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CASE REPORT

Intraorbital ectopic lacrimal gland mimicking malignant orbital tumor

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Abstract

A 6-year-old boy with left proptosis which was realized 2 months earlier was evaluated. The left eye movements were restricted in all gaze positions. The left lacrimal gland was hypertrophic on examination. An orbital magnetic resonance imaging revealed a mass lesion starting from the lacrimal gland region extending through the superior and lateral orbit causing a pressure on the lateral rectus muscle. An incisional biopsy from both the lacrimal gland and the orbital part of the mass revealed no tumor cells but minimally inflamed lacrimal gland tissue which supported an ectopic lacrimal gland in the orbit. Although rare, ectopic lacrimal gland of the orbit might mimic orbital malignancies in children. Histopathologic confirmation is mandatory for differential diagnosis.

Keywords: Ectopic lacrimal gland; lacrimal gland; orbit; proptosis.

"Ectopic lacrimal gland" is defined as lacrimal gland located elsewhere rather than its normal location.[1] Ectopic lacrimal gland – which is a choristoma – is an uncommon condition and can be detected in conjunctiva, limbus, cornea, and rarely in the orbit.[1–3] It occurs due to sequestration of a portion of normal lacrimal gland at an abnormal site during embryogenesis. These lesions, especially the orbital located ones, can clinically simulate a neoplasm.[4]

Herein, a 6-year-old boy with a histopathologically proven ectopic orbital lacrimal gland that mimicked malignant orbital tumor is presented.

Case Report

A 6-year-old boy with left proptosis was brought to the clinic with the initial diagnosis of lymphoproliferative tumor of the orbit. The proptosis was evident for 2 months and it was told to be progressive. On ophthalmologic examination, the visual acuity was 20/20 in both eyes. The lacrimal gland on the left side was hypertrophic (Fig. 1a). The anterior and posterior segment examinations, intraocular pressures were otherwise within normal limits. In the Hertel exophthalmometry measurement (100 mm), right eye was

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16 mm, and left eye was 20 mm. The left eye movements were restricted in every gaze position. The contrast sensitivity was 0.9 in the right, and 0.7 in the left eyes. The visual evoke potential was within normal limits in both eyes.

Orbital magnetic resonance imaging (MRI) was performed to detect possible orbital lesion. T1- and T2-weighted images demonstrated left lacrimal gland mass which showed similar signal intensity with the right lacrimal gland and diffuse contrast enhancement. An obvious mass effect to left ocular bulbus and lateral rectus muscle was seen (Fig. 1b-d]. The possible differential diagnosis as suggested by MRI was lymphoproliferative, infectious, and inflammatory diseases.

Under local anesthesia, through a lateral skin incision the lacrimal gland and the orbital part of the lesion were exposed. Incisional biopsy was performed. The skin was sutured with 6/0 nylon sutures. Local antibiotic ointment twice a day for a week until the sutures were removed was used.

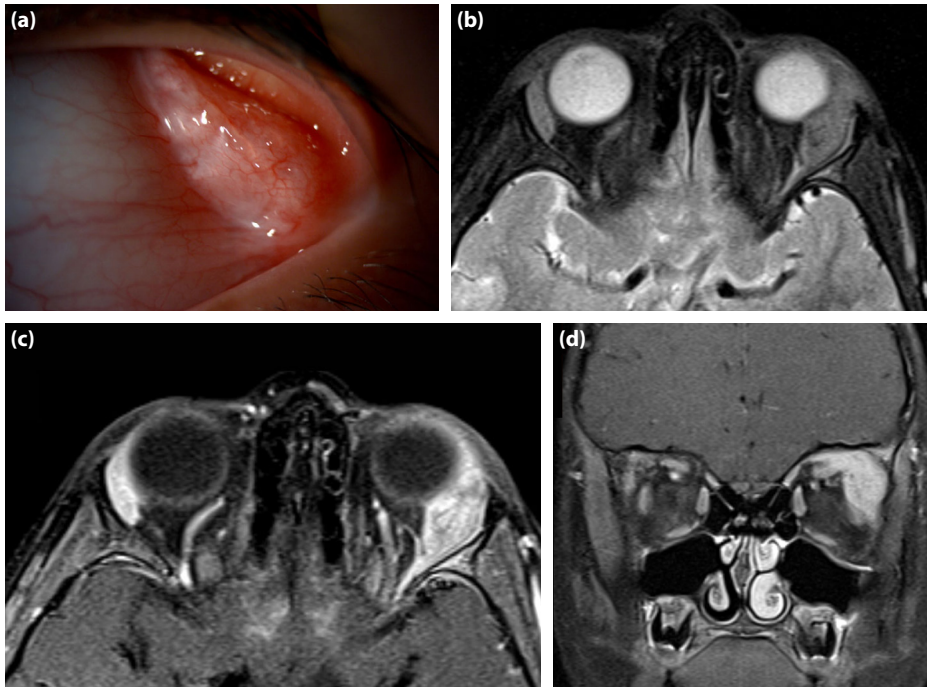

Fig. 1. **(a)** The appearance of the hypertrophic left lacrimal gland at presentation. Axial fat-sat T2W **(b)** axial **(c)** and coronal post-contrast **(d)** fat-sat T1W magnetic resonance imaging demonstrates left lacrimal gland mass that shows similar signal intensity with the right lacrimal gland and diffuse contrast enhancement.

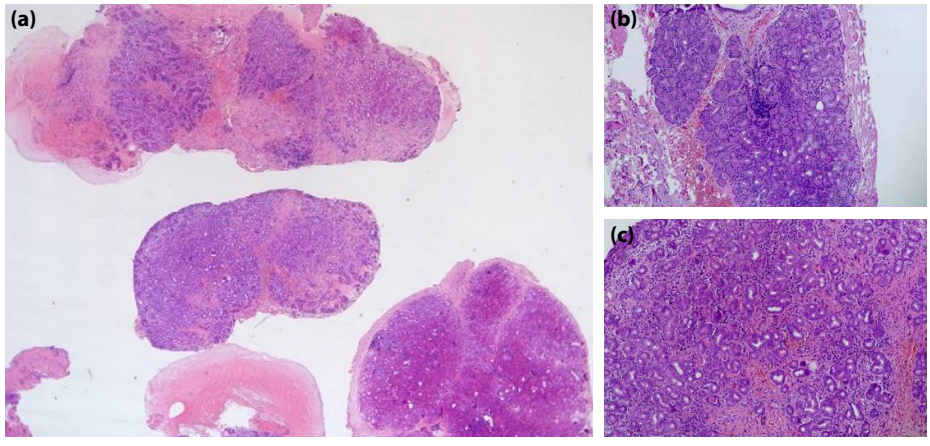

Fig. 2. **(a)** Hamartomatous lacrimal gland tissue at the orbital part of the lesion (H and E ×20), **(b)** Biopsy from the lacrimal gland revealed minimally lymphoplasmacytic cell infiltration between the glandular acini (H and E ×100), **(c)** Hamartomatous lacrimal gland tissue at the orbital part of the lesion (H and E ×200).

Histopathologic examination of the biopsy from lacrimal gland revealed minimally lymphoplasmacytic cell infiltration between the acini and ducts of the lacrimal gland (Fig. 2a and b). The biopsy from the orbital part of the lesion revealed hamartomatous lacrimal gland tissue with some inflammatory cells (Fig. 2c). No tumoral infiltration or no IgG4 related disease was detected. Consequently, the pathology report supported an ectopic lacrimal gland in the orbit.

The proptosis decreased after the surgery with no complications. No need for permanent artificial tear drop use was required. Written informed consent was obtained from the parents of the patient.

Discussion

Rapidly progressive unilateral proptosis with no adjunctive pain in children arises the idea of orbital malignancies such as rhabdomyosarcoma, neuroblastoma, and hemangioma.[5] However, many other pathologic entities which may have atypical features that can mimic malignancy should also be kept in mind. Ectopic lacrimal gland of the orbit is an uncommon condition that might resemble these life-threatening malignancies.[1–3] However, the differential diagnose needs histopathologic evaluation in most cases.

In a review of 35 lacrimal gland choristomas by Green and Zimmerman,[1] 18 were found to be at the bulbar conjunctiva, six in the outer canthus, two in the lower lid, one in the bulbus, and eight in the orbit. Although these lesions are congenital, delayed manifestation is not rare.[5] The reason of this delay is probably an inflammatory process that initiates due to a missing normal ductal system of the ectopic gland.[5,6] Supporting this thesis, an inflammatory process on histopathological examination was detected in the present case as well.

The most common symptom of the disease is unilateral proptosis caused by various degrees of secondary inflammation.[1] Sometimes, ectopic lacrimal gland might even cause destruction of the bones due to severe inflammatory reaction.[5] Moreover, pleomorphic adenoma formation in an ectopic lacrimal gland tissue that can be histopathologically discriminated should also be kept in mind.[7]

Although rare, ectopic lacrimal gland of the orbit might mimic orbital malignancies in children. Histopathologic confirmation is mandatory for differential diagnosis.

Informed Consent: Written informed consents were obtained from the parents for publication of this case report and accompanying images.

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