A case with corneal perforation due to bacterial concretion derived from lacrimal canaliculitis

Sho Ishikawa*, Naoko Kato

Department of Ophthalmology, Saitama Medical University, Saitama, Japan

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ABSTRACT

Purpose: To report a case with corneal perforation, presumably due to ocular cicatricial pemphigoid (OCP) and bacterial concretion derived from lacrimal canaliculitis.

Observations: A patient with OCP demonstrated adherence of concretion to the cornea of her right eye. She also demonstrated canaliculitis in this eye. We removed a whitish precipitate from the bottom of the corneal ulcer.

Conclusions and Importance: Both lacrimal canaliculitis and OCP can cause corneal perforation, and adherence of bacterial concretion onto the cornea is very rare. However, once it occurs, corneal perforation can rapidly follow. OCP sometimes causes corneal epithelial damage, which may influence adherence to concretion. Canaliculitis in patients with OCP should be managed carefully.

1. Introduction

Corneal perforation is an emergency condition that is caused by both infectious and noninfectious corneal disorders. Canaliculitis, a disease of the lacrimal pathway, which accounts for 2% of all lacrimal diseases, rarely causes corneal perforation,1 and is sometimes misdiagnosed and inadequately treated.2 The most common pathogenic agent in canaliculitis is Actinomyces sp.; such infections are difficult to treat with antibiotic drops and tend to form bacterial concretions within the canaliculus. The most effective treatment for canaliculitis is surgical extraction of the bacterial concretion.2,3

Ocular cicatricial pemphigoid (OCP) is a rare autoimmune disease characterized by subepidermal blistering and progressive cicatrization that affects the skin and mucosa. OCP is considered to be one of the most severe ocular surface disorders, and is accompanied by severe dry eye, chronic inflammation, and damage to the epithelium of the corneal and conjunctival mucosa. Subconjunctival fibrosis and contraction of the tissue, and subsequent shortening of the lower conjunctival fornix, are typical findings in OCP. The treatment for OCP requires immunosuppressive agents, including steroid drugs.4

Both canaliculitis and OCP are well-known pathological conditions affecting the integrity of the ocular surface, and can sometimes occur simultaneously in a given patient; however, no case that developed corneal perforation has been reported to date. Here, we report a patient with concurrent OCP and canaliculitis, who developed a corneal perforation presumably due to corneal melting caused by adherence of bacterial concretion to the corneal surface.

2. Case report

An 82-year-old woman was referred to our hospital due to eye pain and decreased visual acuity of her right eye. She had visited another clinic the previous day and had been diagnosed with conjunctivitis and prescribed antibiotic eye drops. She had a history of corneal perforation from an unknown cause 2 years earlier. Her best corrected visual acuity was 20/200 OD. By slit lamp examination, her right cornea revealed a corneal ulcer accompanied by dense whitish precipitate at the mid-central to peripheral area, at 7 o’clock and 9 o’clock (Fig. 1A). Her left cornea demonstrated severe superficial punctate keratitis. Her both lower conjunctival fornix had shortened due to subepithelial fibrosis and inflammation (Fig. 1B). We removed a conjunctival sample for histological examination. The superior and inferior lacrimal puncta of her right eye were swollen and dilated (Fig. 1C and D), and the superior and inferior lacrimal puncta of her left eye were obstructed. The Schirmer-1 test indicated tear secretion was 1 mm for 5 min for both eyes. When we irrigated the lacrimal pathway, the saline passed into...
the nasal cavity and throat of her both eyes; however, a marked amount of concretion and dense mucus was refluxed from both puncta of her right eye.

We diagnosed corneal ulcer and lacrimal canaliculitis of the right eye, and suspected that she had concurrent OCP or drug induced conjunctival cicatrization (ocular pseudopemphigoid) according to the clinical criteria for OCP.6 We removed the whitish precipitate from the bottom of the corneal ulcer, which was a hard concretion that had refluxed from the lacrimal pathway, and subjected this to bacterial examination. We checked Seidel test when we removed concretion plug, and confirmed that no aqueous leakage was observed. We placed a bandage soft contact lens onto her right eye, irrigated the lacrimal pathway with iodine solution, and prescribed oral clarithromycin (Clarith®, Taishotoyama, Toshima, Japan) 200 mg, twice a day.

When she returned the next day, her right cornea showed perforation in the area from which the precipitate had been removed (Fig. 2). While the iris was prolapsed and the anterior chamber was maintained by the bandage contact lens, we performed surgical removal of the lacrimal concretion. A vast amount of concretion was extracted. After removing the concretion, the conjunctive hyperemia resolved, and the corneal perforation healed without requiring surgical intervention.

Histopathological examination showed that the conjunctival epithelial cells contained a blister with basement membrane zone and infiltration of lymphocytes into the conjunctival stroma, consistent with OCP. There were no immune deposits in the basement membrane zone, so it didn’t fulfilled immunopathology criteria for OCP.6 Both the whitish precipitate removed from the corneal ulcer and the surgically extracted canalicular concretion revealed identical features: according to hematoxylin–eosin staining, these samples contained a marked infiltration of neutrophils and eosinophilic granular materials, which were presumably sulfur granules produced by Actinomycetes sp. (Fig. 3).

After removal of the canalicular concretion, we prescribed steroid eye drops (fluorometholone) for her right eye, for 3 months. Her right visual acuity improved to 20/40, the right punctum was spontaneously obstructed, and no recurrence of canaliculitis was observed.

The patient provided written consent for publication of this report, including record details and photographs.

3. Discussion

We here reported a patient with OCP, who developed corneal melting and perforation presumably caused by adhesion of bacterial concretion derived from lacrimal canaliculitis.

Upon histopathological examination, the concretion was shown to be an aggregation of infiltrated neutrophils and bacterial threads, which is characteristic of Actinomycetes concretions. Actinomycetes and neutrophils produce elastase, a protease, which may play a role in melting the corneal stroma.

Bacterial concretions do not normally adhere onto the corneal surface; however, this may occur in some pathological conditions, such as under chronic inflammation or a continuously compromised epithelial integrity. We consider that, in the present case, persistent...
inflammation, caused by severe dry eye and untreated OCP, could bring about adherence of the bacterial concretion onto the bare corneal stromal surface, and subsequent melting of the corneal stroma below, and subsequent corneal perforation.

Corneal perforation caused by canaliculitis is a rare condition; however, this has previously been reported by a few investigators. Yokogawa et al. have reported two cases with corneal perforation associated with canaliculitis, and mention that aerobic bacteria were detected in both cases. They speculated that allergies against toxins produced by some bacteria may be involved in the mechanism, and that perforation occurs mostly at the peripheral cornea in such cases. In contrast, corneal perforation in the present case occurred in the mid-central cornea, within the area where the concretion directly adhered to the cornea. Therefore, we hypothesize that the corneal melting was due to enzymes released by *Actinomycetes*, rather than to an allergic reaction.

Since topical or systemic antibiotic treatment is not effective, the most adequate treatment for canaliculitis is thought to be surgical treatment. In the present case, we performed only punctoplasty and removal of the concretion, because the patient had severe dry eye. Her punctum was obstructed spontaneously several weeks after the procedure and required no further treatment. Albeit rarely, it has been reported that patients with OCP might present with lacrimal obstruction, which is usually punctal or canicular.6

4. Conclusions

If bacterial concretions adhere to the cornea, it may quickly progress to corneal perforation. We should pay special attention to patients with OCP with infection of the lacrimal pathway.

Patient consent

The patient’s legal guardian consent to publication of the case in writing.

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Conflicts of interest

The following authors have no financial disclosures.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

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References

1. Yokogawa H, Kphayashi A, Yamazaki N, Masaki T, Sugiyama K. Surgical therapies for corneal perforations: 10 years of cases in a tertiary referral hospital. *Clin Ophthalmol*. 2014;8:2165–2170.
2. Vujancevic S, Meyer-Rusenberg HW. Therapy for actinomycosis in the lacrimal pathway. *Klin Mochl Augenheilkd*. 2010;227(7):568–574.
3. Yokogawa H, Kphayashi A, Yamazaki N, Masaki T, Sugiyama K. Surgical therapies for corneal perforations: 10 years of cases in a tertiary referral hospital. *Clin Ophthalmol*. 2014;8:2165–2170.
4. Vujancevic S, Meyer-Rusenberg HW. Therapy for actinomycosis in the lacrimal pathway. *Klin Mochl Augenheilkd*. 2010;227(7):568–574.
5. Yokogawa H, Kphayashi A, Yamazaki N, Masaki T, Sugiyama K. Surgical therapies for corneal perforations: 10 years of cases in a tertiary referral hospital. *Clin Ophthalmol*. 2014;8:2165–2170.
6. Vujancevic S, Meyer-Rusenberg HW. Therapy for actinomycosis in the lacrimal pathway. *Klin Mochl Augenheilkd*. 2010;227(7):568–574.
7. Yokogawa H, Kphayashi A, Yamazaki N, Masaki T, Sugiyama K. Surgical therapies for corneal perforations: 10 years of cases in a tertiary referral hospital. *Clin Ophthalmol*. 2014;8:2165–2170.
8. Vujancevic S, Meyer-Rusenberg HW. Therapy for actinomycosis in the lacrimal pathway. *Klin Mochl Augenheilkd*. 2010;227(7):568–574.
9. Messer EM, Hintschich CR, Partscht K, Messer G, Kampik A. Ocular cicatricial pemphigoid. Retrospective analysis of risk factors and complications. *Ophthalmologie*. 2000;97(2):113–120.
10. Satchi K, McNab AA. Conjunctival cicatrizing disease presenting with lacrimal obstruction. *Orbit*. 2016;25:1–3.
11. Chan LS, Ahmed AR, Ashalt GJ, et al. The first international consensus on mucous membrane pemphigoid: definition, diagnostic criteria, pathogenic factors, medical treatment, and prognostic indicators. *Arch Dermatol*. 2002;138:370–379.