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CASE REPORT

Marginal zone B-cell lymphoma of minor salivary gland representing tumor-forming amyloidosis of the oral cavity. A case report

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We report here a case of mucosa-associated lymphoid tissue (MALT)-type lymphoma arising from the minor salivary gland of the oral cavity exhibiting tumor-forming amyloidosis. The patient was a 64-year-old Japanese woman who presented with 4-year history of a left soft palate mass. Despite multiple and multifocal recurrences including the lip, soft palate, tongue, oral base and vocal code and soft palate, the tumor remained localized in the upper aerodigestive tract, and the patient did not develop multiple myeloma during the course of disease. Histologically, the majority of the lesion was occupied by amyloid deposition. Only the periphery of the lesion contained numerous plasmacytoid cells, along with occasional centrocyte-like cells. In addition, lymphoepithelial lesion and follicular colonization were noted. The present case indicates that primary minor salivary gland MALT-type lymphoma appears to be the cause of tumor-forming amyloidosis of the upper aerodigestive tract including the larynx.

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Keywords: amyloidosis; immunohistochemistry; marginal zone B-cell lymphoma; minor salivary glands; oral cavity

Case report

A 64-year-old Japanese woman presented with a left soft palate mass of several months duration. There was no lymphadenopathy detected on physical examination. In May 1992, a left soft palate tumor (Ø = 2 cm) was resected and a diagnosis of amyloid tumor was made. There was no paraproteinemia noted pre-operatively. Staging workup did not detect any other evidence of disease. The patient was followed without treatment. In February 1996, tumor on the left tongue edge was resected and diagnosed as amyloid deposition. In May 1999, a left soft palate tumor (Ø = 2 cm) was resected. Multifocal recurrence in the aerodigestive tract including the lip, soft palate oral base and vocal code was noted in May 2003, and the vocal cord tumor was biopsied in December 2003. Although, bone marrow examination was not performed during the course of disease, there were no significant of progression into multiple myeloma. She is alive with disease in May 2005.

Macroscopically, the lesion was well circumscribed and a firm mass, and cut surface showing a gray appearance suggested of fibrous connective tissue.

Microscopically, the initial resected specimen and third relapsed specimen showed similar histopathological findings. At low magnification, the majority of the

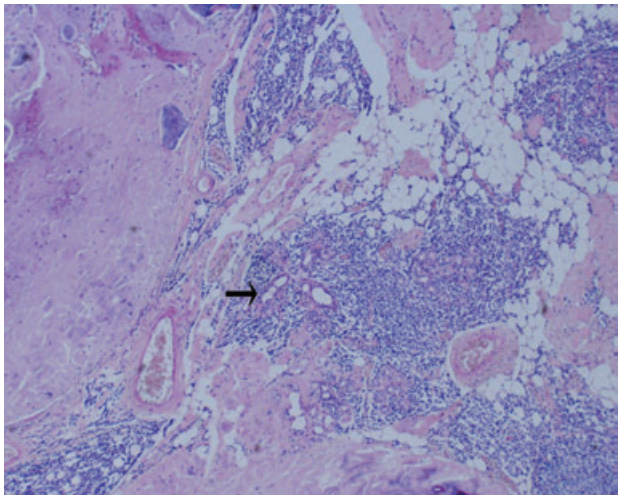

Figure 1 At low magnification, the central portion of the nodule is occupied by the amyloid deposition. The dense lymphoid infiltrate was noted at the periphery of the nodule. Note the residual salivary gland ducts (arrow; Giemsa ×10).

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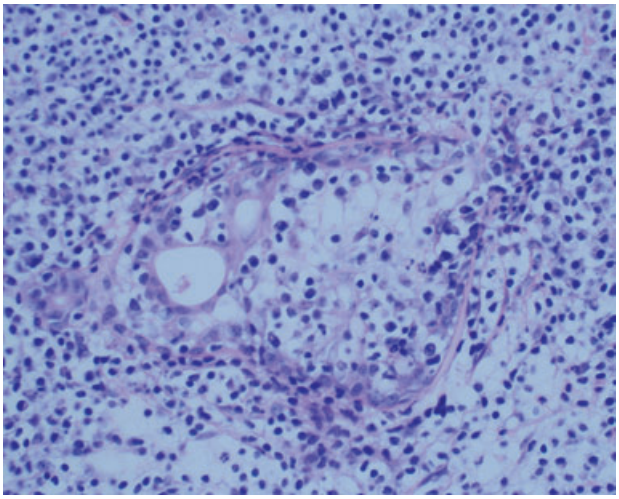

Figure 2 High-power field at the periphery of the nodule. Note numerous plasmacytoid cells and plasma cells and centrocyte-like cells with indented or round nuclei and scant cytoplasm (Giemsa ×250).

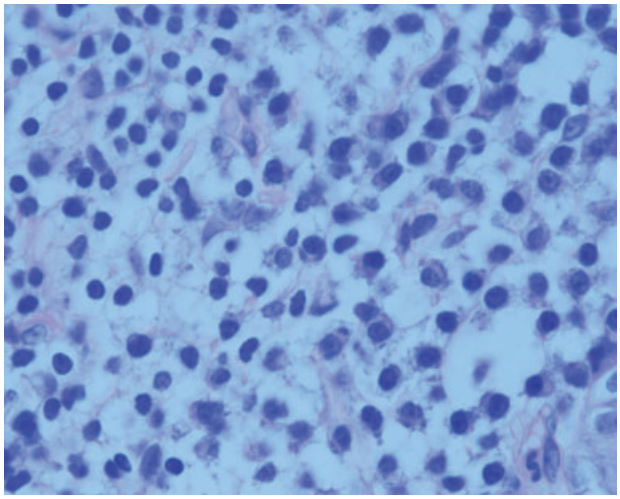

Figure 3 Medium-power field at the periphery of the lesion. There was an evidence of destructive lymphoepithelial lesions (Giemsa ×100).

nodule was occupied by amorphous eosinophilic materials, and dense lymphoid infiltrate was observed at the periphery of the lesion (Fig. 1). The materials were positive for Congo Red stain with associated 'apple-green' birefringence under the polarized light. A few foreign body giant cells were observed around the amyloid deposit. The periphery of the lesion contained numerous plasmacytoid cells and plasma cells, and occasional centrocyte-like (CCL) cells with indented or round nuclei (Fig. 2). The plasmacytoid cells, plasma cells and CCL cells invaded the residual salivary gland duct resulting in a lymphoepithelial lesion (Fig. 3). However, there were no lymphoid follicles detected.

Immunohistochemistry was performed on the paraffin sections using a Ventana automated stainer (BenchMark^{TM}: Tucson, AZ, USA) according to the manufacturer's instructions. The plasma cells and plasmacytoid cells were stained positive for intracytoplasmic immunoglobulin (Ig)G/κ (Fig. 4). The CCL cells were CD20+, CD79a+, Bcl-2+, surface IgD–

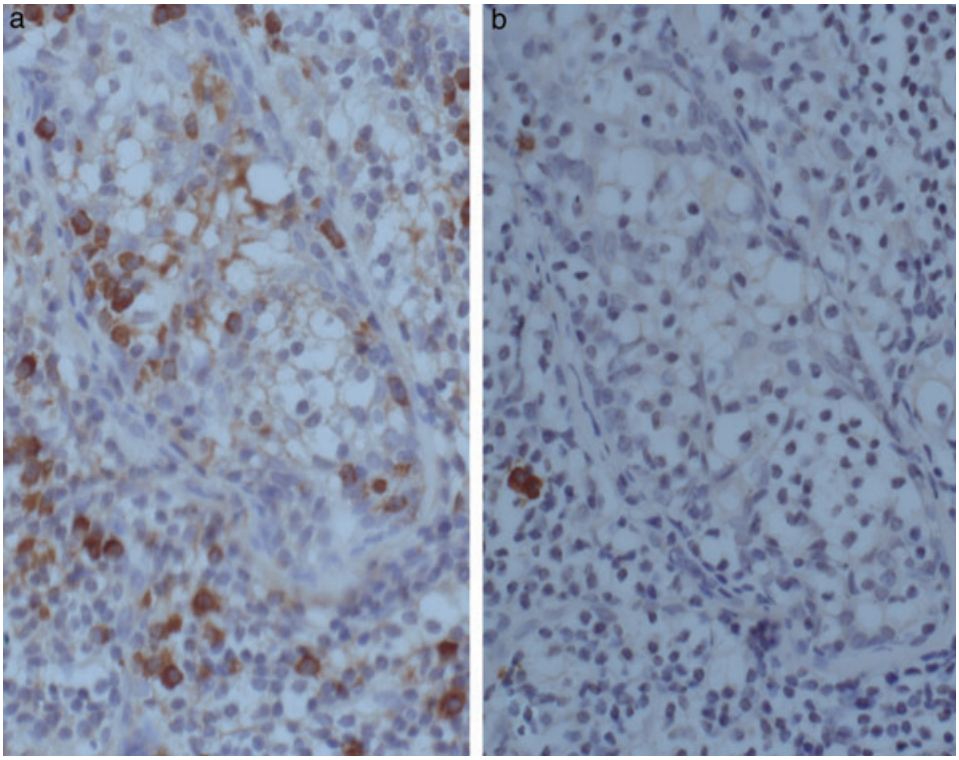

Figure 4 The plasma cells and plasmacytoid cells were intracytoplasmic κ light chain-positive (a), lambda chain-negative (b; ×100).

and IgM−, CD3−, CD5−, CD23−, CD43−, CD45RO−, CyclinD1−. CD23 immunostaining highlighted the residual follicular dendritic cell network colonized by the tumor cells.

The amyloid reacted positivity with antiamyloid P component antibody and IgG/κ, but was negative for antiamyloid A component antibody.

The small biopsied specimens from the tongue (second relapse) and vocal cord (third relapse) contained only amyloid deposition.

Discussion

Recently, marginal zone B-cell lymphoma of mucosa-associated lymphoid tissue (MALT)-type has been reported as a cause of localized amyloidosis (1–3). However, little is known about an association of primary salivary gland MALT-type lymphoma and localized amyloidosis (1–3). An association between amyloidosis and MALT-type lymphoma can have diagnostic relevance as the amyloid can displace lymphomatous proliferation, particularly in a small biopsy specimen (2). In the present case, CCL cells were difficult to detect due to prominent infiltration of the plasma cells and plasmacytoid cells. However, a few lymphoepithelial lesions were detected in the routinely stained section. Moreover, immunohistochemical study demonstrated characteristic pathological findings of MALT-type lymphoma including monotypic intracytoplasmic immunoglobulin of the plasma cells and their precursors as well as follicular colonization (2).

Interestingly, in this case, the tumor relapsed at the vocal cord 15 years after disease onset. Among upper aerodigestive tract, the larynx is the most common site for localized amyloid deposit (4, 5). Laryngeal amyloidosis is characterized by monoclonal light-chain deposition (4). Recurrent respiratory tract disease is not uncommon, but the usual clinical course is relatively indolent (4, 5). Thompson et al. examined 11 cases of laryngeal amyloidosis (5). They found an associated monoclonal lymphoplasmacytic infiltrate and recurrent/multifocal disease in the respiratory or gastrointestinal tract in a few cases, and there was an absence of systemic plasma cell dyscrasia or overt systemic B-cell lymphoma. They suggested that a portion of the laryngeal amyloid might be the result of MALT-type lymphoma (5). The present case clearly indicated that MALT-type lymphoma appears to be the cause of laryngeal amyloidosis representing a recurrent/multifocal disease.

References

1. Caulet S, Robert I, Bardaxoglou E, et al. Malignant lymphoma of mucosa associated lymphoid tissue: a new etiology of amyloidosis. *Pathol Res Pract* 1995; **191**: 1203–7.
2. Goteri G, Ranaldi R, Pileri SA, Bearzi I. Localized amyloidosis and gastrointestinal lymphoma: a rare association. *Histopathology* 1998; **32**: 348–55.
3. Lim JK, Lacy MQ, Kurtin PJ, Kyele RA, Gertz MA. Pulmonary marginal zone lymphoma of MALT type as a cause of localized pulmonary amyloidosis. *J Clin Pathol* 2001; **54**: 642–6.
4. Lewis JE, Olsen KD, Kurtin PJ, Kyle RA. Laryngeal amyloidosis: a clinicopathologic and immunohistochemical review. *Otolaryngol Head Neck Surg* 1992; **106**: 372–7.
5. Thompson LDR, Derringer GA, Wenig BM. Amyloidosis of the larynx. A clinicopathologic study of 11 cases. *Mod Pathol* 2000; **13**: 528–35.