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## Case Report

# Mantle cell lymphoma presenting as a symmetrically hard palatal swelling: A case report and review of the literature



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## ABSTRACT

Mantle cell lymphoma (MCL) is a rare B-cell neoplasm accounting for approximately 3–10% of all non-Hodgkin lymphomas (NHLs). Initial clinical manifestation in the oral cavity is extremely rare. We report a case of MCL involving the hard palate. The patient, a 92-year-old man, presented at the department of oral surgery of our hospital because of swelling of the palate, causing instability of his maxillary denture. On examination, the hard palate was symmetrically swollen, and computed tomography revealed an extensive soft tissue mass with osteolysis. Biopsy of the mass disclosed monotonous diffuse infiltrates of small to intermediate-sized lymphoid cells underneath the squamous epithelium. Immunohistochemistry revealed that the cells were positive for CD5, CD20, CD79a, bcl-2, cyclin D1, ki-67 (MIB-1 index, 68.5%) and (weakly or strongly) CD43, and negative for CD3, CD10, CD21, CD23, bcl-6, TdT and EBV-latent infection membrane protein-1. *In situ* hybridization also showed negativity for EBV-encoded small RNA. These findings were histologically compatible with MCL. To our knowledge, this is the first reported case of MCL initially manifesting as a symmetrical mass on the hard palate with osteolysis in such an elderly patient.

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## 1. Introduction

Mantle cell lymphoma (MCL) is a lymphoid malignancy of B-cells of the mantle zone of the primary lymphoid follicle [1]. MCL accounts for approximately 3–10% of all NHLs, and affected patients are typically older male adults usually presenting with stage III or IV disease [1,2]. Lymphadenopathy is the first clinical sign in approximately 75% of patients, while extranodal sites are initially affected in the remaining 25% [2]. The commonest extranodal

sites of involvement are the gastrointestinal tract, Waldeyer's ring, spleen, bone marrow and peripheral blood [1]. Few cases of MCL arising within the oral cavity have been reported. In the nine reports published to date, involvement of the oral cavity was rare, and only 15 such cases were described [3–11] (see Table 1).

In fact, little is known about the clinicopathological features of extranodal MCL at oral sites. In a literature review, we were able to identify only two cases of MCL exhibiting symmetrical involvement of the hard palate [8,9], and none of them were associated with osteolysis. Here we report a rare case of MCL manifesting as a symmetrical swelling of the hard palate osteolysis in a very elderly Japanese patient.

## 2. Materials and methods

### 2.1. Case report

A 92-year-old Japanese man was referred to the department of oral and maxillofacial surgery of our hospital because of a painless swelling in the hard palate. He had first become aware of swelling

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**Table 1**  
Summary of clinicopathological findings of oral mantle cell lymphomas.

| Site             | Number of cases | Age [average] | Sex (M/F) | Initial symptom  | Size (mm)  | Osteolysis | Stage                                   | Morphology                               | Cyclin D1       | EBV             | Genetics        | Treatment   | Outcome months [average]                           | Author [ref.]  |
|------------------|-----------------|---------------|-----------|--|--|------------|---|--|-----------------|-----------------|-----------------|---|--|--|
| Palate           | (1)             | 92 [92]       | (1/0)     | Swelling with denture irritation                                     | N/A  | +          | IIIA (Finally)                          | Classical                                | +               | -               | -               | Pred  | AWD 10 [10]  | Our case   |
| Palate           | (9)             | 41–87 [66.6]  | (7/2)     | Swelling (6) with bilateral (2) or denture irritation (2)<br>N/A (3) | 80 × 30 × 30 (1)<br>50 × 30 × 20 (1)<br>40 × 40 × 30 (1)<br>30 × 15 (1)<br>N/A (5) | N/A (9)    | IAE (2)<br>III (1)<br>IV (1)<br>N/A (5) | Classical (5)<br>Blastoid (1)<br>N/A (3) | +(6)<br>N/A (3) | N/A (9)         | +(2)<br>N/A (7) | Chem (3)<br>Chem/Rad (1)<br>Chem/Pred (2)<br>Rad (2)<br>N (1) | AWD (5)<br>16–48 [29.3]<br>DOD (4)<br>10–48 [35.2] | Chang et al. [3], Tan et al. [4], Guggisberg and Jordan [7], Kyo et al. [8], Scheller et al. [9], Milgrom and Yahalom [11] |
| Tongue           | (4)             | 62–74 [66.7]  | (2/2)     | Swelling (3), dysphagia (1)  | 40 (1)<br>N/A (3)  | N/A (4)    | IA (1)<br>N/A (3)                       | Classical (3)<br>Blastoid (1)            | +(3)<br>N/A (1) | -(1)<br>N/A (3) | +(2)<br>N/A (2) | Chem/Rad (2)<br>Chem/Pred (1)<br>N/A (1)                      | AWD (1)<br>54 [54]<br>DOD (3)<br>1.2–48 [22.4]     | Guastafierro et al. [6], Guggisberg and Jordan [7]   |
| Floor of mouth   | (1)             | 72 [72]       | (0/1)     | Swelling in floor of mouth   | 15   | N/A        | IVA                                     | Classical                                | +               | N/A             | N/A             | Sur   | AWD 12 [12]  | Rockacy et al. [5], Guggisberg and Jordan [7]  |
| Maxillary mucosa | (1)             | 55 [55]       | (1/0)     | N/A  | N/A  | N/A        | I                                       | N/A                                      | +               | N/A             | N/A             | N/A   | AWD N/A [N/A]                                      | Triantafillidou et al. [10]  |

M, male; F, female; ( ), (total number); N/A, data not available; Classical, classical MCL; Blastoid, blastoid variant of MCL; EBV, Epstein-Barr virus; Genetics, t(11;14)(q13;q32); Pred, prednisolone; Chem, chemotherapy, Rad, Radiotherapy; N, none-treatment; Sur, surgical resection; AWD, alive without disease; DOD, died of disease; +, positive; -, negative.

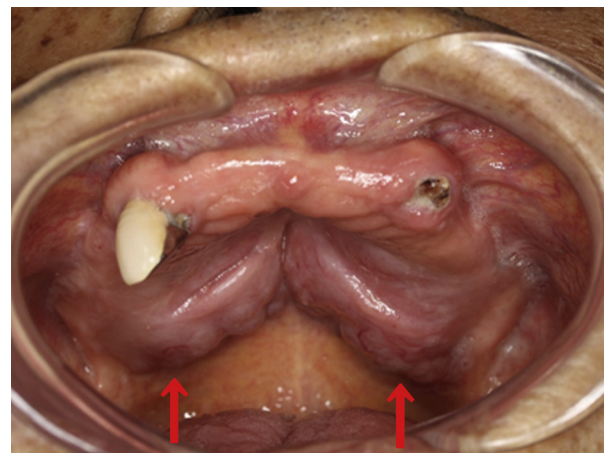
6 months previously, as it had caused instability of his maxillary denture, and subsequently showed slow growth thereafter.

On examination, the hard palate showed a normal coloration and no ulceration, but was symmetrically swollen (Fig. 1). Computed tomography (CT) revealed a palatine tumor (Fig. 2A and B) and osteolysis with a “moth-eaten” appearance (Fig. 2B). Although cervical lymphadenopathy was noted (Fig. 3), generalized lymphadenopathy was not evident. The results of blood tests were all within the normal ranges, except for the level of soluble interleukin-2 (sIL-2) receptor (1559 U/ml). Both the patient and his family had an unremarkable medical history. The clinical diagnosis was a suspected lymphoma in view of the markedly increased level of the sIL-2 receptor, and biopsy of the palatine tumor and a cervical lymph node was performed. The biopsy sample from the palatine tumor disclosed diffuse infiltrates of small to intermediate-sized lymphoid cells underneath the oral mucosal epithelium (Fig. 4A–C). Immunohistochemistry revealed that the lymphoid cells were positive for CD5 (Fig. 5A), CD20 (Fig. 5B), CD79a, bcl-2, cyclin D1 (Fig. 5C) and CD43 (Fig. 5D: weakly or strongly positive), ki-67 (MIB-1 index, 68.5%), and were negative for CD3, CD10, CD21, CD23, bcl-6, TdT and EBV-latent infection membrane protein-1. *In situ* hybridization also showed negativity for EBV-encoded small RNA. The lymph node biopsy sample revealed similar findings (data not shown). On the basis of these data, MCL of the hard palate seemed the most likely diagnosis. The patient was transferred to the hematology department of a local medical center, where the diagnosis of MCL was confirmed by additional examination (not including chromosomal analysis). In view of the patient’s advanced age, chemotherapy was not administered, and he received only prednisolone. Thereafter, the size of the tumor mass decreased markedly, and the patient had a good clinical course. However, at the family’s request, any further treatment was discontinued due to progression of the patient’s pre-existing dementia.

The case study protocol was reviewed and approved by the Research Ethics Committee of Meikai University School of Dentistry (A0832).

2.2. Immunohistochemistry

Deparaffinized sections were immersed in absolute methanol containing 0.3% H<sub>2</sub>O<sub>2</sub> for 15 min at room temperature to block endogenous peroxidase activity. After washing, the sections were immersed in 0.01M citrate buffer, pH 6.0, and heated in a microwave oven for 5 min at high voltage and then for 10 min at



**Fig. 1.** The hard palate lesion. The hard palate was showing bilateral symmetrically swollen non-ulcerative palatal mucosa (red arrows). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

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