



Case report

Case of bilateral Mooren's ulcers following filtering surgery using EX-PRESS glaucoma filtering devices

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ABSTRACT

Purpose: To report our findings in a case of bilateral Mooren's ulcer that developed after filtering surgeries using the EX-PRESS glaucoma filtering device (EX-PRESS surgery).

Patients and methods: A 71-year-old Japanese man with primary open angle glaucoma underwent EX-PRESS surgery first in his left eye and 1 month later in his right eye. He developed Mooren's ulcer in his right eye at 7 months and in his left eye at 10 months after the initial EX-PRESS surgery. Systemic examinations showed no collagen vascular disease, and he did not have a history of bacterial or viral infections. He was not allergic to metallic materials. Before the EX-PRESS surgery, he had undergone cataract surgery combined with trabeculectomy in both eyes, and a reoperation of trabeculectomy in his left eye. He had not developed Mooren's ulcer after these surgeries. The Mooren's ulcer after the EX-PRESS surgery was treated with oral prednisolone (30 mg tapering) in combination with topical 0.1% betamethasone sodium. The ulcers were responsive and healed well in three months.

Conclusions: The EX-PRESS devices were most likely the cause of the Mooren's ulcers considering that they were located close to the site of EX-PRESS insertion and no peripheral corneal ulcer developed after prior intraocular surgeries.

1. Introduction

Mooren's ulcer is a rare, painful ulceration of the peripheral cornea with conjunctival and episcleral injection.^{1,2} The course is progressive, and the entire cornea can be involved. A wide variety of systemic diseases including herpes zoster,³ hepatitis C,⁴ and parasitic infection⁵ have been associated with Mooren's ulcers. In addition, there have been other causes associated with Mooren's ulcer, e.g., physical trauma,⁶ cataract surgery,⁷ pterygium surgery,⁸ penetrating keratoplasty,^{9,10} and epikeratoplasty.¹¹

We report our findings in a case of bilateral Mooren's ulcers that developed following filtering surgery using the EX-PRESS glaucoma filtering device.

2. Case report

A 71-year-old Japanese man underwent EX-PRESS surgery for primary open angle glaucoma first in his left eye and 1 month later in his right eye. The bleb size was small in the left eye and moderate in the right eye, and the intraocular pressures were well controlled bilaterally.

Seven months after the EX-PRESS surgery, he complained of redness and a foreign body sensation in his right eye. A corneal ulcer with epithelial defect was detected near the site of the EX-PRESS insertion, and limbitis with severe scleral injection was observed (Fig. 1). An allergic reaction to the nylon sutures was suspected, and they were removed. Then, 0.5% levofloxacin and 0.1% fluorometholone were applied topically, and there was a slight reduction in the size of the ulcer and infiltration, however a complete resolution was not observed. Topical 1% betamethasone sodium phosphate was started, and the corneal ulcer, limbitis, and scleral injection gradually decreased.

Ten months after the surgery in his left eye, a similar corneal ulcer developed near the site of EX-PRESS insertion (Fig. 2). After the sutures around the limbus were removed, topical 0.1% betamethasone sodium phosphate was applied to his left eye and continued in his right eye. However, the corneal ulcers in both eyes responded less favorably, and they progressed with thinning and a steep undermined leading edge (Fig. 3).

Gram staining of corneal scrapings did not show any microorganisms. Cultures for bacteria and fungi were negative, and there was no mucopurulent eye discharge. We diagnosed this case with Mooren's

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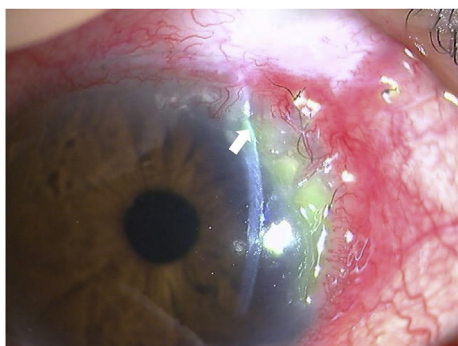


Fig. 1. Slit-lamp photograph of the right eye at 7 months after the EX-PRESS surgery. A marginal corneal ulcer can be seen close to the site of the EX-PRESS implantation with severe conjunctival injection. Arrow indicates the site of EX-PRESS implantation.

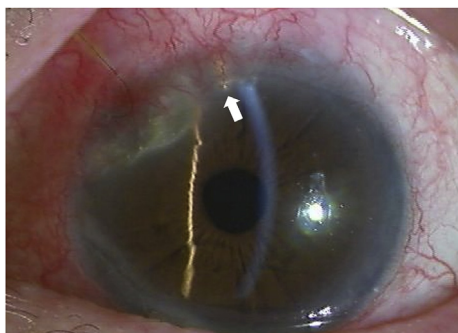


Fig. 2. Slit-lamp photograph of the left eye at 10 months after the EX-PRESS surgery. A marginal corneal ulcer close to the site of the EX-PRESS implantation with conjunctival and scleral injection. Arrow indicates the site of EX-PRESS insertion.

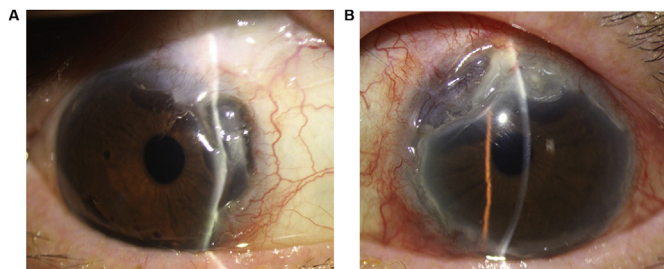


Fig. 3. Slit-lamp photograph of the right eye (3a) and left eye (3b) taken during topical steroid therapy. Bilateral progressive thinning of the ulcers with a steep undermined leading edge can be seen.

ulcers bilaterally.

Then, 30 mg of oral prednisolone combined with topical 0.1% betamethasone sodium was started and tapered gradually as the signs and symptoms decreased. The corneal ulcers were responsive and healed well in three months. The 0.1% topical betamethasone sodium has been continued to prevent recurrences. There have been no recurrences of the ulcerations in either eye during the 2-year follow-up (Fig. 4).

Nine years before the EX-PRESS surgery, he had cataract surgery combined with trabeculotomy bilaterally, and a reoperation of trabeculotomy four years previously in his left eye. He had not developed any complications after these surgeries. He was being treated with conventional medications for the diabetes mellitus and hypertension.

His laboratory data showed that he had mild liver dysfunction, and the hepatitis C antibody was negative. He had no history of allergy including that to metallic materials. Blood tests, including complete blood cell, platelet count, antinuclear antibodies, rheumatoid factor, C-

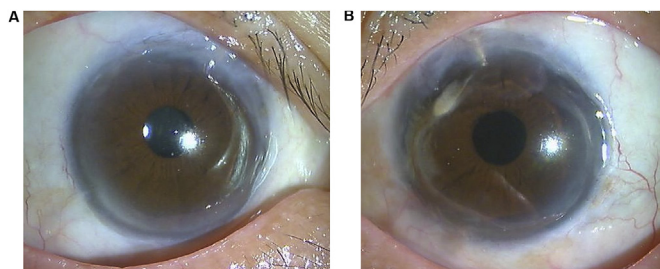


Fig. 4. Slit-lamp photograph of the right eye (3a) and left eye (3b) of the patient at 2 years after the onset of the ulcer in the right eye. The corneal epithelium has healed with thinning of corneal stroma after the systemic steroid therapy. Betamethasone sodium phosphate 0.1% is being continued.

reactive protein, liver function tests, blood urea nitrogen and creatinine tests, fasting blood sugar tests, revealed no abnormal findings associated with collagen vascular disease. Patch testing for metallic allergy was negative for the metal components of the EX-PRESS glaucoma filtering device.

3. Discussion

Although EX-PRESS surgery has a minimum insult to the cornea, it has been recently shown that there is a decrease in the corneal endothelial cell density after the EX-PRESS surgery.¹²⁻¹⁴ Our report is the first to show Mooren's ulcers developing following the EX-PRESS surgery. The differential diagnoses for Mooren's ulcer are bacterial corneal ulcer, rheumatoid arthritis (RA), scleritis, and dellen associated with conjunctival chemosis. The patient had no mucopurulent eye discharge. He also did not show the common symptoms of RA such as joint inflammation and abnormal laboratory finding associated with RA. Although scleritis sometimes accompanies corneal peripheral ulcer in the late phase, he had no history of scleritis. As for a dellen, it represents an area of localized corneal thinning without epithelial defect. Thus, we diagnosed his condition as Mooren's ulcers developing following the EX-PRESS surgery.

Mooren's ulcer is believed to be caused by an autoimmune mechanism¹⁵ with both humoral- and cell-mediated autoimmune phenomena associated with it.^{2,10,11,15-19} The triggers for the hyperactive immune reactions might be trauma, surgery, and systemic or local infections.²⁻¹¹

Gottsch et al. suggested that prior corneal surgery may play a role in the pathogenesis of Mooren's ulcer.¹⁰ In addition, some studies have implied that the epithelial basement membrane of corneal epithelium and especially the corneal stem cells are the specific target of this disease.²⁰ The limbal epithelium may be a more specific target.²¹ We suggest that the glaucoma implant device may have increased the risk of an autoimmune reaction against the peripheral cornea. The question remains, however, whether the implanted device material or the surgical trauma was the cause of the Mooren's ulcers. It must also be considered that Mooren's ulcer might be a multifactorial disorder.

We recommend that clinicians pay special attention to marginal ulcer or limbal injection after EX-PRESS surgery.

Patient consent

Consent to perform the surgery and to publish this case report was obtained. This report does not contain any personal information that could lead to the identification of the patient.

Authors' contributions

All authors agree to be accountable for all aspects of the work, have drafted and revised the manuscript critically, and have final approval of

the final version.

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Declaration of competing interest

No conflict of interest exists.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.ajoc.2020.100588>.

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