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Case Report

A case report of Extranodal natural killer / T-cell lymphoma presenting as extensive myiasis

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ABSTRACT

We described a 56-year-old Indonesian man with one-month history of nasal obstruction and rapidly increasing nasal swelling and two weeks history of severe facial pain with foul smelling discharge, bleeding and noticing live maggots emerging from his nasal and oral cavity. On examination, he appeared cachexic with a markedly swollen, erythematous and deformed external nose. Live maggots, pus and necrotic tissues were found in both nasal cavities, with erosion of upper gingiva and hard palate. Patient was managed initially with tracheostomy under local anaesthesia, followed by removal of around 350 live maggots from the nasal cavities and debridement of necrotic tissues. A midline nasal cavity mass which extended laterally into the nasal cavity was found, along with a large defect over the gingival labial sulcus with necrotic hard palate and a communication between oral and nasal cavities. Biopsies were taken from the nasal mass, gingiva and hard palate. Histopathological results from the biopsies showed diffuse, aggressive infiltrative malignant lymphoid cells with widespread angionecrosis, consistent with features of extranodal NK/T-cell lymphoma. To our knowledge, there is only one other reported case where the diagnosis of ENKTCL was made after patient presented with oro-nasal myiasis. Clinicians should have raised awareness on this atypical presentation so that further investigation and management can be implemented promptly.

1. Introduction

Myiasis in the oro-nasal region is uncommon [1] and represent an opportunistic infection of dipterous larvae feeding on hosts' viable or necrotic tissues [2]. Some authors have reported oro-nasal myiasis in patients with known non-Hodgkin lymphoma due to a combination of immunocompromised state and poor hygiene. Here, we describe a case of patient with extensive myiasis as the first presentation of underlying extranodal NK/T-cell lymphoma (ENKTCL). This paper has been reported in line with the SCARE criteria [3].

2. Report of a case

A 56-year-old Indonesian man with no known medical illness presented with one month history of nasal obstruction and nasal swelling, which rapidly increased in size causing difficulty in mouth opening. He also complained of two weeks history of severe facial pain as well as foul smelling discharge, bleeding and noticing live maggots emerging from his nasal and oral cavity

On examination, he appeared cachexic (weighing 38kg) with a markedly swollen, erythematous and deformed external nose. Live maggots, pus and necrotic tissues were found in both nasal cavities, with erosion of upper gingiva and hard palate (Fig. 1a and b). Other physical examinations were normal with no maggot infestations of the tongue, floor of mouth, nasopharynx and larynx. Skull X-ray showed a midline erosion of the alveolar process of the maxillary bone (Fig. 2).

Patient was managed initially with tracheostomy under local anaesthesia, followed by removal of around 350 live maggots from the nasal cavities and debridement of necrotic tissues at the columella, philtrum and upper lip. A midline nasal cavity mass which extended laterally into the nasal cavity was found, along with a large defect over the gingival labial sulcus with necrotic hard palate and a communication between oral and nasal cavities. Biopsies were taken from the nasal mass, gingiva and hard palate. A Ryles tube was inserted for feeding and patient was started on IV antibiotics and Turpentine dressing [4].

Histopathological results from the biopsies showed diffuse, aggressive infiltrative malignant lymphoid cells, which were medium to large sized displaying pleomorphic vesicular nuclei, coarse chromatin clumps

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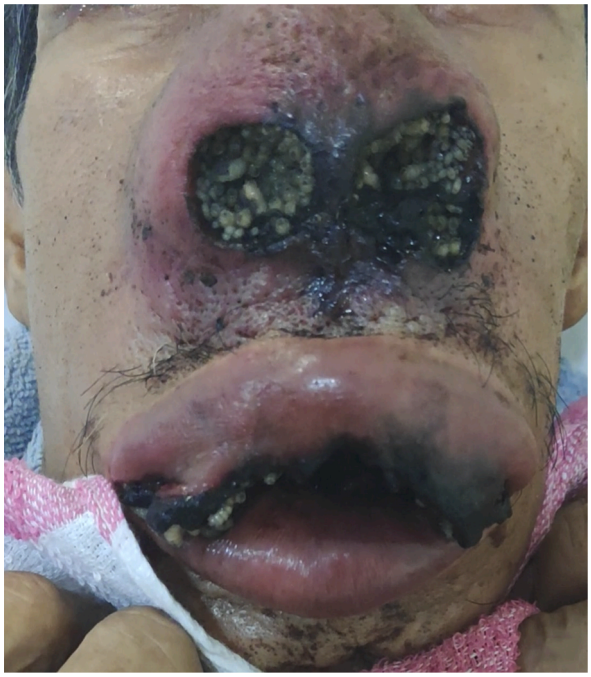

Fig. 1a. Myiasis and necrosis of nasal and oral cavities.

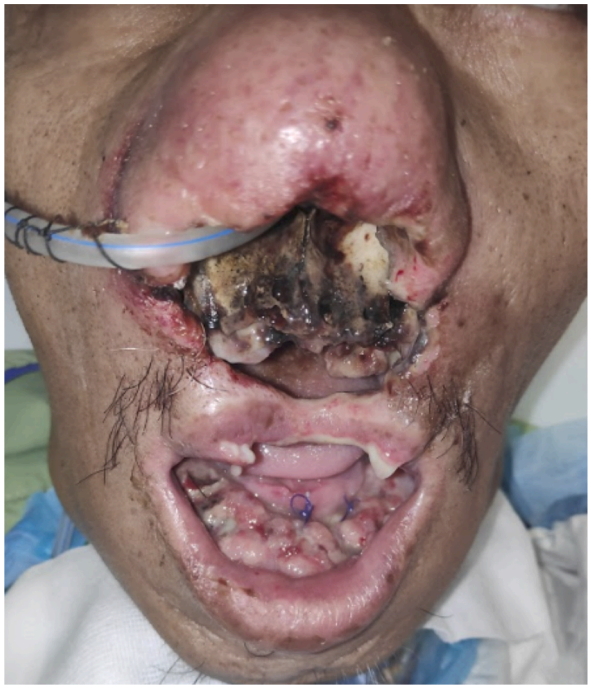

Fig. 1b. Erosion of upper gingiva and hard palate.

to conspicuous nucleoli, mixed with small lymphocytes, with widespread angionecrosis. Immunohistochemistry of the biopsy sample was positive for CD2 and CD56 but negative for CD3. These findings were consistent with features of nasal ENKTCL.

Unfortunately, patient could not afford further imaging investigation or treatment here and was transferred back to Indonesia for further haematological and ENT management.

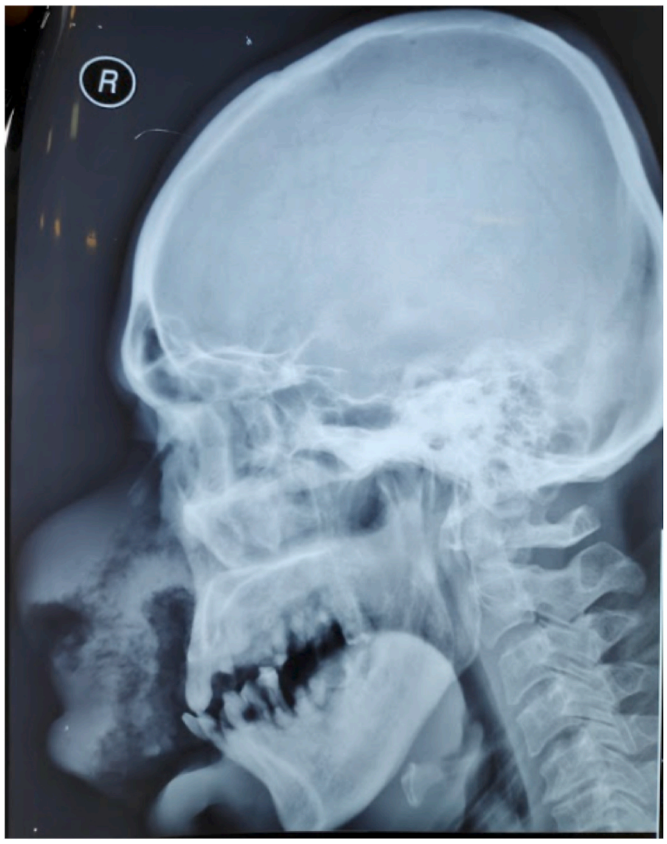

Fig. 2. Skull X-ray showing erosion of maxillary bone.

3. Discussions

Extranodal NK/T-cell lymphoma (ENKTCL) is a rare form of non-Hodgkin lymphoma, more commonly found in Asia and South America, with strong association with EBV infection [5]. This disease most commonly involves the sinonasal cavity with male predilection (male to female ratio of 3:1) and a mean age of onset of 52[5].

Patients with ENKTCL (nasal type) usually present with non-specific symptoms such as nasal obstruction, epistaxis and facial swelling [5]. However, as the disease progresses, patients may have midline facial destruction with eventual necrosis of upper anterior aerodigestive tract [1]. These necrotic and ulcerated areas are at risk of myiasis without proper hygiene and wound dressing [1].

Diagnosis of ENKTCL requires large biopsies of suspicious areas where the histopathology usually shows variable cell sizes with zonal necrosis and polymorphic infiltration of small lymphocytes, plasma cells and eosinophils [6]. Immunohistochemistry staining of nasal ENKTCL are typically positive for CD2, cytoplasmic CD3e and CD56 but negative for surface CD3 [6]. Extent of disease are evaluated with computer tomography (CT) scan while magnetic resonance imaging (MRI) are used for visualization of soft tissue involvement [5].

Treatment of ENKTCL includes chemotherapy with or without adjuvant radiotherapy [5]. On the other hand, treatment of myiasis includes injection of lidocaine, occlusive coating of Vaseline ointment, clear fingernail polish and surgical extraction of larvae [7]. Patients with underlying ENKTCL should also be educated about personal hygiene, adequate wound care and sanitation of surroundings including control of fly population, especially in those living in rural areas and from lower socio-economic class [1].

To our knowledge, there is only one other reported case [6] where the diagnosis of ENKTCL was made after patient presented with oro-nasal myiasis. Clinicians should have raised awareness on this atypical presentation so that further investigation and management can

be implemented promptly.

Ethical approval

No ethical approval was required as this is a retrospective case report.

Sources of funding for your research

None.

Author contribution

Eugene Hung Chih WONG: data interpretation, writing paper, final editing, Gabriel Xia Peng QUAH: data collection, writing paper, Doh Jeing YONG: writing paper, final editing.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Registration of research studies

1. Name of the registry: N/A
2. Unique Identifying number or registration ID:
3. Hyperlink to your specific registration (must be publicly accessible and will be checked):

Guarantor

Eugene Hung Chih WONG.

Declaration of competing interest

None.

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None.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.103419.

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