Complication of Ectopic Lacrimal Gland Tumor on Long-term Follow-up: A Case Report

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Dear Editor,

Ectopic lacrimal gland tissue is reported to be located at epibulbar conjunctiva, other locations including orbital, eyelid, intraocular, lacrimal sac, and nasal mucosa sites [1]. Although ectopic lacrimal glands in the ciliary body are quite rare, there have been more than 10 cases reported worldwide [2,3]. But, studies on long-term follow-up outcomes and complications are limited, although accompanying proptosis at the time of diagnosis has been frequently reported. Herein, we report a single case of ectopic lacrimal gland on ciliary body, resulting in secondary glaucoma during 20 years of follow-up. We obtained informed consent from the patient for the case report publication.

A previously healthy 10-month-old female patient presented with right upper eyelid swelling and redness. And she was admitted to an emergency room in 2003. B-scan ultrasonography and orbit computed tomography demonstrated right orbital cellulitis and a round-shaped 0.8cm ciliary body mass. Fine needle aspiration and incisional biopsy were done, which revealed ectopic lacrimal gland tumor in histology. The size of residual mass gradually increased and there was no sign of malignant transformation during 17 years of follow up period. The patient was transferred to the glaucoma clinic due to intermittent high intraocular pressure and corresponding ocular pain of the right eye once or twice a month in 2020. Thus, she started dorzolamide 2%/timolol maleate 0.5% fixed combination (Cosopt®, Merck & Co., Inc., NJ., USA). No other abnormal findings were observed on slit lamp examination.

On 10, January 2021, she was admitted to an emergency room due to ocular pain and decreased vision in the right eye that started a day ago. Best-corrected visual acuity was 2 / 20 and intraocular pressure (IOP) measured by Goldman applanation tonometer was 44mmHg. Slit lamp examination showed corneal edema and moderate anterior chamber inflammation (cell grade from +2 to +3). Anterior chamber was deep enough and there was no peripheral angle closure. Pupil was fixed and fundus was normal in the right eye. The left eye was normal. Under the impression of secondary glaucoma associated with uveitis, she was treated with additional oral acetazolamide, topical prednisolone acetate 1% (Prednilone®; Daewoo Pharm, Seoul, Korea), brimonidine 0.15% (Alphagan–P®, Allergan, Inc., Irvine, CA) and intravenous mannitol injection. IOP was in the normal range after 3 days. Although she used Cosopt and Alphagan prophylactically, there were 2 more additional acute glaucoma attacks in 2021. Slit lamp examination revealed new cystic lesion in supero-temporal iris in 2, March, 2022 (Fig. 1A). On 6, July, 2022, the cystic lesion became larger than 4 months before (Fig. 1B), but there was no
other abnormal sign such as increased IOP or anterior chamber inflammation.

On 29, July, 2022, she was admitted to emergency room with 3rd attack, presented with decreased visual acuity and 33mmHg of intraocular pressure in the right eye. The size of ciliary body mass was slightly decreased but there was flare reaction and inferior floating cells in the anterior chamber (Fig. 1C). The next day cyclitic membrane was observed more clearly in the anterior chamber (Fig. 1D). As diagnosed with uveitis associated with iris cyst rupture, on 4, August, 2022, she underwent anterior chamber and iris cyst aspiration biopsy of the right eye. The results of cytology revealed suspicious findings of lacrimal gland secretion, but no malignant cells were observed and some pigment laden macrophages existed. During two months of follow up period, IOP fluctuated from 10 to 38mmHg. Therefore, short-term follow up and additional anterior chamber irrigation to control IOP is considered.

To conclude, we report a case of secondary glaucoma associated with ectopic lacrimal gland tumor that eventually ruptured during a 20 year of long-term follow-up period. Although rare, long-term follow up revealed ectopic lacrimal gland may cause uveitis and glaucoma attacks. Our limitations are that examinations showing glaucoma status such as optical coherence tomography or visual field examination were rarely performed during the follow up period. Although optical coherence tomography did not show RNFL thinning nor macular thinning yet, follow up examinations are needed to confirm that recent increase in IOP does not lead to structural and functional damage in the future.
References


Fig. 1. Anterior segment photos of the patient with ectopic lacrimal gland tumor in the right eye. (A) On 2nd, March, 2022, a newly developed iris cyst was observed on the supero-temporal side. (B) Four months later (6th, July, 2022), the size of the iris cyst increased, but there was no IOP increase or anterior chamber inflammation. (C) She visited emergency room 3 weeks later due to increased IOP and decreased visual acuity on 29th, July, 2022. There was anterior chamber flare reaction. (D) The cyclitic membrane was observed the next day.