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CASE REPORT

Incidental Langerhans cell histiocytosis of the parotid gland resembling marginal zone B-cell lymphoma: a case report

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Langerhans cell histiocytosis (LCH) manifesting as a parotid gland mass is an extremely rare clinical presentation. We report a case of LCH involving the bilateral parotid glands in an 81-year-old Japanese female. Pathologically, the lesion was characterized by numerous lymphoid follicles, dense lymphoplasmacytoid infiltrate and cystic dilatation of the parotid gland duct in addition to nodular and diffuse proliferation of LCs. Moreover, both LCs and small T-lymphocytes invaded the ductal epithelium forming a lymphoepithelial lesion-like morphology. The present case indicates that LCHs should be added to the different diagnosis for marginal zone B-cell lymphoma of mucosa-associated lymphoid tissue type involving the salivary glands.
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Case Report

An 81-year-old Japanese female presented with a 4-year history of bilateral enlarged parotid glands. Under a diagnosis of chronic sialoadenitis, a bilateral parotidectomy was performed in August 2003. There was no other evidence of disease. She received no medication after parotidectomy and is alive and disease-free 3 months postoperatively at the last follow-up.

The resected bilateral parotid glands measured 4 cm in diameter. Macroscopically, the cut surface was homogenous, and demonstrated variable cystic structures. Histologically, low-power field demonstrated that there is an ill-defined nodule measuring 7 mm in the right parotid gland (Fig. 1). In the contralateral parotid gland, there was a diffusely infiltrating lesion containing numerous lymphoid follicles accompanied by a loss of acini and remnants of salivary gland ducts (Fig. 1). Some of the dilated salivary ducts showed a multicystic appearance. The ill-defined nodule contained numerous Langerhans cells (LCs) and mature eosinophils (Fig. 2a). The LC had grooved or contorted nuclei with delicate chromatin, inconspicuous nucleoli and an abundant pale or eosinophilic cytoplasm (Fig. 2a). In the remaining area, the lesion was diffusely infiltrated by numerous small lymphocytes and mature plasma cells, scattered LCs and a few eosinophils. Many small vessels with swollen endothelial nuclei and thickened basement membranes were also present. LCs and small lymphocytes showed prominent invasion of the ductal

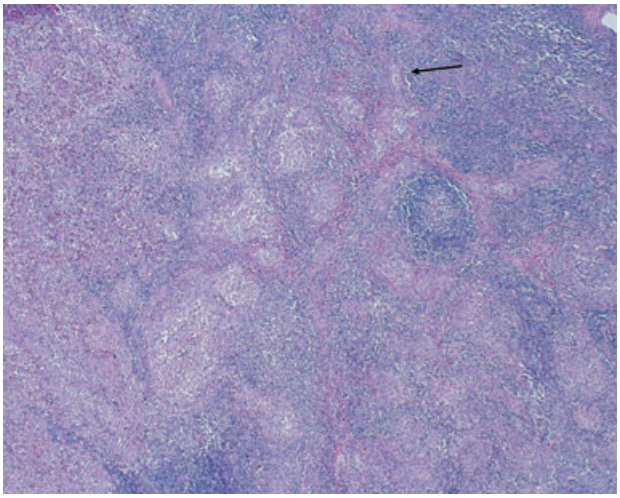

Figure 1 Low-power field of the right parotid gland showed an ill-defined pale staining nodular lesion. Reactive lymphoid follicles and remnant of salivary gland ducts (arrow) were also present. Note the two dilated salivary gland ducts (HEx 12.5).

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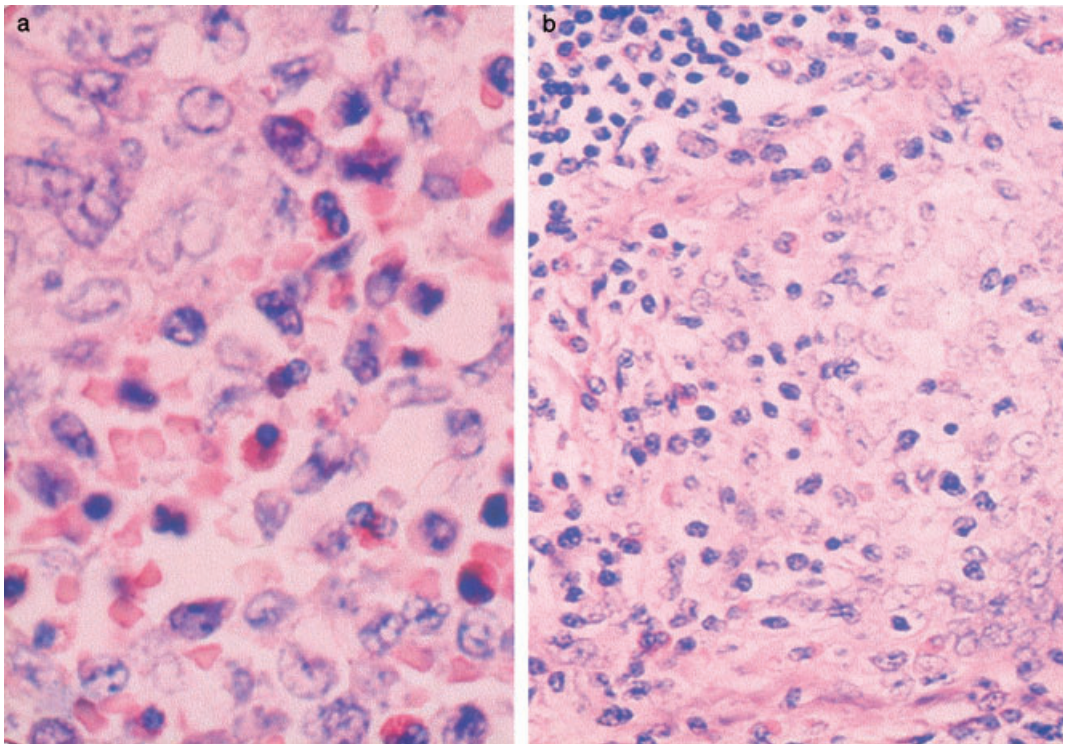

Figure 2 High-power field of Fig. 1. (a) The nodular lesion contained numerous LCs and mature eosinophils (Giemsa ×200). (b) LCs and small lymphocytes showed prominent invasion of the ductal epithelium forming an epimyoepithelial island-like structure. Note the squamous metaplasia of the duct epithelium (Giemsa ×132).

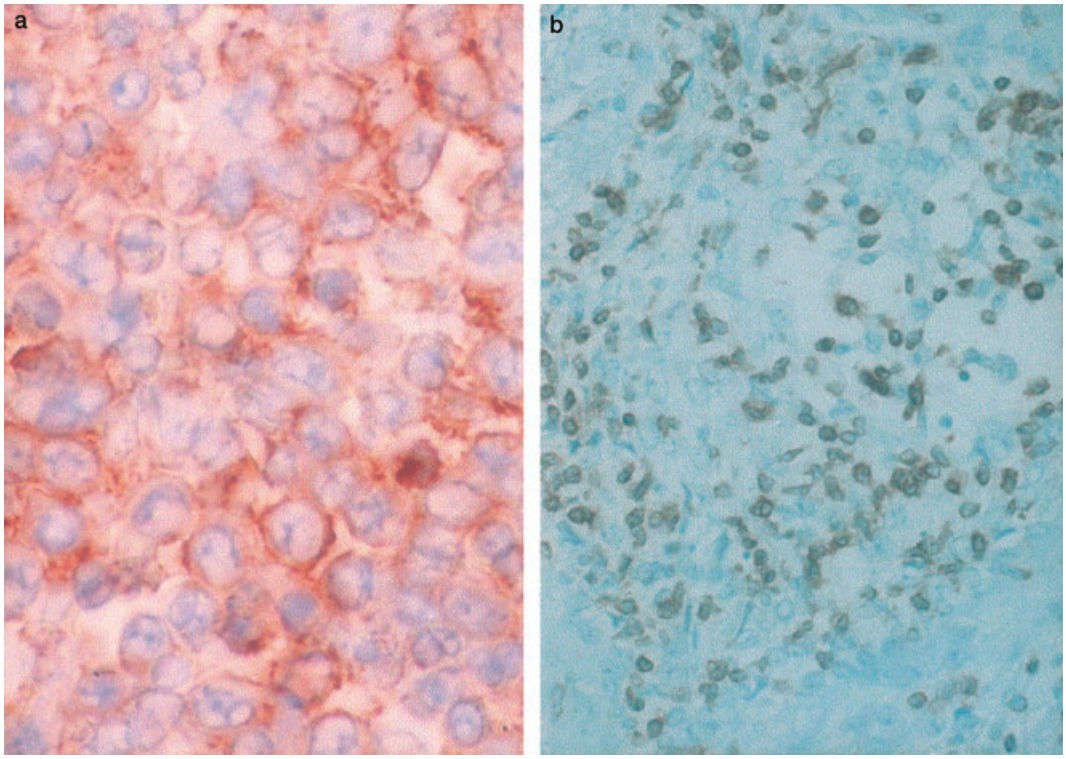

Figure 3 (a) Langerhans cells were strongly expressed CD1a protein (×200). (b) Most of the intraepithelial small lymphocytes were CD3+ (×100).

epithelium forming an epimyoepithelial island-like structure (Fig. 2b). Ductal epithelium invaded by LCs occasionally showed squamous metaplasia (Fig. 2b).

For immunohistochemistry, the sections were stained using a Ventana automated (BenchMark^{TM}) stainer.

Langerhans cells showed strong reactivity to S-100ß-protein and CD1a (Fig. 3a), and a minority of LCs showed weak staining with CD68. The plasma cells were demonstrated to be polytypic with a kappa/lambda, ratio of 1:1. Staining with CD3 and CD20 showed the mixed nature of lymphocytes admixed with LCs. The majority of the small lymphocytes invading the ductal epithelium were CD3+ T cells (Fig. 3b). Polymerase chain reaction analysis demonstrated that there was no clonal rearrangement of the immunoglobulin heavy chain rearrangement was detected (1).

Discussion

Langerhans cell histiocytosis (LCH), formerly known as histiocytosis X (2, 3), rarely involves the salivary gland, even in patients with multifocal disease (2, 4). Moreover, LCH confined to the salivary gland is an extremely rare condition (5).

The present case somewhat resembled primary marginal zone B-cell lymphoma of mucosa-associated lymphoid tissue (MALT) type of the salivary gland (6), i.e. (i) dense lymphoplasmacytoid infiltration accompanied by loss of acini; (ii) numerous reactive lymphoid follicles; (iii) dilatation of the salivary gland ducts lead to a multicystic appearance; and (iv) presence of lymphoepithelial lesion-like structure. However, the majority of the intraepithelial lymphocytes showed T-cell markers. The polytypic nature of B-lymphocytes was demonstrated by immunohistochemistry and polymerase chain reaction. Moreover, the presence of characteristic LCs with S-100 and CD1a expression confirmed the diagnosis of LCH (2).

Interestingly, an association of reactive lymphoid hyperplasia and incidental LCH has been reported in the thyroid gland (chronic thyroiditis) (7) and thymus (myasthenia gravis) (8). The present case indicates that an association of reactive lymphoid hyperplasia and incidental LCH also occurs in the salivary gland. However, the pathogmonic significance of association of reactive lymphoid hyperplasia and incidental LCH remains unclear.

References

1. Wan JH, Trainor KJ, Brisco MJ, Morley AA. Monoclonality in B cell lymphoma detected in paraffin wax embedded sections using the polymerase chain reaction. *J Clin Pathol* 1990; **43**: 888–90.
2. Lieberman PH, Jones CR, Steinman PM et al. Langerhans cell (eosinophilic) granulomatosis. A clinicopathologic study encompassing 50 years. *Am J Surg Pathol* 1996; **20**: 519–32.
3. Weiss LM, Grogan TM, Müller-Hermelink HK et al. Langerhans cell histiocytosis. In: Jaffe ES, Harris NL, Stein H, Vardiman JW, eds. *Pathology and genetics of tumours of haematopoietic and lymphoid tissues*. Lyon: IARC Press, 2001; 280–2.
4. Green I, Beher A, Shanon E, Grosky M. Multifocal extraosseous eosinophilic granuloma of the head and neck. *Arch Otolaryngol Head Neck Surg* 1988; **114**: 561–3.
5. Darvishian F, Hirwat S, Teichberg S, Wolk D, Allen SL, Hajdu SI. Langerhans' cell histiocytosis in the parotid gland. *Ann Clin Lab* 2002; **32**: 201–6.
6. Isaacson PG, Norton AJ. *Extranodal lymphomas*. Edinburgh: Churchill Livingstone, 1994; 67–83.
7. Thompson LDR, Wenig BM, Adair CF, Smith BC, Heffess CS. Langerhans cell histiocytosis of the thyroid: a series of seven cases and review of the literature. *Mod Pathol* 1996; **9**: 145–9.
8. Pescarmona E, Rendina EA, Ricci C, Baroni CD. Histiocytosis X and lymphoid follicular hyperplasia of the thymus in myasthenia gravis. *Histopathology* 1989; **14**: 465–70.