining epithelia of odontogenic cysts.[5]

References

1. Barnes L, Eveson JW, Reichart P, Sidransky D. *World Health Organization classification of tumors pathology and genetics of head and neck tumors*. Lyon, France: IARC Press; 2005. p. 302—3.
2. Chiang CT, Hu KY, Tsai CC. Central granular cell odontogenic tumor: the first reported case in oriental people and literature review. *J Formos Med Assoc* 2014;**113**:321—5.
3. Cheng SJ, Wang YP, Chen HM, Chiang CP. Central granular cell odontogenic tumor of the mandible. *J Formos Med Assoc* 2013; **112**:583—5.
4. Wu YC, Wang YP, Chang JYF, Chen HM, Sun A, Chiang CP. Langerhans cells in odontogenic epithelia of odontogenic fibromas. *J Formos Med Assoc* 2013;**112**:756—60.
5. Wu YC, Wang YP, Chang JYF, Chiang CP. Langerhans cells in the lining epithelia of odontogenic cysts. *J Formos Med Assoc* 2013; **112**:725—7.