Langerhans cell histiocytosis (LCH) of the mandible in an adult: a rare case

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DESCRIPTION
Langerhans cell histiocytosis (LCH) is a clonal proliferation of Langerhans cells (immature dendritic cells) occurring as an isolated lesion or as part of a systemic (multifocal) proliferation. LCH is classified under dendritic cell-related disorders. It can affect bone, skin, lymph nodes, thymus, bone marrow, viscera (spleen and liver), nervous system and the gastrointestinal tract.1 It has been diagnosed in all age groups, but is predominantly seen in children. In adults the incidence is 1–2 cases per million.2

A 29-year-old male patient presented with a history of painless swelling in the lower jaw since 2 months. On palpation a single palpable, non-tender, freely movable right submandibular lymph node was evident. Intraoral examination was not remarkable. Mandibular occlusal radiograph revealed buccal cortical expansion (figure 1). Orthopantamogram revealed multiple, ill-defined radiolucencies in the mandible (figure 2).

Haematological investigations showed a significant increase in eosinophil count—17%. Microscopic examination (H&E), revealed cellular proliferation in the form of sheets, with cells characterised by unique reniform nuclei with pale eosinophilic cytoplasm representing Langerhans cells (figure 3), interspersed with numerous acute and chronic inflammatory infiltrate. Immunohistochemical evaluation of CD1a (figure 4), S-100 were positive. Entire body imaging was performed, which revealed no evidence of lesion in other organs. Based on these findings we came to a diagnosis of LCH affecting the mandible.

Figure 1 Occlusal radiograph shows buccal cortical expansion.

Figure 2 Orthopantamogram shows multiple ill-defined radiolucencies.

Figure 3 Proliferation of neoplastic Langerhans cells (×40 view, H&E).

Figure 4 CD1a was positive (×20 view, immunohistochemistry).
Learning points

▸ Although a rare lesion in an adult, Langerhans cell histiocytosis (LCH) should be considered as a differential diagnosis among the multilocular radiolucencies of the jaw.
▸ A meticulous clinicopathological correlation with immunohistochemistry as an adjunct is the gold standard for diagnosis of LCH.

Competing interests None.

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REFERENCES