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Case report

Cystic mucosa-associated lymphoid tissue lymphoma of lacrimal gland associated with vision loss: A case report

Jacqueline Mupas-Uy [a], Yoshiyuki Kitaguchi [a], Yasuhiro Takahashi [a], Emiko Takahashi [b], Hirohiko Kakizaki [a, *]

[a] Department of Oculoplastic, Orbital & Lacrimal Surgery, Aichi Medical University Hospital, 1-1 Yazako-Karimata, Nagakute, Aichi, Japan
[b] Department of Pathology, Aichi Medical University Hospital, 1-1 Yazako-Karimata, Nagakute, Aichi, Japan

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ABSTRACT

Purpose: To report an atypical case of cystic extranodal marginal zone B-cell lymphoma of mucosa-associated lymphoid tissue (MALT lymphoma) of lacrimal gland associated with vision loss.
Observations: An 89-year-old woman was presented with a rapidly progressing proptosis, lagophthalmos, and vision loss. Endophthalmitis was also present. Computed tomography scan images showed a hyperdense mass with hypodense cystic areas occupying the superolateral orbit, which displaced the globe antero-inferiorly with optic nerve compression and stretching. An erosion to the adjacent superior and lateral orbital walls was also demonstrated. Complete tumor excision was performed via upper transconjunctival orbitotomy concurrently with enucleation. The immunohistopathological diagnosis was MALT lymphoma.
Conclusions and importance: This case emphasizes the importance of considering lymphoma in the differential diagnosis of a cystic superolateral orbital mass.
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1. Introduction

Malignant lymphoma is a neoplasm derived from clonal proliferation of lymphocytes which leads to formation of solid tumors.[1] It is the most common malignant tumor of the orbit in patients older than 60 years.[1] Extranodal marginal zone lymphoma of mucosa-associated lymphoid tissue (MALT lymphoma) is a low grade lymphoma and it is the most frequently observed subtype in the orbit.[2] It often arises from the lacrimal gland, which usually presents as indolent, slow-growing, solid mass without a cystic lesion, or bone erosion or destruction.[3,4] This study reports a case of lacrimal gland MALT lymphoma with atypical presentation, which showed a rapid enlargement of cystic lesion, associated with vision loss.

1.1. Case report

An 89-year-old woman presented with a right upper eyelid palpable mass for 6 months, which was rapidly enlarging in a month. The patient had severe dementia.

At the first examination, her visual acuity was no light perception and direct pupillary light reflex was lost in the right eye. The mass was firmly elastic with a smooth surface. There was proptosis and inferior displacement of the globe. Lagophthalmos was present. Ophthalmologic examination showed chemosis, corneal ulcer in the interpalpebral zone and hypopyon in the right eye (Fig. 1). Contrast-enhanced computed tomography (CT) showed a hyperdense, well-defined mass with a hypodense cystic area with enhanced wall and septa measuring 35 × 40 × 45mm in the superolateral orbit (Fig. 2). The mass followed the globe contour, but there was globe displacement with optic nerve compression and stretching. An erosion to the adjacent superior and lateral orbital walls was also demonstrated. Pre-operative differential diagnosis was schwannoma or adenoid cystic carcinoma, with associated endophthalmitis secondary to exposure keratopathy and subsequent corneal ulceration.

* Corresponding author. Department of Oculoplastic, Orbital & Lacrimal Surgery, Aichi Medical University Hospital, 1-1 Yazako-Karimata, Nagakute, Aichi 480-1195, Japan.
E-mail address: cosme@d1.dion.ne.jp (H. Kakizaki).

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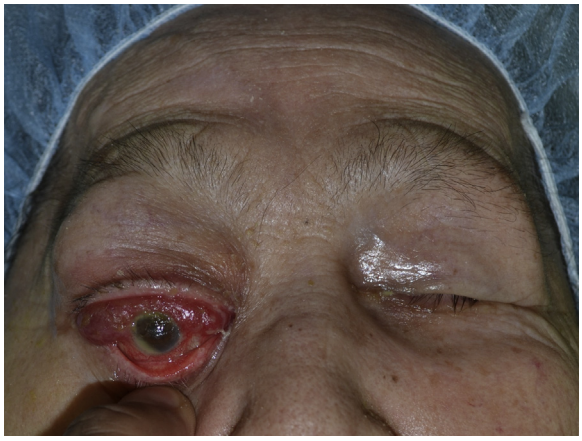

Fig. 1. Pre-operative photograph. This shows a mass in the upper eyelid with proptosis, inferior globe displacement, chemosis and hypopyon of the right eye.

She underwent tumor excision via superior transconjunctival orbitotomy. A pale, pinkish, multi-lobulated mass was encountered. Rupture of the mass expressed non-viscous fluid. Although intra-operative frozen section demonstrated the diagnosis of malignant lymphoma, the tumor was removed completely (Fig. 3) to eliminate necessity of additional management for the orbital lesion. Enucleation was also performed after tumor excision.

Post-operative histopathological examination revealed many lymphocytes and plasma cells infiltrating the lacrimal gland, with few germinal centers. The cyst consisted of dilated ducts with lympho-epithelial lesion, containing fluids with lymphocytes, histiocytes and degenerated substances. Tumor cells were positive for CD20, CD79a, and bcl-2, but negative for CD3, CD5, CD10, bcl-6, cyclin-D1. In situ hybridization showed kappa was greater than lambda (Fig. 4A–C). Immunohistochemical findings supported the diagnosis of MALT lymphoma. Fluid in cysts composed of lymphocytes, histiocytes, and degenerated substances (Fig. 4D). Infiltration of neutrophils in the cornea, anterior chamber, iris and vitreous were present and no involvement of lymphoma.

At the time of last examination, 2 months after surgery, no evidence of recurrence of MALT lymphoma in the orbit was observed. The patient's family refused review of other systemic lesions.

2. Discussion

We presented an atypical case of cystic MALT lymphoma of lacrimal gland associated with vision loss. Lacrimal gland lymphoma is usually identified as diffuse enlargement and axial elongation of the gland, which follows the contour of the globe,[5] and without producing bone erosion or destruction.[4] In this case, lymphoma was not considered pre-operatively because of its cystic appearance, aggressive course and bone involvement, although it followed the globe contour. Cystic appearance on CT scan rather raises the differential diagnosis of schwannoma and adenoid cystic carcinoma.[6] Adenoid cystic carcinoma especially shows aggressive course and bone involvement.[7]

MALT lymphoma rarely appears as cyst in the orbit. A cyst is defined as a closed sac with a distinct membrane compared to the nearby tissue. Whereas, this tumor arises from non-encapsulated clusters of lymphocytes in mucosal tissues and does not form a membrane. The mechanism of cyst-like formation in this case was predominantly due to lacrimal duct obstruction by the tumor, which impeded the outflow and caused accumulation of fluids containing lymphoma cells leading to the atypically rapid enlargement of the tumor.

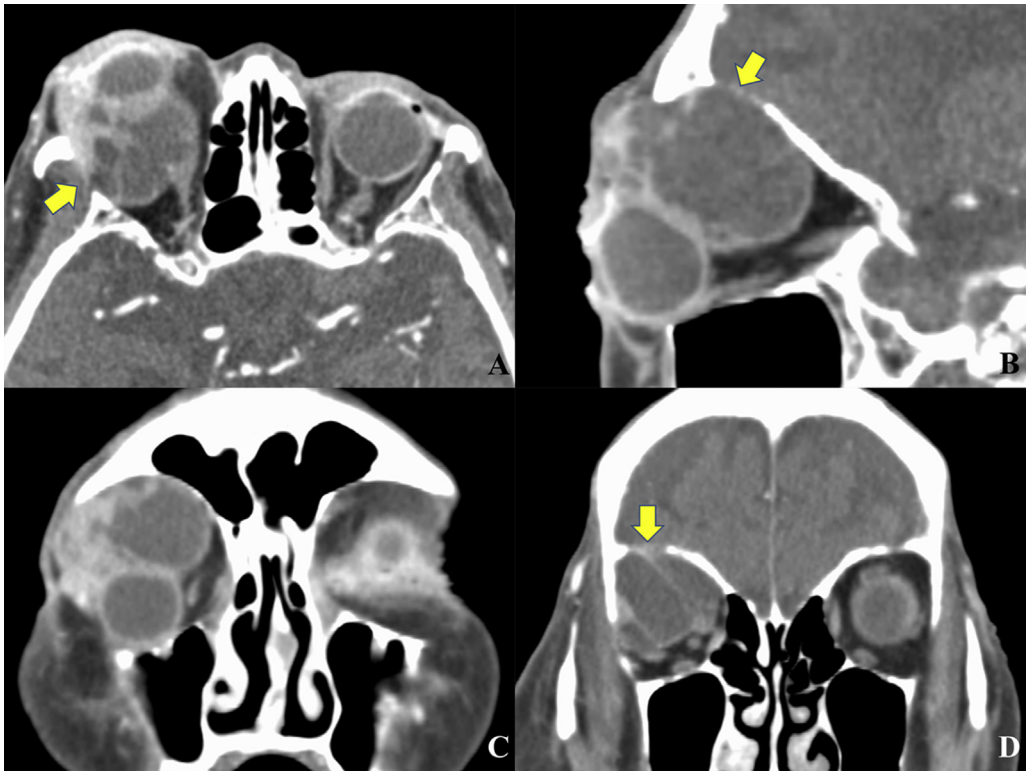

Fig. 2. Pre-operative **(A)** axial, **(B)** saggital, and **(C, D)** coronal contrast-enhanced computed tomographic images. A superolateral hyperdense mass with a hypodense cystic area, which follows the globe contour and erodes the adjacent superior and lateral orbital walls (arrows).

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