An Unusual Presentation of Supraglottic Low-Grade B-Cell Non-Hodgkin's Lymphoma with Tracheostomal Myiasis

Abstract

60-year-old male patient presented with dysphagia and a change in voice for eight months. It was established after Direct laryngoscopy surgery and biopsy, that it was a low-grade B cell non-Hodgkin lymphoma. The primary lesion is resolved with Rituximab, Cyclophosphamide, Doxorubicin, Vincristine, and Prednisolone regimen. Four months later, patient presented with a discharge and maggots at the tracheostomy site. Ifosfamide, Etoposide, Carboplatin was started after a secondary recurrence of disease progression. Hereby we infer this is an unusual case presentation, myiasis with lymphoma recurrence and tough exacting to the otolaryngologist as there are more chances of misdiagnosing as squamous cell carcinoma..

Keywords: *DL Scopy, Biopsy, Tracheostomy, supra glottis low grade B cell NHL, PET CT scan, Maggots, Chemotherapy, R-CHOP regimen, ICE regimen*

Rupa Mehta, Krishna Sasanka KSBS, Mudalsha Ravina[1], Nitin M Nagarkar

Departments of Otolaryngology, Head and Neck Surgery and [1]Nuclear Medicine, All India Institute of Medical Sciences, Raipur, Chhattisgarh, India

Laryngeal neoplasms are rare and, in the larynx, primary non-Hodgkin lymphomas (NHL) the second most common hematopoietic tumor and with the B-cell phenotype which accounts in less than 1% of larynx malignancies. Supraglottic primary laryngeal lymphomas with predominance over left side and in males compared to females in the age group of 4 to 81 years.[1-4] But in our case the lesion is located on the right side of larynx [Figure 1]. Very few numbers of cases in the global texts not more than 100 cases of primary laryngeal lymphoma have been reported.[1,2,5] Normally the laryngeal lymphoma commonly found in supraglottis which presents as submucosal mass without mucosal ulceration and distinguished by without any clinical and gross differential criteria, compared with squamous cell carcinoma (SCC). On review of literature myiasis overspread in live human or animal tissues mainly affects the nasal cavity, ear, cutaneous tissue, exophytic malignant growth and nonhealing ulcers. But as in our case occurrence of myiasis at the tracheostomy site is rare still exists [Figure 2] and only few cases were reported in literature. Our case post-operative Histopathological examination with IHC (Immunohistochemistry) shown low grade B cell non-Hodgkin's lymphoma [Figure 3]. IHC findings were shown both CD20, Bcl2 were positive [Figure 4] for tumor cells and CD10, Bcl6, CD23 were negative for tumor cells. CD30 stains immunoblasts. Ki 67 index was 20-30%. CD3 and CD5 stains reactive T cells. Cellular marrow with trilineage hematopoiesis and reactive changes were seen in bone marrow aspiration. Based on CT staging, biopsy and bone marrow aspiration, disease confined to the larynx rather no evidence of involvement outside the larynx. The patient underwent total six Chemo cycle R-CHOP regimen [Rituximab at a dose of

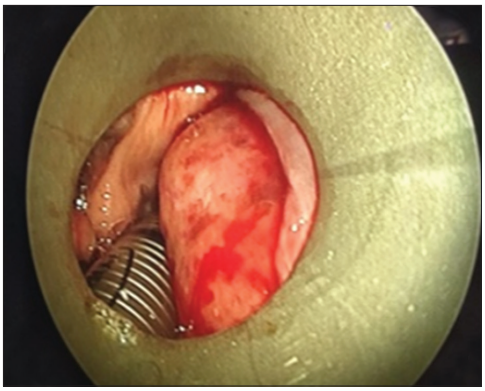

Figure 1: DL (Direct laryngoscopy) pinkish smooth surface mass present at right pyriform fossa and aryepiglottic fold

Address for correspondence:
Dr. Krishna Sasanka KSBS, Flat No. 352, Tulip-3, Parthivi Pacific, Tatibandh, Raipur - 492 099, Chhattisgarh, India.
E-mail: sasankmsentndl@gmail.com

Received: 16-09-2021
Accepted: 12-10-2021
Published: 08-07-2022

Access this article online

Website: www.ijnm.in
DOI: 10.4103/ijnm.ijnm_141_21
Quick Response Code:

How to cite this article: Mehta R, Sasanka KK, Ravina M, Nagarkar NM. An unusual presentation of supraglottic low-grade b-cell non-hodgkin's lymphoma with tracheostomal myiasis. Indian J Nucl Med 2022;37:196-8.

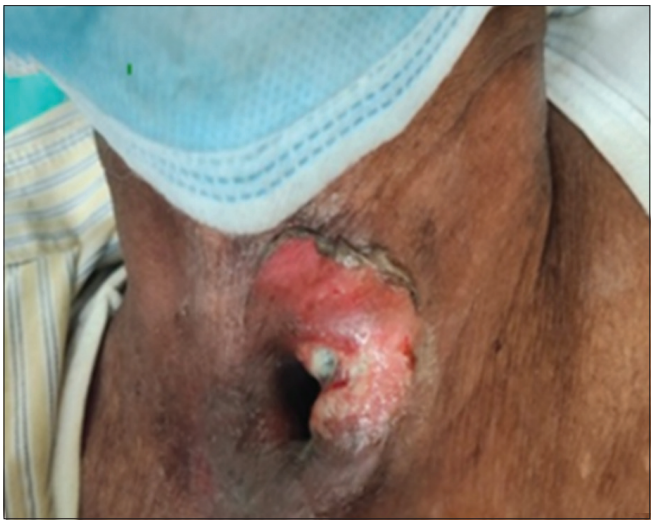

Figure 2: Tracheal stoma after removal of maggots

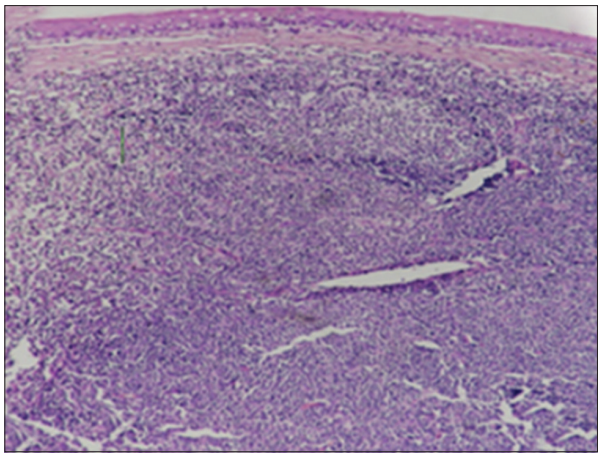

Figure 3: H and E section shows squamous epithelium and subepithelium shows lymphoid infiltrate in a vaguely nodular to diffuse pattern composed of small cleaved lymphoid cells and few large cells with round nuclei and prominent nucleoli (×10)

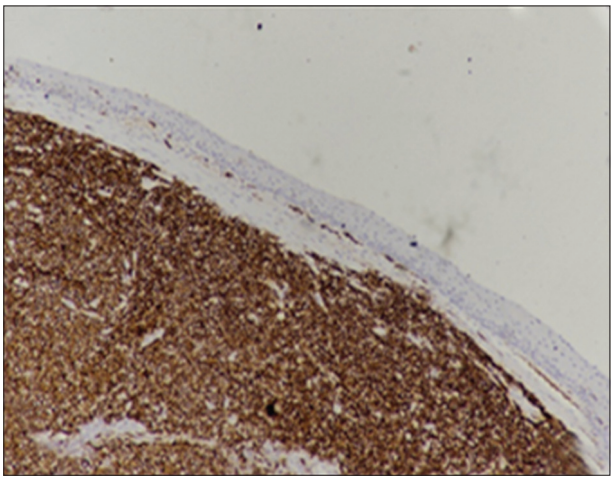

Figure 4: IHC-tumor cells show strong and diffuse positivity of CD2

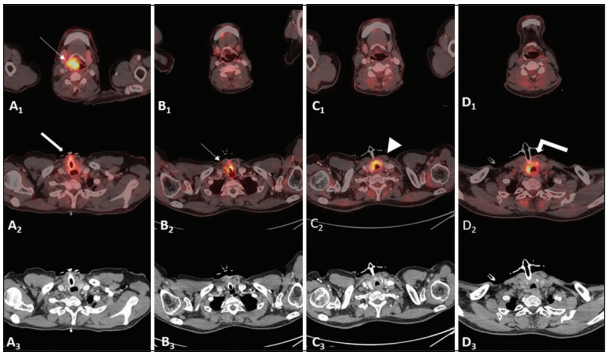

Figure 5: ^{18}F-labeled fluoro-2-deoxyglucose (FDG) positron emission tomography/computed tomography (PET/CT): (a1, white arrow), Baseline PET-CT show FDG-avid soft-tissue lesion (measuring approximate - 2.6AP cm × 2.7TR cm × 3.9CC cm, maximum standard uptake value [SUV max]-9.6) in the hypopharynx involving the right pyriform fossa with partial luminal compromise, (a2, solid arrow) show diffuse uptake around the tracheostomy tube (SUV max-5.0)-likely inflammatory. No abnormal thickening was noted on the CT image (a3). PET-CT (mid-cycle chemotherapy response assessment): (b1) complete metabolic resolution of the primary lesion (Deauville Score 1), (b2, white arrow) reveals diffuse uptake around the tracheostomy tube which is likely to be inflammatory, (b3) no abnormal thickening noted at the tracheostomy site. PET-CT (postcompletion of 6 cycles of chemotherapy): (c1) no abnormal uptake at the primary site, (c2, arrow head; c3) FDG uptake with mild thickening noted at the tracheostomy site. (d1-d3, curved arrow) is the dual time point imaging obtained after 2 h of injection showing the increase in uptake – suggestive of disease progression (Deauville 5)

375mg/m2body surface area/ IV, Cyclophosphamide - 750 mg/m2/ IV, Doxorubicin - 50 mg/m2/IV, Vincristine - 1.4 mg/m2/IV, Prednisolone - 100 mg /oral/ days to 5, for each cycle –Inj. Pegfilgrastim- 6mg/ subcutaneous]as per radiotherapist instructions. In our case, PET-CT showed complete metabolic response post chemotherapy as per the Deauville's criteria [Figure 5]. The Deauville criteria is a five-point scale. The scale scores the most intense uptake in a site of initial disease, if present as follows: score 1 (no uptake), score 2 (Uptake ≤ mediastinum), Score 3 (Uptake > mediastinum but ≤ liver), score 4 (Moderately increased uptake compared to the liver), score 5 (Markedly increased uptake compared to the liver and/ or new lesions), Score X (New areas of uptake unlikely to be related to lymphoma).[6,7] On follow up PET-CT (post mid cycle and completion chemotherapy) there was no abnormal tracer uptake noted in the hypopharynx, suggestive of complete metabolic response. After 4 months patient came with complaints of purulent discharge and foul smell at tracheostomy site. On examination at the operated site, the Portex-cuffed tracheostomy tube (size 8.0) completely blocked by secretions, crusts and Maggots creeping all over the tracheal stoma. Tracheostomy tube was changed and treated with intravenous antibiotics, multiple wound debridement's and removal of nearly 230 live maggots within 72 hours using turpentine oil by taking appropriate precautions to prevent turpentine aspiration. Only few cases were found in literature with myiasis and the predisposing factors such as psychiatric illness, immunocompromised individuals, exposed wound with foul smelling discharge, infective dermatitis, Hansen's disease and low socioeconomic status.[8] On

repetition of PET CT FDG uptake with thickening was noted around the tracheostomy which showed significant increase on Dual time point imaging taken after 03 hours of FDG injection. This appearance is due to new lesion at tracheostomy opening suggestive of disease progression (Deauville 5). Second line chemotherapy ICE regimen [Ifosfamide-7000mg, Etoposide - 140 mg, Carboplatin - 370 mg, Mesna - 7000 mg (3500 mg 4 hr before& 4 hr after Ifosfamide)] has been started. NHL of larynx in view of its rarity, more chances of misinterpreting the case as SCC. By this case it is clear to be aware of lymphoma as a key differential diagnosis for various types of laryngeal tumours. Even though tracheostomal myiasis is rare but still exists in our case so there is need to educate the patient in aspects of tracheostomy tube care which prevents further complications..

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.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Siddiqui NA, Branstetter BF 4th, Hamilton BE, Ginsberg LE, Glastonbury CM, *et al*. Imaging characteristics of primary laryngeal lymphoma. AJNR Am J Neuroradiol 2010;31:1261-1265.
2. Horny HP, Kaiserling E. Involvement of the larynx by hemopoietic neoplasms. An investigation of autopsy cases and review of the literature.Pathology Research and Practice, 1995; 191:130-138, 1995.
3. Ansell SM, Habermann TM, Hoyer JD, Strickler JG, Chen MG, *et al*. Primary laryngeal lymphoma. Laryngoscope 1997;107: 1502-1506.
4. Kato S, Sakura M, Takooda S, Sakurai M, Izumo T (1997) Primary nonHodgkin's lymphoma of the larynx. J LaryngolOtol 111: 571-574.
5. Markou K, Goudakos J, Constantinidis J, Kostopoulos I, Vital V, *et al.* (2010) Primary laryngeal lymphoma: Report of 3 cases and review of the literature. Head Neck 32:541-549.
6. Sally F. Barrington& Regine Kluge. FDG PET for therapy monitoring in Hodgkin and non-Hodgkin lymphomas. Eur J Nucl Med Mol Imaging (2017) 44 (Suppl 1):S97–S110.
7. Min-Li Zhou, Kui Zhao, Shui-Hong Zhou, Qin-Ying Wang, Zhou-Jun Zheng, *et al*. Role of PET/CT in the diagnosis, staging, and follow-up of a nasal-type natural killer T-cell lymphoma in the larynx: a case report and literature review. Int J Clin Exp Med 2014;7(11):4483-4491.
8. M. D. A. Vitavasiri, FICS, M. D. P. Charoenchasri, M. S. S. Kaewmanee, and M. D. M. Bhaibulaya, "Subdermal myiasis caused by maggots of chrysomyiabezziana," Siriraj Hospital Gazzetee, vol. 47, no. 5, pp. 419–422, 1995.