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Serous Retinal Detachment in a Patient with Rhino-Orbital Mucormycosis

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Background: Rhino-orbital mucormycosis is a difficult disease to treat. We report one case of rhino-orbital mucormycosis, complicated by serous retinal detachment, that responded to aggressive treatment.

Case: A 38-year-old man with diabetic ketoacidosis was referred to the emergency department of our hospital with fever, proptotic right eye, and complaint of lethargy.

Observations: Fundus examination showed serous retinal detachment and focal lesions of retinitis with exudate at the inferior portion coincident with the position of opacification in the orbit on MRI. Fluorescein angiography showed pooling of dye in the detached retina and leakage from focal lesions of retinitis. We thoroughly removed the large necrotic materials in the orbit and sinus through the lower conjunctiva without enucleation or exenteration. Microscopic examination and culture of the necrotic materials that were removed from the orbit proved that the patient had mucormycosis. The serous retinal detachment improved 10 days after orbital debridement combined with intravenous and local (intraorbital) amphotericin B treatment. Visual acuity recovered to 20/50.

Conclusion: We propose that inflammation of the sclera in close contact with necrotic fungi materials may cause serous retinal detachment. **Jpn J Ophthalmol 2001;45:301–304** © 2001 Japanese Ophthalmological Society

Key Words: Rhino-orbital mucormycosis, serous retinal detachment.

Introduction

Mucormycosis is an often fatal fungal infection in diabetic and immunocompromised hosts. Inhalation of fungi of the family *Mucoraceae* leads to infection of the palate, nose, or paranasal sinuses. This process often progresses from the sinonasal cavity to the orbit, and later to the cavernous sinus. Multiple clinical symptoms, including rhinocerebral, pulmonary, gastrointestinal, cutaneous, and disseminated lesions, have been described.[1] Fungal hyphae invade vessel walls, particularly arteries, resulting in thrombosis and tissue infarction.[2] Clinically, rhino-orbital-cerebral mucormycosis might present with blurred vision, an orbital apex syndrome consisting of complete ophthalmoplegia, ptosis, chemosis, and pain in the periorbital region.[3,4]

In this article, one case of rhino-orbital mucormycosis complicated by serous retinal detachment and retinal necrotic lesions is reported.

Case Report

A 38-year-old man with diabetes, complaining of lethargy, was treated at another hospital. There was proptosis in his right eye with limitation of ocular motility, conjunctival chemosis, right periorbital edema, and facial swelling (Figure 1). The serum glucose concentration was 340 mg/dL and diabetic ketoacidosis was diagnosed. The patient was transferred to our institution 7 days after onset.

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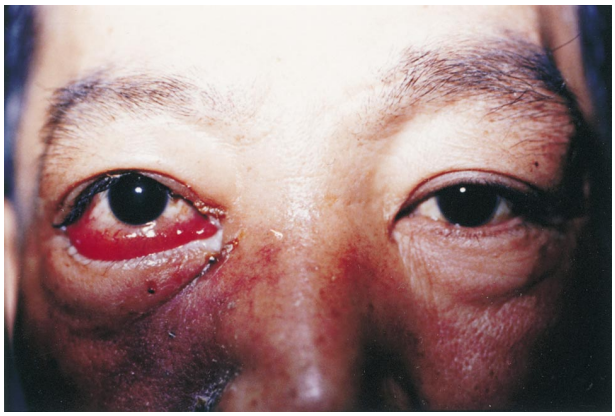

Figure 1. Preoperative photograph: Marked proptosis, conjunctival chemosis, and facial swelling can be seen in patient's right eye and face.

Magnetic resonance imaging (MRI) (Figure 2) confirmed soft-tissue opacification of the ethmoid and sphenoid sinuses on the right side. Orbital involvement extended along the retrobulbar fat plane and inferior rectus muscle to the orbital apex. Optic nerve involvement was strongly suggested. There was no significant dural enhancement and no involvement of the cavernous sinus or the cavernous carotid artery. Endoscopic biopsy with ethmoidectomy led to a diagnosis of rhino-orbital mucormycosis.

Systemic intravenous amphotericin B (1 mg/kg per day) was started on day 1, in collaboration with the infectious disease department. Best-corrected visual acuity was 20/100 in the right eye and 20/20 without correction in the left eye. The pupil responded sluggishly to light in the right eye. Fundus examination demonstrated several horizontal choroidal folds across the macula. Although systemic intravenous amphotericin B was administered for 2 weeks, the patient developed sudden visual loss. Proptosis in the right eye progressed to 25 mm in contrast to 14 mm for the left eye, as measured with Hertel's exophthalmometer. Fundus examination of the right eye (Figure 3) showed serous retinal detachment and focal lesions of retinitis with exudate in the inferior portion coinciding with the opacification in the orbit on MRI. Fluorescein angiography (Figure 4) of the affected eye showed pooling of dye in the detached retina, leakage from focal lesions of retinitis, and filling of retinal arteries 22 seconds after injection, indicating a delay in retinal arterial filling.

Fourteen days after admission, a large necrotic mass (8.5 × 5 cm) and many necrotic fragments were surgically removed through a wide incision in the lower conjunctiva of the right eye without enucleation or exenteration. The large mass had several branches that extended to the orbital apex, into the ethmoid sinus, and between the maxillary bone and facial tissue (Figure 5, left). A modified spinal needle (18-gauge, Quincke; Hakko Medical, Nagano) modified was inserted in the orbit for postoperative periodic irrigation according to the method of Luna et al.[5] A few specimens were selected for microscopic examination and culture. Extensive broad nonseptate and branching hyphae with mixed inflammatory response in the microscopic examination were compatible with mucormycosis (Figure 5, right). Every 12 hours for 9 days, the operated area in the orbit was irrigated with 10 mL of amphotericin B solution (1 mg/mL) using the spinal needle. The patient has survived and no other symptom has developed during the postoperative period. The serous

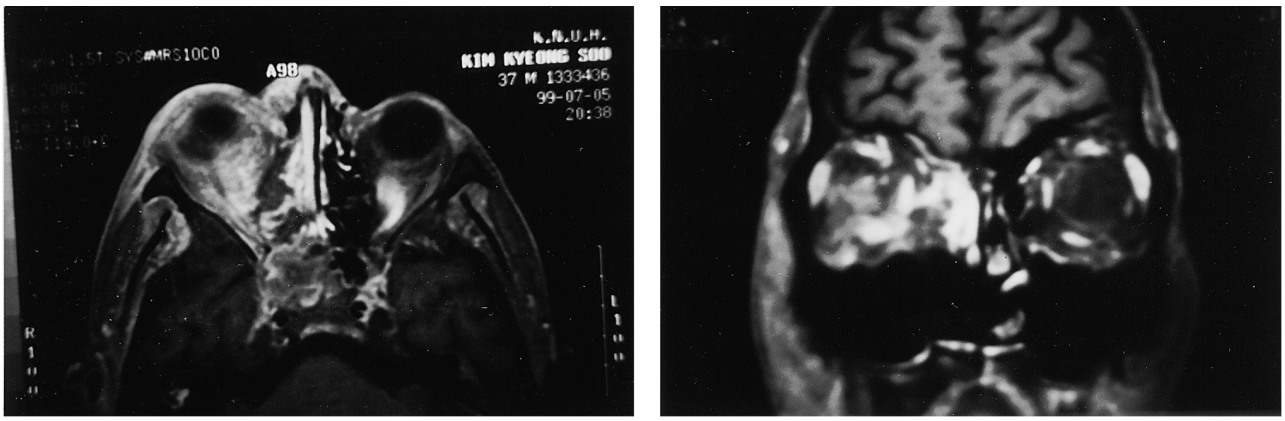

Figure 2. Axial and coronal T_1-weighted magnetic resonance imaging confirm soft-tissue opacification of ethmoid and sphenoid sinuses on right side. Orbital involvement extends along retrobulbar fat plane and inferior rectus muscle to orbital apex.

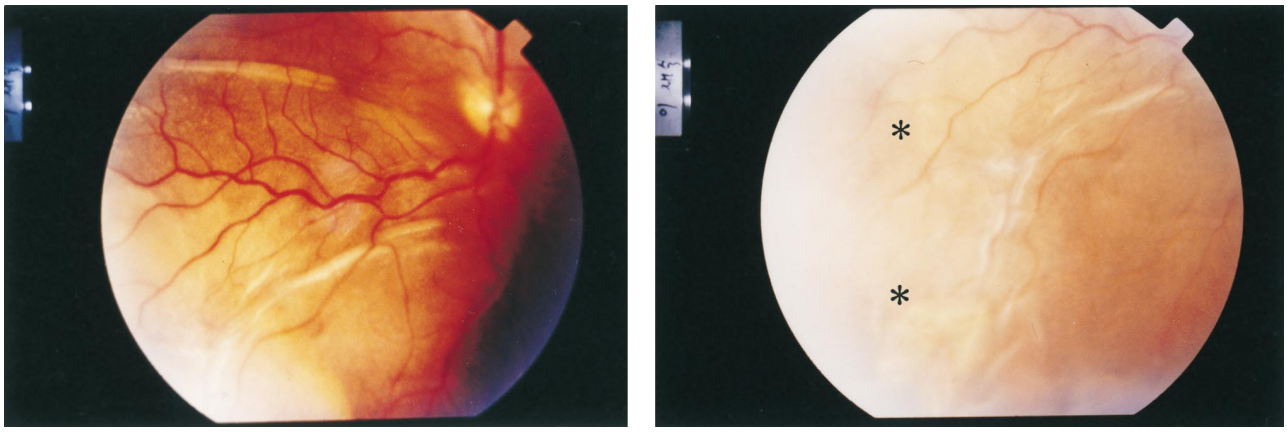

Figure 3. Fundus photograph shows serous retinal detachment and focal lesions of retinitis with exudate (*) at inferior portion of fundus.

retinal detachment improved 10 days after orbital debridement combined with intravenous and local amphotericin B treatment (Figure 6). Visual acuity improved to 20/50 and no delay in retinal arterial filling was observed by fluorescein angiography.

Discussion

Rhino-orbital-cerebral mucormycosis has a distinct predilection for patients with diabetes.[1] Early diagnosis, systemic antifungal therapy, control of the underlying disease, and aggressive surgical debridement are important in the treatment of mucormycosis.[3,4,6,7] Amphotericin B is the drug of choice for this disease. The survival rate for individuals who have this infection ranges from 44% to 85%.[3,6–8] This case was successfully treated with orbital debridement combined with intravenous and local amphotericin B treatment, without enucleation or exenteration.

From the paranasal sinuses, *Mucor* fungi often invade the orbit and progress posteriorly into the cavernous sinus. *Mucor* fungi have a propensity for growing along the walls of blood vessels, so these fungi have been called angiotropic.[1] Thrombosis of the cavernous sinus and cavernous carotid artery may occur, and intracranial extension may be in the form of distal mycotic emboli or direct meningeal inflammation.[9]

The pathogenetic mechanisms of serous retinal detachment and retinal necrotic lesions are not yet established. We do not exclude possibilities of the following in the development of the serous retinal detachment: relative impairment of blood supply to the choroid and retina because of the compression of blood vessels due to necrotic fungi materials in the orbit; or choroidal and retinal inflammation caused by mycotic emboli; or direct invasion of blood vessels resulting in thrombosis and tissue infarction. We sug-

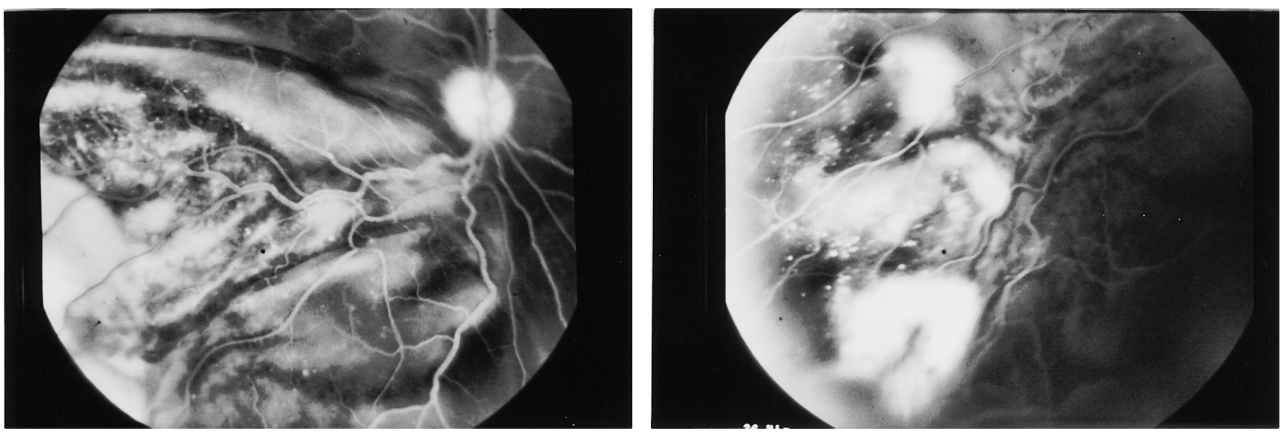

Figure 4. Fluorescein angiograph shows pooling of dye in detached retina and leakage from focal lesions of retinitis.

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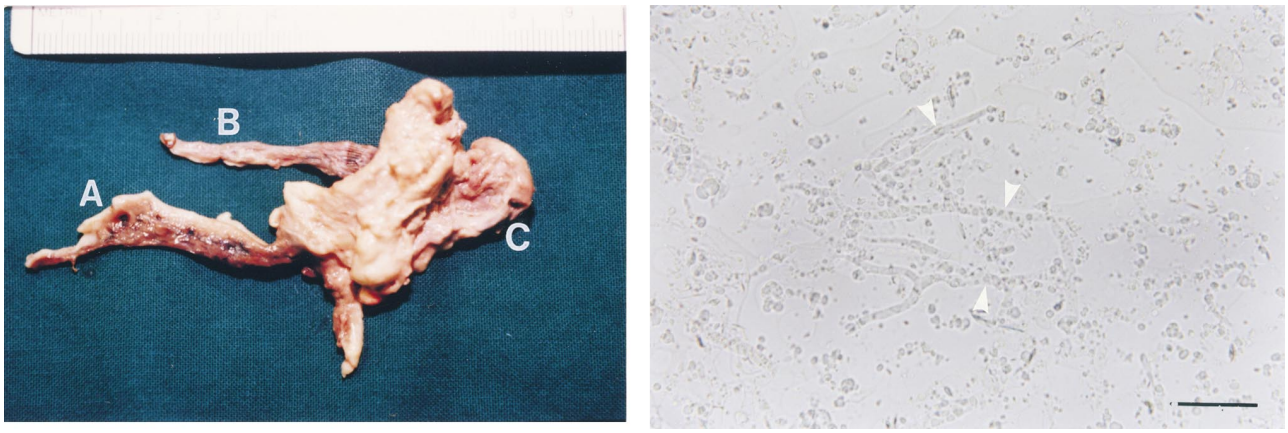

Figure 5. Mass of 8.5 × 5 cm in size removed from orbit, Left: Several branches of necrotic fungi mass extended to orbital apex (A), into ethmoid sinus (B), and between maxillary bone and facial tissue (C). Wet smear. Right: Orbital specimen demonstrates multiple broad, nonseptate hyphae (arrowheads) within necrotic orbital contents. Bar = 100 μm, KOH method.

gest that the inflammation of the sclera in close contact with necrotic fungi materials is a probable cause that may develop into serous retinal detachment.

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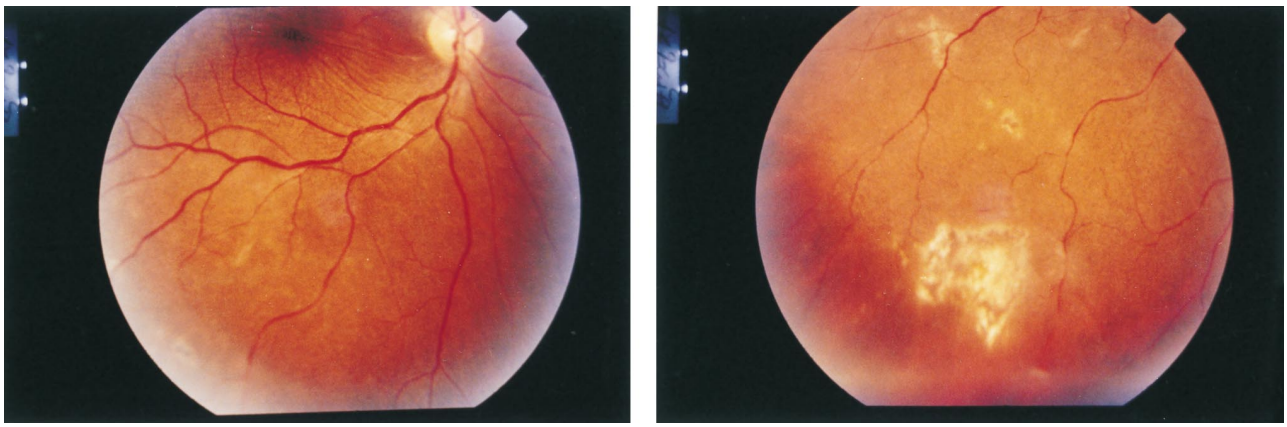

Figure 6. Serous retinal detachment improved 10 days after orbital debridement combined with intravenous and local amphotericin B treatment.