Journal of Infection (2005) **51**, 325–328

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Keratitis caused by *Trachipleistophora anthropopthera*

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Accepted 18 August 2004
Available online 30 September 2004

KEYWORDS
Stromal keratitis;
Microsporidium;
Trachipleistophora anthropopthera;
Keratoplasty;
Electron micrograph

Abstract We report a case of intrastromal keratitis in a 42-year-old male with underlying human immunodeficiency virus-1 infection. Numerous microsporidial spores were found from corneal biopsy. Ultrastructural studies of corneal tissues revealed dimorphic sporophorous vesicles containing characteristic spores belonging to *Trachipleistophora anthropopthera*. Infection could be controlled by penetrating keratoplasty but not by topical fumagillin and systemic albendazole per se. This is the first report of human keratitis caused by this organism.

Introduction

Microsporidia are groups of obligate intracellular protozoa known to infect a variety of hosts, ranging from invertebrates to vertebrates. At least seven well-documented genera have been identified as human pathogens, i.e. *Enterocytozoon, Encephalitozoon, Nosema, Pleistophora, Vittaforma, Brachiola* and *Trachipleistophora*.[1,2] Although microsporidia are well recognized as opportunistic agents, the majority of infected cases occur in patients with underlying human immunodeficiency virus-1 (HIV-1) infection. Microsporidiosis may present with multi-organ involvement or localized symptoms, probably depending on infecting parasite species and mode of transmission.[1–3] The most common sites of infections are in alimentary system, causing chronic diarrhoeal illness. Meanwhile, corneal infection by microsporidia is uncommon and thus may be overlooked. To date, six species and two unclassified microsporidia have been known to cause keratoconjunctivitis.[1] Here we report the first case of corneal infection due to *Trachipleistophora anthropopthera* in an HIV-infected patient.

Case report

A 42-year-old Thai male patient was referred to our hospital in April 2000 with a history of redness, pain, irritation and slowly progressive decreased vision in the left eye over the period of 1-year and 4 months. His visual acuity before the present illness was normal. He could not recall experiencing

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0163-4453/$30.00
doi:10.1016/j.jinf.2004.08.030

any eye trauma prior to symptoms. He was previously treated with topical steroids to control inflammation of the left eye. Six months thereafter, secondary glaucoma developed in the left eye, which required antiglaucoma drugs administration. He was known to be positive for HIV-1 since 1999. His $CD4^+$ T-lymphocyte count was 5 cells/μl and HIV-1 RNA 121 869 copies/ml.

Physical examination revealed a slightly thin man. Visual acuity in the right eye was 20/30, and light perception in the left eye. Slit lamp examination showed dense stromal infiltration of left cornea, about 8×6.5 mm^2, with spicule-like border but the corneal epithelium remained intact and the anterior chamber was clear. His right eye was normal. Repeated conjunctival and corneal scrapings gave negative results for bacteria, fungi and acanthamoebae by staining and culture methods. Microsporidia could not be detected by modified trichrome stain of scraped corneal tissues, urine and stool samples.

In January 2001, corneal biopsy was performed. Small portions from biopsied materials were used for bacteriologic and fungal cultures, which yielded negative results. However, abundant oval-shaped spores measuring $2\text{-}2.5\times3.8\text{-}4.0$ μm^2 were observed by modified trichrome stain of corneal tissue (Fig. 1). Therapy was commenced with albendazole 400 mg twice daily and topical fumagillin (fumagillin bicyclohexylammonium salt 0.07 mg/ml, Fumidil B) applied 2-hourly as described.[5-7] Three weeks after biopsy, corneal infiltration increased in both diameter and intensity. Slit lamp examination also revealed flattened anterior chamber with prolapsed iris. Therefore, penetrating keratoplasty was performed.

The corneal button was found to harbor abundant microsporidial spores upon modified trichrome stain method. Histopathologic studies under light microscope revealed numerous microsporidial spores in desquamated corneal epithelium with some inflammatory responses. Transmission electron microscopy demonstrated various developmental stages of microsporidia surrounded by sporophorous vesicles. Two distinct types of spores were observed, i.e. (1) thick-walled spores measuring 3.7×2 μm^2 with mammilated exospore containing 8–9 coils of polar filaments and 3–4 coils of thinner filaments at the posterior portion lying somewhat inward, and (2) thin-walled spores measuring 2.4×2 μm^2 with 2–3 coils of polar filaments (Fig. 2). Thick-walled spores were more abundant in corneal tissue than thin-walled spores. These characteristics have previously been described for *T. anthropopthera* found at autopsy of two patients with acquired immunodeficiency syndrome (AIDS).[4]

The patient received oral albendazole and topical fumagillin for 6 months after keratoplasty. Two and a half years after keratoplasty, there was no recurrence of keratitis. However, the visual acuity in the left eye remained unchanged from that before treatment because of optic nerve damage from steroid induced glaucoma prior to penetrating keratoplasty.

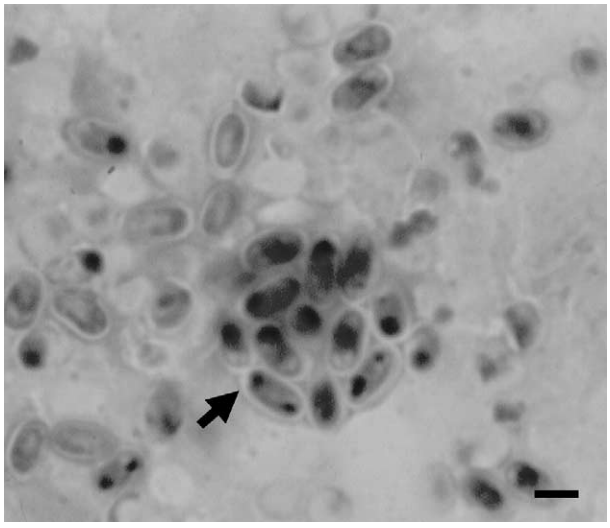

Figure 1 Microsporidial spores in corneal tissue stained with modified trichrome method. An arrow indicates a cluster of spores. Bar = 2 μm.

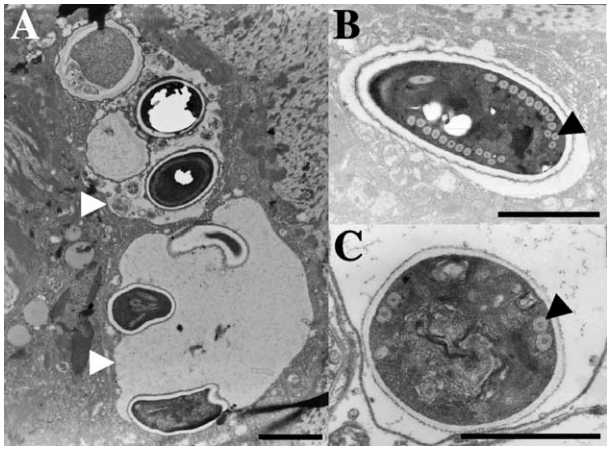

Figure 2 Transmission electron micrographs depicting polysporoblastic vesicles (white arrowheads) containing immature and mature spores of *T. anthropopthera* (A), a thick-walled spore with 8–9 coils of polar filaments and 3–4 coils of thinner filaments (black arrowhead) (B), and a thin-walled spore with 2–3 coils of polar filaments (black arrowhead) (C). Bars = 1 μm.