continued study is indicated to better understand which patients will benefit from infliximab and who will encounter toxic effects.

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Recalcitrant Candida Endophthalmitis Associated With Mannose-Binding Lectin Deficiency

Mannose-binding lectin (MBL) is an acute-phase reactant that binds microbial surface carbohydrates and participates in opsin-mediated phagocytosis. An important component of the innate immune system, MBL also activates the lectin complement pathway.1 Low serum levels of MBL have been associated with serious bacterial, fungal, and viral infections.2 We describe an otherwise healthy man with MBL deficiency and recalcitrant Candida endophthalmitis.

Report of a Case. A 43-year-old man of Middle Eastern descent had a 1-week history of blurred vision in the right eye. His and several siblings had a history of recurrent oral thrush but no diagnosed immunodeficiency. He had no history of intravenous drug abuse or recent symptoms of oral thrush. On examination, vision in the affected eye was 20/200. A 1.5-mm focal white retinal infiltrate was seen along the superior-temporal arcade with neurosensory macular detachment. He was treated with a varying regimen of sulfadiazine, d-arabin, leucovorin calcium, clindamycin hydrochloride, and prednisone in the ensuing 7 weeks (Figure 1A). Results of rapid plasma reagin, fluorescent treponemal antibody absorption, human immunodeficiency virus enzyme-linked immunosorbent assay, blood cultures, and Bartonella and anti-Toxoplasma IgG and IgM titers were either within the normal range or negative. Five weeks after the patient’s initial visit, his visual acuity decreased and the retinal lesion was nearly twice its original size (Figure 1B). The prednisone dosage was increased but the lesion continued to enlarge (Figure 1C). Vitrectomy performed 7 weeks after his initial visit revealed yeasts and pseudohyphae on Gram staining. At the conclusion of surgery, 50 µg of voriconazole in 0.1 mL of sterile water was injected intravitreally. Cultures grew Candida albicans sensitive to voriconazole and fluconazole. Oral voriconazole at a dosage of 200 to 300 mg by mouth twice a day and later oral fluconazole at a dosage of 800 mg by mouth daily failed to slow progression. Treatment with intravitreal voriconazole at a dose of 100 µg in 0.1 mL of sterile water was started, but the lesion progressed despite 6 more injections every 3 to 4 days during the next month. A second pars plana vitrectomy with excision of the entire white lesion in the macula was followed by endolaser and gas-fluid exchange. Cultures yielded Candida albicans still sensitive to voriconazole and fluconazole. Urine and blood culture results were negative. Postoperatively, the patient was treated with intravenous fluconazole and micafungin. Further studies were performed, including transesophageal echocardiography, lymphocyte subset analysis, thyroid stimulation hormone level, serum cortisol level, and lumbar puncture with cerebral spinal fluid analysis and culture, all of which had unremarkable results. The patient was not anergic to a Candida skin test; lymphocyte transformation to Candida antigen and neutrophil function test results were normal. One month later, a new retinal infiltrate was seen in the midperiphery, followed by a second focus the next week. The patient received a series of 16 injections of intravitreal voriconazole every 1 to 3 days with alternating doses of 100 µg and 200 µg in 0.1 mL of sterile water. Four satellite lesions developed, followed by a partial retinal detachment, lens capsular opacification, and anterior chamber inflammation. The left eye remained healthy. The affected eye was enucleated and examined histologically in its entirety. The specimen revealed persistent growth of yeasts in the cortical vitreous (Figure 2). Extensive
testing of immune function disclosed a severe deficiency of MBL (6 ng/mL; reference range, 100 ng/mL).

Comment. Deficiency of MBL has been correlated with a variety of infections in otherwise healthy individuals but may also enhance susceptibility to certain infections in persons receiving chemotherapy or those with secondary immunosuppression for other reasons. Genetic polymorphism explains the range of serum concentrations of MBL found in the general population, with severe serum MBL deficiency associated with homozygosity of yet unnamed minority alleles. We are unaware of any previous reports of recalcitrant Candida endophthalmitis associated with MBL deficiency in an otherwise healthy individual. Mannose-binding lectin replacement therapy using plasma-derived or recombinant lectin is under investigation but is not currently available for clinical use.

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Complex Choristoma Masquerading as a Dacryocystocele

Orbital dermoid cysts are choristomas believed to arise during development from sequestered ectodermal rests. The cysts are lined by keratinized stratified squamous epithelium, contain adnexal structures, and are often attached to superotemporal orbital bone. A variant of the orbital dermoid cyst, the conjunctival dermoid cyst, is lined by nonkeratinized epithelium with goblet cells and tends to be found anteronasally in orbital soft tissue without bony attachment.

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REFERENCES


Correction

Error in Text. In the Letters: Research Letters article titled “Recalcitrant Candida Endophthalmitis Associated With Mannose-Binding Lectin Deficiency” by Oliva et al, published in the June 2009 issue of the Archives (2009;127[6]:822-823), a dosage error occurred in the “Report of a Case” paragraph on page 822. The 22nd sentence should have read, “The patient received a series of 16 injections of intravitreal voriconazole every 1 to 3 days with alternating doses of 100 µg and 200 µg in 0.1 mL of sterile water.”