Ocular Sporotrichosis Mimicking Mucormycosis in a Diabetic
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Primary sporotrichosis of the eye is very rare; most infections are limited to the conjunctiva or adnexa. We report a case of Sporothrix endophthalmitis associated with necrotizing ethmoid sinusitis developing in a young diabetic man with ketoacidosis. The infection clinically resembled rhino-ophthalmic mucormycosis. Cure followed evisceration and an abbreviated course (215 mg) of amphotericin B. Sporothrix must now be regarded as another fungal agent capable of causing primary rhino-ophthalmic infection similar to Mucor.

Sporotrichosis is a well-known but relatively rare fungal infection caused by Sporothrix schenckii. Most clinically recognized infections are limited to the skin and follow traumatic inoculation of the agent which is distributed worldwide, most often in association with vegetative organic matter such as wood, plants, or dried moss. Centripetal lymphocutaneous spread from the primary site produces a characteristic linear pattern of lesions which is usually easily recognized.

In contrast, extracutaneous sporotrichosis has been recognized and reported much less frequently. Most cases have involved the lung or bone and joints. Sporotrichosis involving the eye is relatively rare; in the vast majority of cases it has followed traumatic inoculation of the conjunctiva or periorbital tissues. Such instances are considered pathophysiologically to be mucous membrane variants of cutaneous disease. Eight previous cases of global infection have been reported without adnexal or conjunctival involvement, either as part of multifocal systemic infection or more rarely as isolated (unifocal) endophthalmitis.

This report describes a case of unifocal ocular sporotrichosis in a young diabetic man. The infection, an endophthalmitis associated with necrotizing sinusitis, resembled rhino-ophthalmic mucormycosis clinically. It was successfully controlled by evisceration and a relatively limited course of amphotericin B. This clinical pattern of ophthalmic sporotrichosis has not previously been reported.

Case Report

A 27-year-old diabetic man was admitted to the hospital with severe right-sided ocular pain. A juvenile diabetic for
17 years, he was severely incapacitated by generalized peripheral neuropathy and long-standing proliferative retinopathy (completely blind OD: light perception only, OS). Laser photocoagulation had been used 2 years previously. Up to the time of admission his diabetes had been satisfactorily controlled with 35 units of Lente insulin administered once daily.

Approximately 24 hours prior to admission the patient noted stabs of right-sided ocular pain which rapidly became excruciating and associated with nausea and vomiting. Generalized headache, chills, fever, dental pain, and nasal congestion were denied. He was unable to recall a history of ocular trauma or a foreign body.

On examination the patient was a well-nourished white man in severe pain. The blood pressure was 130/80 mm Hg, pulse rate 88 beats/min, respirations 16/ min, and the temperature 98.6°F (37°C) orally. The right eye was proptotic and surrounded by periorbital edema and overlying ecchymosis (Fig 1). No conjunctival purulence or abrasions were noted. A dense cataract, OD, was unchanged from previous examination and precluded internal examination of that eye. Multiple hemorrhages, soft exudates, proliferative changes, and extensive scarring from photocoagulation therapy were seen in the left retina. Tonometry OD disclosed a pressure of 70 mm Hg; the left eye was not tested but was normotensive by digital examination. Pretibial necrobiosis diabeticorum and motor and sensory signs of severe peripheral neuropathy were present. Examination of the heart, lungs, and abdomen revealed no abnormalities.

The hematocrit value was 36%, hemoglobin 11 gm/100 ml, and the white blood cell count, 11,000/cu mm (55% adult polymorphonuclear cells, 31% band forms, and 7% lymphocytes). The BUN was 30 mg/100 ml, serum uric acid 10 mg/100 ml, and blood glucose 464 mg/100 ml. Large amounts of acetone were detected in undiluted serum and the arterial blood pH was 7.34. Chest roentgenogram showed no abnormalities. X-rays of the paranasal sinuses revealed faint clouding of the right anterior ethmoid sinus.

Mild ketoacidosis was easily reversed with supplemental crystalline zinc insulin and fluid repletion. Initial ophthalmologic diagnosis was acute secondary glaucoma and treatment was initiated with retrobulbar injection of 2% lidocaine (Xylocaine) followed by 1.5 ml of absolute alcohol.

Severe throbbing orbital pain persisted unabated and the patient began to experience nasal congestion over the next 14 days; proptosis and pronounced periorbital inflammation were also unchanged. Yet he remained afebrile. A repeat roentgenographic study of the paranasal sinuses on the 14th hospital day showed increased opacification of the right anterior ethmoid sinus and suggestion of erosion of the lateral wall (Fig 2).

The following day, the medial right orbit was explored surgically. Because no communication between the orbit and

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**Fig 1.** Proptosis, periorbital edema, and inflammation, OD.

**Fig 2.** Opacification of the right anterior ethmoid sinus with erosion of the lateral wall (arrow).
ethmoid sinus could be identified, the sinus was not entered surgically. The markedly inflamed eye was exsiccated by section and curettage. After exsiccation the orbit was irrigated with a 2% solution of chloramphenicol and packed open.

Ocular fragments histopathologically showed marked necrosis and polymorphonuclear infiltration of the choroid, retina, and vitreous humor. Numerous 1 to 2 micron ellipsoidal cigar-shaped bodies typical of *Sporothrix schenckii* were seen in Gram’s and Gomori silver-methenamine-stained smears of the vitreous humor (Fig 3). However, bacterial and fungal cultures of this material and other global fragments were negative, as were multiple cultures of bone marrow, blood, and urine.

Following exsiccation, amphotericin B was begun. Between the 18th and 28th hospital days the patient received a total of 215 mg. Development of progressive azotemia (BUN and creatinine rising from 15 to 53 mg and 0.8 to 2.1 mg/100 ml, respectively) and the appearance of hyperkalemia (6.7 mg/100 ml) necessitated discontinuance of the drug. During therapy the patient’s periorbital pain and swelling and nasal congestion gradually resolved. He was discharged on the 41st hospital day markedly improved with minimal residual orbital swelling and inflammation.

Four days later the patient returned with acute staphylococcal pneumonia. Sinus x-rays were essentially unchanged from those obtained in the first hospitalization. Antral washings were obtained and yielded only scant growth of *Staphylococcus epidermidis* and *Corynebacterium* sp in bacterial culture. However, fungal cultures gave heavy growth of tan, glabrous colonies of fungus microscopically consistent with *S. schenckii*. Slide cultures at 25°C to elicit the mycelial phase revealed delicate branching, septate hyphae bearing aortal to spherical microconidia in bouquet-like arrangement, compatible with *Sporothrix* (Fig 4). This fungus could not be converted back to a yeast phase because of bacterial overgrowth. Animal inoculation was not performed.

The patient’s fever and respiratory symptoms resolved rapidly following institution of parenteral methicillin therapy. Since the original orbital inflammation had not exacerbated and there was clinically no evidence of active fungal infection of the orbit or sinus, it was believed that continued presence of *Sporothrix* represented colonization rather than infection and a second course of

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**Fig 3.** Ellipsoidal cigar-shaped bodies approximately 1 to 2 microns in length, characteristic of the yeast phase of *Sporothrix schenckii* (Gomori silver methenamine-stained smears of vitreous humor, original magnification X1,000).

**Fig 4.** Slide culture of sinus washings showing septate hyphae with microconidia in bouquet-like clusters, compatible with the mycelial phase of *Sporothrix schenckii*. 
antifungal chemotherapy was withheld. The patient has had no signs of Sporothrix infection in the intervening 14 months.

A serum specimen obtained in the 15th hospital day of the initial hospitalization for endophthalmitis was tested by the Mycology Serology Laboratory at the Center for Disease Control in Atlanta, Georgia. A 1:8 titer against Sporothrix schenckii was obtained, both by tube agglutination and slide latex agglutination. The presence of agglutinating antibody to S. schenckii antigen in this titer or higher is very suggestive of and specific for active deep infection.5

Discussion

Most Sporothrix infections of the eye are superficial and follow conjunctival or periorbital inoculation.1,2 Of only 8 previously reported cases of endophthalmitis,1,2,5 5 were associated with multifocal systemic disease. Our patient's uniocular ocular infection is particularly unique in its clinical resemblance to mucormycosis.

Mucormycosis caused by Rhizopus sp—which are unrelated to Sporothrix—is a very rare, but well-known infection of the paranasal sinuses and orbit which often culminates in lethal cerebral spread by direct extension.3 Most cases have occurred in diabetics with severe ketoadosis or in patients with acute leukemia. Inhalation of environmental spores followed by mucosal invasion in the paranasal sinus is thought to be the probable route by which rhino-ophthalmic mucormycosis develops in diabetics. Why diabetics, particularly with ketoacidosis, appear to be most susceptible is unclear but may be related to associated hyperglycemia and acidosis, impaired mobility, and phagocytic function of polymorphonuclear leukocytes and generally compromised inflammation.6 Similar pathophysiologic derangements may predispose to deep infection with Sporothrix schenckii and account for the mucormycosis-like features which characterized our patient's infection.

Our patient probably acquired Sporothrix initially by inhalation: infection began as acute ethmoiditis and spread secondarily to the eye. Although cultures of intraocular contents at the time of florid endophthalmitis did not yield the microorganism despite clear-cut microscopic evidence of infection, antral sinus washings obtained after the orbital infection had resolved yielded a fungus morphologically consistent with S. schenckii. Moreover, the diagnosis of Sporothrix infection was supported by serologic data. The fact that the patient's diabetes remained under control probably has prevented residual colonization from progressing to recurrent active infection. Noteworthily, fungal cultures of the enucleated eye were also negative in the case of primary endophthalmitis reported by Cassady4 and diagnosis was based on the histopathologic features.

Amphotericin B had to be prematurely discontinued in our case because of the appearance of renal insufficiency and hyperkalemia. A total dose of only 215 mg is unusually small to cure deep fungal infection. However, this abbreviated regimen was undoubtedly augmented by evisceration and orbital debridement. The patient has been without evidence of recurrent disease for 14 months but is being closely observed for signs of recurrence.

Recent reports suggest that certain systemic fungal infections including endophthalmitis caused by organisms highly sensitive to amphotericin B such as Candida albicans can be successfully eradicated with limited courses of the drug ranging from 10 to 355 mg.5 These total dosages are far less than the 1 to 2 gm or more usually required to reliably cure most systemic mycoses such
as histoplasmosis, coccidiomycosis, blastomycosis, or cryptococcal meningitis. Further studies are needed, however, before "low dose" amphotericin can be advocated for systemic Sporothrix infections. Documented systemic sporotrichosis, including of the eye or sinuses, should be treated with conventional regimens, generally aiming for a total dose of 1 to 2 gm. Only if extenuating circumstances intervene, such as renal insufficiency, and cure seems probable, should an abbreviated course be accepted. If Sporothrix infection recurs in our patient, amphotericin B will probably be reinstalled; if again poorly tolerated, iodides might be considered although attempts at treating systemic Sporothrix infection with potassium iodide alone have generally not been successful. Other antifungal agents such as 5-fluorocytosine are also ineffective against Sporothrix.11

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