CASE REPORT

An Unusual Case of Non-Hodgkins Lymphoma: A Case Report

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Abstract

Primary non-Hodgkin lymphoma (NHL) is rare in the oral cavity. When it does occur, mandibular NHL typically manifests similarly to an odontogenic process. This results in delayed diagnosis and treatment. Here we present a case of a 40-year-old female who came with the complaint of a bony hard swelling in the anterior region of the mandible. Although radiographic features were suggestive of a malignant bone tumour, incisional biopsy revealed sheets of malignant small round blue cells, suggestive of lymphomas, which were later confirmed as large B-cell lymphoma based on immunohistochemical markers. Fortunately, the solitary bony lymphoma had not disseminated and hence, management by chemoradiation and BIPPS pack placement allowed for disease eradication and resulted in new bone formation post-surgically. The purpose of this report is to describe a rare case of NHL of the mandible, explore the diagnosis and workup and discuss treatment strategies.

Keywords: lymphoma, Non-hodgkins, Oral.

Introduction

Lymphoma is a heterogeneous malignant disease of the lymphatic system, characterised by a proliferation of lymphoid cells or their precursors.[1] Lymphomas make up about 14% of all head-neck malignancies, out of which 97% are non-Hodgkin lymphomas (NHL). NHL is a malignant tumour characterised by the proliferation of either B or T lymphocytes.[2] NHL is usually uncommon in the oral cavity; of the reported cases, only 0.6% of the cases were found in the mandible (mostly the body). Diffuse large B-cell lymphoma is an aggressive subtype of NHL where the malignant cells are B lymphocytes.[3] The clinical courses, treatment responses, and prognoses of NHLs vary with different subtypes and anatomic sites.[4]

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Case Report

A 40-year-old female patient visited the department of OMFS with the chief complaint of swelling in the lower region of the face. Intraorally, a swelling extending from the 31–47 region was noted, extending from the gingival margin to the vestibular depth. Mobility of the tooth from 31-47 was also observed. The covering mucosa was normal in colour without ulceration or inflammatory signs. The lesion was well defined, firm in consistency and non-tender on palpation (Figure 1A).

Cone beam computerised tomography (CBCT) was taken, which showed periosteal lifting on the buccal side from the 31–47 region, featuring a Codman's triangle. The 3-D construction showed a typical moth-eaten appearance. No pathological evidence of fracture was noted. The differential diagnosis included osteosarcoma and metastatic tumour (Figure 1B). A PET-CT was taken in which an FDG-avid diffuse permeative pattern of rarefaction was seen in the mandible, with widening of the body and ramus. A mild periosteal reaction was noticed in the angle of the right mandible with associated soft tissue thickening in the surrounding planes,

indicating a metabolically active primary malignant tumour of the bone.

An incisional biopsy of hard and soft tissue was taken. Hematoxylin and eosin-stained sections showed round neoplastic cells arranged in sheets separated by a minimal connective tissue stroma. The neoplastic cells had scant cytoplasm with prominent hyperchromatic nuclei (Figure 2). The histopathological features were suggestive malignant round cell tumour. Lymphoma, Ewing's sarcoma and metastatic tumours were considered in the differential diagnosis. Neoplastic cells were positive for LCA, CD20, BCL2, and MUM1 and were negative for CK, CD3 and CD10. Ki67 proliferation index was 50–60%, confirming the diagnosis of non-Hodgkin's diffuse large B-cell lymphoma, activated B-cell subtype.

The patient was referred to the cancer centre for further management, where 6 cycles of RCHOP followed by local radiotherapy were planned. However, after 6 cycles of chemotherapy, the lesion responded very well and began to subside, and hence, radiotherapy was not done. The lower anterior teeth were extracted under local anaesthesia due to extreme mobility. After the extraction, the patient reported osteonecrosis of the alveolar bone and chronic pus discharge. Sequestrectomy and debridement of the necrotic bone were performed, followed by placement of the BIPPS pack. The patient reported back for review after 1 week, and healing was found to be satisfactory (Figure 3A). Follow-up was done after 3 months. Clinical and radiographic examination showed good healing and new bone formation (Figure 3B).

Discussion

Lymphomas are malignant neoplastic proliferations of lymphocytes and represent the second most common primary malignancy in the head and neck.2 Based on the Revised European-American Classification of Lymphoid Neoplasm (REAL), lymphomas are currently classified using the World Health Organisation (WHO) classification that recognises over 20 different subtypes of NHL and considers morphology, immunophenotype, genetic, and clinical features as the premise for classification. Between 85% and 90% of all NHLS derive from B lymphocytes.[3]

The aetiology of NHL is still unknown. However, patients exposed to pesticides and radiation or affected by autoimmune diseases such as rheumatoid arthritis, systemic lupus erythematosus, and the Sjögren syndrome, viral infection with Epstein-Barr virus (EBV), human T-cell lymphotropic virus

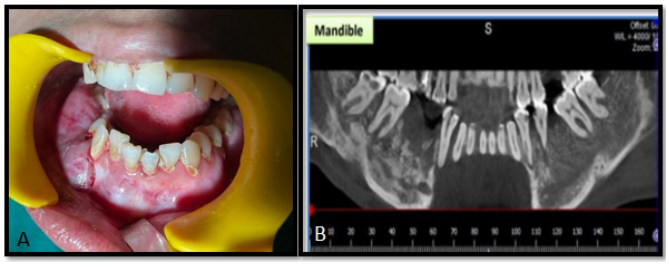

Fig. 1A – Clinical photograph showing intraoral swelling with expansion of cortical bone in the anterior mandibular region.

Fig. 1B- CBCT showing moth-eaten appearance in the anterior region of the mandible.

Table 1: Histological differential diagnosis of Lymphoma

Ewing's sarcoma	Sheets or nests of monotonous round blue cells with minimal cytoplasm and prominent nuclei, pseudo-rosettes are present.
Plasmacytoma	Sheets of monoclonal plasma cells exhibiting cart wheel appearence of the nuclei.
Malignant melanoma	Sheets and nests of atypical melanocytes.
Undifferentiated carcinoma	Sheets of round or oval malignant cells exhibiting high-grade features such as cellular and nuclear pleomorphism.

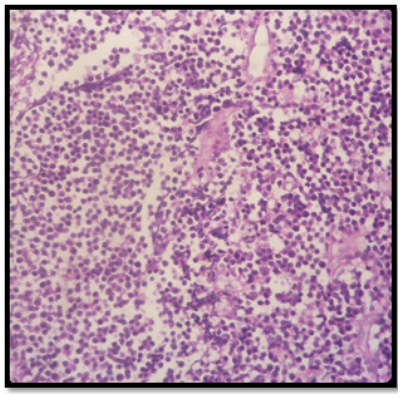

Fig. 2- Photomicrograph showing round neoplastic cells arranged in sheets (H&E, 10x).

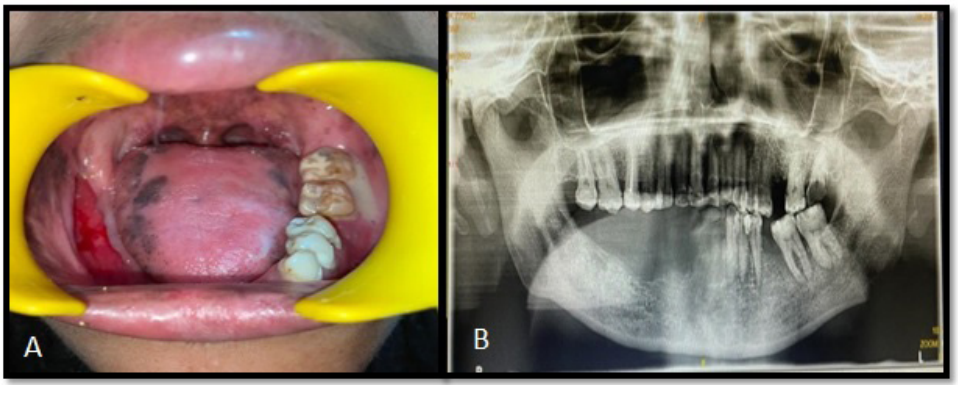

Fig. 3A- Clinical photograph showing postoperative healing after 1 week of sequestrectomy.

Fig. 3B- OPG taken 3 months after sequestrectomy showing new bone formation.

1 (HTLV-1), human immunodeficiency virus (HIV), human herpes virus type 8 (HHV-8), and hepatitis B and C viruses (HBV and HCV), have been related to a greater risk of NHL.5,6 In our case patient had undergone a serology test and was found to be negative for HIV and Hepatitis C infection.

The clinical features of NHL are significant swelling, limited mouth opening and movement associated with pain. The clinical manifestations include mucosal swelling, tooth mobility, and numbness-chin syndrome.[7] In this case, the patient had almost the same clinical features, including bone swelling, tooth mobility and restricted mouth opening. However, the radiographic features were highly suggestive of Osteosarcoma due to periosteal elevation and presence of "Codman's triangle". In such cases, where the clinical and radiographic features overlap with other bony tumours, histopathological examinations remain the gold standard for final diagnosis.

The diagnosis of NHL is based on histological examination, and a majority of the intraoral lymphomas are predominantly of B-cell lineage. The most common histopathological subtypes are diffuse large B-cell lymphoma (DLBCL) and plasmablastic lymphoma. Diffuse large B-cell lymphoma has an increased prevalence in elderly patients, occurring on average in the seventh decade of life.[8,9]

The diagnosis of oral NHL is established by tissue biopsy, and an adequate specimen should be obtained to ensure an accurate diagnosis. Histologically, the differential diagnosis of DLBCL includes undifferentiated carcinomas, sarcomas, plasmacytomas, and malignant melanomas (Table 1). Therefore, immunohistochemistry is pivotal for a definitive diagnosis. DLBCL is negative for epithelial markers, CD3, CD138, myeloperoxidase, desmin, and S100, ruling out the aforementioned lesions included in the histological differential diagnosis.[5]

The therapeutic options for lymphomas of the oral and maxillofacial regions include chemotherapy, radiotherapy, and immunotherapy. In the pre-rituximab era, more than a third of patients with this disseminated DLBCL were cured with chemotherapy alone, with most following the CHOP regimen (a combination of cyclophosphamide, doxorubicin, vincristine, and prednisone). The addition of the monoclonal CD20 antibody rituximab to chemotherapy (R-CHOP) was the most important progress in treatment. A retrospective study in British Columbia, Canada, compared outcomes of patients given CHOP-like chemotherapy in the pre-rituximab era with outcomes of patients treated in the rituximab era. The addition of rituximab to CHOP-like chemotherapy improved overall survival at 2 years from 52% to 78%.[10]

In our case, the lesion responded very well to chemotherapy and began to subside after the extraction of teeth at the affected site. The case was planned for 6 cycles of RCHOP followed by local radiotherapy, but surprisingly, after 6 cycles of chemotherapy, the lesion responded very well and began to subside. Ansell et al in their study stated that R-CHOP is a standard therapy for DLBCL and R-CHOP is superior to CHOP. 10 Diffuse large B-cell lymphoma is considered an aggressive yet treatable neoplasm with a variable clinical course.

Conclusion

Although non-Hodgkin lymphoma is rare in the oral cavity, it should always be considered in the differential diagnosis of malignant intraoral disease. When a tissue diagnosis is confirmed as NHL of the mandible, a determination must be made regarding the orientation and spread of the tumour. A good medical history, detailed clinical and imaging evaluations, and attention to the patient's signs and symptoms are crucial for the correct diagnosis and appropriate treatment, which in turn can lead to a better patient prognosis.

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