Case Report

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Dendrite-like anterior stromal keratitis coinfected with *Acanthamoeba* and *Pseudomonas* in an orthokeratology contact lens wearer

Chih-Chien Hsu[1,2]

Abstract:

Acanthamoeba species can cause a keratitis misdiagnosed as herpes keratitis or fungal keratitis. We report an unusual dendrite-like anterior stromal keratitis coinfected with *Acanthamoeba* and *Pseudomonas aeruginosa* in an orthokeratology contact lens wearer in Taiwan. Topical 1% voriconazole and 0.5% levofloxacin were prescribed because besides *Acanthamoeba* keratitis, fungal keratitis was also highly suspected initially. Topical 0.02% chlorhexidine was added after the culture of the scraped cornea showed positive results of *Acanthamoeba* and *P. aeruginosa*. The lesion subsided using this triple combination therapy for 1 week. Both *Acanthamoeba* and *P. aeruginosa* are potentially devastating causes of infectious keratitis. Our case highlights the importance of considering the possibility of a concurrent infection and atypical presentation in cases with contact lens-related keratitis. The use of topical levofloxacin combined with voriconazole should be considered as the first-line treatment in such patients.

Keywords:

Acanthamoeba keratitis, coinfection, levofloxacin, orthokeratology, *Pseudomonas*

Introduction

Acanthamoeba keratitis (AK) is a severe but unusual infectious disease of the cornea. The pathogen, *Acanthamoeba*, is a free-living cyst-forming protozoan that is distributed in diverse environments including air, soil, dust, and water. The active form of Acanthamoeba is trophozoite which has an amoeboid shape with pseudopodia, phagocytoses any encountered small sized particle and feeds on keratocytes in the cornea. The cystic form can survive in a difficult environment, and an effective treatment of AK needs cysticidal drugs.[1] Unlike AK, *Pseudomonas aeruginosa* keratitis usually progresses rapidly and presents with significant suppurative stromal infiltrate and mucopurulent exudate. It is thought that AK can develop in eyes with advanced bacterial keratitis. Coinfections with other microorganisms have been reported in patients with culture-proven AK.[2-5] Herein, we report a case of dendrite-like anterior stromal keratitis coinfected with *Acanthamoeba* and *P. aeruginosa*, who was an orthokeratology contact lens wearer in Taiwan.

Case Report

A 20-year-old male orthokeratology contact lens wearer presented complaining of eye pain and redness in the right eye for 2 days. The patient reported that he had worn orthokeratology contact lens continuously for 30 h. After he removed the contact lens, photophobia of the right eye was noted. He went to a clinic where topical 0.25% chloramphenicol eye drops and 0.5% erythromycin eye ointment were prescribed. He went to our clinics due to the symptoms

[1]*Department of Ophthalmology, Taipei Veterans General Hospital,* [2]*Institute of Clinical Medicine, National Yang-Ming University, Taipei, Taiwan*

Address for correspondence:
Dr. Chih-Chien Hsu, Department of Ophthalmology, Taipei Veterans General Hospital, No. 201, Sec. 2, Shih-Pai Road, Taipei 112, Taiwan.
E-mail: chihchienym@gmail.com

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progressed 1 day later. The slit-lamp examination showed a paracentral dendrite-like anterior stromal infiltration with feathery border. There is a partial epithelial defect at the central part of the lesion [Figure 1]. Besides AK, fungal keratitis was highly suspected and topical 0.5% levofloxacin and 1% voriconazole each hour were prescribed first. The culture of the scraped corneal tissue was positive for *P. aeruginosa* and *Acanthamoeba* 3 days and 6 days later, respectively. The lesion subsided in a week after adding topical chlorhexidine 0.02% each hour [Figure 2]. Except paracentral faint corneal scar, the patient had final best-corrected visual acuity 20/20 in both eyes in 1-month follow-up.

Discussion

Biguanides (i.e., polyhexamethylene biguanide and chlorhexidine) and diamidines (i.e., propamidine and hexamidine) are currently available and most often used antiamoebic agents offering both trophozoicidal and cysticidal effects. Recently, an *in vitro* study shows that voriconazole, an antifungal agent, has good cysticidal effect against *Acanthamoeba*.[6] Another study even believes that topical voriconazole has better corneal and anterior chamber penetration and lower cellular toxicity on the cornea than topical chlorhexidine.[7] It may be the first drug of choice of atypical AK that presents like fungal keratitis when the culture result cannot be reached.

Endosymbiont bacteria has been known to exist in *Acanthamoeba* hosts and influence *Acanthamoeba* virulence, AK clinical features, and susceptibility to antiamoebic drugs.[2,8] Iovieno *et al*. found *Acanthamoeba* hosting bacterial endosymbionts in half of AK patients. Half of AK patients with endosymbiont belonged to the genus *Pseudomonas*. Previously, it was assumed that *P. aeruginosa* and *Acanthamoeba* were mutually exclusive ocular pathogens.[9] However, *in vitro* studies have showed that *Pseudomonas* can increase the resistance of *Acanthamoeba* to contact lens disinfecting solutions and create a biofilm on contact lens surface enhancing *Acanthamoeba* retention.[10] The presence of the endosymbiont may modify *Acanthamoeba* phenotype making the protozoa more pathogenic or resistant to therapy.[11] Changes in gene expression and protein profiles have also been observed resulting from the amoeba/bacteria interaction in *Hartmannella*.[12] Nakagawa *et al*. found that the presence of bacteria is essential and a critical number of bacteria is required for the development of AK. The time of coexistence with bacteria may also be an important determinant of the severity of AK.[13] Besides, cases of contact lens-related microbial keratitis caused by *P. aeruginosa which* presented with perineural infiltrates, a typical characteristic of AK, were also reported.[14] It is thus reasonable to use topical levofloxacin in suspected AK patients before the acquirement of culture result. In our case, topical levofloxacin and voriconazole were chosen as the first line treatment. We added topical chlorhexidine after the culture results showed *P. aeruginosa* and *Acanthamoeba*. Ortillés *et al*. found that combined chlorhexidine, voriconazole, and ciprofloxacin showed the greatest amoebicidal activity *in vitro* although monotherapy may still have its effects.[15] The lesion in our case subsided in 1 week after the adding of topical chlorhexidine.

Conclusion

Both *P. aeruginosa* and *Acanthamoeba* are potentially devastating causes of infectious keratitis. Our case highlights the importance of considering the possibility of a concurrent infection and atypical presentation in cases with contact lens-related keratitis. Topical voriconazole should be considered as the first-line treatment for patients with AK suspected of fungal keratitis. The use

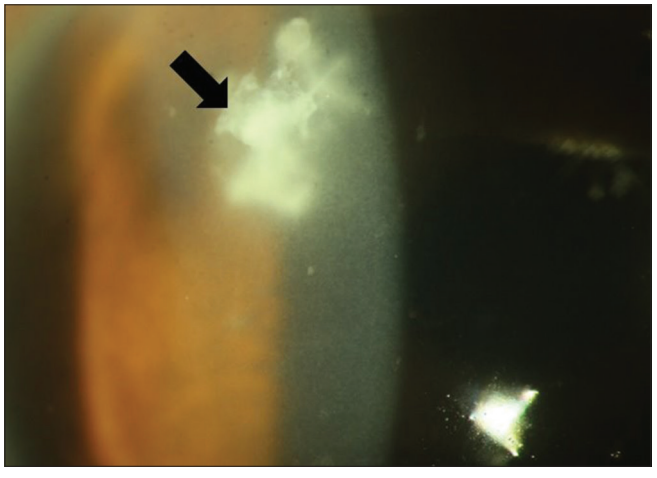

Figure 1: Dendrite-like anterior stromal infiltration with feathery border was found at the paracentral cornea. The black arrow indicated the site with epithelial defect

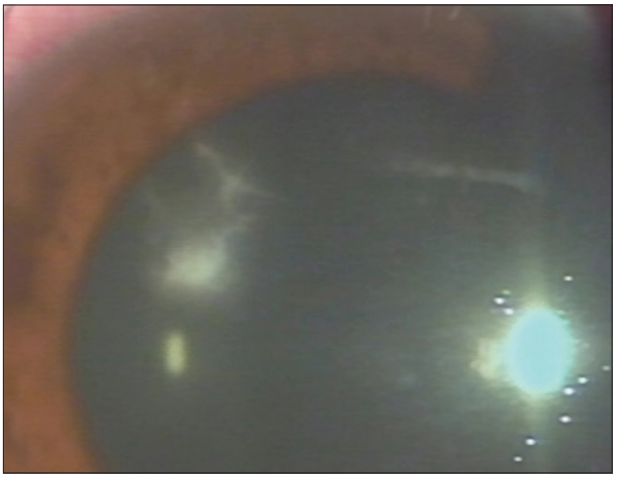

Figure 2: The cornea became silent and fibrosis after 1-week treatment

of topical levofloxacin cannot only control *Pseudomonas* but also help to treat *Acanthamoeba* in such patients who are already under antiamoebic treatments.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

The authors declare that there are no conflicts of interests of this paper.

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