Disseminated Subcutaneous Phaeohyphomycosis caused by *Exophiala oligosperma* in a Patient with Wegener’s Granulomatosis

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Phaeohyphomycosis (PHM) is a rare infectious disease caused by dematiaceous fungi in cutaneous, subcutaneous or visceral organs. Most cases have been reported in immunocompromised patients (1, 2), such as those with autoimmune diseases and post-transplant patients. Various kinds of fungi, including *Alternaria*, *Phialophora* and *Exophiala* species, are known aetiological agents (3). However, few cases of cutaneous or subcutaneous PHM caused by newly recognized *E. oligosperma* have been reported (3, 4) and no treatment for this infection has been established. We report here a case of subcutaneous PHM caused by *E. oligosperma* in a patient with Wegener’s granulomatosis that was treated successfully with voriconazole (VRCZ).

CASE REPORT

A 71-year-old woman with Wegener’s granulomatosis presented with an 8-month history of multiple subcutaneous nodules, accompanied by cutaneous erythema and discharge of pus, on her left forearm (Fig. 1A). A small nodule had appeared on the dorsal part of her left hand, and similar lesions had gradually expanded on her left forearm. She had been treated with prednisolone and methotrexate (MTX) for Wegener’s granulomatosis, with an 8-month history of multiple subcutaneous nodules, accompanied by cutaneous erythema and discharge of pus, on her left forearm (Fig. 1A). A small nodule had appeared on the dorsal part of her left hand, and similar lesions had gradually expanded on her left forearm. She had been treated with prednisolone and methotrexate (MTX) for Wegener’s granulomatosis.

Fig. 1. (A) Multiple subcutaneous nodules with pus discharge on the left forearm. (B) Nine weeks after initiation of voriconazole treatment, the nodules have decreased significantly. (Broken lines indicate the extent of the subcutaneous lesions).
Lopez et al. (3) mentioned a case of subcutaneous PHM in a post-renal transplant patient who presented multiple subcutaneous nodules on the right leg, and was treated with 3-month administration of oral ITCZ. Tokuhisa et al. (4) also reported a case of cutaneous PHM of the face in a healthy patient without relevant history, who was treated with topical terbinafine cream. In our case, the patient, taking corticosteroid and immunosuppressant for Wegener’s granulomatosis, presented multiple subcutaneous nodules on her forearm, and was treated successfully with oral VRCZ combined with surgical excision. The MIC of our isolate indicated susceptibility to MCFG, AMPH-B, 5-FC, ITCZ, MCZ and VRCZ, but not to FLCZ. Bossler et al. (7) also reported that E. oligosperma from their case of olecranon bursitis was very susceptible to 5-FC, ITCZ, VRCZ and AMPH-B.

Dematiaceous fungal infections are classed into PHM and chromoblastomycosis on the basis of the morphology of causative agents in lesions. The form of the “hyphae” and “sclerotic cells” can be characteristic in the lesion of PHM and chromoblastomycosis, respectively (10). In our case, fungi in the biopsy specimen were observed in the form of “hyphae”, which changed to “sclerotic cells” after treatment of the fungal infection (Fig. 2C, D). Exophiala species in PHM tissues are generally present in the form of hyphae, but may be sclerotic cells in inactive and durable parasitic conditions. Both treatment with VRCZ and the host defence recovery may reduce the activity of fungi and shift its morphology to a strong durable parasitic form of sclerotic cells. However, the tissue with sclerotic cells obtained by operation did not reveal signs of recovered host defence, such as maturation of granulomas or a reduction in abscesses.

REFERENCES