## T-cell lymphoma of oral cavity: A rare entity

Preethi Sharma, Madhuri Gawande, Minal Chaudhary, Rajul Ranka

Departments of Oral Pathology and Microbiology, Sharad Pawar Dental College,
Datta Meghe Institute of Medical Sciences, Wardha, Maharashtra, India

## **Abstract**

Lymphomas are heterogeneous malignancies of the lymphatic system characterized by lymphoid cell proliferation. They can broadly be divided into Hodgkin's lymphoma (HL) and non-HL (NHL). NHL can originate from B, T or natural killer lymphocytes. Extranodal presentation of T-cell NHL is extremely rare and is often seen in immunocompromised individuals. Here, we report a rare case of T-cell lymphoma of the oral cavity in a 13-year-old patient. The patient was diagnosed to have T-cell lymphoma on the basis of biopsy and immunohistochemistry and was referred to the oral surgery department for definite treatment.

Keywords: Immunohistochemistry, lymphoma, oral cavity, palate

Address for correspondence: Dr. Asst.prof. Preethi Sharma, Department of Oral Pathology and Microbiology, Sharad Pawar Dental College, Datta Meghe Institute of Medical Sciences, Sawangi (Meghe), Wardha, Maharashtra, India.

E-mail: dr.preethisharma@gmail.com

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#### INTRODUCTION

Lymphomas are defined as heterogeneous malignancies of the lymphatic system characterized by a proliferation of lymphoid cells or their precursors. According to the WHO definition,[1] lymphomas are classified as non-Hodgkin's lymphoma (NHL) or Hodgkin's lymphoma (HL). NHLs represent 86% of all lymphomas. NHL is the second-most common oropharyngeal malignancy after squamous cell carcinoma. Oral lymphomas account for 2.2% of all head and neck malignancies, 5% of salivary gland tumors, 3.5% of intraoral malignancies and 2.5% of all cases of lymphomas.<sup>[2]</sup> There is an increasing incidence of NHL in patients with autoimmune disease, including coeliac disease, immune suppression from HIV, rheumatoid arthritis and Sjogren's syndrome.[3] Diagnosis of NHL in the oral cavity may result from mucosal tissue or gingival swelling, or masses.[4]

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#### CASE REPORT

A 13-year-old male patient [Figure 1] reported to the Outpatient Department of Sharad Pawar Dental College and Hospital, Sawangi (Meghe), Wardha, with a chief complaint of swelling with pain for 1 month. The patient was apparently alright 1 month back when he noticed a swelling on the left side of the face which was initially small in size and has gradually increased to present size of 6 cm × 4 cm. The lesion was firm, ill-defined, tender, nonmobile and was associated with pain which was pricking in nature. No history of fever, trauma and bleeding or pus discharge had been reported for the patient. Medical and dental history was not contributory. On inspection, the face was bilaterally asymmetrical due to diffuse swelling on the left side of size 6 cm × 4 cm approximately, roughly oval in shape, smooth in surface, color same as adjacent skin with ill-defined margins. The swelling was extending anterior posteriorly (A/P) from the left corner of mouth

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Figure 1: Extraoral swelling on the face

to left preauricular region, superior inferiorly (S/I) from infraorbital region to level of the left corner of the mouth. Temperature was not raised; tenderness present, consistency was soft to firm. Intraorally, a single-diffuse swelling seen in upper left palatal region of the jaw of size 4 cm × 3 cm approximately, roughly oval in shape, surface was ulcerated, color was same as that of adjacent mucosa, the swelling was extending A/P – from upper left 1–7 region, S/I – from mid-palatal region to depth of vestibule with ill-defined borders [Figure 2].

Orthopantomogram did not show any relevant findings[Figure 3]. On further examination, the patient was found to be systemically fit with all the blood count values to be in normal range. Biopsy was performed under local anesthesia which led to the diagnosis of round cell malignancy.

Under microscopic examination, low-power view showed sheets of uniform, monotonous round cells separated by thin connective tissue septa at places [Figure 4]. The round cells are diffusely arranged throughout [Figure 5]. Numerous endothelial lined blood vessels of varying shape and sizes are seen with intravasated red blood cell counts. The neoplastic round cells are seen invading into muscle tissue suggestive of muscle invasion. Under high-power view, the round cells showed hyperchromatic nuclei occupying the entire cell with a thin rim of eosinophilic cytoplasm. At places, cells show nuclear pleomorphism [Figure 6]. With these features, a diagnosis of round cell malignancy was made.

Round cells can be seen in Ewing's sarcoma family tumors, rhabdomyosarcoma, osteosarcoma and lymphoma. To rule out Ewing's sarcoma, Periodic acid—Schiff stain was performed which came out negative. Immunohistochemistry



Figure 2: Picture showing intraoral swelling on the left side of the palate

was performed to rule out other small round cell tumor. Leukocyte common antigen/CD45 came out positive suggesting the diagnosis of lymphoma [Figure 7]. For further typing, CD3 and CD20 markers were applied for T-cell and B-cell lymphoma, respectively. CD3 came out positivity suggesting the final diagnosis of T-cell lymphoma [Figure 8].

#### **DISCUSSION**

Lymphomas are heterogeneous malignancies characterized by proliferation of lymphoid cells or their precursors. They can be classified as HL or NHL.[1] NHL can further be subclassified into B-cell, T-cell or natural killer/T-cell types. NHL presents more commonly than HL, representing about 86% of all lymphomas, and B-cell lymphomas are seen more frequently than T-cell lymphomas.<sup>[5]</sup> HL rarely shows extranodal disease (1% cases), whereas NHL presents as extranodal disease in approximately 23%-30% of cases.<sup>[6]</sup> Extranodal NHL is commonly seen to involve gastrointestinal tract, Waldeyer's ring, skin, bones and others. Oral cavity involvement is seen in only 2% of cases. [7] Due to the rare presentation of extranodal T-cell NHL in oral cavity, clinicians often find difficulties in proper management of such patients. Various oral subsites involving T-cell NHL as mentioned in literature include labial commissure, gingiva, palate, maxilla, buccal mucosa, floor of the mouth, gingivobuccal sulcus, tongue and uvula.<sup>[5,7,8]</sup> These tumors show a wide spectrum of behavior ranging from relatively indolent to highly aggressive and potentially fatal course. [9] It has slowly grown from a rare cancer to the fifth-most common cancer in the world over a period of 30 years. [10] On reviewing the literature, mixed treatment outcomes in oral T-cell NHL were seen, and it was observed to be unrelated to the site of the tumour. Complete remission with chemotherapy or radiotherapy, though less commonly seen, has been reported in patients



Figure 3: Orthopantomogram showing no relevant findings

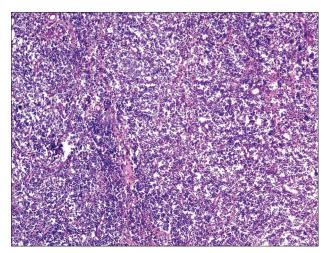


Figure 5: Picture showing diffusely arranged round cells throughout

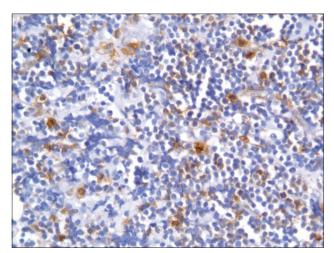


Figure 7: Picture showing leukocyte common antigen-CD-45 positive

with involvement of labial commissure and oral mucosa by Villa et al., and May et al.[5,11,12]

NHL has commonly been associated with HIV and is considered the second-most common HIV-associated malignancy after Kaposi's sarcoma. The risk of NHL is 60 times greater in patients with HIV disease than in otherwise healthy persons.<sup>[13]</sup> T-cell lymphoma has

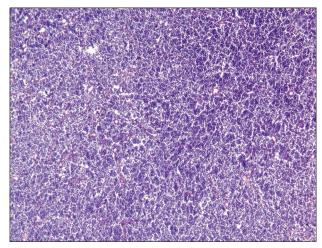
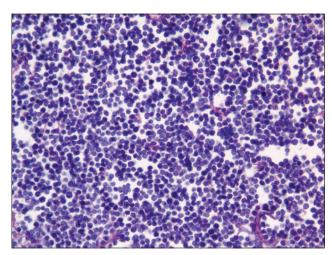


Figure 4: Low-power view showing sheets of uniform, monotonous round cells separated by thin connective tissue septa at places



**Figure 6:** Picture showing round cells with hyperchromatic nuclei occupying the entire cell. Nuclear pleomorphism is also seen at places

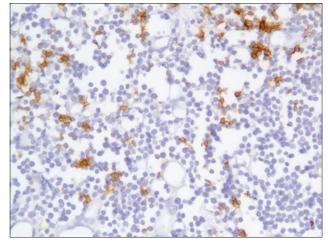


Figure 8: Picture showing CD3 positive

occasionally been reported with systemic conditions such as celiac sprue, Crohn's disease and several autoimmune diseases.<sup>[14]</sup> However, the patient in our case report was

not found to be suffering from HIV-AIDS, any other systemic illness or immune compromising condition. Immunohistochemistry plays an important role in the diagnosis and classification of hematolymphoid neoplasms. The peripheral T-cell lymphomas are about 90% positive for CD3 and CD45RO while B-cell markers include CD19, CD20, CD30 and CD 79. New cell and lineage markers are constantly being discovered and added to the existing list of antibodies. [7,10]

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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#### Conflicts of interest

There are no conflicts of interest.

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