Dere, Özgeç, Eğilmez and Öztürk

Diffuse Large B-Cell Lymphoma of Anterior Mandible: A Case Report

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Abstract

Diffuse Large B Cell Lymphoma (DLBCL), which is rare in the oral cavity, poses diagnostic challenges due to its similarity to benign lesions. In this report, a case of DLBCL presenting as an asymptomatic swelling in the anterior region of the mandible that gradually grows in a 58-year-old female patient is presented. Although it was thought to be a benign lesion in the preliminary diagnosis, it was diagnosed as DLBCL in the histopathological examination performed after excisional biopsy. This case highlights the importance of considering DLBCL in the differential diagnosis of swellings in the oral cavity and emphasizes the importance of making the diagnosis and treatment quickly.

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Key words: Oral, cancer, lymphoma, mandible.

INTRODUCTION

Lymphomas malignant neoplasms are originating from lymphocytes and can be observed in lymphatic tissue, bone marrow or extranodal sites. They are classified into different subtypes based on morphological, genetic, immunological, molecular and clinical features (1). They are mainly classified as Hodgkin's lymphoma (HL) or non-Hodgkin's lymphomas (NHL) (2). Approximately 25% of NHL cases seen at extranodal areas; skin, central nervous system, gastrointestinal system and are the most common of them (3). Oral NHLs are rare compared to other sites, accounting for only 2-3% of all reported lymphomas (4).

Diffuse large B-cell lymphoma (DLBCL) is a type of NHL characterized by diffuse proliferation of large neoplastic B cells and can involve the oral cavity and jaw bones (5).

The most commonly affected areas in the oral and maxillofacial region are the Waldeyer ring, followed by the buccal mucosa, base of the tongue, floor of the mouth and posterior molar region (2). Apart from Waldeyer's ring, DLBCL can also involve the maxillary alveolus, maxillary vestibule and posterior palate in the oral cavity (4).

DLBCL can be misdiagnosed as benign or inflammatory lesions such as periodontitis,

osteomyelitis, and pyogenic granuloma, as well as different malignant tumors (6). DLBCL treatment usually consists of chemotherapy, radiotherapy or a combination of these (7).

This article reports a case of DLBCL affecting anterior part of mandible in a 58 years old female patient.

CASE-REPORT

A 58-year-old female patient came to our clinic with a complaint of swelling in the anterior region of mandible for 2 months. History revealed that the swelling started as a small lesion and progressively enlarged in size. There was no toothache or pain associated with it. There was no history of any purulent discharge from the swelling.

On examination, a single well-circumscribed swelling was seen on the anterior mandible, ovoid in shape, measuring about 2,5 cm in greatest diameter, with the surface color resembling that of normal mucosa, margins were well defined and the swelling crossed the midline on the left side without surface ulceration. (Figure 1) On palpation there was slight tenderness, the swelling was firm in consistency, sessile, attached to the underlying bone, not yielding to pressure, non-fluctuant. The patient reported that she did not feel any pain when the submental area was palpated.

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Dere, Özgeç, Eğilmez and Öztürk

Vitality testing was performed for both lower canines and incisors, and all teeth were vital.



Figure 1. Preoperative intraoral examination.

Panoramic radiographs revealed generalized destruction of alveolar bone in the anterior mandible region of teeth 42-33. A cone beam computed tomography (CBCT) without contrast was ordered to rule out concerning features and showed a diffuse bone destruction and nonspecific 9,66x35.25x27,64 mm enhancing soft tissue mass. (Figure 2) No mobility was observed in the teeth in the lesion area. Submandibular lymphadenopathy was not detected in the patient. The patient had diabetes in her medical history. Fine-needle aspiration cytology was done, but no aspirate was obtained.



Figure 2. Preoperative radiographic findings.

Central giant cell granuloma, myxoma and ameloblastoma were considered as preliminary diagnoses. Under local anesthesia, the mandibular incisors associated with the lesion were extracted, and an excisional biopsy was performed. Collected specimens were sent for a histopathological test. Ten days after the surgery, the sutures were removed and the healing status was examined (Figure 3).



Figure 3. Intraoperative and postoperative view.

The histological examination revealed a diffuse proliferation of atypical round cells with lymphoid features positive for CD79a, CD20, Pax5, CD138, CD3, CD5, CD68, CD138, LCA and PanCK. (Figure 4.) Negative for Bcl-2, Bcl-6, MUM1, CD10, RCC, S100, SOX10, MelanA, CD21, CD23, D1 cyclin and Lambda, Kappa. Ki-67 index was %75-80 positive. (Figure 5.)



Figure 4. An infiltration of medium/large lymphoid cells with diffuse involvement was observed. According to the immunohistochemistry results, CD45, CD20, CD79a and PAX5 were positive.

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Figure 5. In the immunohistochemical examination, CD10, Bcl6 and MUM1 were negative. Ki-67 proliferation index was 75-80%. Diffuse large B-cell lymphoma non-germinal center origin was diagnosed with morphological and immunohistochemical findings.

The final diagnosis was NHL compatible with extranodal DLBCL and the patient was referred to the Department of Oncology. In the PET/CT evaluation, the operated intraosseous area was observed to be clean. A chemotherapy regimen of five sessions was arranged upon the discovery of a suspicious area in the submental region. Control PET/CT scans were planned to determine the effectiveness of the treatment and possible recurrences.

DISCUSSION

The incidence of extranodal lymphoma is 24-27% in North America, 37-48% in Europe and 44.5% in Turkey (8) DLBCL has an unclear etiology that includes immunosuppression, autoimmune diseases, exposure to pesticides and radiation. Additionally, viruses like Epstein-Barr virus, human immunodeficiency virus, human T-cell lymphotropic virus, hepatitis C and B, human herpes virus and some microorganisms play a role in the etiology of DLBCL (9).

Diagnosis of intraosseous lymphoma of the jaws is difficult and is often delayed due to its characteristics similar to other pathologies. Extranodal lymphomas occurring in the mandible often present with nonspecific signs, such as painless swelling. Paresthesia, teeth mobility and cervical adenopathy are not common (10). Early lesions can be confused with inflammatory odontogenic or periodontal diseases, resulting in unnecessary treatments for patients (i.e., endodontic therapy, tooth extraction, antibiotic therapy) and delays before biopsy diagnosis (11).

Lymphomas can cause clinically indeterminate pain and discomfort and

can easily be misdiagnosed as an endodontic lesion resulting from an odontogenic infection. Radiological changes in the early stages of the tumor may not be very obvious and are usually detected as a radiolucent lesion late in the disease, similar to a dental abscess. Histologically, NHLs can easily resemble a periapical granuloma or odontogenic cyst due to cells that appear to be lymphocytic proliferation (12).

Severe jaw pain and teeth mobility caused by rapid destruction of bone can be an early sign of malignancy in the bones (13). The clinician should be aware of jaw tumor symptoms, which may aid in early diagnosis and treatment. Severe local destruction of bone without visible odontogenic infection is an important feature of jaw tumors (14).

The best way to manage lymphoma is correct diagnosis; clinically an excisional or incisional biopsy should be performed to obtain sufficient tissue for morphological and molecular analysis (15).Differential diagnoses of DLBCL include periodontal disease, some benign tumors of hard and soft tissues, squamous cell carcinoma, osteosarcoma, multiple myeloma, Ewing sarcoma, bone metastasis. Langerhans cell histiocytosis, leukemia, and osteomyelitis (16).

There are few case reports of oral and maxillofacial DLBCL in the literature. This makes diagnosis, prognosis, pathological behavior and treatment options difficult. Current treatment for DLBCL generally consists of chemotherapy (17). In the treatment of DLBCL, the chemotherapy regimen consisting of the combination of rituximab. cyclophosphamide, doxorubicin. vincristine and prednisone (R-CHOP) has been used as the standard of treatment in the first line for more than 20 years. In resistant cases, radiotherapy is also used in addition to treatment (18). The initial remission rate of the disease with treatment has been reported as 60-80%, and the 5year survival rate of cases with bone involvement is seen to be around 50%, which is a sign of poor prognosis (19).

Diagnosis of oral DLBCL is challenging for oral surgeons due to its rarity and nonspecific clinical and radiographic features. Maxillofacial surgeons play an important role in the early diagnosis and prognosis of oral NHL. Therefore, they need to have sufficient clinical and pathological knowledge and careful examination in such diseases. Biopsy should be considered without delay.

An informed consent form was obtained from the patient. The treatment and publication process was explained.

HRU IJDOR 2024;4(3)

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References

- Swerdlow SH, Campo E, Pileri SA, et al. The 2016 revision of the World Health Organization classification of lymphoid neoplasms. Blood. 2016; 127(20):2375-2390.
- Epstein JB, Epstein JD, Le ND, Gorsky M. Characteristics of oral and paraoral malignant lymphoma: a population-based review of 361 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2001;92(5):519-25.
- Vannata B, Zucca E. Primary extranodal B-cell lymphoma: current concepts and treatment strategies. Chin Clin Oncol 2015;4:10.
- Kolokotronis A, Konstantinou N, Christakis I, Papadimitriou P, Matiakis A, Zaraboukas T, et al. Localized B-cell non-Hodgkin's lymphoma of oral cavity and maxillofacial region: A clinical study.Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2005;99:303-10.
- Roschewski M, Staudt LM, Wilson WH. Diffuse large B-cell lymphoma—treatment approaches in the molecular era. Nat Rev Clin Oncol. 2014;11(1):12-23.
- Pereira DL, Fernandes DT, Santos-Silva AR, et al. Intraosseous non-Hodgkin lymphoma mimicking a periapical lesion. J Endod 2015;41:1738–42.
- Gustavsson, A., Osterman, B., & Cavallin-ståhl, E. (2003). A systematic overview of radiation therapy effects in Non-Hodgkin's Lymphoma. Acta Oncologica, 42(5–6), 605–619.
- Sağnak Yılmaz, Z., & Çobanoğlu, Ü. (2020). Distribution of lymphoid neoplasms in northeast Turkey: A retrospective analysis of 1136 cases according to the World Health Organization Classification. Konuralp Tıp Dergisi, 12(2), 208– 215.
- Guevara-Canales JO, Morales-Vadillo R, Sacsaquispe-Contreras SJ, et al. Malignant lymphoma of the oral cavity and the maxillofacial region: overall survival prognostic factors. Med Oral Patol Oral Cir Bucal 2013;18:e619–26.
- Kirita, T., Ohgi, K., Shimooka, H., Okamoto, M., Yamanaka, Y., & Sugimura, M. (2000). Primary non-hodgkin's lymphoma of the mandible treated with radiotherapy, chemotherapy, and autologous peripheral blood stem cell transplantation. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology, 90(4), 450–455.
- Rinaggio J, Aguirre A, Zeid M, Hatton MN. Swelling of nasolabial area. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2000;89:669-73.

HRU IJDOR 2024;4(3)

University Faculty of Dentistry Şanlıurfa, Turkey https://ijdor.harran.edu.tr/tr/

- 12. Neville BW: Oral and Maxillofacial Pathology, ed 3. St. Louis, Saunders/Elsevier, 2009.
- 13. Bugshan, A., Kassolis, J., & Basile, J. (2015). Primary diffuse large B-cell lymphoma of the mandible: Case report and review of the literature. Case Reports in Oncology, 8(3), 451–455.
- 14. Regezi JA, Sciubba JJ, Jordan RCK: Oral Pathology: Clinical Pathologic Correlations. St. Louis, Saunders/Elsevier, 2008.
- Jiang M, Bennani NN, Feldman AL. Lymphoma classification update: Bcell non-Hodgkin lymphomas. Expert Rev Hematol 2017;10:405–15
- Zou, H., Yang, H., Zou, Y., Lei, L., & Song, L. (2018). Primary diffuse large B-cell lymphoma in the maxilla: a case report. Medicine, 97(20), e10707.
- Jayapalan, C. S., Pynadath, M. K., Mangalath, U., George, A., Aslam, S., & Hafiz, A. (2016). Clinical diagnostic dilemma in an uncharacteristic rapidly enlarging swelling of the anterior maxilla: extranodal diffuse large B-cell lymphoma. Case Reports, 2016, bcr2015213141.
- Vodicka, P., Klener, P., & Trneny, M. (2022). Diffuse large Bcell lymphoma (DLBCL): Early patient management and emerging treatment options. OncoTargets and Therapy, Volume 15, 1481–1501.
- Triantafillidou K, Dimitrakopoulos J, Iordanidis F, Gkagkalis A. Extranodal non-hodgkin lymphomas of the oral cavity and maxillofacial region: a clinical study of 58 cases and review of the literature. J Oral Maxillofac Surg 2012;70:2776–85.