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Primary non hodgkin's lymphoma of lacrimal sac presented as recurrent acute dacryocystitis: A case report

Noor Shirlyna Irma Ngah[1], Ramiza Ramza Ramli[2], Adil Hussein[1*], Hasnan Jaafar[3]

[1]*Department of Ophthalmology, School of Medical Sciences, UniversitiSains Malaysia, 16150 KubangKerian, Kelantan, Malaysia.*

[2]*Department of Otorhinolaryngology, School of Medical Sciences, UniversitiSains Malaysia, 16150 KubangKerian, Kelantan, Malaysia.*

[3]*Department of Pathology, School of Medical Sciences, UniversitiSains Malaysia, 16150 KubangKerian, Kelantan, Malaysia.*

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ABSTRACT

Primary non Hodgkin's lymphoma of the lacrimal sac is uncommon but potentially delay in diagnosis as it may mimic the presentation of primary post saccal nasolacrimal duct obstruction. In this article, we reported a case of primary non Hodgkin's lymphoma of the lacrimal sac presented with recurrent acute dacryocystitis in a young lady. A 28 years old Malay lady had history of persistent epiphora on left eye for 4 years. Prior to presentation to our clinic, it was preceded by progressive recurrent painful medial canthal swelling for 6 months duration. Clinical diagnosis of post saccal naso lacrimal duct obstruction was made and otorhinolaryngology assessment revealed intranasal mass. Endoscopic excision was done showed diffuse large B cell lymphoma on histology. The patient was started on 6 cycles of chemotherapy which subsequently result in no recurrence of the tumour post chemotherapy. Any case of post saccal nasolacrimal duct obstruction should be investigated thoroughly as it may become as a presentation of other sinister pathology.

1. Introduction

Dacryocystitis is an infection of the lacrimal sac. It is usually due to nasolacrimal duct obstruction and present with epiphora, redness and swelling of medial canthus. Primary nasolacrimal duct obstruction is the most common with histologic finding of chronic inflammation and fibrosis leading to occlusion of the lacrimal drainage system. Neoplasm, systemic inflammatory disease, infection or trauma may result in secondary obstruction of the lacrimal drainage system.

Primary non Hodgkin's lymphoma of the lacrimal sac is rare. Majority of the lymphomas involving lacrimal sac represents secondary involvement[1]. The presenting features are usually nonspecific, including epiphora, medial canthal swelling and redness which may be misleading. It may delay in diagnosis as it mimic primary nasolacrimal duct obstruction[2, 3].

We reported a case of primary non Hodgkin's lymphoma of the lacrimal sac clinically presented with recurrent acute dacryocystitis.

2. Case report

A 28 years old Malay lady presented with left eye persistent epiphora for the past 4 years. It was preceeded by progressive

*Corresponding author: Associate Professor Dr Adil Hussein, Senior Lecturer, Department of Ophthalmology, School of Medical Sciences, UniversitiSains Malaysia, 16150 KubangKerian, Kelantan, Malaysia.
Tel: +6016-7976977
Fax: 09-7653370
E-mail: adilh@usm.my

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recurrent painful medial canthal swelling 6 months prior to the presentation to our clinic. On further history, she revealed episodes of nasal block and epistaxis from left nostril. Ocular examination revealed visual acuity of both eyes 6/6. Anterior and posterior segment examinations of both eyes were unremarkable. There was presence of firm non tender, non-fluctuating swelling at the left medial canthus about 3cm x 3cm in size with discharging sinus (Figure 1). Lacrimal system probing and syringing revealed a hard stop with negative post nasal drip. Clinical diagnosis of post saccal naso lacrimal duct obstruction was made.

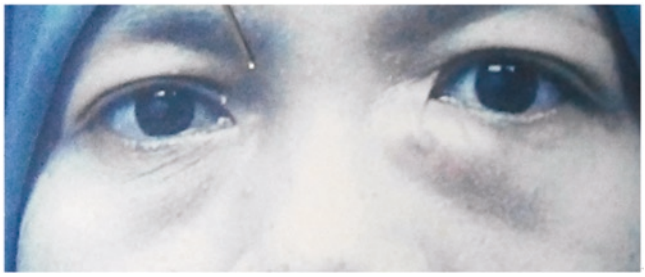

Figure 1. The clinical appearance of the left medial canthal swelling

She was referred to otorhinolaryngology team to rule out any intranasal pathology before external dacryocystorhinostomy surgery. Rigid rhinoscopy revealed polypoidal mass occupying the whole left nostril, originating from the middle meatus and pushing the inferior turbinate laterally (Figure 2). Computed Tomography of the orbit and paranasal sinus showed ill-defined enhancing soft tissue mass within left nasal cavity extended into left maxillary sinus, vomer bone, ethmoid bone, nasolacrimal duct and lacrimal sac (Figure 3). It showed features of aggressive soft tissue mass of left nasal cavity.

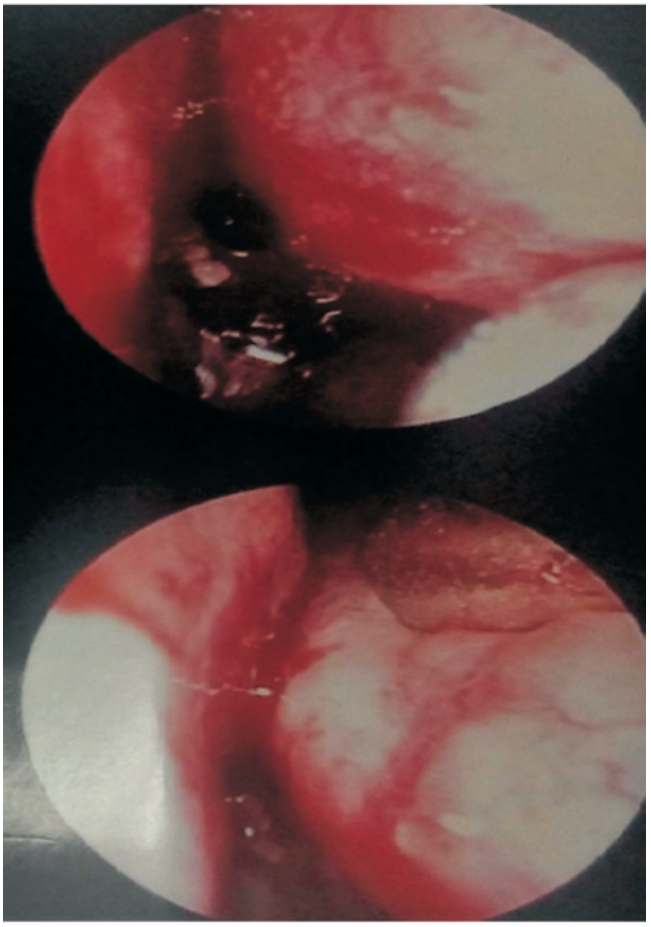

Figure 2. The nasal mass at left middle meatus viewed from rigid rhinoscopy

Patient then underwent endoscopic excision of nasal mass, uncinectomy, middle meatal antrostomy and total inferior turbinectomy. The tumor mass biopsied from left nasal cavity, left lacrimal sac, left maxillary ostium and left floor of nose. The histopathologic study of

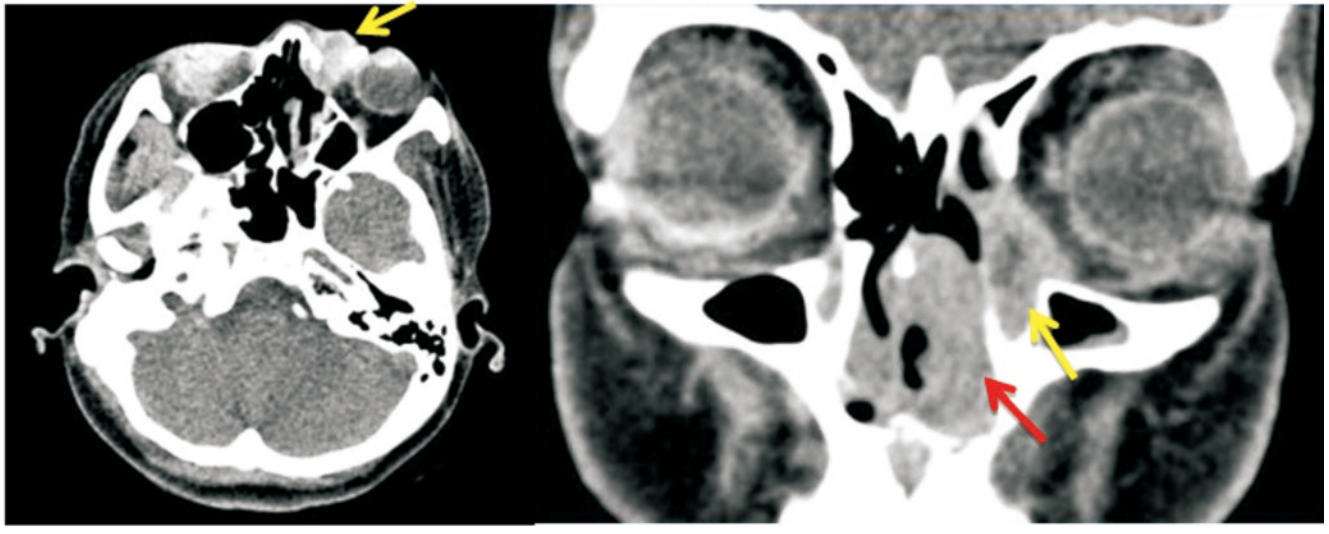

Figure 3. Computed tomography (axial and coronal) of the left orbit with soft tissue mass of left nasal (red arrow) and left lacrimal sac (yellow arrow)

the masses revealed diffuse neoplastic lymphoid cells predominantly large cells (Figure 4 and 5). The malignant cells expressed a B-cell antigen (CD20) and lacked T-cell (CD3). Interpretation of diffuse large B cell lymphoma (DLBCL) was given. For clinical staging, in addition to initial physical examination and biochemical tests, CT scan of the chest of the abdomen and bone marrow biopsy did no show evidence of lymphomatous involvement elsewhere. Patient was classified as stage 2E non Hodgkin's lymphoma by the Ann Arbor staging system.

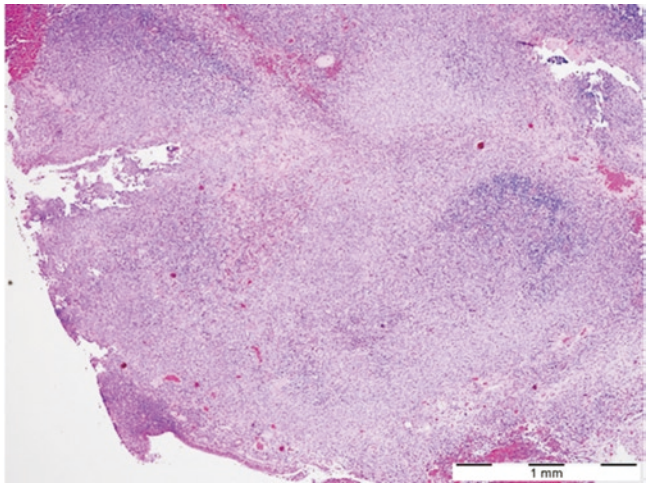

Figure 4. Left lacrimal sac biopsy exhibiting diffuse sheet of large neoplastic lymphoid cells partially covered by respiratory epithelium. (H&E, 40X magnification).

She was treated with 6 cycles of chemotherapy R-CHOP protocol (Rituximab, Cyclophosphamide, Doxorubicin hydrochloride, Vincristine and Prednisolone) plus triple intrathecal chemotherapy (Dexamethasone, Methotrexate and Cytarabine). Post completed 6 cycles of chemotherapy, the medial canthal swelling reduced in size and patient had no more epiphora. Follow up CT of the paranasal sinuses, brain, thorax and abdomen post third and sixth cycles of chemotherapy revealed no recurrence of the tumour or systemic involvement (Figure 6).

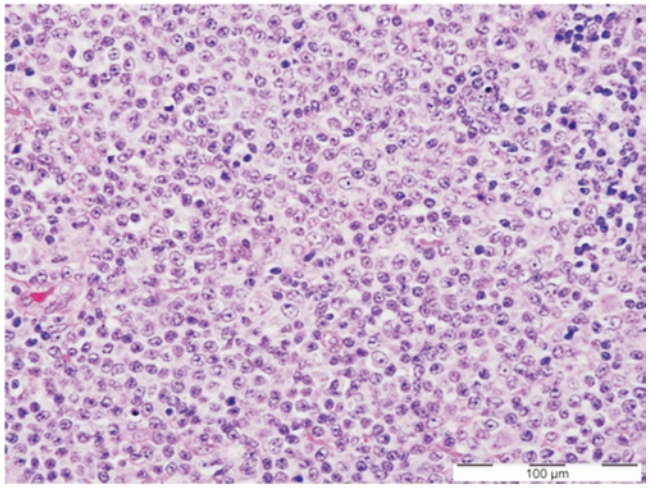

Figure 5. High power view of the tumour cell shows large cells with round vesicular nuclei with admixture of centroblast and immunoblast. (H&E, 400X magnification).

4. Discussion

Lacrimal sac tumors are uncommon which more than 90% of the tumors in various series are malignant and epithelial in origin[1]. Lymphoproliferative lesions of nasolacrimal duct are exceedingly rare. Most of case reported representing secondary involvement. The peak incidences of lacrimal sac tumor are in fifth decade of life[4]. Localized painless or painful medial

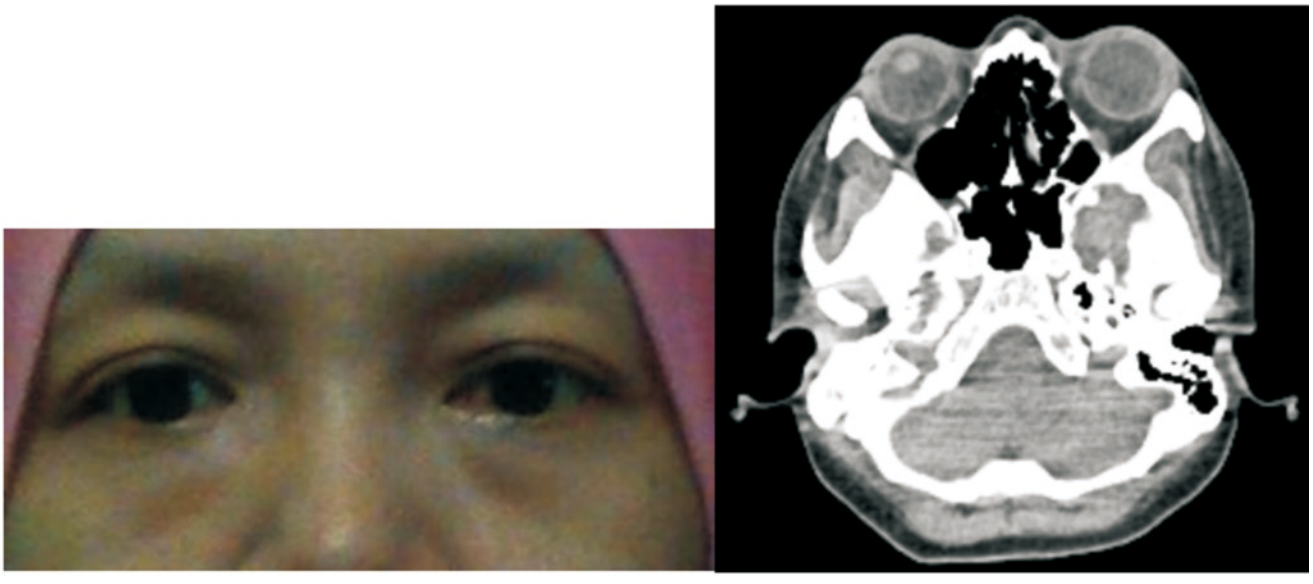

Figure . The clinical appearance of improving medial canthal swelling and Computed tomography (Axial) of the orbit showed no recurrence of tumour.

canthal swelling with nasolacrimal duct obstruction are typical presentation of a lacrimal sac tumor. Other common initial presentations are epiphora and dacryocystitis . These nonspecific clinical manifestations may mimic to primary acquired lacrimal duct obstruction[3-6].

Our patient presented at the age of 28 years old which is younger compare to other cases reported before[2-6]. Review done by Gao et al[7]showed mean age at the time of diagnosis was 56.4 years old. She presented with typical symptoms and signs of chronic naso lacrimal sac obstruction. However history of nasal blockage and occasional epistaxis raise our suspicious of more sinister intranasal pathology. Nasolacrimal sac tumour may arise from the sac or structure surrounding the sac or duct involving nasolacrimal duct or paranasal sinuses. Previous reported cases of primary non Hodgkin's lymphoma of the lacrimal sac showed involvement of nasolacrimal duct and adjacent paranasal sinuses and nasal cavity [8]. Rajendran Punitan et al[5]also described CT scan findings of large mass of extraconal lesion extending to the nasal cavity.

Endoscopic surgical debulking and chemotherapy are choices of treatment in our patient. The study done by SN Madge et al[9] revealed various types of treatment for ocular adnexal DLBCL including chemotherapy, local radiotherapy or combination of both or with surgical debulking. Radiotherapy and chemotherapy had favourable prognosis of lacrimal sac lymphoma[10]. High survival rate of localized DLBCL relate with large proportion of patients receiving chemotherapy as treatment and used of Rituximab in addition to CHOP. Some patients received combination with local radiotherapy[9].

4. Conclusion

Otorhinolaringology evaluation is mandatory in any case of post saccal nasolacrimal duct obstruction to rule out any intranasal or sinus pathology. Prompt referral and treatment will improve the overall prognosis of the patients.

Conflict of interest statement

The authors report no conflict of interest.

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