Microsporidial stromal keratitis in an immunocompromised patient: Successful management with medical therapy

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Microsporidial stromal keratitis is refractory to topical drugs and is classically described in immunocompetent hosts. A 55-year-old patient with renal transplant and oral immunosuppressants, presented with a 15-day history of redness, pain, and diminution of vision in the right eye. Slit-lamp examination revealed epithelial defect and mid-stromal infiltrate. On corneal scraping, microsporidial spores were observed. The patient was started on topical 0.02% polyhexamethylene biguanide (PHMB) and the infiltrate resolved after 6 weeks of initiation of topical therapy.

Key Words: Medical therapy, microsporidia, polyhexamethylene biguanide, stromal keratitis

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Ocular microsporidiosis occurs in two forms: keratoconjunctivitis and stromal keratitis. Former is reported in immunosuppressed and latter in immunocompetent individuals.[1] *While* microsporidial keratoconjunctivitis is usually self-limiting in nature, there is no definitive medical management for stromal keratitis. Several antimicrobial treatments have been tried in stromal keratitis, but to no avail.[2] There have been a few case reports where microsporidial stromal keratitis has been treated with medical management. Herein, we report the first case of microsporidial stromal keratitis in an immunosuppressed host that responded to medical management with topical monotherapy of 0.02% polyhexamethylene biguanide (PHMB).

Case Report

A 55-year-old male presented with redness, watering, and diminution of vision in his right eye for 15 days. There was

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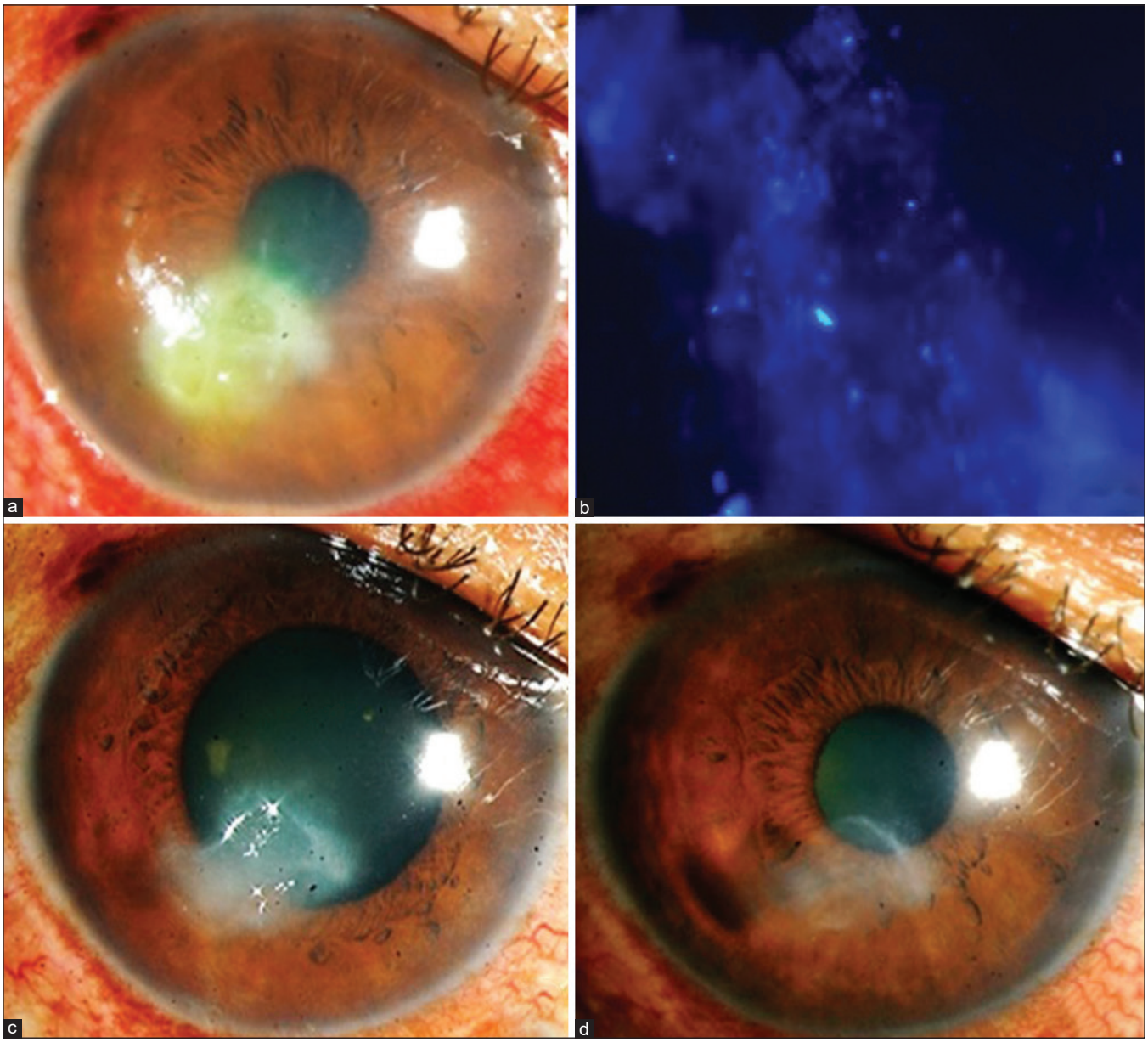

Figure 1: (a) Slit-lamp picture in diffuse illumination on the day of presentation (b) Microsporidial spores on calcofluor white stain (40 ×) (c) Slit-lamp picture demonstrating resolving stage in the 3rd week (d) Slit-lamp picture demonstrating resolved with a scar in the 6th week

no history of ocular trauma or usage of the contact lens. The patient was diabetic, hypertensive, and had undergone a kidney transplant in 2016. He was on systemic tacrolimus (0.5 mg) twice daily, azathioprine (50 mg) once daily, and prednisolone (5 mg) once daily. He was also on insulin as well as antihypertensive medications. On presentation, his visual acuity of the right eye was 20/200 and examination revealed 3 × 3 mm mid-stromal infiltrate with overlying epithelial defect of 2 × 2 mm and streak of hypoyon [Fig. 1a]. The patient underwent routine microbiological workup. Microsporidia spores were seen in potassium hydroxide wet mount with calcofluor white stain [Fig. 1b]. The culture was sent on blood, chocolate, and Sabouraud dextrose agar and no growth was observed at 14 days.

The patient was counseled about the refractory nature of the disease and hourly instillation of 0.02% PHMB eye drop was started. On a 1-week follow-up, there was a marginal improvement and medical treatment was continued as the infiltrate was small in size. During the 2-week follow-up, the infiltrate was decreasing with the absence of hypopyon [Fig. 1c]. The infiltrate resolved with a scar and the visual acuity improved to 20/25 at the last follow-up, 6 weeks later [Fig. 1d].

Discussion

Microsporidia are eukaryotic, intracellular, and spore-forming protozoa belonging to the phylum Microspora and family Microsporidia. A total of 7 out of 150 genera as well as unclassified microsporidia (collectively referred to as microsporidium) have been associated with human disease.[3]

Microsporidial keratoconjunctivitis is usually caused by the genus Encephalitozoon and has been classically described to

affect immunosuppressed patients in the literature, though most of the patients in our routine practice are immunocompetent. Microsporidial stromal keratitis is caused by the genus Nosema and has been observed in immunocompetent patients. It presents as insidious, suppurative or nonsuppurative inflammation, and vascularization of the cornea, mimicking herpes simplex, or fungal keratitis.[4] Ours is the first case report of microsporidial stromal keratitis in a patient who is on immunosuppressant medication.

While microsporidial keratoconjunctivitis is usually self-limiting in nature, there is no definitive medical management for stromal keratitis.[2] There have been a few case reports where microsporidial stromal keratitis has been treated with medical management. Sangit *et al*. had treated one case with topical chlorhexidine gluconate (0.02%) hourly and oral albendazole (400 mg) twice daily. Later, topical loteprednol etabonate (0.5%) was given for a total of 12 weeks.[5] Coca *et al*. had successfully treated one such patient with topical voriconazole (1%) and oral itraconazole (200 mg) twice daily.[6] Shabhapandit *et al*. had treated five out of 34 patients with combination therapy of topical 0.02% PHMB and 0.02% chlorhexidine, with/without oral albendazole. Three of these medically treated patients were lost to follow-up, leading to the ambiguity of treatment.[7]

Since stromal keratitis has such a low success rate with topical management, excision of the infected corneal tissue (in the form of keratoplasty), is the most suitable way of eradicating the microbe. We also explained the option of therapeutic keratoplasty to the patient in the first visit but he responded to 0.02% PHMB. Therefore, he was continued on medical management, making it the first case of microsporidial stromal keratitis responding to topical monotherapy of 0.02% PHMB. PHMB is an antiparasitic drug that is used as first-line therapy for *Acanthamoeba* keratitis. It acts by disrupting membrane integrity and leakage of cellular contents.[8]

Despite being immunosuppressed, our patient responded well to medication. This might be due to early presentation as well as early diagnosis. One large case series on microsporidial stromal keratitis had described the mean duration of presentation as 288 days. Moreover, >80% of cases in their series presented with intact epithelium and nonsuppurative infection.[7] In our case, the patient presented with acute features of pain and redness (15 days). The early presentation might be due to the immunosuppressed condition of the patient. Moreover, the presence of relatively superficial infiltrates with an overlying epithelial defect prompted us to scrape on the 1st day. Besides, the size of the infiltrate was small and restricted up to mid-stroma; making the drug permeation easy.

Conclusion

In nutshell, microsporidial stromal keratitis may occur in immunosuppressed patients. Medical management with topical monotherapy of 0.02% PHMB may be successful in cases of early presentation.

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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Conflicts of interest

There are no conflicts of interest.

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