An unusual localization: Lymphoma of the tongue

Nadir bir lokalizasyon: Dilde lenfoma

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ABSTRACT

Non-Hodgkin's lymphomas may affect extranodal areas but oral cavity is a rare localization. In this article, we reported a 53-year-old male patient who applied with complaint of dysphagia and pain on tongue. A biopsy result showed that the mass on the tongue was an anaplastic lymphoma. Lymphomas should be kept in mind for the differential diagnosis of the masses of the tongue. Diagnosis should be established and treatment should be started rapidly due to aggressive behavior and poor prognosis of the disease.

Keywords: Anaplastic; large-cell; lymphoma; tongue.

ÖZ

Non-Hodgkin lenfomalar ekstranodal alanları etkileyebilir, fakat oral kavite nadir bir lokalizasyondur. Bu yazıda, disfaji ve dilde ağrı yakınması ile başvuran 53 yaşında bir erkek hasta sunuldu. Biyopsi sonucu dildeki kitlenin anaplastik lenfoma olduğunu gösterdi. Dildeki kitlelerin ayırıcı tanısında lenfomalar akılda tutulmalıdır. Hastalığın agresif davranışı ve kötü prognozu nedeniyle hızlı bir şekilde tanı konulmalı ve tedaviye başlanmalıdır.

Anahtar sözcükler: Anaplastik; büyük hücreli; lenfoma; dil.

Lymphomas are solid malignant tumors with variable clinical and pathological features. Basically, there are two subtypes of lymphomas: Hodgkin's lymphoma (HL) and non-Hodgkin's lymphoma (NHL). Despite HLs (1%), NHLs frequently affect extranodal areas (20-30%).^[1] Waldeyer's ring is the most affected extranodal area at the head and neck region, which was affected in 50% of patients with head and neck lymphoma.^[2] The tongue is a very rare localization for the lymphoma. In this article, we report a case of anaplastic large-cell lymphoma of the tongue.

CASE REPORT

A 53-year-old male patient presented with a history of dysphagia, speaking disorder, and pain at the right side of the tongue for one month. There was no weight loss, fever or night sweats. He was treated for B-cell chronic lymphocytic leukemia in 2006 and

classic HL in 2013. He was in remission. During physical examination, an asymmetry was seen at anterior right side of the tongue. With palpation; a solid, hard, and large submucosal mass was detected. There was no significant pain, necrosis, or ulceration (Figure 1). The movements of the tongue were normal. No pathologic condition was detected on the examination of the buccal mucosa, gingiva, oropharynx, tonsils, and head and neck region. Nasal and laryngeal endoscopies were normal. A written informed consent was obtained from the patient.

Magnetic resonance imaging study revealed a mass $(2.5\times3\times4$ in size) at the anterior part of the tongue with low signal intensity on T_1 weighted images and high signal intensity on T_2 -weighted images. Lesion was homogeneous on contrast enhancement (Figure 2). There were no pathological cervical lymph nodes on both sides of the neck.



Figure 1. Solid, hard, large submucosal mass.

Fluorine-18 fluorodeoxyglucose positron emission tomography/computed tomography (PET-CT) scan revealed a hypermetabolic mass which suggested malignancy at the anterior part of the tongue and which was not seen on previous PET-CT scans of the patient (Figure 3).

Incisional biopsy was performed under local anesthesia. On histopathological examination with hematoxylin and eosin staining, diffuse infiltrate consisting of large pleomorphic cells and plenty of apoptotic cells was detected (Figure 4). Immunohistochemical study findings were consistent



Figure 3. Positron emission tomography/computed tomography scan revealed a hypermetabolic mass.

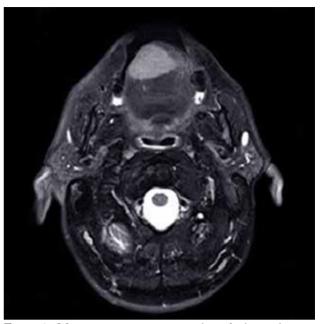


Figure 2. Magnetic resonance imaging showed a lesion that was homogeneous on contrast enhancement.

with anaplastic large-cell lymphoma. After biopsy, a large ulcer occurred on patient's tongue, which was reduced with chemotherapy. Patient died 16 months after the treatment because of distant metastasis and complications of the disease.

DISCUSSION

Lymphoma is the third most frequent neoplasia on a worldwide scale and 3% of all malignant tumors are lymphomas.^[3] Hodgkin's lymphomas typically do not involve extranodal areas. However, NHLs may originate from extranodal sites (20-30%).^[1] Lymphoma

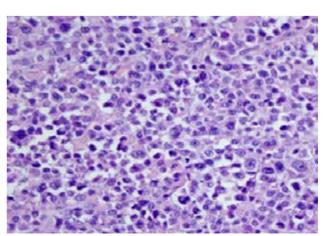


Figure 4. Large pleomorphic cells and plenty of apoptotic cells (H-E×400).

30 KBB Uygulamaları

of oral cavity is rare and constitutes only 2% of all extranodal lymphomas.[4] Sirsath et al.[5] reported only seven cases of primary extranodal lymphoma of oral cavity over a 10-year period. Tongue was affected in three of seven patients. It generally affects elderly patients, particularly those in fourth or fifth decade of life and male gender is affected more than females. [5,6] The symptoms of disease vary according to the site of lesion. There are no characteristic clinical features of NHL of the oral cavity. It presents with a soft, elastic, asymptomatic lesion. Local swelling, pain or discomfort may be seen at the affected area. Involvement of the intrinsic tongue muscles may cause dysphagia, dysarthria and movement failure. Large lesions may cause dyspnea and bleeding.^[7] Magnetic resonance imaging shows the features and borders of the lesion. Diagnosis can be confirmed with fine-needle aspiration biopsy or incisional biopsy. Immunohistochemical studies can be performed in all cases for subtyping of the tumors. Squamous cell carcinomas, melanomas, metastatic tumors, rhabdomyosarcomas, and adenocarcinomas should be considered for differential diagnosis. [8] Treatment of extranodal lymphomas is chemotherapy firstly and then radiotherapy. Treatment protocols change according to subtype of the lymphoma. The prognosis of the diseases depends on the type of malignancy, stage of the tumor, and response to treatment.^[5]

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REFERENCES

- Hanna E, Wanamaker J, Adelstein D, Tubbs R, Lavertu P. Extranodal lymphomas of the head and neck. A 20-year experience. Arch Otolaryngol Head Neck Surg 1997;123:1318-23.
- Ezzat AA, Ibrahim EM, El Weshi AN, Khafaga YM, AlJurf M, Martin JM, et al. Localized non-Hodgkin's lymphoma of Waldeyer's ring: clinical features, management, and prognosis of 130 adult patients. Head Neck 2001;23:547-58.
- 3. Inchingolo F, Tatullo M, Abenavoli FM, Marrelli M, Inchingolo AD, Inchingolo AM, et al. Non-Hodgkin lymphoma affecting the tongue: unusual intra-oral location. Head Neck Oncol 2011;3:1.
- 4. Kobler P, Borcic J, Filipovic ZI, Nola M, Sertic D. Primary non-Hodgkin's lymphoma of the oral cavity. Oral Oncol 2005;41:12-4.
- Sirsath NT, Lakshmaiah KC, Das U, Lokanatha D, Chennagiri SP, Ramarao C. Primary extranodal non-Hodgkin's lymphoma of oral cavity--a single centre retrospective study. J Cancer Res Ther 2014;10:945-50.
- 6. Shah GH, Panwar SK, Chaturvedi PP, Kane SN. Isolated primary extranodal lymphoma of the oral cavity: A series of 15 cases and review of literature from a tertiary care cancer centre in India. Indian J Med Paediatr Oncol 2011;32:76-81.
- 7. Hmidi M, Touiheme N, Elboukhari A, Kettani M, Elmejareb C, Messary A. Primary B cell lymphoma of the tongue: a case report. Pan Afr Med J 2012;12:5.
- 8. Murthy S, Panduranga C. Fine-needle aspiration diagnosis of extranodal non-Hodgkin's lymphoma of the tongue. J Cytol 2011;28:81-3.