Epicorneal Aspergilloma After Penetrating Keratoplasty

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Purpose. To report an epicorneal aspergilloma in a postkeratoplasty patient with a bandage soft contact lens. Methods. Case report of a 77-year-old postkeratoplasty patient who presented with an epicorneal mass under a bandage soft contact lens placed for a nonhealing epithelial defect. On removal of the contact lens, a 4 × 4-mm placoid mass fell free from the corneal surface and was sent for culture and histopathology. Results. Gram stain demonstrated septated branching hyphae, and cultures confirmed the diagnosis of Aspergillus versicolor. Histopathologic sections of the epicorneal mass revealed a full-thickness aspergilloma. Conclusion. Contact lens removal and aggressive antifungal treatment resulted in epithelial healing and resolution of the keratomycosis. The patient remained stable for 6 months followed by a successful repeat penetrating keratoplasty.

Key Words: Aspergillus keratitis—Fungal keratitis—Penetrating keratoplasty.

Fungal keratitis after penetrating keratoplasty (PK), although rare, may be difficult to diagnose and treat and may result in devastating consequences. Fungi are unable to penetrate an intact epithelium; however, patients are at increased risk of a keratomycosis once an epithelial defect occurs. Broad-spectrum topical antibiotic use may alter ocular surface flora and allow fungal species to grow in a noncompetitive environment, while topical corticosteroids suppress the normal immune response and enhance fungal replication. Furthermore, there is a documented increased incidence with therapeutic soft contact lens use. The following case report describes a post-PK patient on topical corticosteroid treatment for a nonhealing epithelial defect. On removal of the contact lens, a 4 × 4-mm placoid mass fell free from the corneal surface and was sent for culture and histopathology.

CASE REPORT

A 77-year-old man with a history of pseudophakic bullous keratopathy complicated by Pseudomonas aeruginosa keratitis underwent a PK with anterior vitrectomy, intraocular lens exchange, anterior segment membrane removal, and pupilplasty in the left eye. One month postoperatively, the patient developed a 1.8 × 1.5-mm epithelial defect secondary to two aberrant eyelashes located at the 7:00 o’clock graft-host junction. The primary surgeon removed the two eyelashes and started the patient on ocufloxacin four times daily, erythromycin ointment hourly, and continued prednisolone acetate 1% three times daily. The patient had a stable persistent epithelial defect after 4 weeks, despite a trial of tobramycin (three times daily), Neosporin (polymyxin B), and bacitracin ointment. The primary surgeon placed a bandage soft contact lens (BSCL) on the patient for 2 weeks. The primary surgeon then replaced the BSCL and continued the ocufloxacin four times daily, erythromycin ointment hourly, Timoptic (timolol maleate) 0.5% twice daily, and Refresh Plus four times daily while increasing the prednisolone acetate 1% to five times daily. After wearing the second BSCL for 8 weeks without any changes in management, the patient presented with a complaint of acute painless loss of vision (from a baseline corrected visual acuity of 20/80 to hand motions at 2 ft) and a “white spot” on his cornea. Initial slit-lamp examination with the BSCL in place revealed a round, white central opacity (Fig. 1). During removal of the BSCL, a 4 × 4-mm dome-shaped white crystalline mass dislodged en bloc from the interface of the posterior BSCL and the corneal epithelium (Fig. 2). Visual acuity in the left eye immediately improved to 20/200. Inspection of the cornea revealed an inferonasal 3 × 1-mm epithelial defect underlying the previous central opacity. Within this epithelial defect was an elevated crystalline opacity that appeared “stuck on” the corneal surface with anterior stromal invasion. The initial treatment included corneal scrapings with epithelial debridement and Gram and Giemsa stains plus fungal cultures. The Gram stain revealed numerous septate hyphae. The corneal infiltrate on follow-up examination displayed grayish-white stromal opacity limited to the anterior stroma with feathery borders and two satellite lesions. Corneal scrapings plated on Sabouraud’s dextrose culture medium revealed Aspergillus versicolor. Histopathologic cross-sections of the epicorneal mass revealed a full-thickness aspergilloma (Fig. 3).

Upon diagnosis, the patient was initially treated with topical amphotericin B, 0.15% solution, alternating with topical natamycin, 5% suspension, every 30 minutes around the clock for 72 hours. The patient was also started on oral fluconazole, 400 mg loading dose, on the day of initial diagnosis followed by 200 mg every day until his epithelial defect healed at 2 months. The patient also had a baseline liver function test prior to starting oral fluconazole and liver function testing performed every 2 weeks.
Topical amphotericin 0.15% and natamycin 5% suspension were continued every hour day 4 through 7. The topical antifungals were then slowly tapered until the epithelial defect resolved at 2 months. At this point in our treatment, we discontinued amphotericin 0.15% and oral fluconazole and continued the topical natamycin 5% suspension an additional 30 days. After 3 months of antifungal treatment, the patient’s Aspergillus keratitis had resolved; however, the PK failed secondary to stromal scarring.

The patient remained asymptomatic and infection free over the next 6 months with hand motion visual acuity. He subsequently underwent repeat PK with pathology of the corneal button revealing no organisms or fungal elements. Two years postoperatively, the patient had a clear graft with a best corrected visual acuity of 20/30.

COMMENT

Fungal keratitis after PK, although rare, may be difficult to diagnose and treat resulting in graft failure and poor visual prognosis. The incidence of a fungal endophthalmitis after a PKP has been shown to be about 0.001%. In cases of keratitis after PKP, the incidence of fungal keratitis has been reported to be between 6% and 36%. Aspergillus infections have presented in numerous clinical scenarios. The epidemiology of keratomycosis varies with Aspergillus common in northern climates where it is found in soil and moist organic matter. Risks factors for orbital aspergillosis include total neutrophils less than 1,000/mm³, T-cell defects, defects of phagocytosis, hematologic malignancy, the use of steroids or other immunosuppressive agents, diabetes mellitus, prosthetic devices, trauma, or excessive environmental exposure. The Aspergillus species prefers warm moist environments, which serve as incubators for fungal replication such as the sinus cavities, lungs, lacrimal gland, or cavernous sinus.

Atypical presentations of Aspergillus keratitis are documented throughout the literature. Case reports describe Aspergillus keratitis after radial keratotomy, photorefractive keratectomy, and laser in situ keratomileusis. A review of the literature has found presentations of fungal lens infiltration simulating crystalline deposits, a case series of fungal growth with extended-wear soft contact lenses, and a retained Aspergillus-contaminated contact lens inducing a conjunctival mass.

The A. versicolor species is rarely implicated in fungal keratitis. Anderson et al. reported the first case report of A. versicolor keratitis. They successfully treated one patient with A. versicolor superficial keratitis with topical amphotericin. However, the majority of fungal keratitis cases are caused by Aspergillus fumigatus and on rare occasions by other Aspergillus species.

The literature does not describe an aspergilloma between a BSCL and the corneal epithelium interface. We hypothesize that the combination of topical corticosteroid immunosuppression combined with a persistent epithelial defect and BSCL provided favorable conditions for development of an epicorneal aspergilloma. An epicorneal aspergilloma is an unusual complication of a therapeutic soft contact lens in a postkeratoplasty eye. Such masses should be evaluated with culture and histopathologic analysis to ensure adequate antimicrobial therapy. Any PK patient with a persistent epithelial defect and a BSCL on topical corticosteroids is at high risk of developing a fungal keratitis. The clinician must follow these patients closely and consider early Gram and Giemsa stains with fungal cultures for any nonhealing epithelial defects.
REFERENCES