

## CASE REPORT

# Ill-fitting dentures as primary presentation of mantle cell lymphoma: A case report and literature review of the primary mantle cell lymphomas of the hard palate

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**ABSTRACT**

Mantle cell lymphoma (MCL) is a subtype of B-cell non-Hodgkin's lymphoma seen predominantly in males. Common extra-nodal sites of involvement of MCL are Waldeyer's ring, gastrointestinal tract, bone marrow and peripheral blood. The extra-nodal palatal localization of MCL is quite uncommon. MCL is seen in predominantly older patients, therefore undiagnosed MCL patients are likely to have total prosthesis. In this study, a case of MCL, initially presenting as palatal swelling was reported with relevant literature review and the possible role of dental professionals in the diagnosis of this rare entity was discussed.

**Key words:** Hard palate, mantle cell lymphoma, non-Hodgkin lymphoma, oral lymphoma

**INTRODUCTION**

Mantle cell lymphoma (MCL) is a B-cell malignant lymphoid tumor which is defined as a separate entity and included in lymphoma classification by World Health Organization (WHO).<sup>[1]</sup> MCL includes small-medium sized lymphoid cells and accounts for 6–10% of all B-cell lymphomas.<sup>[2,3]</sup>

MCL patients are predominantly older males and usually present as stage IV (Ann Arbor) disease. The prognosis of MCL is reported to be poor with a mean survival of 3 years.<sup>[1,4,5]</sup>

In most of the cases the primary presentation site is the lymph nodes. However, extra-nodal sites of involvement such as oral cavity, gastrointestinal tract, Waldeyer's ring, peripheral blood are well documented.<sup>[6]</sup> Primary hard palate involvement of MCL is reported several times.<sup>[7-16]</sup> Palatal MCL is mostly seen in elderly people and may be masked with the presence of prosthesis. In this study, a case of palatal MCL is presented with treatment outcome and a literature analysis was performed to analyze the importance of dental examination in the primary diagnosis of the disease.

**CASE REPORT**

A 71-year-old male patient was referred to Eskişehir Osmangazi University, Faculty of Dentistry, Department of Oral and Maxillofacial Surgery with a complaint of ill-fitting dentures. An intra-oral examination was performed and symmetric mucosal enlargements were observed on hard palate [Figure 1]. The mucosal swellings were asymptomatic. There were no pathologic findings on radiological examination. Patient was uncomfortable about the mobility of his dentures and had tried to fix them by sticking dish rags or other foreign materials into the prosthesis. Medical history was insignificant. Therefore, a provisional diagnosis of reactive mucosal lesion was made and incisional biopsy was performed under local anesthesia.

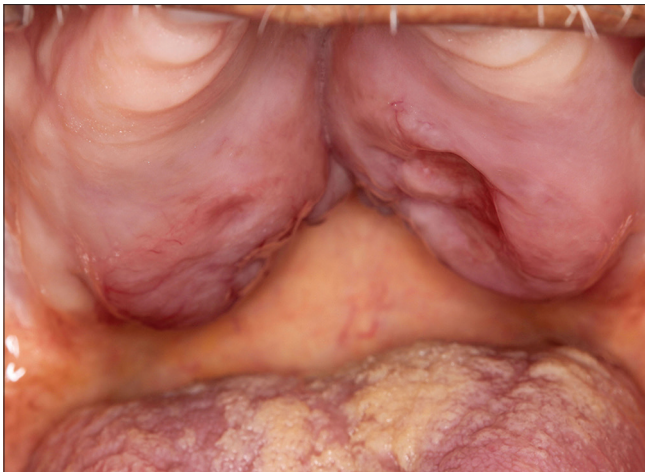
Microscopic examination revealed diffuse abundant small cells with hyperchromatic nuclei in the subepithelial region [Figure 2]. Mucosal ulceration was evident. Mild nuclear enlargement and pleomorphism were observed in

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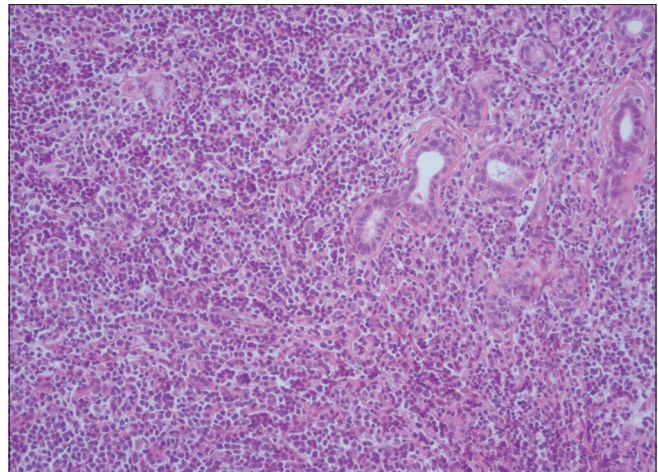
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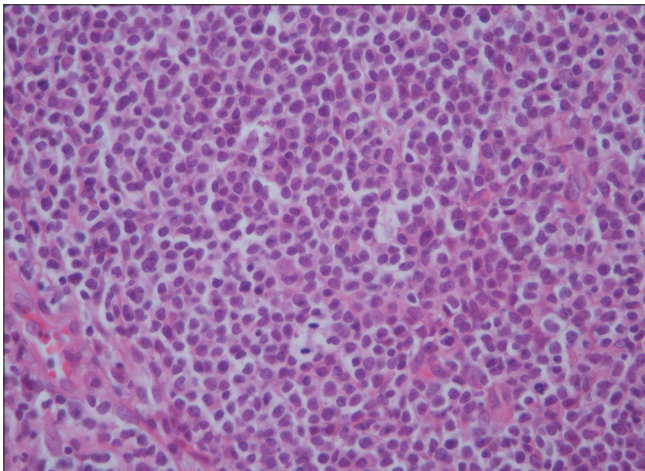
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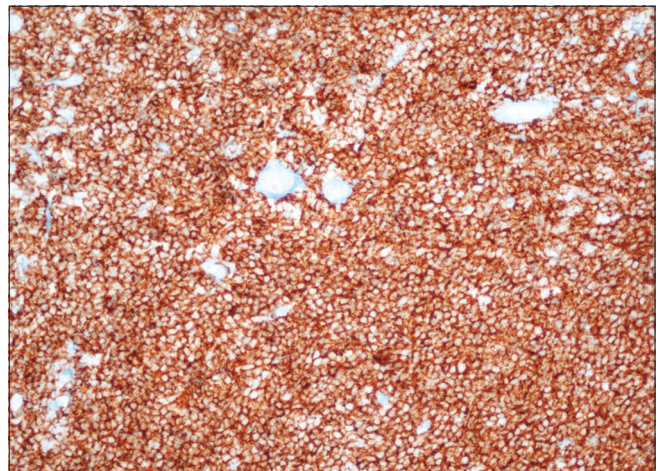
**Figure 1:** Ulcerated palatal symmetric mucosal swelling responsible for the mobility of maxillary dentures



**Figure 2:** Diffuse small cell lymphocytic infiltration is the predominant component on histology. Glandular structures can be seen at the upper-right quadrant (H&E stain,  $\times 100$ )



**Figure 3:** Nuclei of small cells have distinct hyperchromatism. Mild nuclear pleomorphism is also evident (H&E stain,  $\times 400$ )



**Figure 4:** Tumoral cells stained diffusely positive with CD20 (IHC stain,  $\times 200$ )

small cells [Figure 3]. Immunohistochemistry was performed for differential diagnosis.

After the immunohistochemistry, tumoral cells showed strong positive staining with CD20 (L26, Ventana, Tucson) [Figure 4], pax-5 (SP34, Ventana, Tucson), CD5 (SP19, Ventana, Tucson), cyclin D1 (SP4-R, Ventana, Tucson) [Figure 5], bcl-2 (124, Ventana, Tucson) and IgM (poliklnal, Ventana, Tucson). Tumoral cells were partially stained with CD43 (L60, Ventana, Tucson). CD21 (SP104, Ventana, Tucson) and CD23 (SP23, Ventana, Tucson) immunostaining showed follicular dendritic cells. CD10 (SP67, Ventana, Tucson) and IgD (policlonal, Ventana, Tucson) were negative. Ki-67 proliferation index was 50%. The case was diagnosed as MCL with these findings. Patient was referred to Hematology Department of Eskişehir Osmangazi University, Faculty of Medicine for further evaluation and treatment.

Ultrasonography of abdomen revealed multiple lymphadenopathies at mesenteric and para-aortic regions. After informed consent was taken, patient underwent

cyclophosphamide, oncovin, prednol (COP) chemotherapy regimen. However, remission was not observed and the patient died after four chemotherapy cycles.

## MATERIALS AND METHODS

English dental and medical literature search was conducted using the combination of terms such as hard palate, MCL, lymphoma and extra-nodal lymphoma in PubMed. Cases which were reported under the strict diagnosis of MCL and primarily located on the hard palate were extracted and data regarding treatment, follow-up and demographics was reviewed. Cases with secondary involvement were excluded from the study.

## RESULTS

Nine publications with 14 cases defining primary hard palate MCLs published between 1990 and 2014 were identified. The mean age of earlier published cases was 67.1 years. Data



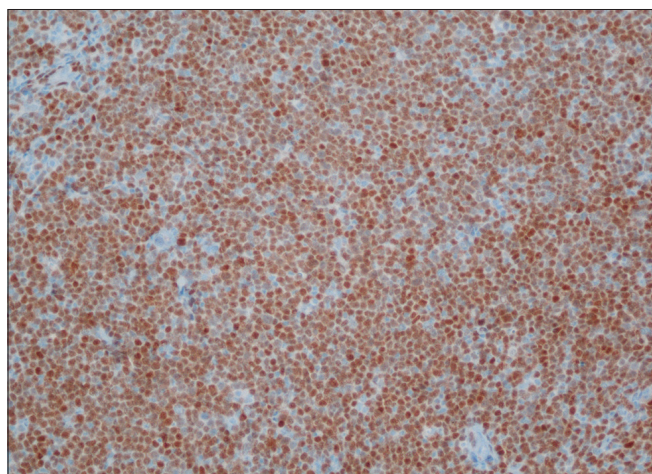
regarding demographics and treatment of published cases are shown in Table 1.

## DISCUSSION

MCL is a counterpart of non-Hodgkin lymphoma family and was first included in lymphoma classification in 2001.<sup>[1]</sup> It originates from peripheral B-cells of mantle zone.<sup>[10]</sup> MCL is relatively uncommon and occurs in middle aged adults with a male predilection.<sup>[1,2,10]</sup>

Although MCL is mainly located in the lymph nodes, involvement of extra-nodal sites such as Waldeyer's ring, peripheral blood, bone marrow and gastrointestinal tract are also reported.<sup>[6]</sup>

Definite diagnosis of MCL is achieved with immunohistochemical staining with or without the assistance of molecular techniques. MCL tumoral cells commonly express CD20, CD5, CD43 antigens and BCL2 and cyclin



**Figure 5:** Tumoral cells showed nuclear positivity with cyclin D1 staining (IHC stain, ×200)

D1 protein. However, they are CD10 and BCL6 negative and show negativity/weak positivity for CD23 antigen expression.<sup>[1]</sup> Histologic examination reveals abundant monomorphic small-medium sized lymphoid cells with slightly/markedly irregular nuclear contours.<sup>[10]</sup> The first impression on histologic appearance reminds of small cell lymphoma. Blastoid and pleomorphic subtypes of MCL have been defined by WHO and it is suggested that blastoid or pleomorphic morphology are aggressive variants; similar histology may be seen in some cases at relapse.<sup>[18]</sup>

Hard palate is an unusual site for primary MCL and this has been reported in the English literature several times.<sup>[7,8,10-13,17]</sup> Kyo *et al.*<sup>[7]</sup> reported that long term use of maxillary prosthesis might have provoked the lymphoid tissue accumulation and proliferation leading to a mass of MCL in the submucosal region of the hard palate. Guggisberg and Jordan<sup>[10]</sup> reported that most oral MCLs occur in an elderly male population and have a predilection for the palate. The duration of the prosthesis usage of MCL patients could not be analyzed in the current literature review due to the unavailable data in the case reports. In the current case, patient was using total prosthesis for 10 years and the only complaint of the patient was the mobility of his dentures. The clinical appearance of the tumor suggested that the palatal accumulation of lymphoid mass might have been initiated by the long term use of the total prosthesis.

Other lymphoid tumors may primarily infiltrate hard palate similar to MCL. Milgrom and Yahalom reported nine cases of primary indolent lymphomas of the hard palate including MCL, follicular lymphoma and marginal zone lymphoma.<sup>[11]</sup> There were two cases of MCL in their series and it is suggested that MCL cases showed more aggressive behavior compared to other lymphomas of the hard palate.<sup>[11]</sup>

Polychemotherapy is mainly used to treat aggressive non-Hodgkin lymphomas including

**Table 1: Demographic and follow-up data of previously reported cases**

Authors	Publication year	Number of cases	Age/sex	Treatment	Follow-up (months)
Kyo <i>et al.</i> <sup>[7]</sup>	2010	1	71, male	CT	NAD
Chang <i>et al.</i> <sup>[8]</sup>	2003	1	62, female	CT	6
Aguilera <i>et al.</i> <sup>[13]</sup>	1998	1	55, male	NAD	NAD
Guggisberg and Jordan <sup>[10]</sup>	2010	2	67, male	Treatment declined	1-24
			87, male	CT	2-8
Scheller <i>et al.</i> <sup>[12]</sup>	2011	1	41, male	RT + CT, COD	48
Milgrom and Yahalom <sup>[11]</sup>	2013	2	75, female	RT + CT, DOD	41
			56, male	RT, COD	16
Kolokotronis <i>et al.</i> <sup>[17]</sup>	2005	1	84, male	CT, DOD	17
Fitzpatrick <i>et al.</i> <sup>[15]</sup>	2012	4	57, male	CT, COD	NDD
			67, female	RT, COD	NDD
			93, male	NAD	NAD
			63, female	CT, COD	NDD
Costa <i>et al.</i> <sup>[16]</sup>	2014	1	61, male	CT, DOD	NDD

RT: Radiotherapy, CT: Chemotherapy, NAD: Nonavailable data, COD: Clear of disease, DOD: Died of disease, NDD: Nondefinite data

MCL. (CHOP; cyclophosphamide, doxorubicin, vincristine, prednisolone<sup>[19]</sup>) Chang *et al.*<sup>[8]</sup> reported a polychemotherapy regimen consisting of Cytosan, doxorubicin, vincristine and prednisone, in conjunction with rituximab administered every 3 weeks. Different chemotherapeutics achieve approximately high treatment response rate of 70%.<sup>[20-22]</sup> Meusers *et al.*<sup>[23]</sup> reported that the CHOP regimen did not show superiority to COP regimen in the treatment of MCL. Although CHOP-like anticycline containing chemotherapeutic combinations do not have distinct advantage on survival, they are currently chosen for standard therapeutic approach.<sup>[3]</sup> Radiation therapy is still a therapeutic option in low-stage disease (Ann-Arbor stage 1,2) In advance stage disease, the combination of radiotherapy and chemotherapy can be used, however, the efficacy of this combination have not been proven yet.

## CONCLUSION

Primary MCL of the hard palate is a rare entity which is mostly seen in elder people. In this study, a case of palatal primary MCL which presents with ill-fitting total prosthesis is reported with clinical and histopathological features. Extensive literature review revealed 14 previously reported cases. Dental total prosthesis may mask the existing lesion on the hard palate, therefore, dental practitioner should be alert at the time of the clinical examination of patients with total or partial prosthesis.

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## Conflicts of interest

There are no conflicts of interest.

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