

Bilateral non-resolving punctate keratitis in a keratoplasty patient

Nikhil S Gokhale, Jyoti Prakash Panda¹

This study aimed to report a case of non-resolving bilateral coarse punctate keratitis in a patient with prior bilateral penetrating keratoplasty. In view of non-response to antivirals, corneal epithelial scraping was carried out, which revealed the presence of microsporidial cysts. The infection resolved after a period of 12 days following the diagnosis, during which steroids were discontinued. Microsporidial keratitis needs to be considered in non-resolving coarse punctate keratitis and microbiologic evaluation is essential to establish the diagnosis.

Key words: Keratitis, Keratoplasty, Microsporidia

Ocular microsporidial infection can occur as a superficial keratoconjunctivitis or as a deep stromal keratitis. Keratoconjunctivitis commonly presents as a red eye with coarse punctate keratitis and is being increasingly reported due to increased awareness. We report a case of bilateral microsporidial keratitis in a patient who had penetrating keratoplasty in both eyes and was on once daily steroid. The case represents a diagnostic dilemma for the clinician because the presentation was unusual and rare.

Case Report

A 65-year-old patient reported with ocular irritation in both eyes since 2 weeks. He had previously undergone a penetrating keratoplasty with lens implant in both eyes (right eye 9 years prior and left eye 4 years prior) for Fuch's endothelial dystrophy. His left eye had also undergone a trabeculectomy 2 years prior and had a clear graft albeit with glaucomatous optic atrophy and a vision of finger counting at 1 m. His right eye had a clear graft with a best spectacle corrected vision of 6/9. He was on Latanoprost Timolol combination (Latocom drops, Sun Pharma, Mumbai, India) for his glaucoma in the right eye. In both eyes, he was using prednisolone acetate 1% (Predforte drops, Allergan, Bangalore, India) once a day. Systemically the patient was a well-controlled diabetic with no other systemic health problem.

On examination at presentation (day 1), there were bilateral coarse punctate epithelial lesions involving mainly the recipient rim and the graft host junction with a few lesions centrally on the graft. The patient was clinically diagnosed as bilateral herpetic epithelial keratitis, and oral acyclovir 400 mg five times a day was added to his treatment plan and he was advised to review after a week.

At day 7 follow-up, the patient reported worsening with blurred vision and increased ocular irritation. In his right eye, there were serpiginous coarse lesions along the graft host junction with an increase in central punctate lesions. His left eye showed serpiginous lesions both along the graft host junction and across the corneal graft along with graft edema [Figs. 1 and 2]. Corneal epithelial scrapings were gently taken from both eyes separately and sent for microbiologic smear examination. A differential diagnosis of non-responsive viral keratitis, microsporidial keratitis, and acanthamoeba keratitis were considered. All drops were stopped and the patient was put on moxifloxacin 0.5% eye drops (Vigamox, Alcon Labs Inc, Fortworth, Texas, USA) four times daily and preservative-free sodium hyaluronate 0.1% drops four times daily (Hyla-PF, Entod Pharma, Mumbai, India).

Smears report was available the same day and revealed the presence of plenty of microsporidial spores in both eyes in KOH, Gram's and 1% acid fast staining (Kinyoun's modified ZN staining) [Fig. 3]. On the basis of the smears, the patient was advised to continue the same therapy.

On follow-up on day 10, the right eye also showed graft edema with a drop in vision to 6/36 and a rise in intraocular pressure to 40 mm Hg. Acetazolamide tablet (Diamox, Pfizer, Mumbai, India) 250 mg three times a day was added to control the intraocular pressure. The punctate epithelial lesions in both eyes were gently debrided under topical anesthesia on days 10 and 12. There was a gradual reduction in punctate lesions at each subsequent follow-up. By day 19, the punctate lesions had nearly resolved; however, the grafts were edematous possibly ascribed to inflammatory reaction and steroid withdrawal. Prednisolone acetate 1% eye drops were restarted on day 19 in both eyes four times a day. Patient improved slowly and by day 30 had clear grafts in both eyes and was continued only on prednisolone acetate 1% drops four times daily and preservative free sodium hyaluronate 0.1% drops four times daily in both eyes and Latanoprost Timolol drops once daily in the right eye [Fig. 4].

Discussion

Microsporidial keratoconjunctivitis is being increasingly reported in immunocompetent patients due to the increased awareness about the entity and the ability to diagnose them

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Department of Cornea, Gokhale Eye Hospital, Mumbai, ¹Department of Microbiology, P.D. Hinduja Hospital, Mahim, Maharashtra, India

Correspondence to: Dr. Nikhil S Gokhale, Gokhale Eye Hospital, Anant Building, Gokhale Road South, Dadar West, Mumbai - 400 028, Maharashtra, India. E-mail: niksgokhale@gmail.com

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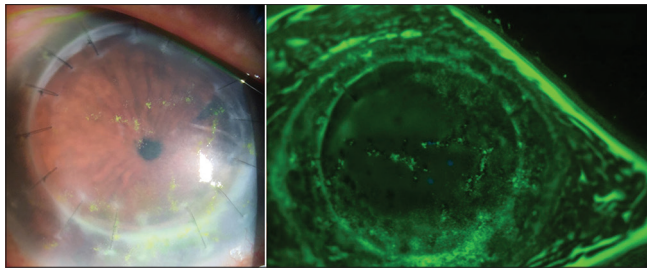


Figure 1: Right eye: Clear graft with coarse punctate epithelial keratitis on slit lamp view and with Fluorescein staining

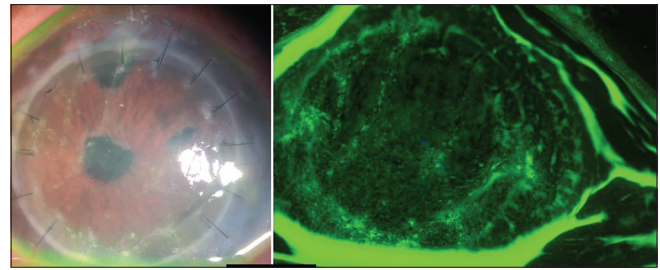


Figure 2: Left eye: Mild graft edema with coarse punctate epithelial keratitis on slit lamp view and with Fluorescein staining

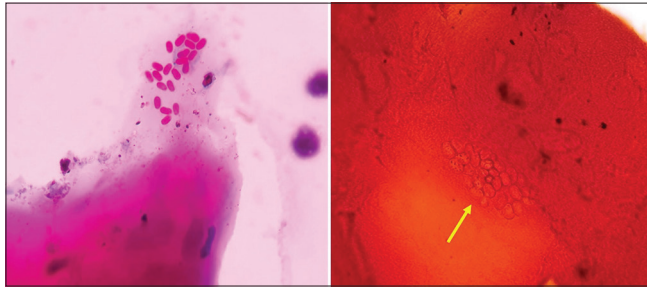


Figure 3: Microsporidial spores can be seen as bright red to pink oval spores on 1% acid fast staining ($\times 100$) and as clusters of unstained haloes in Gram's stain ($\times 100$)

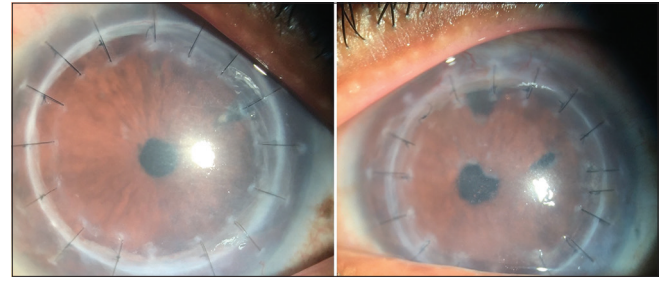


Figure 4: Clear grafts in both eyes after resolution

easily with routine laboratory staining methods. The first case reported following keratoplasty was in 2005 and was unilateral and presented with confluent epithelial lesions along with mild conjunctivitis.^[1] Devi *et al.*^[2] reported a case of bilateral microsporidial keratoconjunctivitis in a post-endothelial keratoplasty patient masquerading as a graft rejection. The clinical appearance of serpiginous lesions and pseudo-dendritic lesions in our case is different from the classical multifocal whitish raised lesions reported in routine infections. The possible use of steroids may have delayed the presentation and allowed a more confluent and contiguous epithelial spread of infection.

In the post-keratoplasty situation, new-onset viral epithelial keratitis is common even in the absence of documented previous herpes.^[3] However, in view of no response to oral antiviral therapy alternative diagnosis of acanthamoeba, microsporidial epithelial keratitis, adenoviral keratitis and Thygeson's keratitis were considered and epithelial scraping was done. Adenoviral infections usually present initially with conjunctivitis and only when it starts resolving corneal punctate keratitis appears. These usually spontaneously resolve over a few days and subepithelial infiltrates evolve. Our patient presented with conjunctivitis and epithelial keratitis simultaneously and these did not resolve or evolve into subepithelial infiltrates. Thygeson's keratitis presents with recurrent episodes of coarse punctate keratitis, which can be bilateral but often asymmetric. These lesions resolve with mild steroid drops. Our patient had bilateral presentation, was already on prednisolone drops, and had no previous similar episodes.

Anecdotal reports of specific drug treatment for microsporidiosis include albendazole, antifungal azoles, propamidine, fluoroquinolones, chlorhexidine, PHMB, and fumagillin, but their effectiveness is debatable as the condition

is usually self-limiting.^[4] Stopping of steroids is recommended to allow the immune system to clear the infection faster.^[4] However, this carries the risk of triggering inflammation and graft rejection in a patient with keratoplasty. There is no reported optimal time frame for which steroids should be stopped. The average resolution time in literature is about 6 ± 2.9 days.^[5] In our patient, there was graft edema and rise in intraocular pressure due to inflammation following stoppage of steroids. After 12 days of steroid withdrawal, most epithelial lesions had resolved and we restarted steroids. The grafts cleared up fully with complete resolution of inflammation. It is a tricky situation as waiting longer could have resulted in triggering a graft rejection while reinstatement early could lead to recurrence of infection.

Conclusion

Microsporidial keratitis should be considered in non-resolving coarse punctate keratitis. A good microbiologic evaluation is essential to establish the diagnosis.

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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Nil.

Conflicts of interest

There are no conflicts of interest.

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