

Case Report

Base of tongue as an unusual site for Non Hodgkin Lymphoma

Chenthilnathan P¹, Mohamad P², Razini F³, Samarendra M⁴

Abstract

Background: Non Hodgkin Lymphoma (NHL) has about 3-7% of oral manifestation. Most of the lesions are from the palate, gingival, tongue, buccal mucosa, floor of the mouth and lips. These site consist approximately 2% of all extra nodal lymphomas as primary origin. The prognosis of NHL depends to the tumour stage, tumour aggressiveness and their response to treatment. The oral lesions appear to respond quite well to irradiation or chemo radiation. **Methods:** We present a case of a 68 year-old Malay male presented with symptoms of voice change, poor oral intake and bleeding from oral cavity, which turns out to be NHL of the base of tongue without visceral involvement.

Keywords: Non Hodgkin Lymphoma, lymphoma, tongue

Introduction:

Lymphomas are malignant neoplasm of the lymphocyte which mainly involves lymph nodes, spleen and other non haemopoietic tissues¹. They are mainly classified as Hodgkin or Non Hodgkin Lymphoma. It can be either of B-lymphocyte or T-lymphocyte in origin¹.

NHL comprises a heterogeneous group of lymphoid neoplasm with a wide spectrum ranging from indolent to highly aggressive and fatal form². About 25% of NHL are of extra nodal origin arise in head and neck region but very rare in Hodgkin's disease³. Among NHL's extra nodal presentations which typical location are stomach, bowel, lung, orbital tissue, thyroid, tonsil, salivary glands, breast, testis and kidney². Waldeyer's ring is the second in common after gastrointestinal tract in the incidence of extra nodal NHL. Furthermore primary lymphomas of the oral cavity with base of tongue as site of origin are still rare and uncommon.

Oral manifestations are seen in 3-5% of cases of NHL³. The oral cavity, including the palate, gingival, tongue, buccal mucosa, floor of the mouth and lips is the primary site of approximately 2% of all

extra nodal lymphomas². Location of oral lymphomas is more frequent in masticator mucosa (hard palate, gingival, dorsum of the tongue) than in movable mucosa (soft palate, floor of mouth, lips, ventral part of tongue, buccal mucosa)⁴. The lingual and buccal mucosa is rarely involved whereas the gingival vestibule and Waldeyer's ring is the most frequent site of occurrence⁴.

Case Report:

A 68 year-old Malay male patient presented with mild throat discomfort for the past 6 months duration. It was associated with left neck swelling which was present and increasing in size over the same duration. He denies any constitutional symptoms such as fever, night sweat and fatigue. He was chronic smoker and had stopped the habit 10 years ago. Initially there was small swelling at back of tongue, ulcer like lesion which progressive increased in size. The ulcer was traumatized during mastication. He denied any history of betel nut or leaf chewing, tooth extraction and radiation. He had loss appetite and loss of weight about 10 kg in last two month and also developed muffled voice. Occasionally he also noted to have blood-stained saliva.

1. P Chenthilnathan, Department of ORL-HNS, School of Medical Sciences, Universiti Sains Malaysia.
2. Irfan Mohamad, MMed (ORL-HNS), Department of ORL-HNS, School of Medical Sciences, Universiti Sains Malaysia.
3. Ireen Razini, Department of Pathology, School of Medical Sciences, Universiti Sains Malaysia.
4. Mutum Samarendra, Department of Pathology, School of Medical Sciences, Universiti Sains Malaysia.

Corresponds to: Dr Irfan Mohamad, Department of Otorhinolaryngology-Head & Neck Surgery, School of Medical Sciences, Universiti Sains Malaysia Health Campus, 16150 Kota Bharu, Kelantan, Malaysia.
Email: irfan@kb.usm.my

Base of tongue as an unusual site for Non Hodgkin Lymphoma

Clinical examination revealed fungating mass arising from base of tongue (Fig.I). The tongue movement was mildly restricted. The mass extended posteriorly into oropharynx. Occasionally he had mild tachypneic on supine position but there was no stridor. The tongue mass was hard, indurated in consistency with ulcerated surface. The mass was not bleeding on touch. The mass measured about 4cm in diameter. Bilateral neck nodes were palpable on the left level II, III and IV, diffused, matted and fixed (the largest was 6x4cm) and right level II and III (the largest was 2x2cm). Neither axillary lymph node nor hepatosplenomegaly were palpable.

A CT scan base of skull till abdomen showed homogeneous mass arising from base of tongue and occupying the whole oropharynx extending superiorly to the level of soft palate, palatine tonsils. The mass caused obstruction to the post nasal space with inferior extension to pyriform fossa, leaving minimal residual airway (Fig. II & III). The nasopharynx, vallecula and parapharyngeal space are clear. The vocal cord is not well visualized. Multiple cervical lymphadenopathy were noted and mainly at the left submandibular region.

Tissue punch biopsy from the base of tongue mass revealed the presence of neoplastic lymphocytes infiltrating the tissues, particularly the epithelium. Multiple fragments neoplastic cells composed of diffuse large lymphoid cells. The neoplastic cells were large with pleomorphic nuclei and mitotic changes. The covering stratified squamous epithelium show focal infiltration by similar tumour cells. Mitotic figures with thin blood vessels are seen in between tumour sheets. The tumour cells are positive for LCA, CD20 and CD79a (Fig IV-VI).

The diagnosis of primary diffuse large B cell NHL was made. The patient was started on chemotherapy and a prophylactic tracheostomy under local anaesthesia was performed before chemotherapy anticipating acute upper airway obstruction. He was started with three cycle of chemotherapy, CHOP regime (doxorubicin, vincristine, cyclophosphamide and prednisolone). No complication were noted during and till the chemotherapy completed. He responded very well after completed three cycle of chemotherapy. The tongue mass and the cervical neck lymph nodes subsided in size and his appetite was improving. Unfortunately he defaulted follow up for further CT scan assessment.

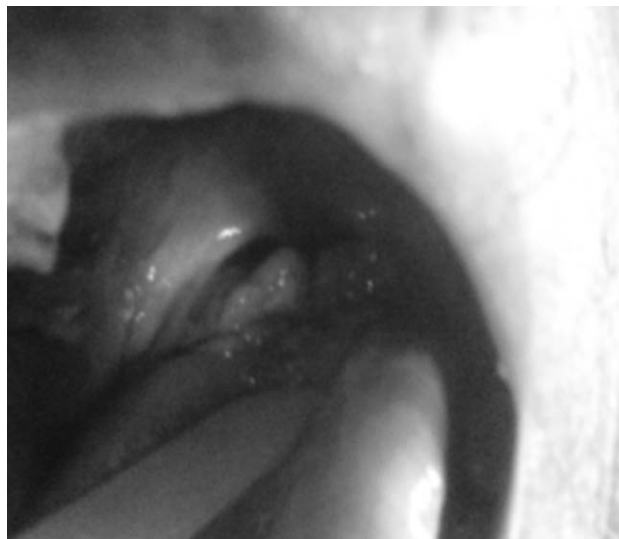


Figure I: A huge fungating mass arising from the base of tongue extending towards the anterior pillar of soft palate

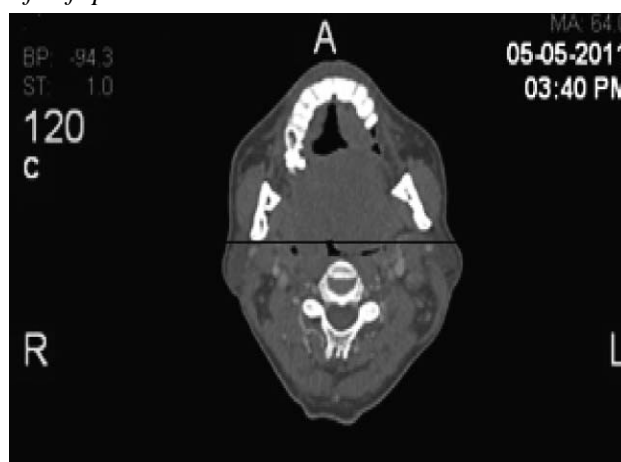


Figure II: Axial view CT scan of oral cavity showing the tumour mass occupying the oropharynx (Arrow) with minimal residual airway.

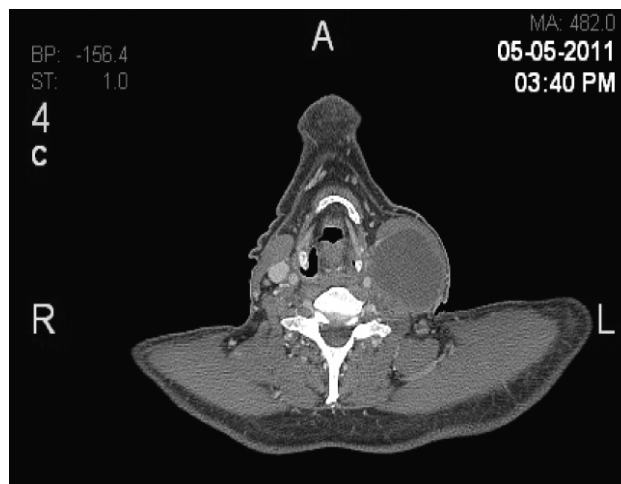


Figure III: axial view CT scan of the neck showing bilateral lymphadenopathy with prominent large left level II, III and IV enlargement

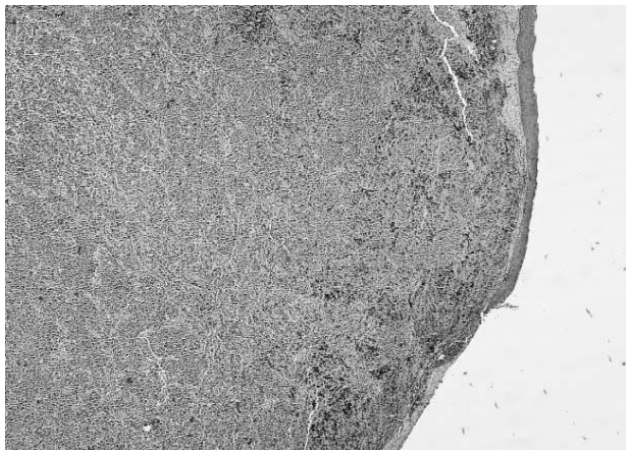


Figure IV: Microscopic view showing diffuse infiltration of the tumour cells with focal infiltration of the stratified squamous epithelium x4 (H & E stain).

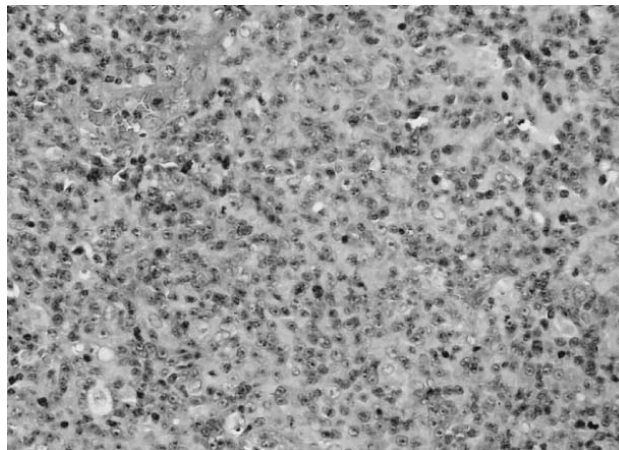


Figure V: Microscopic view showing large tumour cells with vesicular nuclei and prominent single nucleoli with mitosis x 40 (H & E stain)

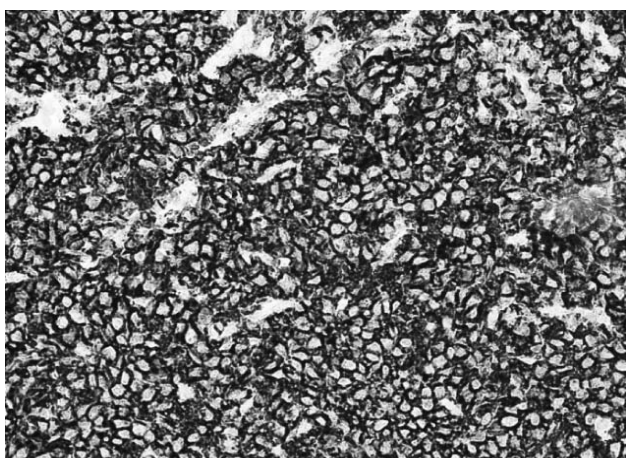


Figure VI: Immunohistochemical stain for CD20 shows diffuse positivity, confirming that this is of B cell immunotype.

Discussion:

Lymphomas represent the 3rd most common group of malignant lesions of oral region, following squamous cell carcinoma and salivary gland neoplasms⁵. In addition, extra nodal NHL in the oral cavity is rare and uncommon even though NHL is about three times more common than Hodgkin's disease (HD)⁶. B-lymphoma is more common than T-lymphoma among oral cavity lymphomas.

The oral cavity contains only small amounts of lymphoid tissue and more commonly involved in NHL than HD. NHL originated from oropharyngeal lymphoid tissue for example from the Waldeyer's ring is the most commonly seen^{6,7}. NHL may affect both osseous and soft tissue of the oral cavity. The favoured sites in the oral cavity are palatal mucosa and 6 can affect bony such as palate and the less common site, the mandible⁷. Thus, lymphoma must

be part of the differential diagnosis in any oral lesion.

The most common clinical appearance of NHL in the mouth is painless non healing ulceration⁸. Most of the malignant oral ulcers are solitary and persistent. The clinical features of primary extra nodal oral NHL are not typical and can be misdiagnosed such as dental abscess or recurrent aphthous ulcers in view of longstanding non healing ulcer⁹. Thus it is important to obtain an early biopsy of oral lesion that does not resolve despite of medical treatment. The diagnosis of NHL can be made only by punch biopsy⁵, as been done in the index case.

The space-occupying effect of a local tumour mass in the oral cavity will be the most frequent presenting symptoms⁹. They include muffled voice (hot potato voice) and throat discomfort as in this particular case owing to the painless progressive swelling on the tongue.

The overall survival for NHL is less than HD¹⁰. The prognosis of NHL is related to the stage of the tumour, the aggressiveness of the malignant cell type and the response to treatment⁵. Oral lesion is relatively sensitive to chemotherapy as evidenced in this case. The mean survival was two years¹⁰. The rate of survival is influenced by the location of the primary site, histology type, stage and the health status of the patient including the age factor. However, lymphoma of the tongue carried the worst prognosis due to the nature of the disease, location of the tumour, painless mass and late symptom presentation, while the best prognosis was in the parotid and tonsillar lesion¹¹.

In conclusion, NHL has a great tendency to affect organs and tissues that do not ordinarily contain abundant lymphoid cells, such as parotid gland, paranasal sinus, oral cavity, larynx and orbit. In the oral cavity, it commonly involves the oropharyngeal lymphoid tissue comprising Waldeyer's ring besides palatal mucosa, mandible or palatal bone, the vestibule and gingival and maxilla. NHL as primary site tumour from the base of tongue as seen in this particular patient is a rare occasion. A non healing oral ulcer not responding to medication, should be investigated earlier and proceed for punch biopsy.

The oral lymphoma responds well to chemo and radiotherapy. Early punch biopsy should be done for any oral ulcer which more than two weeks duration. This diffuse large B-Lymphoma responded well to CHOP regime chemotherapy as to reduce the bulk of tumour and then planned for further chemo irradiation but he defaulted after three cycle of chemotherapy. Tracheostomy should be considered for better outcome and prevent airway complications during the treatment.

References

1. Richards A, Costelloe MA, Eveson JW, Scully C, Irvine GH, Rooney N. Oral mucosal non-Hodgkin's lymphoma-a dangerous mimic. *Oral Oncology* 2000;**36**(6):556-8. [http://dx.doi.org/10.1016/S1368-8375\(00\)00047-6](http://dx.doi.org/10.1016/S1368-8375(00)00047-6)
2. Kobler P, Bovcic J, Filipovic Zore I, Nola M, Sertic D. Primary non Hodgkin's lymphoma of the oral cavity. *Oral Oncology Extra* 2005;**41**(1):12-4. <http://dx.doi.org/10.1016/j.ooe.2004.10.002>
3. Soderholm AL, Lindqvist C, Heikinheimo K, Forssell K, Happonen RP. Non-Hodgkin's lymphomas presenting through oral symptoms. *Int Oral Maxillofac Surg* 1990;**19**(3):131-4. [http://dx.doi.org/10.1016/S0901-5027\(05\)80126-2](http://dx.doi.org/10.1016/S0901-5027(05)80126-2)
4. Zanakis SN, Kambas I, Gourlas PG. A non-Hodgkin's lymphoma in the buccal mucosa. A case report. *Oral Surg Oral Med Oral Pathol* 1992;**74**(3):340-2. [http://dx.doi.org/10.1016/0030-4220\(92\)90072-X](http://dx.doi.org/10.1016/0030-4220(92)90072-X)
5. Griffin TJ, Hurst PS, Swanson J. Non-Hodgkin's lymphoma: a case involving four third molar extraction sites. *Oral Surg Med Oral Pathol* 1988;**65**(6):671-4. [http://dx.doi.org/10.1016/0030-4220\(88\)90006-0](http://dx.doi.org/10.1016/0030-4220(88)90006-0)
6. Wilson TG, Wright JM. Non-Hodgkin's lymphoma of the gingival: review of the literature. Report of a case. *J Periodontol* 1986;**57**(3):155-8. <http://dx.doi.org/10.1902/jop.1986.57.3.155>
7. Barber HD, Stewart JC, Baxter WD. Non-Hodgkin's lymphoma involving the inferior alveolar canal and mental foramen: report of a case. *J Oral Maxillofac Surg* 1992;**50**(12):1334-6. [http://dx.doi.org/10.1016/0278-2391\(92\)90240-Z](http://dx.doi.org/10.1016/0278-2391(92)90240-Z)
8. Shima N, Kobashi Y, Tsutsui K, et al. Extra nodal non-Hodgkin's lymphoma of the head and neck. A clinicopathologic study in the Kyoto-Nara area of Japan. *Cancer* 1990;**66**(6):1190-7. [http://dx.doi.org/10.1002/1097-0142\(19900915\)66:6<1190::AID-CNCR2820660619>3.0.CO;2-U](http://dx.doi.org/10.1002/1097-0142(19900915)66:6<1190::AID-CNCR2820660619>3.0.CO;2-U)
9. Wolvius EB, van der Valk P, van der Wal JE, et al. Primary extra nodal non-Hodgkin's lymphoma of the oral cavity. An analysis of 34 cases. *Eur J Cancer B Oral Oncol* 1994;**30B**(2):121-5. [http://dx.doi.org/10.1016/0964-1955\(94\)90063-9](http://dx.doi.org/10.1016/0964-1955(94)90063-9)
10. Miller RI. Non-Hodgkin's lymphoma of the lip: a case report. *J Oral Maxillofac Surg* 1993;**51**(4):420-2. [http://dx.doi.org/10.1016/S0278-2391\(10\)80359-3](http://dx.doi.org/10.1016/S0278-2391(10)80359-3)
11. Howell RE, Handlers JP, Abrams AM, Melrose RJ. Extra nodal oral lymphoma. Part II. Relationship between clinical features and the Lukes-Collins classification of 34 cases. *Oral Surg Oral Med Oral Pathol* 1987;**64**(5):597-602. [http://dx.doi.org/10.1016/0030-4220\(87\)90066-1](http://dx.doi.org/10.1016/0030-4220(87)90066-1)