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Eosinophilic granuloma of the mandible mimicking a periapical lesion

KEYWORDS
eosinophilic granuloma;
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Langerhans cell histiocytosis includes eosinophilic granuloma (EG), Hand-Shuller-Christian disease, and Letterer-Siwe disease. EG presents as either solitary (monostotic) or multiple (polyostotic) bone lesions without visceral involvement; it usually occurs in patients of 10–30 years of age. Hand-Shuller-Christian disease may involve bone, skin, and viscera of 2–10-year-old children. Letterer-Siwe disease is a rapidly progressing disease in which Langerhans cells proliferate in many tissues including skin, viscera, and bone marrow; it is mostly found in children under age 2.[1] Here, we presented a case of EG occurring in the periapical region of the left mandibular second premolar of a 28-year-old female patient.

This 28-year-old female patient was referred to our hospital for the evaluation and treatment of a radiolucent lesion in the left mandible. The panoramic radiography showed a radiolucent lesion with an irregular border at the periapical region of the left mandibular second premolar (Fig. 1A). The electric pulp test revealed that the left mandibular second premolar had a vital pulp. The patient did not have any major systemic disease. The patient was transferred to the Department of Oral and Maxillofacial Surgery and the lesion and part of its overlying gingival tissue were removed by enucleation and curettage under general anesthesia. Histopathological examination of the excised specimens showed a sheet of tumor cells composed mainly of large, pale-staining mononuclear histiocyte-like cells with coffee-beam or indented nuclei and eosinophils (Fig. 1B). The pale-staining histiocyte-like cells were S100 protein-positive (Fig. 1C and D), indicating that they are Langerhans cells. However, the infiltrated lymphocytes in the lamina propria of the gingiva were negative for S100 protein (Fig. 1D). Therefore, the final histopathological diagnosis was an EG. The patient was regularly followed up in the Department of Oral and Maxillofacial Surgery after operation. No recurrence of the lesion was found 10 year after the initial treatment.

The EG is caused by the monoclonal proliferation of Langerhans cells which are mainly identified by immunohistochemistry using either the anti-CD1a or anti-S100 protein antibody in the recent histopathological laboratories.[2–5] Langerhans cells can also be recognized by the existence of coffee-beam or indented nuclei by light microscopy or the presence of Birbeck granules in the cytoplasm by the electron microscopy. Although plasma cells, lymphocytes, and multinucleated giant cells can also be found in the EG lesion, only Langerhans cells are the main tumor cells. The EG may be treated by enucleation and curettage, low-dose irradiation, and intralesional injection with corticosteroids. Infrequently, the spontaneous regression of a localized EG has been reported.[1] The prognosis of patients with a single EG lesion is generally good.[1] In our patient, there was no recurrence of the EG lesion after a follow-up period of 10 years. In this case, the EG lesion presented as a periapical lesion mimicking either a radicular cyst, a periapical granuloma, or a periapical abscess. The electric pulp testing was necessary to rule out those periapical lesions with the pulp necrosis of the associated tooth.

Conflicts of interest

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

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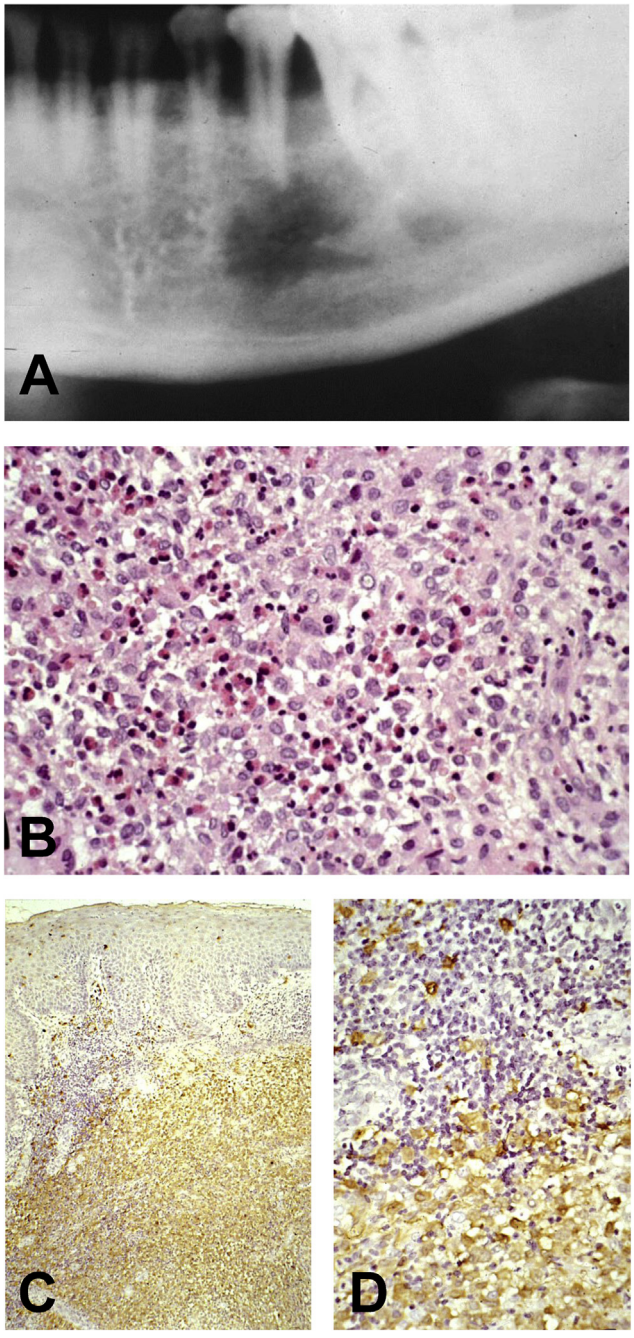

Fig. 1 Radiograph as well as histological and immunostained microphotographs of our case of eosinophilic granuloma. (A) The panoramic radiography showed a radiolucent lesion with an irregular border at the periapical region of the left mandibular second premolar. (B) The tumor contained mainly large, pale-staining mononuclear histiocyte-like cells with coffee-beam or indented nuclei and eosinophils arranged in a sheet pattern (Hematoxylin and eosin stain; original magnification, 20×). (C) The medium-power view showed that the pale-staining histiocyte-like Langerhans cells were S100 protein-positive (immunostain; original magnification, 10×). (D) The high-power view revealed that the superficially-located lymphocytes were S100 protein-negative, but the deeply-situated Langerhans cells were S100 protein-positive (immunostain; original magnification, 20×).

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Ying-Shiung Kuo
Department of Dentistry, Far Eastern Memorial Hospital, New Taipei City, Taiwan

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

Yu-Hsueh Wu
Department of Dentistry, Far Eastern Memorial Hospital, New Taipei City, Taiwan

Graduate Institute of Clinical Dentistry, School of Dentistry, National Taiwan University, Taipei, Taiwan

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

Graduate Institute of Clinical Dentistry, School of Dentistry, National Taiwan University, Taipei, Taiwan

Chun-Pin Chiang*
Department of Dentistry, Far Eastern Memorial Hospital, New Taipei City, Taiwan

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

Graduate Institute of Clinical Dentistry, School of Dentistry, National Taiwan University, Taipei, Taiwan

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

*Corresponding author. Department of Dentistry, Far Eastern Memorial Hospital, No. 21, Section 2, Nanya South Road, Banciao District, New Taipei City 220, Taiwan
E-mail address: cpchiang@ntu.edu.tw

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