

Case report

Stromal microsporidial keratitis successfully treated with medical therapy

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ABSTRACT

Purpose: To report a case of severe stromal microsporidial keratitis successfully treated with oral albendazole and topical voriconazole.

Observations: A 71-year-old man presented with progressive vision loss and corneal opacification for one year. Initial visual acuity was counting fingers attributed to a dense subepithelial opacification. Confocal microscopy and subsequent corneal biopsy lead to the diagnosis of microsporidial keratitis. The patient completed a four-week course of oral albendazole and topical voriconazole which resulted in resolution of the corneal opacification and improvement in visual acuity to 20/250.

Conclusions and Importance: A four-week course of oral albendazole and topical voriconazole was an effective treatment for severe stromal microsporidial keratitis.

1. Introduction

Microsporidia are a group of spore-forming fungi that rarely cause infectious keratitis.¹ While the cornea is the most common site of ocular infection, scleritis and endophthalmitis have been described.² Clinically,

microsporidial keratitis is suspected when culture-negative keratitis does not respond to conventional antibiotic therapy. Methods used to diagnose microsporidial keratitis include microscopic examination of biopsy or scraping with stains such as acid fast, Giemsa, calcofluor white, or potassium hydroxide.^{1,3}

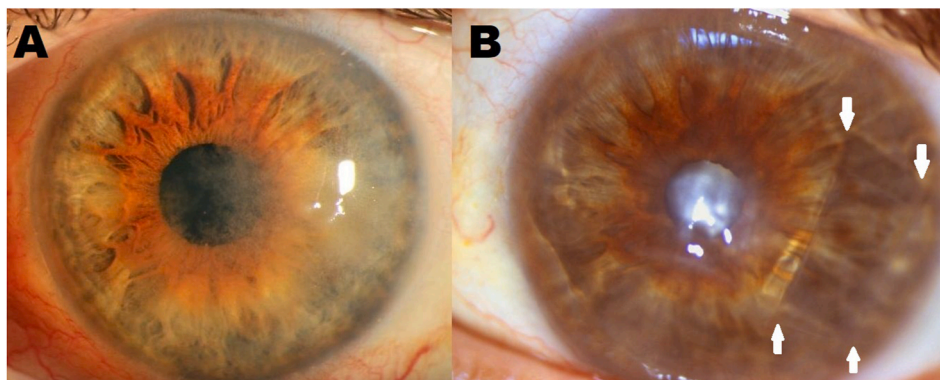


Fig. 1. Initial slit-lamp photograph showing stromal opacity affecting mostly temporal quadrants of the cornea and involving the visual axis (A). Resolution of stromal infiltrates and opacities after treatment. The healed area of biopsy is noted by arrows. A cataract has formed that limited his visual acuity (B).

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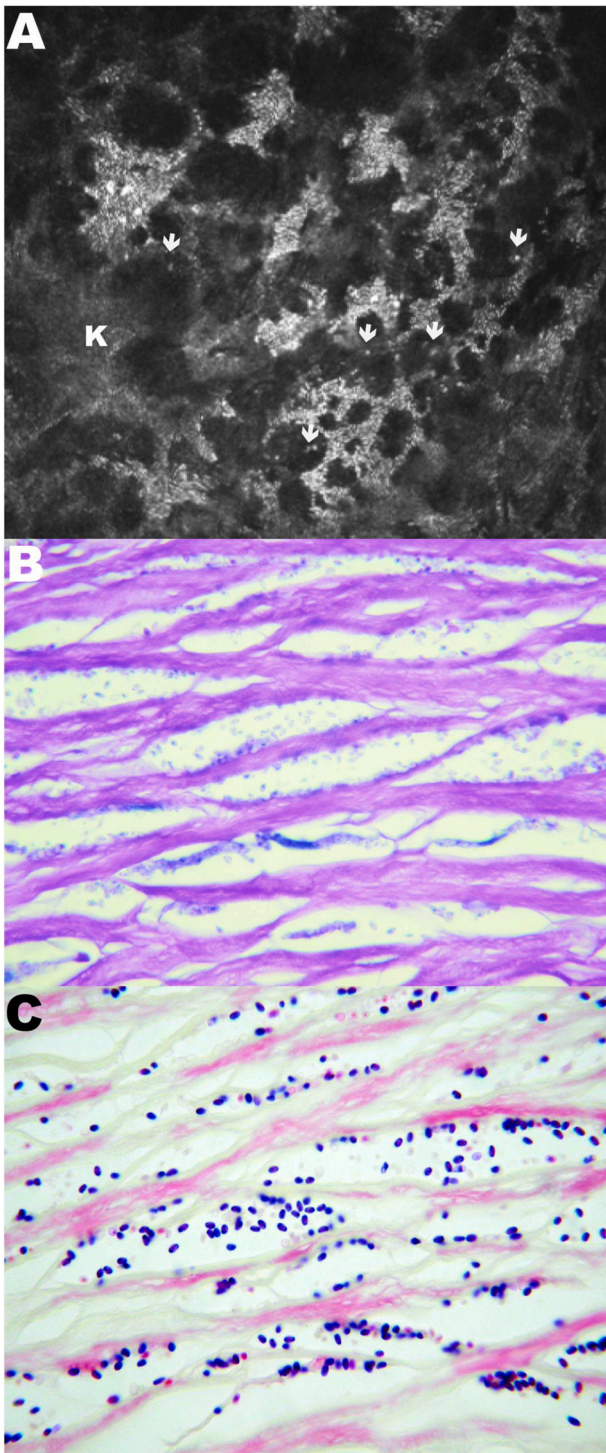


Fig. 2. In vivo confocal microscopy and histological demonstration of microsporidial organisms. Confocal microscopy microphotograph showing keratocytes (K) and small ovoid intercellular objects believed to correspond to microsporidia (white arrowheads) (A). Biopsy stained with hematoxylin-eosin showed hundreds of translucent oval bodies averaging 2.5 to 4 μm in size a majority of which are between collagen lamellae. Spindle-shaped keratocytes also contain slightly smaller translucent spheres (B). Tissue Gram stain shows hundreds of blue oval bodies many of which contain faint pink polar nuclei. A minority of oval bodies are visible but unstained (C). (Original magnification $\times 375$). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

Superficial punctate keratitis secondary to microsporidia can be managed with topical agents or epithelial debridement. Definitive treatment of microsporidial stromal keratitis, however, often requires corneal transplantation.^{3,4} There are several reports of successful medical treatment of microsporidial stromal keratitis.^{4,5} We report a case of microsporidial stromal keratitis documented by confocal microscopy and its successful treatment with a combination of oral albendazole and topical voriconazole.

2. Case report

A 71-year-old man with a past medical history of coronary artery disease, diabetes, and hypertension was referred for management of progressive opacification of the left cornea. There was no history of ocular trauma or contact lens use. He had been treated at various times for over a year with topical antibacterial, antiviral, and corticosteroid regimens, as well as an oral antiviral medication without improvement. At presentation, his vision in the affected eye was counting fingers. No corneal epithelial defects were present. A dense granular opacification of the anterior stroma involved the visual axis (Fig. 1A). Confocal microscopy showed numerous minute oval bodies in the anterior stroma (Fig. 2A). A corneal biopsy revealed myriad translucent oval microorganisms in the stroma without inflammation. The majority of microorganisms were Gram-positive, periodic acid-Schiff negative, and Giemsa stain variable (Fig. 2B and C). The patient was started on topical voriconazole every 2 hours and oral albendazole 400 mg twice daily.

According to the patient, he completed the 4-week course of albendazole and voriconazole. He did not return for follow-up but was seen in consultation when hospitalized for congestive heart failure. At that time, he had been off his medications for four months. Examination showed that his vision improved to 20/250 with correction. The eye was not inflamed. The corneal epithelium was intact with mild subepithelial haze. There were no signs of active keratitis. The granular stromal opacity had completely resolved (Fig. 1B). His visual acuity was attributed to white cataract.

3. Discussion

Microsporidia remains an uncommon cause of infectious keratitis in the United States. It is more often reported in Asian countries. Like other infectious keratopathies, early diagnosis and treatment are vital in minimizing stromal scarring and vision loss. Superficial microsporidial keratitis can be seen in immunocompromised patients when it usually presents as a superficial punctate keratitis or as keratoconjunctivitis. Microsporidial keratoconjunctivitis has been considered a self-limited infection, but there are ample examples of successful treatment with topical voriconazole or fumagillin.^{6–10}

The clinical presentation of microsporidial stromal keratitis overlaps that of other stromal infections. Disease progression is comparably slow and insidious, however. Diagnosis is typically achieved when empirical treatments fail and tissue or exfoliation specimens are obtained.^{4,8,11–13} While superficial infections can be self-limited or treated medically, deeper stromal involvement often requires corneal transplantation to eradicate the infection and improve vision.^{3,4,8} There is no consensus on optimal medical treatment for stromal disease. A few reports in the literature describe successful medical treatment. Among the medications in these reports used alone or in combination are topical polyhexamethylene biguanide, chlorhexidine and voriconazole with or without oral itraconazole or albendazole.^{4,5,8,14} In one retrospective clinical series involving 34 patients, less than half responded to medical therapy consisting of topical polyhexamethylene biguanide and chlorhexidine with or without oral albendazole. Ultimately 25 of the 34 patients required keratoplasty.⁴

4. Conclusions

We present a case of corneal stromal microsporidiosis documented with vivo confocal microscopy. The infection was successfully treated with topical voriconazole and oral albendazole.

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Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

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