Acute Cerebral Phaeohyphomycosis due to \textit{Wangiella dermatitidis} Accompanied by Cerebrospinal Fluid Eosinophilia

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We report a case of cerebral phaeohyphomycosis due to \textit{Wangiella dermatitidis} in an immunocompetent adult man. His cerebrospinal fluid (CSF) showed pleocytosis with a high eosinophil count but without peripheral blood eosinophilia. The present case suggested that this black yeast-like fungus should be included when the causes of CSF eosinophilia are considered, even though it is an extremely rare pathogen.

Central nervous system phaeohyphomycosis due to \textit{Wangiella dermatitidis} (also known as \textit{Exophiala dermatitidis}) is extremely rare, and documented cases have not occurred outside of Asia. So far, only six documented cases of \textit{W. dermatitidis} as a causative organism of brain infections have been reported in the world. There have also been three more probable cases of brain infections, in which \textit{W. dermatitidis} was isolated from body tissues other than the brain (1, 5, 8, 9). However, cases of cerebral phaeohyphomycosis accompanied by cerebrospinal fluid (CSF) pleocytosis and eosinophilia have not been reported as yet, and black yeasts, including \textit{W. dermatitidis}, are not included in the long list of probable causes of CSF eosinophilia (12). We report a case of cerebral phaeohyphomycosis caused by \textit{W. dermatitidis} with a high CSF eosinophil count.

Case report. A 28-year-old male was admitted to the neurology ward of Pusan National University Hospital because of a headache that worsened over 5 days. Five days before his admission to the hospital, he had first noticed a diffuse, pulsating headache upon awakening in the morning. The following day, his headache worsened and was accompanied by nausea and vomiting. One day before his admission to the hospital, a severe headache woke him up at night. He was otherwise a healthy engineer working for a multinational company and had traveled for 3 years to many countries in Southeast and Middle East Asia, North America, South America, and Europe. He had no history of any other systemic diseases, such as tuberculosis, allergies, diabetes mellitus, or hypertension. He denied any history of smoking, habitual drinking, or drug abuse. He had normal blood pressure (120/60 mm Hg), pulse rate (65/ min), and respiratory rate (19/min), and he was afebrile (36.5°C). The results of his general physical examination were nonspecific. A detailed neurological examination also did not reveal any abnormalities, such as neck stiffness, hemiparesis, pupillary change, or pathologic reflexes. A computerized tomography view of the brain showed multiple ill-defined low-density lesions, and a magnetic resonance view showed multiple small enhancing lesions. A spinal tap revealed high opening pressure (290 mm H₂O), and a CSF analysis showed pleocytosis (290/mm³, 60% lymphocytes), an increased level of protein (128 mg/ml), and a normal sugar level (96 mg/ml). A CSF Gram and acid-fast bacillus staining and India ink preparation yielded negative results. A high proportion of eosinophils was noticed on CSF cytology (40%) (Fig. 1). The results of the enzyme-linked immunosorbent assay for \textit{Paragonimus} and \textit{Cryptococcus} were negative. An intravenous osmotic agent, steroid, and oral praziquantel were started 2 days after the patient’s admission, producing significant symptomatic improvement. On day 6 of hospitalization, the patient had a constant, severe headache with nausea and projectile vomiting, which did not respond to intravenous analgesics or osmotic agents. A control brain magnetic resonance view on day 8 revealed obstructive hydrocephalus. On day 10, surgery was performed to relieve the obstructive hydrocephalus. Upon operation, it was revealed that the lateral ventricle was filled with dirty, cloudy CSF, with severe adhesion around the entry of the third ventricle. A biopsy of the ventricular wall was performed, and an extraventricular drainage catheter was placed in the lateral ventricle. A frozen biopsy specimen showed scattered foci of tangled fungi with mold-like features in addition to an intense inflammatory reaction. A fungal culture was performed, and 50 mg of amphotericin B was administered daily. After the operation, the patient was alert and oriented but his headache persisted, with nausea and vomiting. On day 12, the patient suddenly lost consciousness, and after that, he showed a stuporous to comatose mental status. He expired on day 13.

Mycology. Three days after incubation at room temperature and 37°C on Sabouraud’s dextrose agar, the colonies appeared black, reverse black, wet, and mucoid and could be picked up as a string of material from the plate to an inoculating loop. Sparse, septate, and pale olivaceous hyphae were observed under a microscope. Black yeast synanamorphs were abundant. Conidiogenous cells were cylindrical, with rounded apices producing one-celled conidia. Round to ovoid, pale brown conidia accumulated in balls or slipped down the side of conidiophore. The organism was identified as \textit{W. dermatitidis} because of its ability to grow at 40°C and its lack of nitrate assimilation.

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One more interesting point in the present case is CSF eosinophilia. CSF pleocytosis was not described in any of the nine previously reported cases of cerebral *W. dermatitidis* infections, including the three probable cases. In another supposed cerebral *W. dermatitidis* infection, CSF pleocytosis (250 to 1,300/mm³) with 30 to 70% eosinophils was shown (11). However, in the review of Matsumoto et al., the causative organism was not available for a reconfirmation of its identification, and the reviewers were not sure of the cause of the infection (9). Even if the case that was reported by Nakano et al. (11) was caused by *W. dermatitidis*, it is extremely rare for CSF pleocytosis and eosinophilia to accompany cerebral phaeohyphomycosis due to *W. dermatitidis*. To date, the list of main causes of CSF eosinophilia includes inflammatory or infectious diseases, such as cerebral cisticercosis, viral encephalitis, and myelitis, and does not include fungal infection (12). The present case suggests that, in cases of multiple brain abscesses accompanied by CSF eosinophilia, *W. dermatitidis* infection should be included in a differential diagnosis.

REFERENCES