Corneal erforation casused by eyelid margin trichilemmal carcinoma: A case report

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Case report

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Abstract

Background: Trichilemmal carcinoma (TLC) is a rare malignant adnexal tumor predominantly affecting the scalp, eyelids, neck and face of the elderly. Here, we firstly report a rare case of corneal perforation caused by TLC grown in eyelid margin. Case presentation: A 68-year-old female presented with two months history of unprovoked redness, pain and blurred vision in the left eye. On slit-lamp examination, a 1×2mm aseptic corneal perforation embedded by iris prolapsed was noted. After excluding other causes through examinations and case history inquiry, we speculated that the severe MGD and subsequent BKC might be the cause. In order to prevent the ulcer enlargement and intraocular infection, the patient was treated with penetrating keratoplasty immediately. However, a terrible phenomenon has aroused our concern that several small nodules gradually developed on the eyelid margin, accompanied with bleeding, burst and madarosis postoperatiely. The biopsy revealed the eyelid was involved by TLC and then the lesionectomy was performed immediately. There was no evidence of local recurrence and metastasis during 1 year follow-up. Conclusions: The involvement of eyelid margin by TLC, and subsequent BKC and corneal perforation were very rare and it has not been reported yet. The concomitant BKC can be gradually healed after rectifying eyelid margin by total excision of TLC. It is very important to offer careful follow-up to all patients.

Background

Trichilemmal carcinoma (TLC) is a rare malignant adnexal tumor predominantly affecting the scalp, eyelids, neck and face of the elderly. It is a large, solitary, multilobulated lesion that usually originates from the external hair sheath. The involvement of eyelid margin by TLC and subsequent blepharokeratoconjunctivitis (BKC) have not been reported yet. Here, we firstly report the case of corneal perforation caused by a TLC grown in eyelid margin.

Case Presentation

A 68-year-old female presented with two months history of redness, pain and blurred vision in the left eye. Slit-lamp examination revealed that left upper eyelid was slight keratinization and had several aberrant lashes, meibomian gland openings got clogged, the conjunctival was congestions and edema. In addition, diffused corneal epithelial punctate defect and neovascularization, especially an 4×3mm irregular corneal epithelial defect was noted at temporal cornea. The most serious observation was a 1×2mm aseptic corneal perforation existed in the central cornea, which had led to iris incarceration (figure 1A). Patients had not been performed eye surgery, also denied the history of ocular trauma, immune-related disorders and excluded other eye diseases. After excluding other causes through examinations and case history inquiry, we speculated that the rough eyelid margin might be the cause of persistent corneal epithelial defect, neovascularization and perforation by repeatedly rubbing the ocular surface. We did some routine examinations to exclude common causes of eyelid infections including: mites, bacterial or viral infection and non-infectious causes such as allergic blepharositis and seborrheic blepharositis, etc., but all the results were “negative”. Hence, we speculated that the severe MGD might be the cause of

the blepharitis. In order to prevent the ulcer enlargement and intraocular infection, the patient was treated with a penetrating keratoplasty immediately. The size of graft was 7.5mm and bandage contact lens was put on, and all the operation course was successful. At 2 months later, the graft remained clear and no serious complications occurred. However, a terrible phenomenon aroused our concern that several small nodules gradually developed on the rough eyelid margin, accompanied with bleeding, burst and madarosis (figure 1B). Subsquently, we did a biopsy of the eyelid margin, and it revealed an infiltrative, lobulated tumor composed of large, polygonal, clear cells with eccentric nuclei which was differentially diagnosed as TLC (figure 2). Eventually, the cause for changes of eyelid margin came to light and the corneal perforation was caused by TLC inducing the keratinization of eyelid margin. Then the lesionectomy was performed immediately. After that, the patient was transferred to the oncology department for further general examination, and no metastasis was found. Because the characteristic of TLC was rarely metastasizes and low recurrence, the oncologist advised that this patient did not need to get systemic chemoradiotherapy for now. At follow-up 1 year later, there was no evidence of local recurrence and metastasis, and the corneal graft was transparent and no rejection (figure 1C, D).

Discussion And Conclusions

In 1976, Headington firstly described TLC as a histologically invasive, cytologically atypical clear cell neoplasm of adnexal keratinocytes that is in continuity with the epidermis and/or follicular epithelium[1]. It often occurs on the sun-exposed skin of the elderly[2-4], especially affects 40-year-old and older groups[5], without gender pre-dilection[3]. Dailey et al reported the TLC growth in the eyelid margin for the first time[2], however, the involvement of the eyelid margin and the subsequent corneal perforation is rarely reported and the pathogenesis of TLC is not known clearly. Previous studies have postulated that the actinic damage[6], transformation from benign trichilemmoma[7] or long term low dose irradiation[8] could be the etiology of TLC. Histologically, TLC is charecterised by single, exophytic nodular appearance, measuring less than 2cm in diameter, and sometimes it complicates ulceration and keratosis[1, 7, 8]. Microscopically, TLC usually demonstrates proliferative lobules centrered on pilosebaceous , composed of polygonal clear cells and has high-grade mitotic potential[4, 5, 7]. But in this case we noted several diffuse nodules, ulcerations on the upper eyelid margin, and was poorly circumscribed. There was no change in the palpebral conjunctiva.

TLC are often confused with other skin cancer, such as clear cell basal cell carcinoma(BCC), squamous cell carcinoma(SCC), trichilemmoma, and malignant proliferating trichilemmal tumors(PTT)[1, 9] yet can be differentially diagnosed based on their growth pattern and histopathology.

TLC has a benign clinical course, and there is no evidence of recurrence after complete excision[7, 10]. Billingsley[3] and Tze Foon Lai[10] recommended Mohs micrographic surgery as the treatment to ensure complete surgical excision. Besides, full dose irradiation can be also applied to treat TLC. The concomitant BKC can be gradually healed after total excision. It is very important to offer careful follow-up to all patients.

In the early stage of this case, the clinical manifestations of TLC was not apparent. Eyelid margin only showed slight keratinization and roughness, and there was no ulcers or bleeding, it confused us. We excluded common causes of blepharositis, and did not realize it was rare tumor until the ulcer lesions appeared. The involvement of eyelid margin by TLC was very rare and it has not been reported yet. Conjunctival and corneal involvement was considered to be the sequel of BKC, and it affected both the meibomian gland and the ocular surface, leading to keratitis, corneal thinning or perforation, vascularization and scarring. Both mediating chronic inflammatory and rectifying eyelid margin were important in the treatment of BKC.

Abbreviations

BKC: Blepharokeratoconjunctivitis

TLC: Trichilemmal carcinoma

BCC: Basal cell carcinoma

SCC: Squamous cell carcinoma

PTT : Proliferating trichilemmal tumors

Declarations

Ethics approval and consent to participate

This case report was approved by the “Ethics Committee of Xiamen University affiliated Xiamen Eye Center”, and written informed consent was obtained from patient.

Consent for publication

This patient consent to publish the image of eye and other results of her medical examination in this manuscript or paper which may be published in your journal.

Availability of data and materials

The corresponding author (Huping Wu) had full access to all the data in this study and takes responsibility for the integrity of the data.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

All authors have read and approved the manuscript. Specifically, LZ designed this study, performed the literature search, and wrote the manuscript. Both ZL and HW designed this study.

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Figures

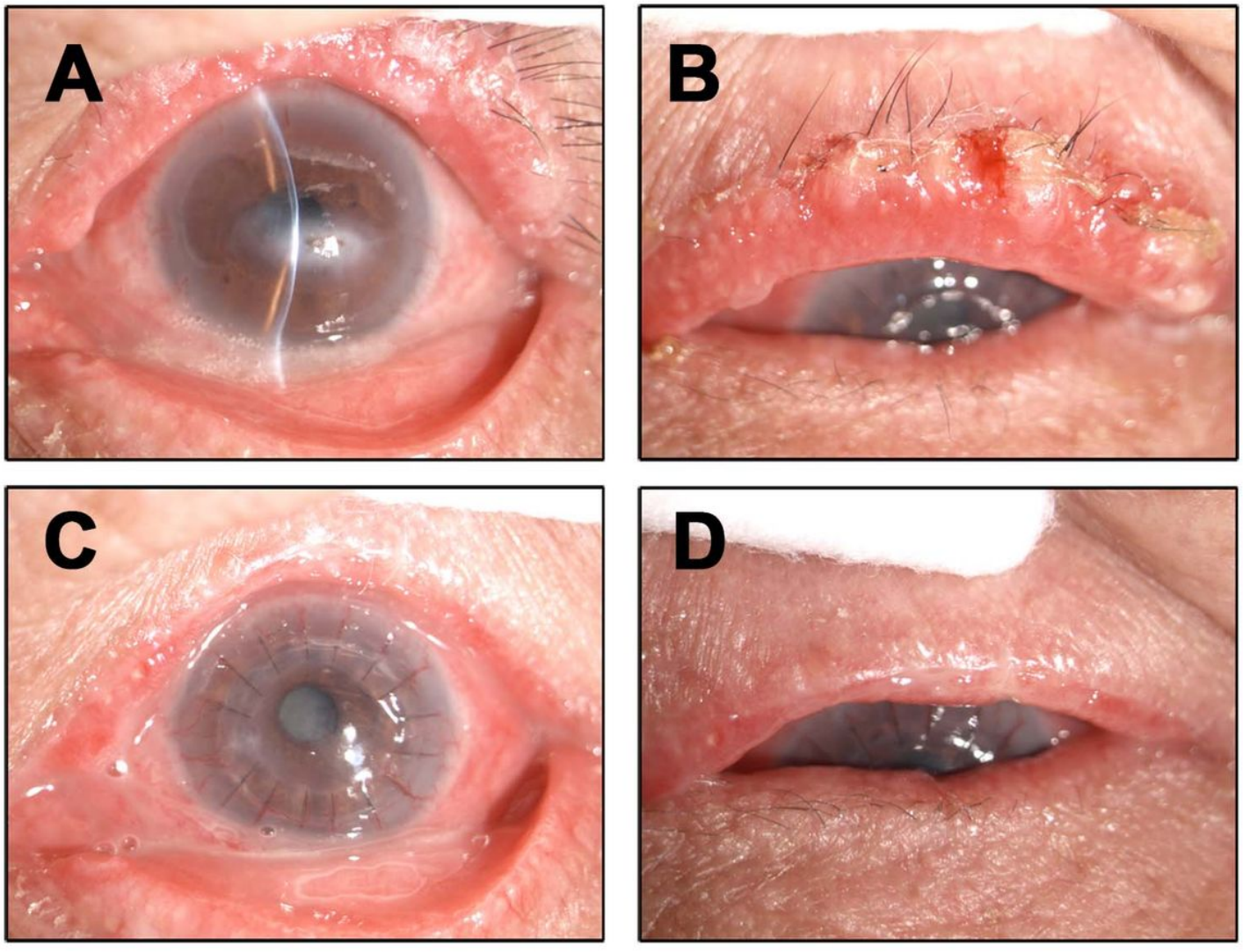

Figure 1

Ocular surface findings. (A) Clinical photographs at the first visit. The left upper eyelid was slight keratinization and had several aberrant lashes. Meibomian gland openings got clogged. The conjunctival

was congestions and edema. In addition, diffused corneal epithelial punctate defect and neovascularization, especially an 4×3mm irregular corneal epithelial defect was noted at temporal cornea. The most serious was a 1×2mm aseptic corneal perforation existed in the central cornea, and the subsequent iris incarceration. (B) Clinical photographs of eyelid at the second month postoperatiely. Several small nodules accompanied with bleeding, burst and madarosis on the left eyelid margin. (C, D) Clinical photographs showed that there was no evidence of local recurrence and metastasis, and the corneal graft was transparent and no rejection after total excision 1 year later.

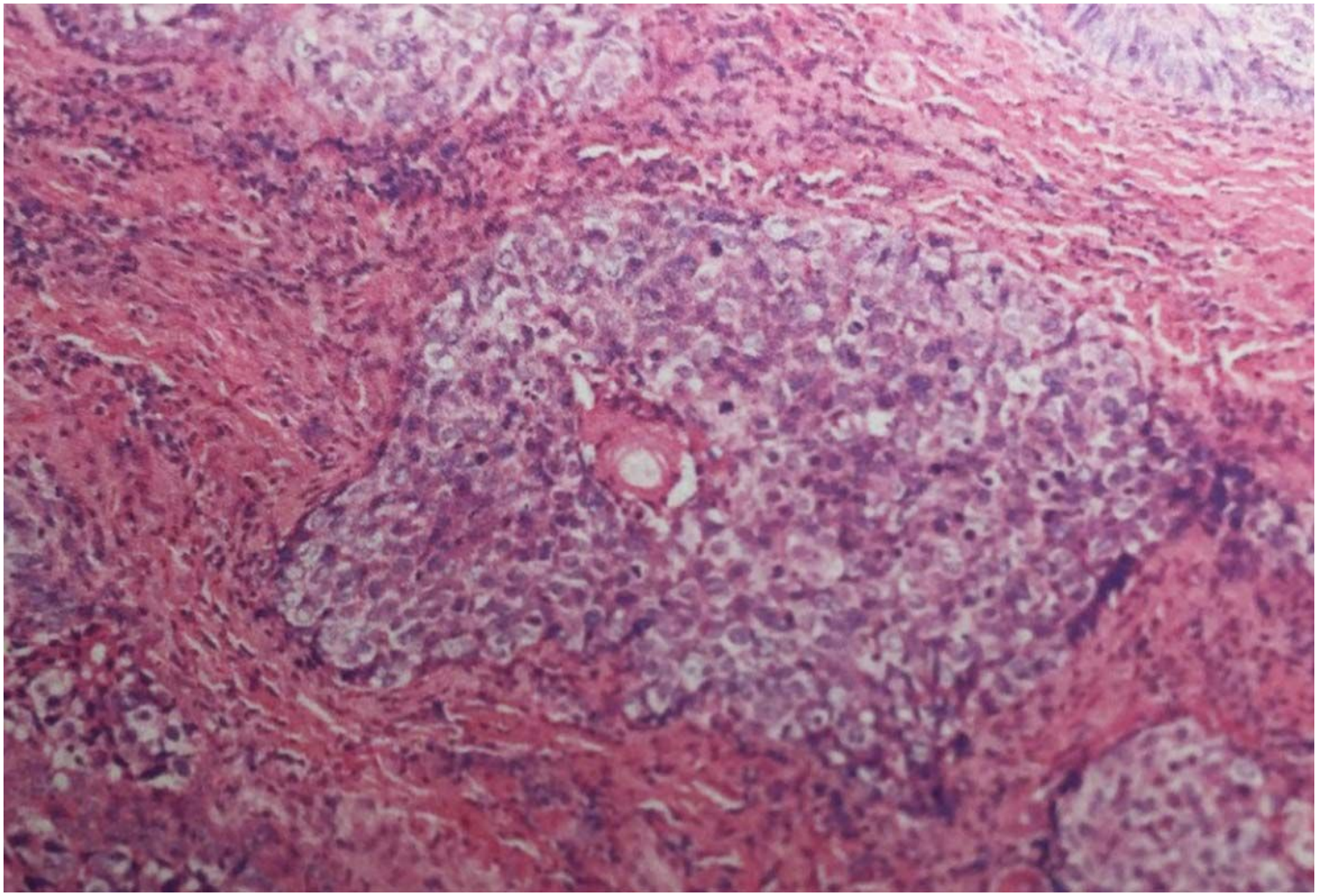

Figure 2

Histological section demonstrating infiltrative lobules of clear cells composed of large, polygonal, clear cells with eccentric nuclei. (Haematoxylin-eosin stain; magnification×200).

Supplementary Files

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