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Langerhans cell histiocytosis in an old man — Case report

**KEYWORDS**

Langerhans cell
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Maxilla;
Skull bone

Two types of diseases may have punched-out osteolytic lesions in the mandible and skull bone: one is multiple myeloma and the other is Langerhans cell histiocytosis (LCH). Multiple myeloma usually affects old men, but LCH frequently occurs in children or young adults.¹ LCH rarely happens in an old man. In this case report, we presented the clinical features and treatment of LCH occurring in a 74-year-old man.

This 74-year-old man was referred to our hospital for treatment of unhealed extraction wounds after multiple extractions of his mandibular teeth. Posterior-anterior skull radiograph and panoramic radiograph revealed multiple punched-out radiolucent lesions in the mandible and skull bone as well as a scooped-out appearance at the alveolar ridge of the mandible, indicating the destruction of the superficial alveolar bone (Fig. 1A and B). Oral examination showed multiple granulation tissue-like masses on the edentulous mandibular alveolar ridge after extractions of the remaining mandibular teeth in another hospital 2–3 months ago (Fig. 1C). The patient was referred to an oral surgeon for further treatment and a granulation tissue-like mass was removed from the left anterior mandibular edentulous alveolar mucosa and sent for histopathological examination. The biopsy specimen showed a sheet of pale-staining histiocyte-like cells

intermixed with numerous lymphocytes, plasma cells, and eosinophils as well as occasional multinucleated giant cells. The histiocyte-like cells possessed coffee bean-like or indented vesicular nuclei and pink cytoplasm (Fig. 1D and E). Immunostaining revealed that the histiocyte-like cells were positive for CD1a and S-100 protein, suggesting that these positively-immunostained cells are Langerhans cells (Fig. 1F and G). Therefore, the histopathological diagnosis of LCH was confirmed. The patient was referred to oncology department for further treatment. A course of vinblastine chemotherapy (6 mg of vinblastine per m² of body surface) was given to the patient. The one-month follow-up oral examination showed a healed edentulous mandibular alveolar ridge mucosa with complete regression of the granulation tissue-like masses (Fig. 1H).

Immunohistochemical staining is usually used for identification of origin of tumor cells.^{2–5} In this study, although the Langerhans cells were characterized by the presence of coffee bean-like or indented vesicular nuclei by hematoxylin and eosin stain, they were usually confirmed as Langerhans cells by CD1a or S-100 protein immunostains.¹ More than 50% of LCH cases are seen in patients younger than age 15.¹ LCH discovered in an old man is unusual and deserved to be reported in detail.

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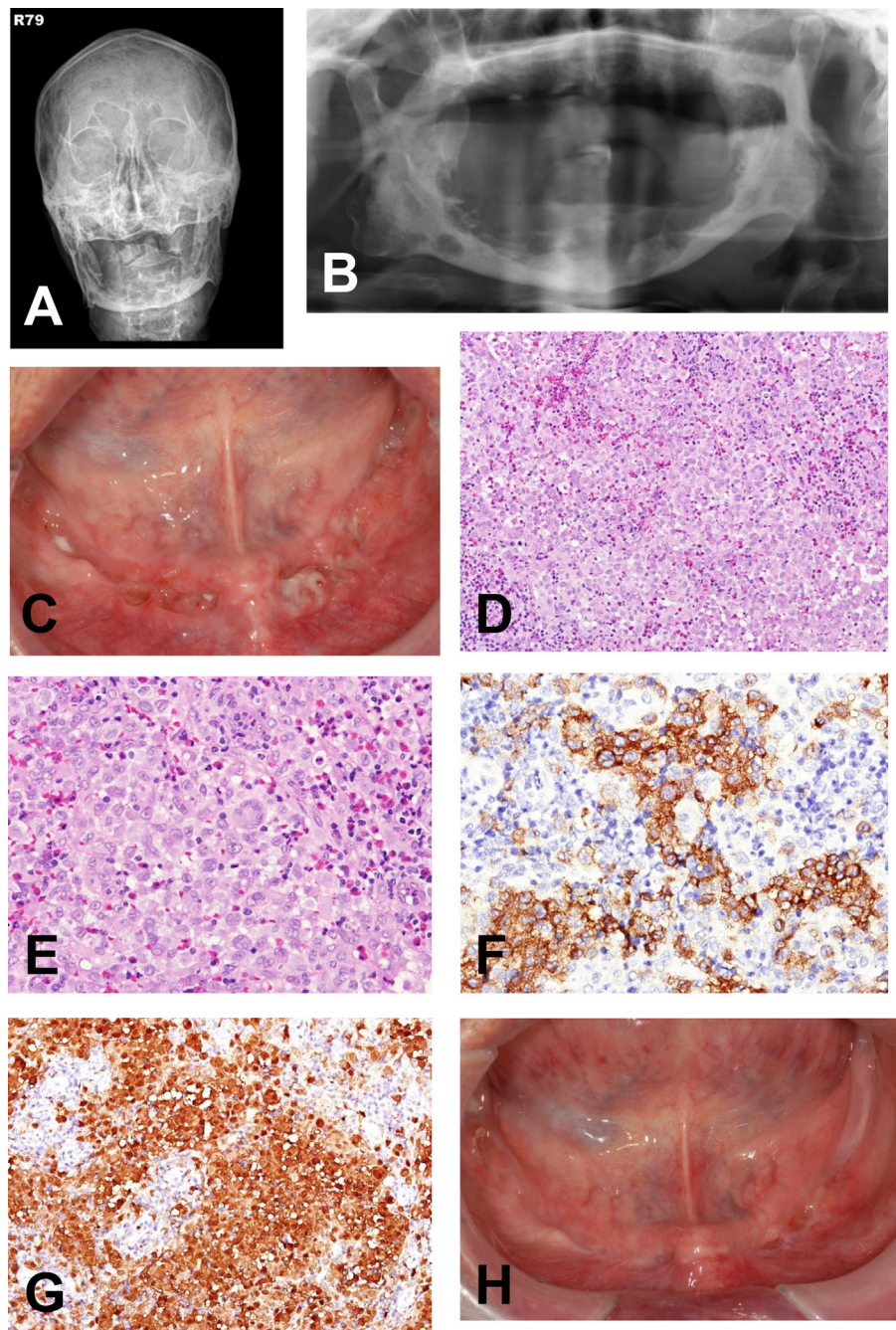


Figure 1 Radiographic, clinical and histological photographs of our case of Langerhans cell histiocytosis. (A and B) Radiographic photographs showing multiple punched-out radiolucent lesions in the mandible and skull bone as well as a scooped-out appearance at the alveolar ridge of the mandible. (C) Clinical photograph demonstrating multiple granulation tissue-like masses on the edentulous mandibular alveolar ridge mucosa. (D and E) Microphotographs exhibiting a sheet of pale-staining histiocyte-like cells intermixed with numerous lymphocytes, plasma cells, and eosinophils as well as occasional multinucleated giant cells. The histiocyte-like cells possessed coffee bean-like or indented vesicular nuclei and pink cytoplasm (Hematoxylin and Eosin stain; original magnification, C, 10 \times and D, 20 \times). (F and G) Immunostained microphotographs revealing that the histiocyte-like cells were positive for CD1a (F, original magnification, 20 \times) and S-100 protein (G, original magnification, 20 \times). (H) Clinical photograph demonstrating a healed edentulous mandibular alveolar ridge mucosa with complete regression of the granulation tissue-like masses.

Declaration of Competing Interest

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

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