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Atypical Bilateral Mooren Ulcer Following Small Incision Lenticule Extraction (SMILE)

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Abstract: A 38-year-old Malay gentleman presented with 1 year history of bilateral eye discomfort and redness with deteriorating vision. He underwent myopic laser refractive surgery and had history of ocular Steven Johnsons syndrome (SJS). He admitted to use regular topical steroid to relieve the symptoms. The visual acuity was 6/24 in the right eye and counting finger in the left eye. There was peripheral corneal melt and thinning which is worse on the left eye and associated with perforation. All investigations to rule out causes of peripheral ulcerative keratitis were negative, and diagnosis of Mooren ulcer was made. A lamellar keratoplasty was performed to seal the leak on left eye, and he was initiated on systemic immunosuppression. At present, his corneal melt has stabilizes and his vision remains at 6/24 in the right eye and 6/60 in the left eye.
Keywords: Mooren ulcer, laser refractive surgery, Steven Johnson's syndrome, ulcerative keratitis, lamellar keratoplasty

INTRODUCTION

Mooren ulcer is peripheral corneal ulceration which previously described as chronic serpiginous ulcer of the cornea or ulcus roden [1,2]. This course is rapidly progressive and painful. Diagnosis is made by excluding other causes of PUK [3] such as rheumatoid arthritis and Wegener granulomatosis.

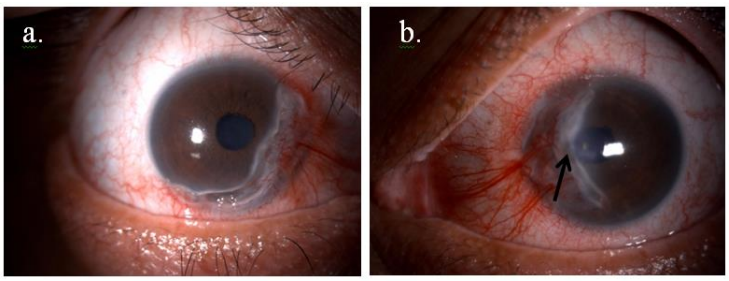
CASE REPORT

A 38-year-old Malay gentleman with underlying diabetes mellitus and hypertension presented with progressive deterioration in vision associated with discomfort since 1 year. He admitted to using topical steroids regularly to relieve the symptoms, without ophthalmologist supervision. He had history of resolved SJS 10 years ago, and had refractive myopic SMILE performed in both eyes 3 years prior.

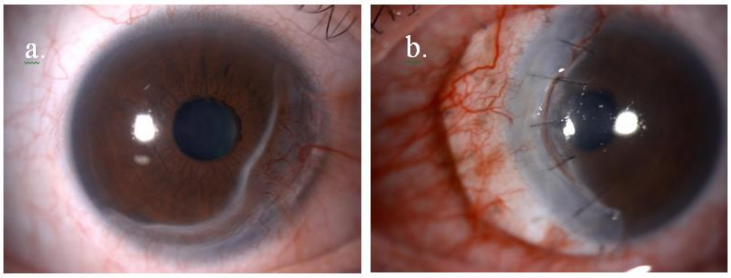
At presentation, the visual acuity was 6/24 in the right eye, and counting finger in the left eye. There was large pannus over a third of cornea nasally, covering area of crescent-shaped corneal thinning with undermining edges in both eyes. (Figure 1 a and b) The anterior chamber was quiet, and other ocular systemic examination were unremarkable. There was leak over the edge of thinning on the left eye, but fortunately anterior chamber was deep and quite, with no signs of infections.

All relevant investigations to rule out different causes of peripheral ulcerative keratitis (PUK) were negative, including hepatitis C, and anti-cytoplasmic antibodies. Other haematologic parameters such as full blood count, renal and liver profile, rheumatoid factors and antinuclear antibody were normal. The erythrocyte sedimentary rate (ESR) was moderately elevated at 46mm/hour and C-reactive protein was 1.24mg/dl.

Topical moxifloxacin was initiated every 4 hourly, along with topical pred forte® four times daily, ointment teramycin on night, oral ciprofloxacin 500mg twice daily and oral doxycycline 100mg twice daily. Additionally , intravenous methylprednisolone (MTP) 500mg daily for 3 consecutive days was started, along with oral cyclosporine of 100 mg bidaily. A banana-patch graft was performed on the left eye to seal the linear perforation along the edge of thinning. The intravenous MTP was transitioned to oral prednisolone of 60 mg/kg bodyweight, with weekly tapering. Visual acuity of the left eye improved to 6/18, N18 and stable in the right eye. Unfortunately, he developed acute renal impairment and hepatitis secondary to cyclosporine, and was changed to oral mycofenolate mofetil 500 mg bidaily. His renal and liver profile normalizes after 1 month cessation of cyclosporine. The ulceration and corneal melting was also halted (Figure 2) and his vision remains stable.



Fig−1 : (a) (Right) and (b) (Left): There was bilateral crescent-shaped ulcer with undermined edges bilaterally and obvious thinning in the right eye and perforation of left eye (arrow)



Fig−2 : (a) (Right) and (b) (Left) : Right eye stabilizes with vascularisation and conjunctivalization of thinned area, and left eye shows stable graft

DISCUSSION

Mooren ulcer has reported following corneal trauma and eye surgery[4], involving over 60% of cases[5]. Severe unilateral corneal melt diagnosed as Mooren has been reported after uneventful phacoemulsification [6]. However to the best of our knowledge, this is the first report of Mooren ulcer in a patient who underwent laser refractive surgery (LRS). It is not yet clearly understood why some patients with Mooren ulcer had histories of corneal surgery, other than an uncontrolled autoimmune process. In this case, history of SJS with resultant subtarsal fibrosis may have adversely affected the ocular surface and the subsequent laser surgery further stimulates an autoimmune process directed against a specific molecule in the altered corneal stroma. As a result, degradative enzymes released primarily by neurotrophils [7, 8] triggers a cascade of autoimmune-mediated keratolysis. Genetic predisposition in susceptible individuals may also play a role, through one of several mechanisms of autoimmune destruction resulting in progressive corneal melting. Comorbidities such as diabetes mellitus and hypertension are not known to be the precipitating factors of Mooren ulcer.

Initial presentation of Mooren ulcer could be redness, tearing, photophobia and severe pain which is disproportionate to the inflammation [9]. Our case presented with only mild symptoms, over a long duration. This could be explained by partial self-treatment with topical over-the counter steroid eyedrops which was used regularly in this case. This also highlights that Mooren cannot be adequately controlled with topical medication alone, and systemic treatment is usually indicated in most cases.

The ulcerative keratitis in Mooren ulcer typically starts from periphery, and progress circumferentially, before spreading to central cornea. Although rare, bilateral disease does occur especially in younger age group, and are usually more severe and difficult to treat [10].

The goal of the treatment is to arrest the destructive immune process and promote healing of the epithelial defect. Management is more challenging in cases of corneal perforation. In this case, ‘banana’ patch graft was performed to seal the linear perforation along the undermined area of corneal melt. In comparison with full thickness penetrating keratoplasty, corneal perforation treated with lamellar keratoplasty has lower risk of subsequent allograft rejection and graft failure[11]. If corneal tissue is not readily available, amniotic membrane should be used sometimes conjunctival flap to temporarily restore ocular surface integrity[12]. If the cornea tissue and amniotic membrane were not available and perforation less than 2.0 mm, application of a bandage contact lens with cyanoacrylate glue can be done[13].

Systemic corticosteroid is the traditional first-line therapy for acute phases of PUK, but is often not suitable to be maintained for long term in high dose sufficient enough to inhibit disease progression or overcome the autoimmune process. Intravenous methylprednisolone for 3 consecutive days, followed by oral therapy, should be initiated in patients with imminent danger of vision loss[14]. It is prudent to consider a second line agent, if the disease is severe, bilateral or it is anticipated that patients may need to be

tapered off oral steroid within a relatively short period of time, due to concurrent diabetes mellitus. Recent study indicate better inflammatory control and fewer side effects with mycophenolate mofetil than with methotrexate or azathioprine as second-line therapy[15]. In our case, oral doxycycline was added to stabilize the keratolysis by inhibition of degradative proteanase.

CONCLUSION

This case highlights occurrence of Mooren ulcer following laser refractive surgery (LRS) in an already compromised ocular surface. Detailed ocular history and careful examination is imperative prior to performing LRS. Systemic immunosuppression is the key treatment in halting the disease process, especially in cases such as this, where ocular surface abnormality is a persistent factor.

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Declaration of interest

We declare that we have no conflict of interest.

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