A Case of Rhinocerebral Mucormycosis in a Diabetic Woman

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We report a case of rhinocerebral mucormycosis in a 63-year-old female with a 1-year history of poorly controlled diabetes mellitus. She had black necrotic ulcers on the hard palate, bloody nasal discharge, swelling of the left side of her face, left blepharoptosis, proptosis, and conjunctival injection.

Histopathological examination of the palatal lesion showed large, nonseptate, right-angled branching fungal hyphae in the dermis, and Rhizopus species was isolated on Sabouraud’s agar media.

The patient was treated with a combination of amphotericin B and surgical debridements but died of asphyxia one month after admission. This is a relatively uncommon, opportunistic infection occurring in a diabetic woman, and only several cases are reported in the Korean literature up to date. (Ann Dermatol 3:2 145–152, 1991)

Key Words: Diabetes mellitus, Rhinocerebral mucormycosis

Mucormycosis is an acute, rapidly developing, often fatal, opportunistic infection caused by the Order Mucorales of the Class Zygomycetes, occurring in patients with a variety of debilitating diseases including diabetes, leukemia, lymphoma, cancer, AIDS, hepatitis, cirrhosis of the liver, anemia, congenital heart disease, burns, severe malnutrition, and iatrogenic immunosuppression. Mucormycosis most commonly involves the rhinocerebral region, pulmonary system, or gastrointestinal tract. The rhinocerebral form of mucormycosis found most commonly in uncontrolled diabetics is caused commonly by the genus Rhizopus, and begins in the nose and progresses through the paranasal sinuses and invades the orbit and CNS secondarily. In the Korean literature, several cases of mucormycosis have been reported but fungus cultures were neither positive nor tried in those cases.

We report, herein, a case of rhinocerebral mucormycosis in a poorly controlled diabetic woman caused by Rhizopus species confirmed by culture, and summarize the reported cases in Korea.

REPORT OF A CASE

A 63-year-old female with diabetes mellitus was referred to our department in October.
1989 because of black necrotic ulcers on the hard palate and swelling of the left side of her face. Two weeks earlier, she noted blood-tinged nasal discharge on the left side and two days later, she also noticed left periorbital swelling, blepharoptosis, conjunctival injection, proptosis on the left, hyperesthesia over the left cheek, and left periorbital pain. The patient had a history of diabetes mellitus for 1 year and received oral hypoglycemic agents intermittently. Her family history was unremarkable.

On physical examination, the patient was afebrile but dehydrated and lethargic. The left side of her face was swollen, and bloody discharge from her left nostril was noted, with macerated necrotic tissue within the left nasal cavity. Ophthalmologic findings included left blepharoptosis, proptosis, conjunctival injection, ophthalmoplegia, and loss of light reflex of the left pupil (Fig. 1, 2). There were also black necrotic ulcers on the hard palate and multiple whitish patches within the oral cavity (Fig. 3).

Laboratory findings were as follows: hemoglobin level, 11.0 g/dl; hematocrit, 35.9%; white blood cell count, 14,400/mm³; and ESR, 110 mm/hr. The serum glucose level was 213 mg/dl; urinary glucose level, 1.0 g/dl; urinary protein level, 30 mg/dl; plasma albumin level, 2.3 g/dl (normal, 3.5 to 5.0 g/dl); blood iron level, 31 µg/dl (normal, 60 to 130 µg/dl); total iron binding capacity, 152 µg/dl (normal, 250 to 350 µg/dl); blood transferrin level, 98.6 mg/dl (normal, 200 to 300 mg/dl). Arterial blood gas analysis revealed pH 7.32 (normal, 7.35 to 7.45), PaCO₂ 32.9 mmHg (normal, 35 to 45 mmHg), HCO₃⁻ 17.3 mmol/L (normal, 23 to 29 mmol/L), and BE -7.0 mmol/L (normal, 0 ± 2 mmol/L). Other laboratory values, including liver function test, renal function test and EKG were within normal limits.

An X-ray film of the chest was normal, but sinus films showed clouding of the left maxillary and ethmoid sinuses (Fig. 4). Sinus and orbit CT scan showed a protruding lesion within the left orbit, mucosal thickening of the left maxillary and ethmoid sinuses, nasal cavity, and proptosis on the left (Fig. 5).

Swabs of black necrotic ulcer on the hard palate yielded irregularly branching, nonseptate, large fungal hyphae on KOH preparation (Fig. 6) and inoculation of necrotic tissue on two Sabouraud’s agar plates revealed a rapid growth of fungus (Fig. 7).

### Legends For Figures

**Fig. 1.** The patient with left blepharoptosis, dark hemorrhagic crust on left nostril, and swelling of the left side of face.

**Fig. 2.** Left blepharoptosis, proptosis, conjunctival injection, erythema and swelling of periorbital region.

**Fig. 3.** Black necrotic ulcers on the hard palate and whitish patches within the oral cavity.

**Fig. 4.** Sinus roentgenogram shows clouding of the left maxillary and ethmoid sinuses (arrows).

**Fig. 5.** CT scan shows protruding lesions (arrows) within the left orbit, mucosal thickening of the left maxillary and ethmoid sinuses, nasal cavity, and proptosis (arrowheads) on the left.

**Fig. 6.** Swab of necrotic ulcer on the hard palate shows irregularly branching, nonseptate, large fungal hyphae (KOH/Parker ink mount, ×100).

**Fig. 7.** White, cottony colonies that soon turned gray-black at the periphery are shown on the Sabouraud’s agar plate at 1 week after inoculation at room temperature.

**Fig. 8.** Round, brown sporangia, sporangiophores, and rhizoids are shown in slide culture at room temperature (lactophenol cotton blue, ×40).

**Fig. 9.** Higher magnification of the Fig. 8 shows sporangia filled with endospores (arrow) and sporangiophores rising at a node opposite the rhizoids (arrowhead) (lactophenol cotton blue, ×400).

**Fig. 10.** Biopsy specimen from a necrotic ulcer on the hard palate shows necrosis and inflammatory cell infiltration in the dermis. Scattered fungal hyphae are also seen in the dermis (H & E stain, ×100).

**Fig. 11.** Large, long, nonseptate hyphae in the dermis (PAS stain, ×100).