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Melanorrhea: Noncontiguous spread of palpebral conjunctival melanoma to the nasolacrimal duct

Raksha Rao, Santosh G Honavar, Michelle De Padua[1], Kaustubh Mulay, Vijayanand P Reddy

A 42-year-old Asian Indian male with a history of conjunctival melanoma in the left eye presented with a recurrent tumor in the upper tarsal conjunctiva. The tumor was completely excised under margin control, followed by two-staged eyelid reconstruction. During the second stage of the eyelid reconstruction, a brown-colored discharge was noted at the punctum, which on cytology was confirmatory of melanoma. Left dacryocystectomy with *en bloc* nasolacrimal duct (NLD) excision was performed. Histopathology demonstrated infiltration of the NLD by the tumor with no presence of melanoma in the lacrimal sac. Lacrimal oncorrhea is a term used to describe tumor spread by free-floating cells in the tear film. All conjunctival tumors carry a risk of tumor spread by oncorrhea.

Key words: Conjunctiva, melanoma, melanorrhea, oncorrhea, tumor spread

Conjunctival melanoma is a rare ocular tumor often complicated by high rates of recurrences and metastasis.[1-3] It can arise *de novo* or secondary to a primary acquired melanosis, or a nevus.[1] The protocol-based management of conjunctival melanoma includes wide excision with 4 mm clear margin and heavy cryotherapy to the free margins.[1] Despite these vigorous measures, conjunctival melanoma has a tendency to recur and spread systemically through lymphatogenous and hematogenous routes, and locally by direct extension.[1,2] Herein, we describe a case of spread of conjunctival melanoma by "oncorrhea," with the free tumor cells in the tear film causing indirect tumor extension into the nasolacrimal duct (NLD).

Case Report

A 42-year-old Asian Indian presented with a pigmented mass in the left eye for 7 months. There was a history of conjunctival melanoma in the same eye arising from the temporal bulbar conjunctiva treated by excision biopsy and subsequently by plaque brachytherapy elsewhere 2 years ago. There was no other significant ocular, medical, and family history.

On examination, the best-corrected visual acuity in both eyes was 20/20. Anterior segment of the right eye was unremarkable. In the left eye, a darkly pigmented mass measuring 18 mm × 16 mm × 6 mm with a nodular surface was present in the upper tarsal conjunctiva [Fig. 1a]. There was no obvious extension into the orbit, medial eyelid, or the lacrimal system. Temporal bulbar conjunctival scarring with lower fornicial symblepharon was present from previous surgery. Regional lymph nodes were unremarkable. Fundus was normal for both eyes. Systemic evaluation including computed tomography (CT) of the head and neck, chest X-ray, and ultrasound abdomen was unremarkable. A diagnosis of recurrent conjunctival melanoma of left eye was made.

The tumor was completely excised with 4 mm margin clearance obtained under frozen section control. Histopathology was confirmative of conjunctival melanoma [Fig. 1b]. Eyelid reconstruction was done by the Cutler-Beard technique Stage I. Eight weeks later during the Stage II procedure, a brown-colored discharge was noted at the punctum and pressure over the lacrimal sac yielded more such discharge [Fig. 1c]. Cytology of the discharge confirmed the presence of melanoma cells.

The patient was evaluated further, and CT of the orbits and paranasal sinuses did not reveal any mass in the lacrimal drainage apparatus. In addition, the patient was referred for a nasal endoscopy to rule out the presence of any intranasal mass. After consultation with the medical oncologist, a conservative surgery was planned for the patient. Left dacryocystectomy with *en bloc* NLD excision up to the inferior turbinate was performed. Histopathology showed the absence of tumor cells in the lacrimal sac with evidence of only subepithelial chronic inflammation [Fig. 1d], whereas the NLD showed atypical epithelioid melanocytes with partial thickness stromal invasion [Fig. 1e]. External beam radiation therapy directed to the lacrimal sac fossa, anterior lacrimal crest, and the bony NLD at a dose of 50 Gy was given. There was no local recurrence at 29-month follow-up [Fig. 1f]. The patient has been advised lifelong 6 monthly ophthalmic and nasal evaluation to note any recurrences.

Discussion

Conjunctival melanoma is a rare ocular tumor with a high tendency for local recurrences and distant spread.[1-3] It arises

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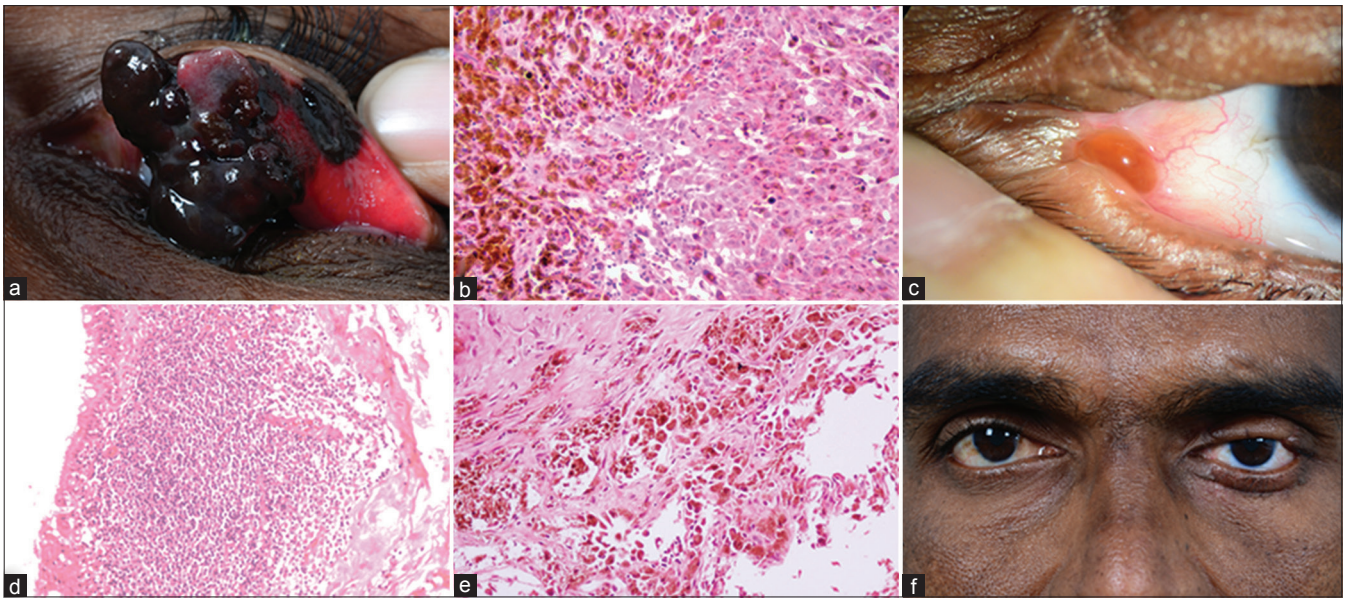

Figure 1: Conjunctival melanoma spread into the nasolacrimal duct by "melanorrhea" (a) External photograph reveals the large pigmented melanoma on the left upper tarsal conjunctiva, (b) histopathology of the tumor biopsy confirming the diagnosis of conjunctival melanoma, with variably pigmented, epithelioid melanocytes in the basal area of the epithelium (Hand E, ×75), (c) external photograph reveals brown-colored discharge on pressure over the left lacrimal sac, (d) section of the lacrimal sac shows subepithelial chronic inflammation, without evidence of the tumor (H and E, ×50), (e) black pigmented lesion from nasolacrimal duct shows atypical pigmented cells, consistent with melanoma (H and E, ×75), (f) external photograph of the patient at final follow-up 29 months after the surgery and external beam radiotherapy without any tumor recurrence

from the melanocytes interspersed among the basal cells of the conjunctival epithelium.[1] Risk factors for recurrence include the thickness of primary tumor >4 mm, subsequent local recurrences, incomplete excision at the time of surgery, nonlimbal tumor location, superior quadrant location, and corneal involvement >2 mm.[2] Metastasis can occur to regional lymph nodes or hematogenously to liver, skin, and bone.[1]

Conjunctival melanoma spread to the lacrimal drainage apparatus by direct extension is also a known entity.[3] In a report by Satchi *et al*., the authors described five cases of lacrimal drainage apparatus melanoma arising in patients with conjunctival melanoma, all of whom had undergone orbital exenteration as a part of their treatment.[3] Melanoma of the lacrimal drainage apparatus was clinically present at the time of exenteration in one case, found unexpectedly in two cases, and had developed subsequent to exenteration in two cases.[3] There are only a very few reported cases of primary sac and NLD melanoma, and the common presentations were a lacrimal sac fossa mass or epiphora.[4,5]

In 1989, Khan *et al*. described the term lacrimal oncorrhea in their report on sebaceous carcinoma and speculated that the tumor cells can dislodge from the main mass and float free in the tear film to get implanted in the lacrimal system.[6] This could explain the multicentricity and noncontiguous involvement by the tumor.[6] Our patient also had noncontiguous involvement of the NLD, and the likely mechanism, in this case, is lacrimal oncorrhea. To the best of our knowledge, this is a novel case of a conjunctival melanoma with possible spread into the NLD by way of oncorrhea, and thus, it can be termed as "melanorrhea."

Conclusion

Conjunctival tumors can seed into the tear film and extend into the lacrimal apparatus. Careful preoperative assessment to rule out the involvement of the lacrimal system must be undertaken. Evaluation of the lacrimal system must also be done in all postoperative follow-ups to detect any tumor spread.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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