Research Article

T-Cell Lymphoma of Oral Cavity: A Case Report

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Abstract

Malignant lymphomas are heterogenous group of neoplasms of lymphoid tissues with different clinical courses and varied prognosis. Non-Hodgkin's lymphoma (NHL) often presents at the extranodal sites of head and neck region, but the intraoral lesions are much less frequent, especially when they are the only manifestation of the disease. The oral cavity including the palate, gingiva, tongue, buccal mucosa, floor of the mouth and lips are the primary sites for approximately 2% of all extranodal lymphomas. We here report the case of a 40 years old male that had initial extra nodal intraoral presentations undiagnosed previously. The clinical, histopathological and immunological examinations revealed T-cell lymphoma.

Keywords: Extranodal, Non-Hodgkin's lymphoma, Oral cavity, Soft palate, Hard palate.

Introduction

Lymphomas are a heterogenous group of clonal malignant neoplasms of lymphocytic cell line known for their spectrum of behaviour ranging from relatively indolent to highly aggressive and potentially fatal course. They are broadly classified as Hodgkin's lymphoma (HL) and Non-Hodgkin's lymphoma. Primary NHL usually arises within the lymph nodes but 20-30% accounts for the extra nodal sites [1]. The incidence of oral NHL is about 0.1% to 5% [2]. Oral NHL can involve the paranasal sinuses but can also arise from within the soft tissue or bone, gingiva, floor of mouth, salivary glands and cheek [3]. Majority of adult NHL (about 79%) are of B-cell origin while the rest belong to T cell or NK cell type but according to other studies the incidence of B cell lymphoma is even higher [4]. Owing to scarcity of reported cases, the diagnosis and understanding of the biological behaviour of oral NHL becomes difficult and therapeutic options are therefore limited. A thorough clinical, histopathological and immunohistochemical evaluation is essential for the diagnosis and management of oral NHL.

Manuscript received: 22nd Feb 2016
Reviewed: 28th Feb 2016
Author Corrected: 10th March 2016
Accepted for Publication: 20th March 2016

Case Report

A 40 years old male presented with a unilateral swelling at the upper left quadrant of oral cavity, at the junction of soft and hard palate. On examination, the swelling was 4 X 4 cms in size, bluish red in colour, sessile, non-tender on palpation, with smooth surface and well-defined margins. There was no other swelling in the oral cavity, no cervical or axillary lymphadenopathy or any other positive findings.

The radiographic and clinical examination revealed no other significant finding. The conclusion that the lesion was primarily in the oral cavity was made. The biopsy of the lesion was taken for confirmation of the diagnosis. The histopathogical and immunohistochemical studies revealed that the lesion was T cell lymphoma of oral cavity.

Discussion

Lymphoma is the third most common neoplasm of head and neck region; the first two being squamous cell carcinoma and salivary gland neoplasms [5]. Lymphomas arise due to mutation of progenitor cells of lymphoid lineage, that can be determined by

immunophenotyping and genetic arrangement studies and various etiologies have been suggested. The incidence of oral lymphoma is rare and is approximately 2% of all extranodal lymphomas [6]. Gulley et al [7] suggested the role of EBV in oral lymphomas. Also, it has been reported that there is an increased rate of lymphoma in patients who are congenitally immunocompromised and in patients who receive immunosuppressive therapy [8].

Almost 23% of patients with NHL presented with involvement of an extranodal head and neck site [9]. The most common site involved in oral cavity is palate and gingiva [10], however, lymphomas of other sites have also been reported [11, 12]. It is uncommon for NHL to appear first or only orally. Primary occurrence of NHL in oral mucosa is rare and when oral soft tissue lesions appear for the first time ,they are generally non-tender, soft to firm swelling of the area often with overlying ulcerations [13], and they are often characterized by an absence of other symptoms [8, 14, 15].

Enrique et al [16] has reported that the incidence of involvement of cervical lymph nodes in HL is 100% and in NHL is 86.6%. Abdominal adenopathies may be found in 50% patients of head and neck NHL [16]. The varied presentation of the disease provides a diagnostic dilemma owing to the protean manifestation of its presentation. It may present with nasal obstruction, rhinorrhoea, hypoacousia and cranial nerve palsies. Most lesions occur in Waldeyer's ring and occurrence in oral cavity is very rare [1]. Our patient had lesion in the palate region including both hard and soft palate.

The signs and symptoms suggestive of lymphoma in the head and neck region are the presence of numbness, tooth mobility, swelling, unexplained dental pain or ill-defined lytic osseous changes [17]. Other differential diagnosis includes a dental abscess, periodontal infection or benign reactive hyperplasia. Our patient presented with a non-tender swelling, gingivitis, tooth mobility but as the swelling was progressive, incisional biopsy of the lesion was done and diagnosis of lymphoma was made which on further confirmation with immunohistochemistry turned out to be T cell lymphoma.

Lymphoma presents mainly in older adults [4] and there is a logarithmic increase in incidence with increasing age [18]. Also, male gender is commonly involved [10]. In our case also the patient was a 40 years old middle aged male.

Various classifications and staging systems have been suggested including working formulation classification, REAL classification, WHO classification, IPI and Ann Arbor staging system, and NCI proposed grading [19, 20, 21].

The diagnosis of oral extranodal NHL is difficult owing to the versatility of its presentation and low index of suspicion. Incisional biopsy coupled with immunological studies of biopsied tissue is a definitive diagnostic modality [22]. CT scan of head and neck, chest, abdomen and pelvis are the mainstay of staging oropharyngeal extranodal lymphomas.

Investigations for immunocompromised patients like HIV and EBV infections should be examined because oral cavity is preferred site for extranodal NHL. Concurrent immunohistochemistry is useful for distinguishing cell types further confirming the diagnosis [1]. In our patient, histopathological examination revealed monoclonal proliferation of lymphocytes in a chronic inflammatory background (Figure 1).

Immunohistochemistry showed that the tumour cells were strongly immunopositive for CD3 (Figure 2) and CD5 (Figure 3), focal positive for CD45 (Figure 4), and negative for CD19 and CD20. The diagnosis of T cell lymphoma of oral cavity was made. The patient was seronegative for HIV. He was asked for follow-up after the investigations but he did not turn up.

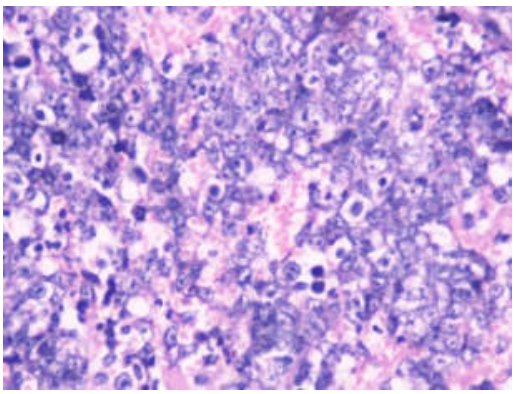

Figure 1: H & E stain 10X: Monoclonal proliferation of lymphocytes

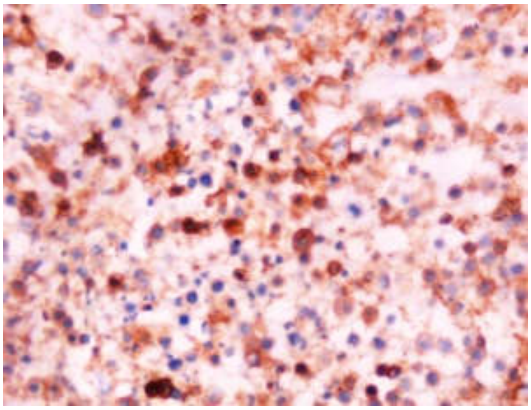

Figure 2: CD3 positive 40X

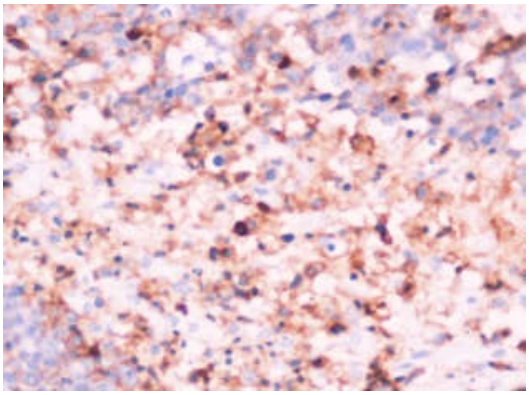

Figure 3: CD5 positive 10X

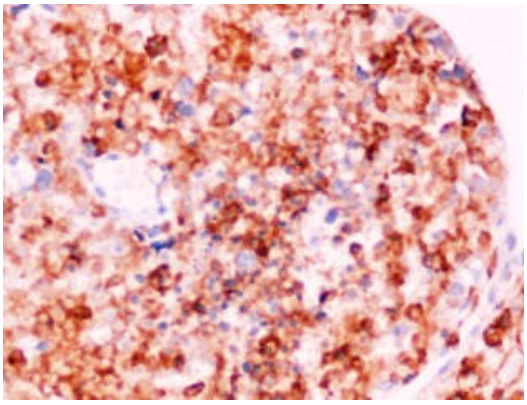

Figure 4: CD45 Focal positivity 10X

Primary oral NHL is a rare entity though it is a preferred site in immunocompromised patients. In immunocometent patients, the diagnosis is difficult due to low index of suspicion. Chemotherapy is the mainstay of treatment. Anti-retroviral therapy (ART) along with chemotherapy is indicated in HIV positive cases. It is the responsibility of the dentists, paediatricians, clinicians and oral & maxillofacial surgeons who treat these patients to be aware of this rare possibility as correct diagnosis is very essential for appropriate treatment at an early stage of the disease.

Funding: Nil, **Conflict of interest:** None.
Permission of IRB: Yes

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How to cite this article?

Prashant D, Fatima S, Tiwari N, Prashant V. T-Cell Lymphoma of Oral Cavity: A Case Report. *Int J Med Res Rev* 2016;4(3):397-400. doi: 10.17511/ijmrr.2016.i03.20

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