CASE REPORT OPEN ACCESS

# Microsporidial Keratitis - First Case Series of a Rare Pathogen in the Wake of Flood Disasters of 2022 in Pakistan

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### **ABSTRACT**

The recent monsoon rains in Pakistan were unprecedented and caused flooding all over Pakistan, especially in Sindh and Balochistan. Following this national disaster, various water-borne and contagious diseases started erupting all over the country. In such a calamity-struck city of Jacobabad, we started receiving cases with a peculiar set of ocular complaints mimicking viral keratoconjunctivitis. Failure to respond to traditional treatment and the unique appearance of these corneal opacities led to a rare diagnosis of Microsporidial Keratoconjunctivitis, which was later confirmed by microscopy and staining of corneal scrapings of the most affected case. In line with published literature, all cases were treated with topical fluoroquinolone and topical anti-fungal therapy, following which the disease was cleared within a week. The disease has seen an upward trend the world over, especially among Asia. To the best of our knowledge, no such cases have been reported in Pakistan as yet. In this case series, we highlight the strong correlation of emergence of microsporidial keratitis in patients following exposure to pooled water bodies after the monsoon rainy season and floods. Moreover, this report will help create awareness in eye professionals regarding the prevention, timely diagnosis and treatment of these rare and emerging cases.

Key Words: Keratitis, Spores, Water-borne diseases, Microsporidia

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# **INTRODUCTION**

Microsporidia is a family of very rare, spore-forming organisms which tend to infect humans and animals. Although a large family of 1,300 formally described species in 160 genera, only 13 species have been known to cause human disease. These organisms most commonly involve the gastrointestinal tract followed by infection of the cornea, which is relatively rare. Being a relatively rare disease, ophthalmologists often face challenges regarding its diagnosis and treatment. We present three cases of microsporidial keratoconjunctivitis that were reported after exposure to flood water.

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# **CASE REPORTS**

We recorded data on three (1 male, 2 females) cases of microsporidial keratoconjunctivitis. They reported over a course of 1 month presenting in Eye OPD at a hospital in Sindh Province of Pakistan, the area which has been heavily affected by the monsoon rains and floods in the region from June to September 2022. All of them were young patients (15 to 36 years) presenting with the similar unilateral presentation of redness, photophobia and non-purulent conjunctivitis. Human immunodeficiency virus (HIV) serology of all three was negative and the rest of the systemic investigations were also within normal limits.

**Case 1:** A 15-year female presented with 4 days history of redness, grittiness and mild discomfort in the right eye. She was residing in makeshift tents community following the destruction of houses by floods. On ocular examination, the visual acuity (VA) of her right eye was 6/9. Slit-lamp examination showed multiple, well-delineated, small and coarse epithelial and subepithelial opacities in the superotemporal quadrant of the cornea (Figure 1). The underlying stroma and endothelium were normal. This superior bulbar conjunctiva was inflamed with mild chemosis. The anterior chamber was quiet and the

rest of the posterior segment examination was also unremarkable. Intraocular pressures were within normal limits. An initial clinical diagnosis of Thygeson Superficial Punctate Keratopathy was made, but later on, the clinical scenario and treatment response favoured the diagnosis of Microsporidial Keratitis. She was started on a combination of topical steroids and antibiotics (Tobramycin-Dexamethasone) 6 hourly along with topical lubricants and was called for follow-up after 1 week. No further investigation was carried out. She responded well to the above treatment.

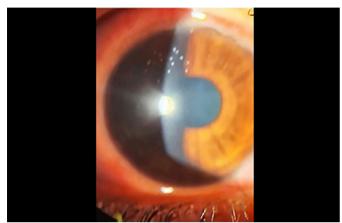


Figure 1: Corneal lesions seen during slit-lamp examination.

Case 2: A 36-year male, optician, presented to eye OPD with a history of pain, blurring, redness and photophobia of the left eye for 1 week, for which, he consulted a local doctor and was instilling acyclovir eye ointment and tobramycin-dexamethasone eye drops but the symptoms were getting worse. On further inquiry, he revealed his symptoms appeared a few days following eye camps in the flooded regions. On ocular examination, VA of the left eye was 6/60 and he was having lid swelling. Corneal examination revealed diffuse, greyish-white, coarse, epithelial and subepithelial, multifocal, well-defined opacities (Figure 2). The posterior segment examination was unremarkable. Diagnostic and therapeutic corneal scraping was carried out followed by gram staining and light microscopy which revealed spores of microsporidia (Figure 2). Once the diagnosis was established, his steroids were immediately tapered down. Topical moxifloxacin was initiated 2 hourly along with topical Fluconazole 2 mg/ml 4 hourly with topical lubricants and oral doxycycline. The patient started improving the very next day with marked visual and clinical improvement in lesions as well. VA improved from 6/60 to 6/9 and the cornea was completely clear by 2<sup>nd</sup> follow-up visit after 1 week (Figure 2).

Case 3: A 26-year female, presented with mild redness, photophobia and grittiness of left eye. She had the exact same corneal opacities like the previous two cases (Figure 3). She was, thus, clinically diagnosed as a case of Microsporidial kerato-conjunctivitis and was given the same antimicrosporidial treatment regimen (Moxifloxacin-fluconazole-flouromethalone). She responded very well and, all the corneal opacities had healed and the VA had improved from 6/36 to 6/6 as reported on her follow-up visit after a week. There was a drastic improvement in symptoms as well.

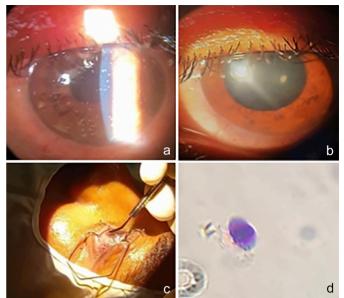


Figure 2: Progress of Case after Corneal debridement and Treatment. (a) Day 1 presentation (b) Day 7 post-treatment (c) Corneal scraping (d) Spore of microsporidia on light microscopy.



Figure: 3 Progress of case 3 before and after treatment. (a) Day of presentation (b) Post-treatment results.

### DISCUSSION

An unprecedented monsoon caused massive flooding in Pakistan this year. Approximately 1,678 people have died, including 555 children, and an additional 12,860 were injured. Around 546,288 people are living in temporary tents because of the floods. The flood situation along with compromised state of hygiene in tents led to the appearance of different pathogens. In the 2010 flooding seen in northern Pakistan, the subsequent disease trend also showed a tendency for eye diseases in camp settings.<sup>3</sup>

Microsporidia are predominantly seen in the humid environments of tropical regions. The exact route for corneal involvement remains unclear. However, trauma, contact lens use, and contact with infested water have been implicated.<sup>4</sup> Whereas authors noted that 50% of patients had ocular exposure to muddy water in one setting, however, an outbreak in Taiwan linked all cases to have followed exposure to infected water of a

swimming pool.<sup>4,5</sup> Although HIV-infected persons have a high prevalence of microsporidiosis, it is also seen in HIV-negative, immunocompetent cases,<sup>6</sup> as in these three patients. There is an increased incidence of disease in July to October corresponding to the monsoon season in Asia, which can cause microsporidial contamination in water bodies,<sup>7</sup> as was the case with our patients which cropped up after the monsoon rains.

The presentation of ocular microsporidiosis can either be in an acute setting as microsporidial keratoconjunctivitis or a rather slow and insidious microsporidial stromal keratitis. The clinical picture of microsporidial keratitis may be confused with atypical adenoviral conjunctivitis and is generally treated as such. A failure to respond to topical steroid treatment may hint towards a microsporidial infection. In addition, the multiple, coarse and raised epithelial infiltrates seen in microsporidiosis distinguish it from nummular infiltrates of adenoviral keratoconjunctivitis, Thygeson's superficial punctate keratitis or acanthamoeba. The prevalence of microsporidial keratoconjunctivitis is increasing worldwide; even though most cases are either misdiagnosed or undiagnosed.

Once clinical suspicion of microsporidiosis is made, it can be confirmed by corneal scrapings. Gram staining and light microscopy will reveal the gram-positive spores in the epithelium,<sup>2</sup> as was seen in our cases. Fluoroquinolone monotherapy has been advocated with promising results. This may be augmented by topicalantifungaltreatment. 4Additionally, fumagillinandalbendazole have also been used successfully.2 Researchers are divided regarding the use of topical steroids. Whereas, a number of authors are in favour of topical steroids to reduce the local inflammation and prevent its sequelae (uveitis, stromal haze and nummular keratitis), others have proposed disease progression by promotion of local immunocompromised state due to topical steroids. 4 Studies suggest that corneal epithelial debridement achieves the best resolution even without topical treatment. 10 As we had also witnessed a dramatic clinical and symptomatic improvement in our patient after the scrapings were performed. Further studies on a larger scale are required to corroborate the dual role of corneal scrapings in debulking the disease and to provide diagnosis as well.

In conclusion, this case series will raise awareness regarding microsporidiosis and its diagnosis. Being an agricultural country, the incidence of this disease may be very high, especially in rural areas of Pakistan. This case series might also help us in finding new insights into the treatment of this emerging pathogen.

## **PATIENTS' CONSENT:**

Informed consent were obtained from the patients to publish the data.

### COMPETING INTEREST:

The authors declared no competing interest.

### **AUTHORS' CONTRIBUTION:**

SM: Diagnosis of cases, clinical follow-up, and manuscript writing.

MI: Proofreading and final approval of the manuscript.

SN: Manuscript writing.

SH: Literature search and clinical follow-up.

All the authors have approved the final version of the manuscript to be published.

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