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# Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: [www.elsevier.com/locate/jomsmmp](http://www.elsevier.com/locate/jomsmmp)

## Case Report

# Intraosseous lymphoma of the oral and maxillofacial regions: Report of our experiences, involving some difficult cases to be diagnosed



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## ARTICLE INFO

### Article history:

Received 10 December 2014

Received in revised form 2 March 2015

Accepted 29 May 2015

Available online 20 June 2015

### Keywords:

Lymphoma

Oral cavity

Jaw

## ABSTRACT

Intraosseous lymphoma in the maxilla or mandible is sometimes difficult to diagnose because of the absence of specific signs and symptoms. This study reports five cases of intraosseous lymphoma in the oral and maxillofacial regions. There were four cases of diffuse large B-cell lymphoma (DLBCL), including EBV-positive DLBCL of the elderly, and one of Burkitt's lymphoma. Regarding the tumor sites, three of the cases were in the maxilla, while two were in the mandible. Two of the conditions were clinically diagnosed as inflammatory diseases at the time of their first examination. Two patients died of their lymphoma during the follow-up period. Based on these events and findings, we clearly realized that careful examination is necessary to be diagnosed early, which results in a much better prognosis. Moreover, maxillofacial surgeons need to keep following up carefully on their patients to immediately recognize any recurrences after completion of treatment.

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## 1. Introduction

Lymphoma which consists of malignant neoplastic mutations of normal lymphoid cells may originate in any region containing lymph tissue [1]. Traditionally, lymphoma has been classified into two subtypes, Hodgkin's lymphoma (HL) and non-Hodgkin's lymphoma (NHL) which depends on the presence or absence of Reed–Sternberg cells. Previous studies have shown that 48% of NHL cases occurred in extranodal sites, whereas HL was frequently diagnosed as a nodal disease [1]. The head and neck is one of the most common site for extranodal lymphomas which occur frequently in Waldeyer's ring, salivary glands, orbit, paranasal sinuses, and thyroid glands [2]. Extranodal lymphoma in the oral cavity represents less than 5% of oral malignant disease commonly arising in submucosal tissues of gingiva, palate, and tongue [3]. However,

intraosseous lymphoma in the maxilla or mandible is uncommon, which is sometimes difficult to diagnose because of the absence of specific signs and symptoms. Therefore, early detection of the lesion is necessary for a better prognosis.

The World Health Organization (WHO) classification system for neoplastic diseases of the hematopoietic and lymphoid tissues, which modified the Revised European-American Lymphoma classification, is currently the most commonly used system for the classification of malignant lymphoma [4]. Regarding B-cell lymphoma, the diffuse large B-cell lymphoma (DLBCL) and the extranodal marginal zone lymphoma of mucosa-associated lymphoid tissue (MALT) subtypes are commonly observed in the oral and maxillofacial regions. However, other types of lymphoma arising from the oral and maxillofacial regions or identified from oral manifestations are uncommon. Of those, Burkitt's lymphoma (BL), which was initially described in Africa, is frequently associated with the Epstein–Barr virus (EBV) and observed particularly in children and patients with the Human immunodeficiency virus (HIV) [5].

Here we describe five cases of intraosseous lymphoma in the oral and maxillofacial regions, including BL and DLBCL. One of them is EBV-positive DLBCL of the elderly which has been reported rarely in the oral and maxillofacial regions.

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## 2. Case reports

### 2.1. Case 1

A 27-year-old woman experienced pain and numbness in the right lower premolar region. She consulted a general dentist who referred her to our hospital.

Clinical examination revealed a spontaneous pain in the right lower premolar region with numbness of the right lower lip. No facial or gingival swelling was observed (Fig. 1A and B). A panoramic radiograph showed that the bone trabeculation on the right side appeared relatively less dense than on the left side (Fig. 1C). A MRI depicted low intensity areas of the right mandible on T1-weighted and T2-weighted, while T2 STIR showed high intensity (Fig. 1D). A CT scan demonstrated a lower density of bone trabecular on the right side of mandible without erosion of the cortex (data not shown). The patient also noticed a small lump on right breast with pain. Needle biopsy revealed that it was possibly BL (Fig. 2A–C). This case presented chromosomal translocation of the c-myc gene: t(8;14)(q24;q32) detected by the fluorescence in situ hybridization (FISH), confirming BL (Fig. 2D). The fluorescence activated cell sorting (FACS) showed CD10, CD19, CD20, and  $\kappa$ -ch positive, but it was  $\lambda$ -ch negative, which verified this case of the matured B-cell lymphoma (data not shown). The patient subsequently underwent an extensive work-up at the hematology department and was diagnosed with stage IV lymphoma.

The hematologist recommended six courses of hyper-CAVD chemotherapy with rituximab. After the full course of therapy, the patient continues to be free of any evidence of lymphoma.

### 2.2. Case 2

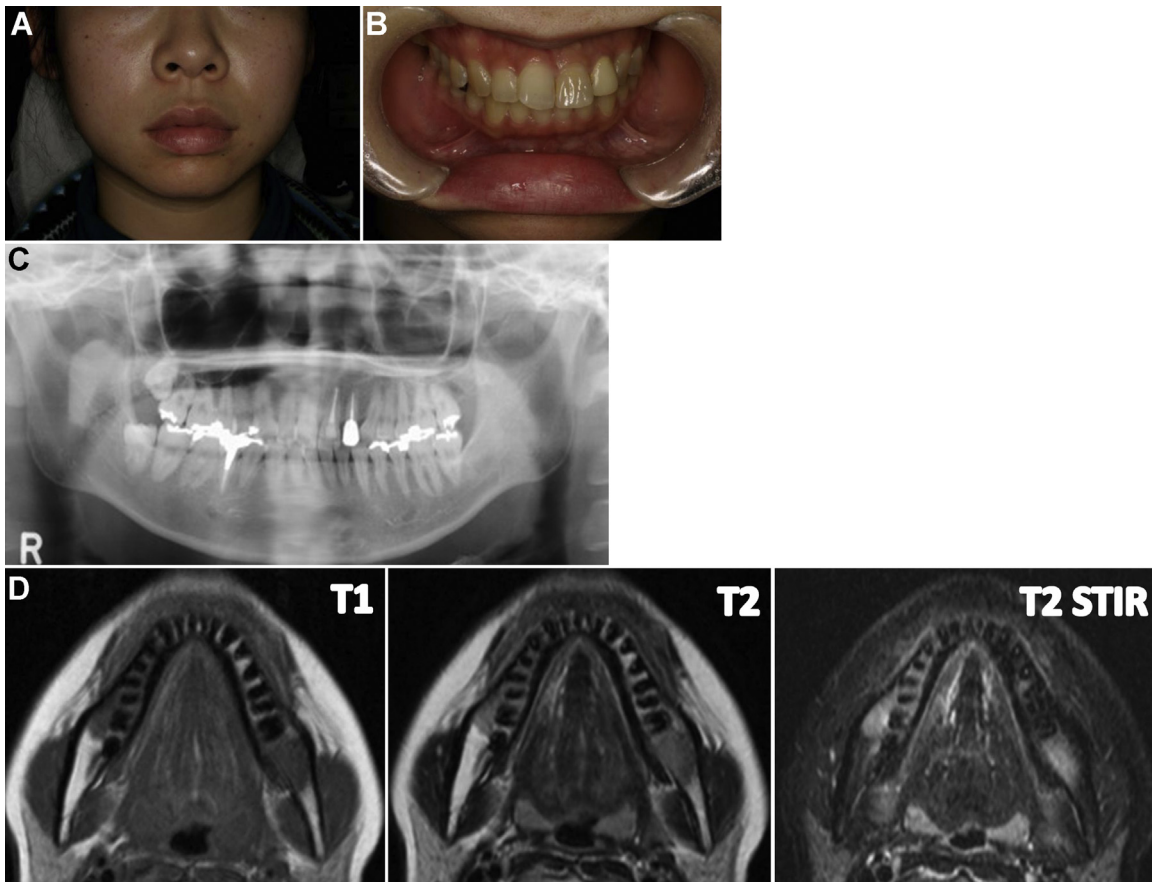
An 81-year-old man experienced pain and swelling of the right mandibular gingiva. He initially saw a general dentist and surgical drainage was done under the diagnosis of subperiosteal abscess. Because the symptoms did not improve after the surgical treatment, the patient was referred to our hospital for examination and treatment.

Clinical examination revealed a swelling with skin reddish of the right lower cheek. Intraoral examination found swelling in the right lower buccal vestibule (Fig. 3A and B). The patient also developed a numbness of the right lower lip. A panoramic radiograph showed no apparent difference in the bone trabeculation between the right and the left side of mandible (Fig. 3C). A CT scan revealed the soft tissue mass around the right mandibular bone with destruction of the lateral cortex (Fig. 3D and E). A biopsy was performed, which was diagnosed as EBV-positive DLBCL of the elderly (Fig. 3F–I).

The patient was then referred to the hematology department for systemic work-up and management. The patient was then diagnosed with stage IV disease identified as lymphoma of the lung and mesentery as well as the mandibular bone. However, he wished to be treated at another facility near his home and family.

### 2.3. Case 3

A 63-year-old man consulted his dentist for the treatment of a toothache of the right upper premolar. Although he received dental treatment, the symptom failed to improve. During the next 2 months, the patient experienced numbness of the right upper



**Fig. 1.** Case 1 reveals Burkitt's lymphoma of the mandible. (A) No facial swelling is observed. (B) Intraoral photograph showing no gingival swelling. (C) A panoramic radiograph showing lower density of the bone trabeculation in the right mandibular bone. (D) MRI showing the mandibular bone images on T1-weighted, T2-weighted, and T2 STIR.

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