



## CORRESPONDENCE

## Langerhans cells in plexiform ameloblastoma



## KEYWORDS

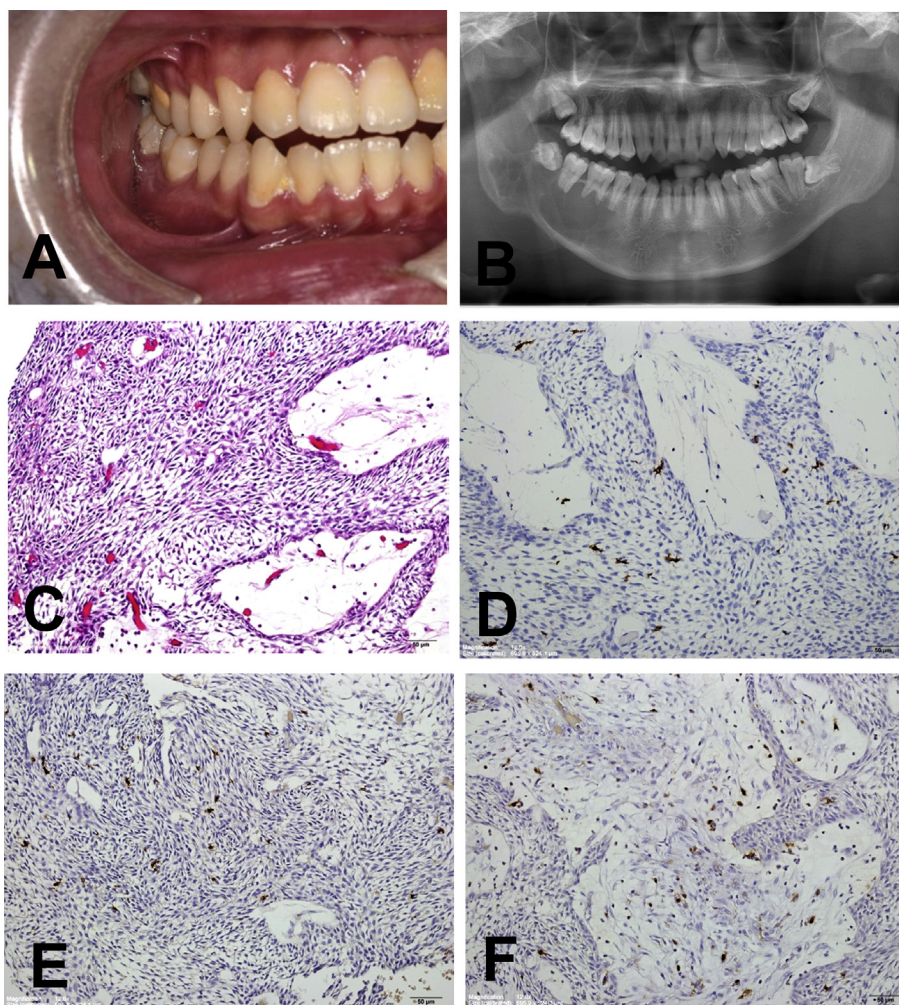
CD1a;  
 langerhans cell;  
 mandible;  
 plexiform  
 ameloblastoma;  
 S-100 protein

Ameloblastoma is a benign odontogenic neoplasm that has locally invasive behavior. Ameloblastomas can be further divided into conventional solid or multicystic, extraosseous or peripheral, desmoplastic, and unicystic types according to the clinical and radiographic features. Histologically, six histologic variants including the follicular, plexiform, acanthomatous, granular cell, desmoplastic, and basal cell ameloblastomas are found. Our previous studies have shown the Langerhans cells in several cysts and tumors of the head and neck region including a unicystic ameloblastoma.<sup>1–5</sup> In this report, we demonstrated the finding of Langerhans cells among tumor epithelial cells of a plexiform ameloblastoma.

This 17-year-old male patient came to the dental department of our hospital with the chief complaint of a swelling over the right lower face for approximately 5 months. Clinical examination showed a slight swelling at the buccal gingiva of teeth 45, 46, and 47 areas with firm consistency and smooth surface (Figure 1A). No palpation pain or numbness of the right lower lip was noted. Panoramic radiography revealed a multilocular radiolucent lesion at the right mandible, extending from tooth 45 to the angle and ascending ramus (Figure 1B). External resorption of apical root portions of teeth 45, 46, and 47 and impaction of tooth 48 were also observed (Figure 1B). The patient was referred to the Oral and Maxillofacial Surgery Department for further treatment. Under general anesthesia, segmental mandibulectomy from tooth 45 to ascending ramus was performed with subsequent bone grafting for the mandibular bone defect. The excised mandibular

bone with the tumor was sent for histopathological examination. Microscopically, the tumor was composed of long, anastomosing trabeculae or sheets of odontogenic epithelia showing the central stellate-reticulum like cells and peripheral columnar or cuboid ameloblast-like cells (Figure 1C). Immunohistochemical staining demonstrated CD1a-positive and S-100 protein-positive dendritic Langerhans cells among the central and peripheral tumor epithelial cells forming a plexiform pattern (Figures 1D and 1E). Langerhans cells were also found in the stromal connective tissues of the ameloblastoma (Figure 1F). These specific histologic findings confirmed the diagnosis of a plexiform ameloblastoma. The postoperative healing of the patient was uneventful.

Langerhans cells are bone marrow-derived antigen-presenting cells that present the antigenic peptides to helper T cells and induce a T cell-mediated immune response.<sup>1–5</sup> In this study, we demonstrated Langerhans cells among the tumor epithelial cells of a plexiform ameloblastoma by anti-CD1a and anti-S-100 protein immunostaining. The peripheral columnar tumor cells are ameloblast-like cells and the central tumor cells are stellate reticulum-like cells. During the tumor growth, some of the tumor cells may undergo apoptosis and release tumor antigens in the tumor epithelial cells forming a network. Because Langerhans cells have epithelial tropism, they may be attracted into the groups of tumor epithelial cells to phagocytose the released tumor antigens. These antigen-carried Langerhans cells may migrate out of the tumor network into the stromal connective tissue and finally reach to the regional lymph nodes, where they process the antigenic proteins into antigenic peptides and further present the antigenic peptides to T cells in the paracortical area of the lymph node. The epithelial tropism and whole antigen-presentation process by Langerhans cell to T cells can explain why we can see Langerhans cells among tumor epithelial cells and in the stromal connective tissues of a plexiform ameloblastoma. However, further studies are needed to elucidate whether the few number of Langerhans cells in ameloblastomas are associated with the locally invasive behavior of ameloblastomas.



**Figure 1** Clinical, radiographic, and histological photographs of our case of plexiform ameloblastoma. (A) Clinical photograph demonstrating a slight swelling at the buccal gingiva of teeth 45, 46, and 47 areas. (B) Panoramic radiograph showing a multilocular radiolucent lesion at the right mandible, extending from tooth 45 to the angle and ascending ramus as well as the external resorption of apical root portions of teeth 45, 46, and 47 and impaction of tooth 48. (C) Hematoxylin and eosin-stained tissue section exhibiting anastomosing trabeculae or sheets of ameloblastoma tumor epithelial cells (original magnification, 20 $\times$ ). (D) Anti-CD1a-stained tissue section showing brown-stained Langerhans cells among tumor epithelial cells (original magnification, 20 $\times$ ). (E) Anti-S-100 protein-stained tissue section demonstrating brown-stained Langerhans cells among a sheet of tumor epithelial cells (original magnification, 20 $\times$ ). (F) Anti-S-100 protein-stained tissue section showing brown-stained Langerhans cells in the anastomosing trabeculae of tumor epithelial cells and in the stromal connective tissue of the ameloblastoma (original magnification, 20 $\times$ ).

### Conflicts of interest

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

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