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Recalcitrant Endogenous Trichosporon Endophthalmitis in 2 Immunocompromised Patients

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Trichosporon is a fungal organism found in soil and freshwater, but it has been associated with infectious complications including endogenous endophthalmitis in immunocompromised patients.^{1,2} We report two cases of endogenous trichosporon endophthalmitis requiring systemic antifungals, repeated intravitreal injections of amphotericin and voriconazole along with pars plana vitrectomy for therapeutic and diagnostic purposes. A waiver of authorization was obtained from the Columbia University Institutional Review Board for this case series.

Case report

Case 1

A 69-year-old man recently underwent a heart transplant for ischemic cardiomyopathy and was admitted with a large pericardial effusion requiring a pericardial window. His immunosuppression regimen included tacrolimus and low dose prednisone. He reported 10 days of worsening vision in the left eye. On ocular examination, his visual acuity was 20/60 OD and 20/200 OS. Slit lamp examination revealed moderate cataracts. On dilated examination the left eye had a low-grade vitritis, a small yellow lesion superior to the fovea with associated macular edema and a two disc-diameter creamy lesion in the periphery with overlying vitritis and three smaller satellite lesions. The patient underwent a vitreous tap and inject with vancomycin and ceftazidime followed by intravitreal voriconazole. A day later, the pericardial fluid revealed *trichosporon inkin*, and he was started on intravenous voriconazole. No organism was isolated from the vitreous tap. Over the next three weeks, the lesion continued to progress in size and eventually broke through the ILM and spread

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into the vitreous despite systemic voriconazole and weekly intravitreal injections of amphotericin and voriconazole. He underwent a pars plana vitrectomy with biopsy of the lesion, which was vascular in the center and fluffy on the edges. Diathermy was applied to the lesion followed by placement of two rows of laser and 20% SF6 gas exchange. His culture showed no growth. Following surgery, he received 3 more weekly injections of voriconazole and amphotericin. His vision slowly improved to 20/50, which was limited by a 3+ nuclear sclerosis cataract. The systemic voriconazole was continued for 8 months in total and he did not develop a recurrence following discontinuation (figure available at <http://www.opthalmology-retina.org>).

Case 2

A 37-year-old man on chemotherapy for acute lymphoblastic leukemia was admitted with *trichosporon asahii* pneumonia and fungemia. Initially managed with intravenous micafungin and voriconazole, he was switched to amphotericin B because of persistent fevers. During initial evaluation, he had bilateral retinal hemorrhages without evidence of fungal endophthalmitis. On re-evaluation two weeks later, the right eye was 20/50 and the left eye was 20/30. The intraocular pressures and anterior segment examination were unremarkable. On dilated examination, the right eye had grade 1 vitreous haze with a 5 mm creamy lesion inferior to the nerve with associated satellite lesions and subretinal fluid. The left eye had an isolated 1 mm creamy round lesion in the nasal periphery without associated vitritis. His examination progressed with new anterior chamber cell and worsening vitritis and subretinal fluid tracking towards the macula in the right eye, so he underwent a vitreous tap and injection of amphotericin and voriconazole with addition of intravenous voriconazole. No organism was isolated from the vitreous tap. The intravitreal amphotericin and voriconazole injections were initially performed bilaterally every five days, but the lesions continued to progress, so the interval was shortened to every two days. This treatment regimen slowed progression in the right eye. The left eye lesion progressed to be a Y shape with movement towards the optic nerve and extension into the vitreous. Given the overall poor response to therapy, a decision was made to perform a diagnostic vitrectomy in the left eye. The lesion was trimmed to the base of its stalk, where it had penetrated through the internal limiting membrane. The specimen had no growth of fungal or bacterial elements. Over the next month, intravitreal injections of amphotericin and voriconazole were administered to both eyes every two days followed by every five days followed by weekly until injections were discontinued. During this time the lesions continued to diminish and scar. In total, he received 67 intravitreal injections. He was monitored closely and maintained on micafungin systemically for 7 months and voriconazole systemically for 10 months. On follow up, he was 20/20 in each eye without recurrence of disease following discontinuation of systemic antifungals (Figure, additional figure available at <http://www.opthalmology-retina.org>).

Discussion

Endogenous endophthalmitis with trichosporon species is very rare. A literature review was performed using PubMed as the information source, restricted to papers published in English. Search terms used were: “Endophthalmitis and trichosporon,” “Endophthalmitis

and fungal,” and “Endophthalmitis and yeast. Three cases published within the literature have used systemic antifungal medications, with diagnostic pars plana vitrectomy performed in one case.^{2,3} One case was treated with systemic micafungin and voriconazole, one was treated with diagnostic vitrectomy followed by intravenous amphotericin, and another was treated with intravenous amphotericin alone.^{2,3} Two of these cases involved *Trichosporon beigelii* and the third case did not mention the species encountered.

The patients in this case series differ from these prior cases in the more recalcitrant nature of their progression and the need for multiple treatment modalities including systemic antifungal medications, numerous intravitreal antifungal medications, and pars plana vitrectomy for diagnostic and therapeutic purposes. The first case is unique in that it is the first reported case of *trichosporon inkin* endogenous endophthalmitis. The second case is unique in that it was a bilateral presentation and required almost 70 intravitreal injections over the course of two and a half months. Both cases maintained systemic antifungal medications for 8-10 months and both cases had good visual acuity outcomes.

In a review of trichosporon infections, *trichosporon asahii* was the main causative species and voriconazole treatment was associated with favorable outcome in those with hematological underlying disease.¹ The use of systemic voriconazole was important in both cases because there is poor intraocular penetration of systemic amphotericin.² We decided to use both amphotericin and voriconazole intravitreally because the half-life of amphotericin is much longer than voriconazole and higher concentrations can be achieved intraocularly compared to systemically.²

In these patients with endogenous trichosporon endophthalmitis, escalating therapy is appropriate and considering a diagnostic and therapeutic vitrectomy in addition to systemic and intravitreal medications is important.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Figure.

Trichosporon asahii endogenous endophthalmitis in the right eye of a 37-year-old man with acute lymphoblastic leukemia. Upper left: At presentation, there is a large subretinal abscess with hemorrhage and associated subretinal fluid inferior to the optic disc. Upper right: 3 weeks later, there is worsening vitreous haze, extension of the subretinal abscess, and extension of subretinal fluid into the macula, prompting more aggressive therapy. Bottom left: 4 weeks later, following every-other-day intravitreal amphotericin and voriconazole injections in addition to intravenous antifungals, there is reduction in vitreous haze, and flattening of subretinal abscesses. Bottom right: 6 months after initial presentation, the subretinal abscesses have resolved, leaving behind subretinal fibrosis and atrophy.