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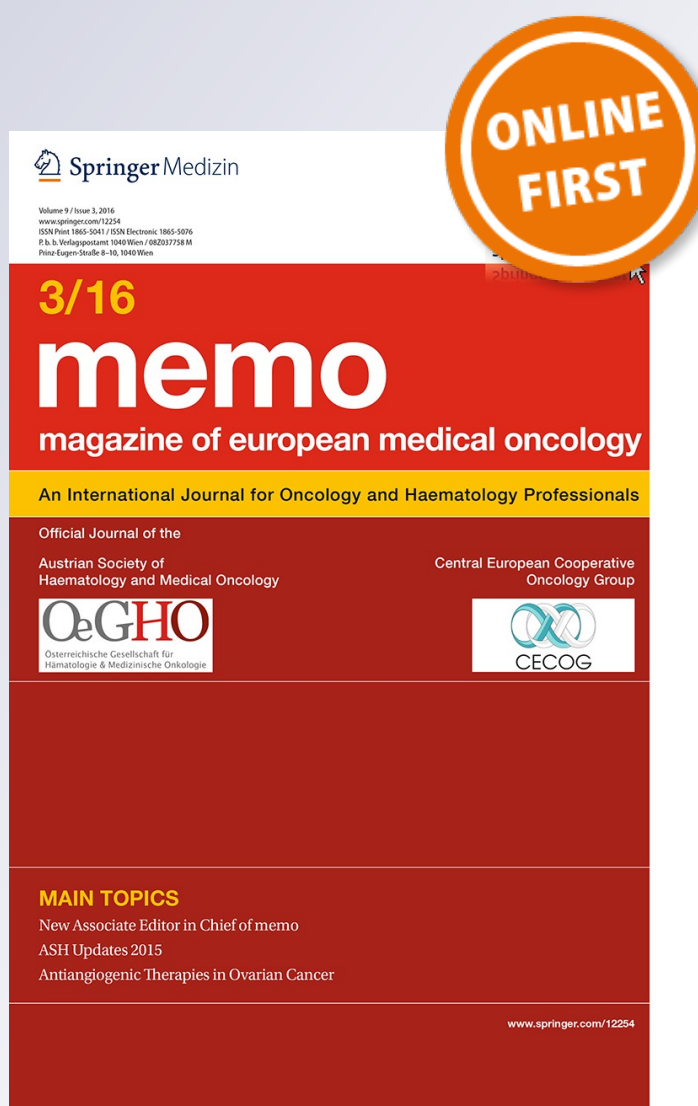
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Primary non-Hodgkin's lymphoma of the gingiva: a case report and review of the literature

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Summary Up to 40 % of cases of non-Hodgkin's lymphomas (NHL) occur with exclusive extranodal presentation, and involvement of the oral cavity is very rare. We report the case of a 40-year-old woman who presented with a gingival mass that was diagnosed as diffuse large B-cell lymphoma (DLBCL). The patient was treated with chemoradiotherapy, thus, achieving a complete remission, and the patient is currently free of disease.

Keywords Computed tomography · Lymphoma · Tumor · Radiotherapy · Chemotherapy

Introduction

Lymphomas are malignant lymphoproliferative neoplasms representing 5 % of malignant neoplasms of the head and neck, ranking second of the most common malignancies of the head and neck after squamous carcinoma [1]. Classically they are clas-

sified into Hodgkin's and non-Hodgkin lymphomas depending on the presence of Reed-Sternberg cells [2]. Unlike Hodgkin lymphoma, non-Hodgkin lymphomas can occur in up to 40 % of cases with exclusive extranodal involvement [3]. The most commonly affected sites are the gastrointestinal tract, skin, thyroid, lungs, kidneys, orbit, testicles, breasts, paranasal sinuses, Waldeyer's ring and oral cavity.

In the head and neck, the most common site of involvement of non-Hodgkin lymphoma is Waldeyer's ring, with primary involvement of the oral cavity being extremely rare and representing between 2 and 5 % of all extranodal lymphomas [4, 5]. Here we present a case report of non-Hodgkin lymphoma of the gingiva.

Case report

A 40-year-old female without a significant medical history presented in August 2014 to the Dental Service complaining of swelling on the alveolar level of the lower left incisors interpreted initially as gingival hyperplasia. The swelling had been present for 6 months without pain. The clinical exam revealed a mass of 2 × 2 cm in the area mentioned above, without ulceration. No other injuries or cervical lymphadenopathy were identified. Also, the patient presented no other symptoms (Fig. 1).

A biopsy of the lesion was performed, and the result revealed a homogeneous proliferation of medium and large round cells arranged in a diffuse pattern with marked nuclear pleomorphism, scant cytoplasm, atypical mitosis and compromised edges of the surgical resection, suggestive of lymphoproliferative malignant lesion. The case samples were sent to the Catholic University of Santiago for immunohistochemical study, which was positive for CD20, CD79a, CD45 and BCL6, and negative for CD3, CD5

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case report



Fig. 1 Clinical presentation of the tumor

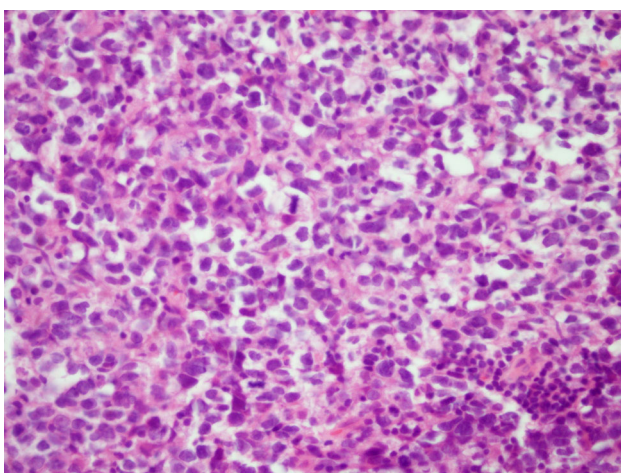


Fig. 2 Hematoxylin and eosin stain: oral mucosa biopsy with dense lymphoid infiltrate in the connective composed of large cells arranged in a diffuse pattern of moderate eosinophilic cytoplasm, large central core, with one or more prominent nucleoli and marked mitotic activity

and CD30. The final diagnosis was a diffuse large B-cell lymphoma (DLBCL) (Fig. 2).

Further study was performed with computed axial tomography of neck, thorax, abdomen and pelvis demonstrated soft tissue injury in the left mandibular body of 1.8 × 1 cm, without bone involvement, without lymphadenopathy and without lesions suggestive of distant metastases. It was evaluated by hematology to complete the study of staging of the lymphoma, showing absence of bone marrow involvement (Fig. 3).

The case was reviewed by the Head and Neck Tumor Board (HNTB) where it was diagnosed as diffuse large B-cell lymphoma I-E. The HNTB recommended chemo- and radiotherapy as the best treatment option. She received two doses of chemotherapy with rituximab, cyclophosphamide, doxorubicin, vincristine and prednisone (R-CHOP). As for the radiation, she received 30 Gy in 3 weeks presenting a complete remission (Fig. 4).

The case has been followed for 1 year 6 months. The patient is currently free of disease and undergoes periodic checks in hematology and oncology, with no clinical or imaging evidence of recurrence.

Discussion

Lymphomas are malignant neoplasms representing 5 % of malignant neoplasms of the head and neck, ranking second of the most common malignancies of the head and neck after squamous carcinoma [1]. Overall, 40 % of non-Hodgkin lymphoma (NHL) may initially present with exclusive extranodal involvement, with the most affected sites being the gastrointestinal tract (stomach and small intestine), skin, thyroid, lungs, kidneys, orbit, testicles, breasts, paranasal sinuses, Waldeyer's ring and oral cavity [3]. The oral cavity is formed by the palate, the anterior 2/3 of the tongue, the floor of the mouth, gingiva, lips and oral mucosa [4].

In the head and neck, the most common site of NHL involvement is Waldeyer's ring, with primary involvement of the oral cavity being extremely rare, representing between 2 to 5 % of all extranodal lymphomas [4, 5]. The main affected structure of the oral cavity is the palate, with exclusive involvement of the gingiva being extremely rare [6]. In a literature review by Angiero et al. [6] 40 cases of primary gingival NHL were reported of a total of 411 cases of oral cavity lymphomas published to date. A series of 381 cases of lymphomas of the head and neck published by Etemad-Moghadam et al. [7] reported a total of 100 NHL with extranodal involvement, with the most frequent site being Waldeyer's ring followed by the salivary glands and only two cases of primary involvement of the gingiva. The most common histological type was diffuse large B-cell lymphoma (DLBCL) in 75 % of cases.

DLBCL originates from mature B cells but its etiology is still unclear. At the molecular level, there have been reports of alterations in BCL6 (B-cell lymphoma 6) protein, a protein encoded by the BCL6 gene. This alteration is unique to this type of lymphoma [8]. It has also been associated with infection by Epstein Barr virus (EBV) and human immunodeficiency virus (HIV), with this association being reported in up to 62.5 % of cases in different series and with a worse prognosis versus uninfected patients [9].

The most common clinical presentation is the appearance of a rapidly growing mass at the compromised site and can be accompanied by pain and/or ulceration [7]. It usually occurs during the fifth decade of life and gender distribution is quite similar, with male to female ratios of 1.16:1 to 3:2 [10]. The presence of B symptoms is rare, occurring in less than 30 % of cases. It is usually diagnosed in advanced stages (III–IV) and bone marrow involvement is reported in up to 30 % of the cases [11].

Fig. 3 CT soft tissue and bone window. **a** Poor enhancement postintravenous contrast injection (*arrow*). **b** No evidence of bone infiltration

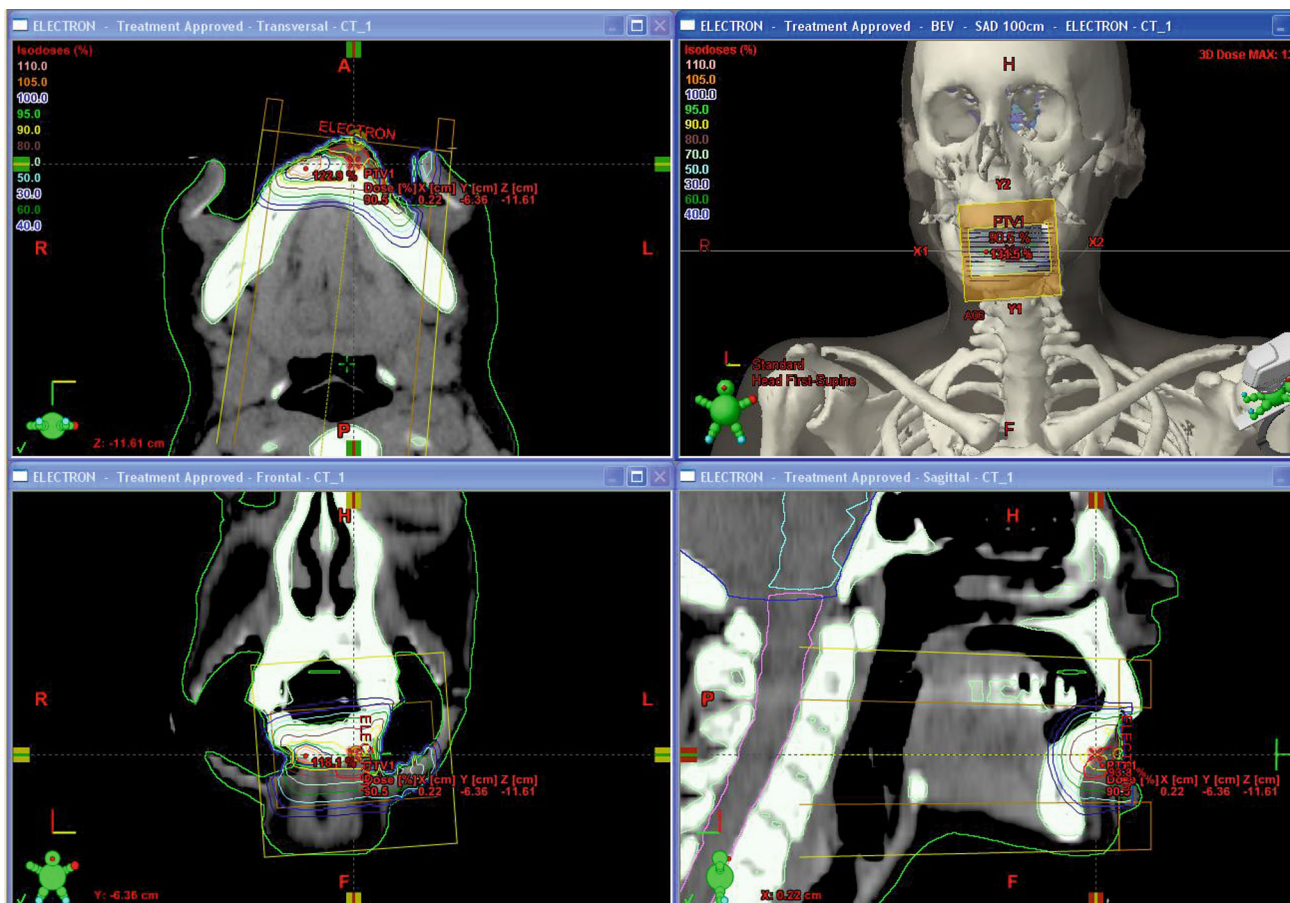


Fig. 4 Three-dimensional conformal radiation plan

There are no characteristic images or lesions in radiology exams. In 10–20% of cases there is no compromise of the bone and in the remaining 80% diffuse bone infiltration is observed. However, even without bony wall destruction, this finding is useful in differentiating it from squamous cell carcinoma [12]. The definitive diagnosis is done with a biopsy of the lesion. Histologically, it is characterized by large

cells with a diameter three times larger than normal lymphocytes with a core and prominent nucleoli. Immunohistochemistry demonstrates expression of B cell markers such as CD20 (usually negative for plasma cell tumors) [5] and CD79a and unique markers of diffuse large B-cell lymphomas such as BCL-6. Also, it is nonreactive for CD3 [8], a common T cell marker.

case report

The differential diagnosis should be done with any inflammatory lesion of the oral cavity such as periodontitis or osteomyelitis and all malignant head and neck lesions such as odontogenic malignant tumors, malignant tumors of mesenchymal (osteosarcoma) and undifferentiated carcinomas [13].

The treatment consists of complete resection of the lesion for biopsy followed later by chemotherapy with or without radiation therapy. The combination of these two treatments is used in patients with high-grade and fast-growing lymphomas, like DLBCL. The used chemotherapy is R-CHOP and it is complemented with a dose of radiation of 28–40 Gy [14]. With this treatment the initial remission rate varies between 60–80 %, but with a 5-year survival of 30–50 % [5]. The prognosis depends on the type of lymphoma, the stage and the association with HIV infection [6], with a poorer prognosis for those with DLBCL versus follicular lymphomas [5], advanced stages compared to the early stages (the average survival rate for I-E stage is 10 years [15]) and those infected with HIV [9].

Conclusion

Primary non-Hodgkin lymphoma of the gingiva is a rare condition characterized by an increase in gingival volume with or without ulceration that should always be included in the differential diagnosis of any lesion of the oral cavity; thus, the role of the dentist is essential to early diagnosis. The diagnosis is done by biopsy of the lesion, with diffuse large B-cell lymphoma being the most common histologic type. The treatment is based on chemotherapy and radiotherapy with an initial remission of up to 80 %, but with an overall survival rate of 30–50 % depending on the type of lymphoma, stage at diagnosis and association with HIV.

Conflict of interest I. Sepúlveda, M. Schorwer, E. Platin, M. Yañez, and F. Fredes declare that they have no competing interests.

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