Case Report

Peripheral T-cell lymphoma of tongue: Report of a rare case and review of literature

Swetha Narla, S Annapurneswari, Ashok Parameswaran, Sheila Nair

Department of Histopathology, Apollo Hospitals, Chennai, Tamil Nadu, India

Abstract

Primary non-Hodgkin lymphomas of the oral region are rare, accounting for 3–5% of all malignant lesions. Of these, peripheral T-cell lymphomas (PTCLs) are extremely rare with only a few cases reported in literature. We describe a case of 50-year-old female who presented with an indurated lesion on the tongue. PTCL was diagnosed after immunohistochemical and T-cell receptor gene rearrangement analysis. Although PTCL of oral cavity is extremely rare, the possibility should always be considered in the differential diagnosis of T-lymphoid proliferations affecting this area.

Key Words: Non-Hodgkin lymphoma, oral cavity, peripheral T-cell lymphoma, tongue

Address for correspondence:
Dr. Swetha Narla, Department of Histopathology, Apollo Speciality Hospitals, 320 Anna Salai, Chennai - 600 035, Tamil Nadu, India.
E-mail: drswetha.gmc2k2@gmail.com
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INTRODUCTION

Extranodal lymphomas account for 20–30% of non-Hodgkin lymphoma (NHL).[1] NHL accounts for 3–5% of all malignant lesions of the oral cavity which most commonly affects the Waldeyer's ring including tonsils (55%), soft palate (30%), tongue (2%), buccal floor (2%), retromolar area (2%) and others (9%).[2] 1% of all lymphomas are oral cavity lymphomas of which diffuse large B-cell lymphoma (DLBCL) is the most common type.[3] Oral T-cell lymphomas are extremely rare, and only a few cases have been reported in literature.[4] Most patients are asymptomatic, and rest of them present with swelling causing pain and discomfort mimicking benign orodental conditions. Hence histopathological examination is essential for accurate diagnosis. The overlying lingual mucosa may or may not be involved. Understanding the biological behavior and therapeutic options of oral cavity lymphomas is difficult due to paucity of cases. A proper clinical evaluation, histopathologic and immunohistochemical evaluation will help in the diagnosis and management of the cases.

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CASE REPORT

A 50-year-old female presented with a nodule on the tongue which she had noticed 45 days before her visit which was gradually increasing in size. The patient did not experience B symptoms (fever, weight loss and night sweats). Local examination revealed a 2 cm × 2 cm submucosal indurated lesion involving lateral margin of the left side of dorsum of tongue. Magnetic resonance imaging showed a well-defined T2-weighted hyperintense lesion in the left half of anterior portion of tongue measuring 3.2 cm × 2.5 cm × 1.7cm [Figure 1]. There was no midline extension. There was no significant cervical lymphadenopathy. A possibility of carcinoma was entertained clinically. Other parts of the oral cavity and neck were normal. Systemic examination including cardiac,

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respiratory, abdominal and central nervous system was normal. Chest X-ray and computed tomography (CT) scan were unremarkable. HIV status was non-reactive. Wide local excision of the tongue lesion was performed. Histopathological examination revealed a diffuse infiltrate of small lymphoid cells with scanty cytoplasm, irregular hyperchromatic nucleus and inconspicuous nucleoli. There were admixed few epithelioid histiocytes, eosinophils and rare multinucleated giant cells. Areas of necrosis were noted. The cells were present below the epithelium and infiltrated into the deep skeletal muscle fibers. The overlying mucosa was not involved [Figure 2]. Immunohistochemical staining was performed on 5μm sections of formalin fixed paraffin embedded tissue samples using the Ventana automated immunostainer. The list of antibodies used is shown in Table 1. The cells were positive for CD3, CD4, CD8 (CD8 > CD4), CD2 (few cells), CD7 (few cells). The cells were negative for CD20, CD30, ALK1 and CD56. There was aberrant expression of CD5. Ki67 was high at the invasive front (around 30–40%) and low in rest of the areas (around 3–5%) [Figure 2]. CD68 was positive in the histiocytes. Postoperatively, whole body positron emission tomography- Computed tomography (PET-CT) scan did not reveal any significant lesion elsewhere in the body. Clonal population of T cells was seen by T-cell receptor gamma chain rearrangement assay [Figure 3]. The patient was categorized as StageI since postoperative PET-CT scan was normal. The final diagnosis was peripheral T-cell lymphoma (PTCL) The lesion was completely excised and since the patient was in Stage I, a close follow was advised without any adjunctive chemotherapy/radiotherapy. One year later, the patient was lost for follow-up.

Table 1: The list of antibodies used in Immunohistochemistry

Antibody	Clone	Source
CD3	Polyclonal	Dako
CD20	L26	Cell Marque
CD4	Polyclonal	Dako
CD8	Polyclonal	Dako
CD2	AB75	Biogenex
CD5	4C7	Cell Marque
CD7	CBC.37	Dako
CD68	PG-M1	Dako
CD30	Ber-H2	Dako
CD56	Polyclonal	Dako
ALK 1	Alk1	Dako
Ki 67	MIB1	Dako

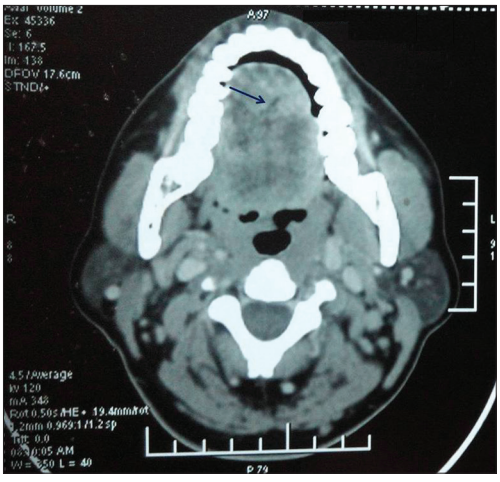

Figure 1: Magnetic resonance imaging scan image showing an ill-defined lesion in the left half of anterior portion of tongue

DISCUSSION

About 20–30% of NHLs arise from extranodal sites.[1] Gastrointestinal tract being the most common extranodal region followed by head and neck region. Lymphomas represent third most common malignant lesion of the oral cavity following squamous cell carcinomas and salivary gland neoplasms and account for

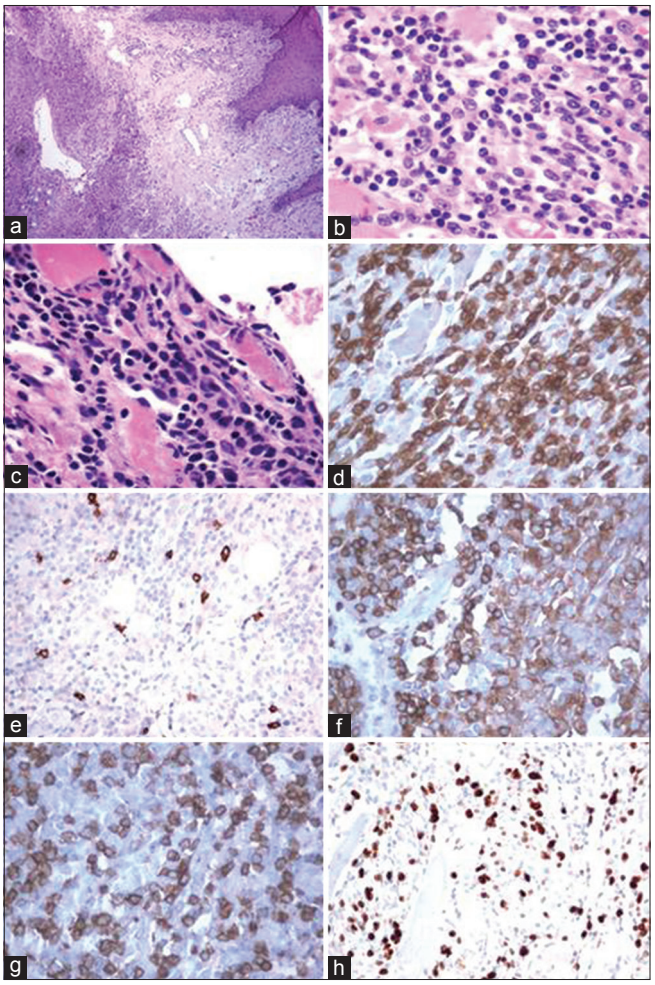

Figure 2: (a) Submucosal lymphoid infiltrate with overlying uninvolved mucosa (H&E stain, ×40). (b) Sheets of small lymphoid cells with hyperchromatic nucleus, scant cytoplasm (H&E stain, ×200). (c) Cells infiltrating skeletal muscle fibres (H&E stain, ×400). (d) Lymphoid cells staining positive for CD3 (IHC stain, ×100). (e) The cells were negative for CD20 (IHC stain, ×100). (f) Strong positivity for CD8 more than CD4 (IHC stain, ×100). (g) The cells showing positivity for CD4 (IHC stain, ×100.(h) High Ki67 proliferation index at the invasive front (IHC stain,×100)

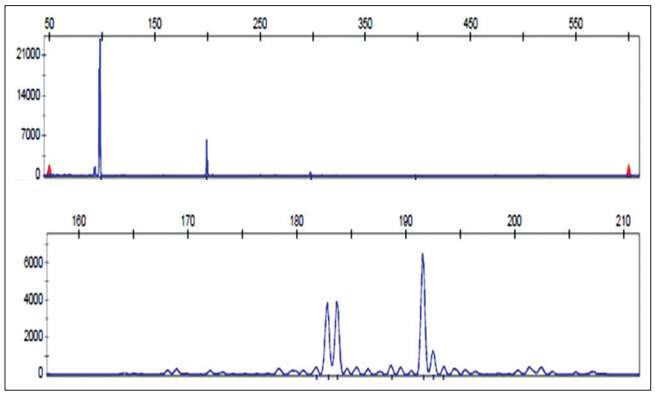

Figure 3: Clonal population of T-cells seen by T-cell receptor gamma chain rearrangement assay

3–5%.[5] DLBCL has been reported as the most common type of primary NHL.[3] Most common sites are tonsils followed by palate, tongue, buccal mucosa and retromolar area.[2] Only a small number of PTCLs of oral cavity have been reported so far.[4]

Little is known about the etiological factors of primary lymphomas of the oral cavity. Apart from age, there are several risk factors associated with NHL.[6-8]

1. Primary or acquired immunodeficiency
2. Autoimmune disease (Sjogren's syndrome, Coeliac disease, treatment with immunosuppressant drugs)
3. Infective agents such as herpetic virus, hepatitis C virus, *Helicobacter pylori*
4. Exposure to chemical agents
5. Hereditary factors.

It usually affects the elderly over sixth decade of life.[9] The most common symptoms are pain, swelling and discomfort in the throat. Oral NHLs may mimic benign oro dental conditions and hence are easily misdiagnosed.[10] The biological behavior and therapeutic options of oral cavity lymphomas are difficult to interpret due to paucity of cases. Similar to the lymphomas of other regions, oral cavity lymphomas are quite sensitive to both chemotherapy and radiotherapy. The prognosis is related to the stage of the tumor, the aggressiveness of the cell type and response to treatment.[11] Went *et al*.[12] reported that Ki67 is prognostically relevant. New predictive score included age >60 years, high LDH, poor performance status and Ki-67 >80% which are associated with poor prognosis. Our patient was 50 -year-old and had normal LDH value. Ki-67 was around 30–40% at the invasive front. These parameters seem to have a good outcome in this case. Wolvius *et al*.[13] reported median survival of 34 months with no difference in prognosis between patients with bone and soft tissue lymphoma. Our patient did not have tumor anywhere else in the body which was confirmed on PET-CT scan and is in good health at the end of 1 year of treatment.

CONCLUSION

NHL of the oral cavity is an uncommon disease and most of the cases reported in the literature are B-cell NHLs.[1] PTCLs are extremely rare with only a few cases reported in the literature.[4] We report a case of primary PTCL of the tongue of low-grade morphology and good prognosis.

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Conflicts of interest
There are no conflicts of interest.

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