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Case Report

Post-Keratoplasty Keratitis Associated with *Sphingobacterium spiritivorum*

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Received: September 24, 2020; **Published:** October 31, 2020

Abstract

Purpose: To describe a patient with *Sphingobacterium spiritivorum* as a co-infective organism in polymicrobial keratitis after corneal transplant.

Observations: A 56-year-old man on systemic immunosuppression for atopic dermatitis, with a history of keratoconus, bilateral herpetic keratitis, and a previous penetrating keratoplasty presented with pain in the right eye. Initial examination revealed a geographic epithelial defect associated with a loose suture at the graft host junction. On follow up examination the next day, a corneal infiltrate was identified, cultures from which revealed coagulase-negative *Staphylococcus*, *Staphylococcus aureus*, *Streptococcus viridians*, and *Sphingobacterium spiritivorum*. The patient responded to empiric therapy with topical moxifloxacin and fortified vancomycin and tobramycin.

Conclusion and Importance: *Sphingobacterium spiritivorum,* a rare aerobic, gram-negative rod, may be a co-infecting agent in microbial keratitis in immunosuppressed patients. Complete resolution can be achieved by the standard topical antimicrobial therapy.

Keywords: *Keratitis; Keratoplasty; Sphingobacterium spiritivorum*

Introduction

Microbial keratitis is an uncommon complication following keratoplasty that may develop at any time after surgery and can reduce graft survival [1]. Risk factors for microbial keratitis include contact lens wear, exposed sutures, topical steroids and immunosuppression among others [1]. The most common causes of microbial keratitis after keratoplasty are *Staphylococcus aureus, Staphylococcus epidermidis,* and *Moraxella* species [1]. Herein we describe a patient who developed a post-keratoplasty corneal infection with multiple organisms including *Sphingobacterium spiritivorum.* This organism, a rare gram-negative rod, has been reported to cause opportunistic infections in immunocompromised patients [2]. To our knowledge, there has been no previous report of this pathogen in microbial keratitis after corneal transplantation.

Case Report

A 56-year-old man had a history of rigid gas permeable contact lens-dependent keratoconus, bilateral herpetic eye disease, and atopic dermatitis. In 2015, he underwent a therapeutic keratoplasty for a perforated corneal ulcer in the right eye, the cultures from which grew Fusarium. There was no recurrence of infection after antimicrobial therapy and keratoplasty. The best corrected visual after surgery was 20/20 with a scleral contact lens.

Three years later, the patient was seen acutely for pain in the same eye while wearing a scleral contact lens. His medications included oral cyclosporine, acyclovir, topical cyclosporine 2% three times a day, and prednisolone acetate 1% daily. Examination revealed a geo-

graphic epithelial defect associated with a loose suture at the graft host junction superotemporally without infiltrate. Corneal smears and cultures were obtained. Treatment with moxifloxacin 0.5% eye drop every 2 hours was started and oral acyclovir was increased to 400 mg five times a day. The next day he was noted to have a 2 x 3 mm gray-white corneal infiltrate. His regimen was modified to include hourly fortified vancomycin (25 mg/ml), fortified tobramycin (14 mg/ml), and moxifloxacin 0.5% eye drops. The corneal cultures subsequently revealed coagulase-negative staphylococcus, *Staphylococcus aureus, Streptococcus viridians* and *Sphingobacterium spiritivorum*. The *Sphingobacterium spiritivorum* culture had intermediate sensitivity to ciprofloxacin, and it was resistant to amikacin and tobramycin. Resolution of the corneal ulcer after approximately ten days was achieved with treatment with topical moxifloxacin and fortified vancomycin and tobramycin, despite apparent limited coverage as shown by culture sensitivities. Topical prednisolone acetate 1% and cyclosporine 2% were continued during this time period. The epithelial defect completely healed in 7 days and the topical antibiotics were tapered to off over a period of three weeks.

Five weeks following presentation, the patient's best corrected visual acuity returned to 20/20 with a scleral contact lens.

Discussion

Sphingobacterium spiritivorum is an aerobic, oxidase-positive, indole-negative, urease-positive, non-motile gram-negative rod [2,3]. It is abundant in nature, particularly in water, plants, and soil [2]. Its name is derived from characteristic sphingophospholipids located in the bacterial cell wall [2]. Most commonly, *Sphingobacterium spiritivorum* is identified in the blood or urine of patients with opportunistic infections [3]. Non-ophthalmic case reports of *Sphingobacterium spiritivorum* have been reported in elderly, immunocompromised patients and can result in bacteremia and death [2]. Previous case reports in general medicine have also reported *Sphingobacterium spiritivorum* in the sputum of a patient with cystic fibrosis and in patients with cellulitis or catheter-related bacteremia [2]. Affected patients often have associated chronic or refractory anemia [2].

In the ophthalmic literature, there have been reports of *Sphingobacterium* species causing or associated with microbial keratitis; however, none have been associated with corneal transplantation. Bouchoucha., *et al*. identified *S. multivorum* as a co-cultured bacterium in a contact lens solution in France [4]. Farias., *et al*. described a *S. multivorum* infection isolated from a corneal ulcer in Brazil [5]. Kilvington., *et al.* isolated *S. spiritivorum* from a contact lens storage case in a patient with a corneal infiltrate in California [6]. Of the above cases, treatment was not reported. Pandita., *et al*. identified a case of *S. spiritivorum* corneal ulcer in Waikato, New Zealand, which responded to treatment with ciprofloxacin [7].

Although our patient responded to empiric therapy with topical moxifloxacin and fortified vancomycin and tobramycin, there are limited clinical reports of successful treatment regimens for this rare bacterium. To our knowledge, there have been only three prior cases of this bacterium associated infectious keratitis and one prior case of a positive contact lens culture in a patient with infectious keratitis. To our knowledge, this is the only reported case of this bacterium co-infecting a corneal transplant. It is important to note that we are reporting an association and not definitively a causal relationship of this organism to the infection.

Conclusion

To our knowledge, there are no previous reports of *Sphingobacterium spiritivorum* keratitis isolated from a corneal ulcer in an eye with a corneal transplantation. Although culture results did not alter therapy in this instance, they are indicated in patients with infectious keratitis, particularly those who are immunosuppressed either locally or systemically. *Sphingobacterium spiritivorum* may be a possible cause of microbial keratitis in immunosuppressed patients. The organism responded to standard topical antimicrobial therapy in our patient.

Patient Consent

Written permission was obtained from the patient.

Funding

No funding or grant support.

Conflicts of Interest

The authors have no financial disclosures.

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Volume 11 Issue 11 November 2020