Subcutaneous phaeohyphomycosis caused by Bipolaris hawaiiensis in an immunocompetent patient

Sir,

Phaeohyphomycetes are dematiaceous (brown pigmented) fungi which derive their pigmentation from melanin in cell walls. Cutaneous and subcutaneous infections due to this group of fungi are reported globally, usually following traumatic implantation of the fungal elements through contaminated soil, wood splinters or thorns. Bipolaris hawaiiensis is a phaeohyphomycete which belongs to division Deuteromycota and genus Cochliobolus. They have been occasionally found in humans, mainly as chronic colonizers and in patients with allergic sinusitis. We present a case of post surgery subcutaneous infection due to Bipolaris hawaiiensis, which has very rarely been reported in the literature.

A 30-year-old lady presented with complaints of recurrent swelling and pus discharge from a laparotomy surgery scar for 3 months. She had undergone surgery for removal of ovarian cysts due to polycystic ovarian disease, two weeks prior to the beginning of these complaints. There was mild to moderate pain at the site in addition to occasional discharge of pus. She later developed multiple similar swellings on the surgery scar and was prescribed antibiotic courses. She would partially respond to these drugs and relapse on stopping the medicines.

Dermatological examination revealed multiple painless nodules on and in the region surrounding the abdominal surgical scar [Figure 1]. There was no evidence of any underlying immunosuppression or other risk factors. Biopsy from the medial and uppermost swelling revealed the presence of dense granulomatous infiltrate and fungal material [Figure 2]. Microscopic examination of nodular tissue after treatment with 10% KOH revealed light brown septate hyphae. The material was cultured on potato dextrose agar (PDA). Colonies on PDA after 7 days were whitish-grey, which later became blackish brown. The growth on Sabouraud’s agar was downy grayish within 3-6 days, which rapidly became woolly gray along with dark pigmentation on the reverse of the colony. A slide culture of the isolate on corn-meal agar stained with Lactophenol Cotton blue (LCB) showed brown septate hyphae with geniculate, sympoidal conidiophores bearing conidia through pores [Figure 3]. Conidia were ellipsoidal and rounded at the ends with 4-6 pseudoseptae which did not extend to the cell wall and cells were enclosed within sacs. The fungus was identified as Bipolaris hawaiiensis on the basis of cultural characteristics and the arrangement of poroconidia on LCB mount.
Anti-fungal susceptibility testing was carried out by agar dilution method (a modification of the Clinical Laboratory Standards Institute (CLSI) broth based methodology) and the isolate was found to be sensitive to itraconazole. Surgical evacuation of the collection was done and oral itraconazole, 400 mg was administered twice daily to which there was a good response after 16 weeks [Figure 4].

*Bipolaris* is a filamentous dematiaceous fungus that usually grows on plant debris and in soil. It is an emerging human pathogen that has been occasionally reported from countries with hot climates like southern states of USA, Australia, Pakistan and India. The increasing incidence is due to the ever increasing population of immunosuppressed patients. Several reports of allergic fungal sinusitis (AFS) due to *Bipolaris* have appeared in the literature.[1] Other human infections reported include allergic bronchopulmonary disease, keratitis, orbital cellulitis, prosthetic heart valve infection, meningoencephalitis and osteomyelitis. Twenty two cases of *Bipolaris hawaiiensis* infections have been reported in humans [Table 1]. Very rarely have cutaneous infections been attributed to *Bipolaris* species.[2] Superficial cutaneous infection in three immunocompetent patients has been reported by Robb et al. A non-healing ulcer in a pancytopenic patient with acute leukemia preceded by trauma has been reported.[3]

Dermatoses caused by *Bipolaris hawaiiensis* infection have been reported twice in the past. A case of subungual hyperkeratosis due to the fungus has been reported.[4] The other report is a case of subcutaneous phaeohyphomycosis caused by *Bipolaris hawaiiensis* manifesting as a verrucous lesion on the big toe.[5]

Amphotericin B, itraconazole, voriconazole and surgical excision were reported to be successful modes of therapy. Surgery is preferred in immunocompromised patients.

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The first lesion appeared over the right medial canthus and bridge of the nose as an erythematous papule. There was a gradual increase in the number of lesions thereafter, involving the nose, upper lip, arms, buttocks and trunk. Some of the lesions were painful with scant bleeding on trauma. There was no history of any systemic complaints or bleeding from visceral sites. No one else in the family was affected.

Cutaneous examination revealed 25-30 non-compressible to partially compressible tender plaques and nodules ranging in size from 1 cm to 5 cm over the trunk, face, arms, and buttocks. [Figures 1 and 2]

There was no associated bruit or thrill. Over the left arm the papules were grouped to form a plaque. [Figure 1b]

Similar lesions were also present over the inner aspect of the upper lip, gingiva, and right vestibule. Keeping the provisional possibilities of glomus tumor, blue rubber bleb nevus, and venous malformation, a skin biopsy was taken from a nodule. Histological examination showed acanthosis and papillomatosis of the epidermis along with multiple ectatic dilated vascular channels in the dermis. [Figures 3 and 4]

These vascular channels were surrounded by multiple layers of cuboidal glomus cells with ovoid nuclei and eosinophilic cytoplasm. These features were consistent with the diagnosis of glomuvenous malformation.

Hemogram, liver function tests, renal function tests, urine routine microscopy, stool for occult blood, coagulation profile, and X-ray paranasal sinuses were within normal limits. At present, the patient has undergone 2 sessions of sclerotherapy with polidocanol for symptomatic lesions with minimal improvement. The patient has been counseled about the nature and usual course of the disease, and an informed decision will be made regarding planning of future sessions.

Glomus tumors are thought to represent neoplastic proliferations of glomus cells which are modified...