Infectious *Pseudomonas* and *Bipolaris* scleritis following history of pterygium surgery

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We report an interesting case of infectious scleritis from coinfection of *Pseudomonas aeruginosa* and *Bipolaris* with no corneal infiltrate. A healthy 60-year-old man with a history of infectious scleritis following pterygium excision presented with purulent material growing *P. aeruginosa* and 1+ colonies of *Bipolaris* species of fungus. Broad spectrum treatment was initiated with hourly topical moxifloxacin, fortified tobramycin, and natamycin along with a subconjunctival injection of voriconazole and topical cyclosporine, with PO ketoconazole. After 10 weeks of aggressive empiric treatment, the patient's symptoms had resolved, and his vision returned to baseline although a scleral patch graft was utilized to stabilize scleral thinning.

Key words: *Bipolaris*, infectious scleritis, *Pseudomonas*, treatment

We present the first report of infectious scleritis secondary to *Bipolaris* fungus species without corneal infiltration. *Bipolaris* is a pigmented dematiaceous fungus which has been reported to induce mycotic keratitis, subcutaneous phaeohyphomycosis, sinusitis, peritonitis, and cerebral and disseminated infections.[1] The three most commonly reported species of *Bipolaris* are *Bipolaris spicifera*, *Bipolaris hawaiiensis*, and *Bipolaris australiensis*.[2] Structurally, these filamentous, septated hyphae molds contain asci that can have up to eight flagelliform or filiform ascospores. The multicellular, possibly pigmented fusoid to cylindrical-shaped conidia are produced through conidiophore wall pores and display a sympodial geniculate or zig-zag growth pattern.[3] The pigmented (dematiaceous) form displays a dark brown melanin-like color within the cell wall, which can be a useful clinical marker for infection.[4] However, as in our case, only 27.2% of eyes with dematiaceous fungal infection actually demonstrate such macroscopic pigmentation, which can provide a challenge for diagnosis. This is likely due to higher levels of inflammation that mask the pigmentation.[4]

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Case Report

A healthy 60-year-old male with a 30 pack-year smoking history presented with 2 months of right eye pain, headache, and photophobia. His ocular history was significant for pterygium excision with mitomycin C in the right eye 5 years ago as well infiltrative infectious scleritis secondary to *Enterobacter cloacae* and *Curvularia* fungus species 2 years ago. His first episode of the right eye infectious scleritis presented as scleral thinning 3 mm nasal to the limbus with overlying infiltrates that resolved with topical moxifloxacin and natamycin over the course of 3 months. However, a residual calcified plaque remained in the area of scleral thinning.

Two years later, the patient returned complaining of several weeks of worsening pain and photophobia in the right eye. On examination, his best-corrected visual acuity was 20/25 in the right eye and 20/20 in the left eye. Slit-lamp examination of the left eye was normal. Examination of the right eye revealed soft, yellow-white purulent material overlying the existing calcified plaque with extensive scleral thinning [Fig. 1]. Fundus examination in both eyes was normal.

A limbal peritomy was performed to define the areas of scleral thinning, and the purulent material was noted to be adherent to the calcific plaque. The plaque and purulent material were excised with gentle traction and sent for culture and histopathology. To avoid scleral perforation, a Tutoplast® scleral graft was sutured over the scleral defect and covered with a conjunctival flap. Initially, the patient was treated with hourly topical moxifloxacin, fortified tobramycin, and natamycin.

Von Kossa stains of the scleral plaque showed marked calcification. Gomori methenamine silver stains of the purulent material eventually revealed a marked amount of branching fungal hyphae (indicated by arrow) with cultures growing *Pseudomonas aeruginosa* and *Bipolaris* fungus [Fig. 2].

Nine days after the procedure, the patient developed moderate discomfort in the right eye. Examination revealed a retracted conjunctival flap with further purulent material overlying the scleral patch graft. A decision was made to inject subconjunctival voriconazole, and QID dosing of topical cyclosporine 2% plus oral ketoconazole (400 mg daily) was added to his treatment plan. After 10 weeks of treatment, the patient's symptoms had resolved, and vision returned to baseline [Fig. 3]. Once clinical evidence of infection resolved, medications were discontinued.

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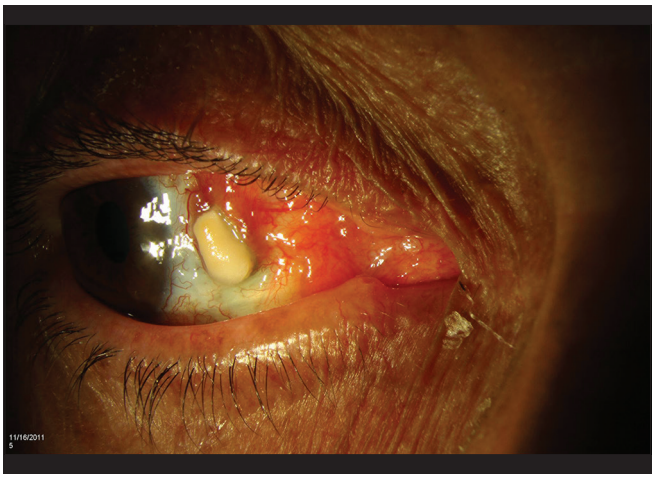

Figure 1: Slit-lamp photograph of the right eye 2 years after initial presentation, now with an elevated mass of purulence overlying the area of scleral thinning

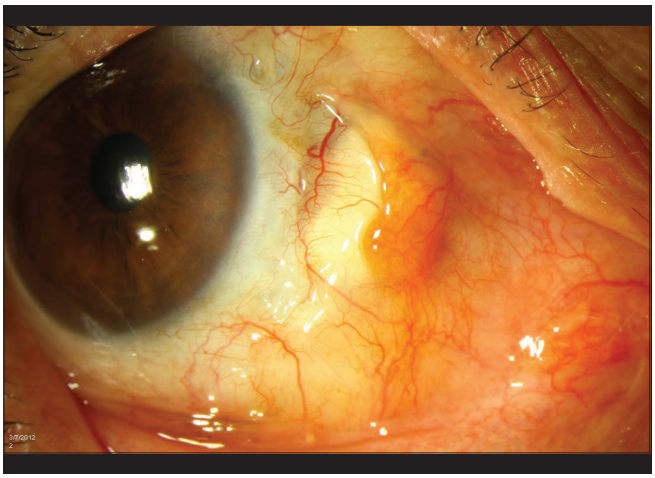

Figure 3: Slit-lamp photograph of the right eye on postoperative month 3 with epithelialization of the scleral patch graft and resolved infection

Discussion

In our patient, aggressive medical and surgical management resulted in complete resolution of the infection. Surgical intervention was elected due to the chronicity and recurrence of the lesion and due to the possibility of impending scleral perforation. *In vitro* strains of dematiaceous fungi have shown to respond well to antifungals such as natamycin and amphotericin B.[1] Hence, a suggested clinical treatment modality includes topical natamycin 5% supplemented with oral ketoconazole.[4] Keratitis has been shown to resolve in over 70% of cases with dematiaceous fungi,[5] but poor penetration of antimicrobials through the collagen-bound scleral layer poses a challenge for effective treatment.[6] The persistence of organisms such as *Pseudomonas* species and fungal hyphae in the sclera despite aggressive medical therapy has been reported.[7] For such cases of infectious keratoscleritis that are refractive to medical treatment such as this particular case, the best option may be to include surgical management such as excision with lamellar corneoscleral graft and/or cryotherapy.[8]

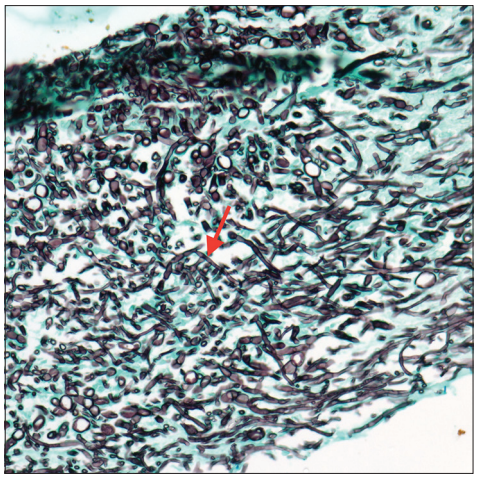

Figure 2: Gomori methenamine silver stain of purulent area demonstrating numerous branching hyphae (arrow) indicative of fungus

Infectious scleritis typically presents as an extension of keratitis resulting from ocular injury, often from trauma, foreign bodies, radiation, or surgery. Infectious scleritis following pterygium removal has been reported, and is often related to postoperative beta irradiation and/or intraoperative antimetabolites.[9] The events resulting in an infection involve direct pathogenic invasion of the sclera that triggers an immune-mediated vasculitis and necrosis at the site of pterygium excision.[9] Hence, if a presumed inflammatory scleritis does not respond as expected to steroid treatment, an infectious etiology should be considered. Such cases can be diagnosed through smear/culture, scleral biopsy, or anterior chamber aspirate. Poor prognosis in infectious scleritis includes corneal involvement, inappropriate antimicrobial therapy, and presence of fungal infection. Thus, mycotic scleritis should always be considered in the differential diagnosis, and prompt diagnoses and treatment are critical to salvage vision.[1] To our knowledge, this is the first report of infectious scleritis without corneal involvement caused by the *Bipolaris* species.[10]

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Conflicts of interest

There are no conflicts of interest.

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