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Phaeohyphomycosis of the dorsum of the hand: A rare case report

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Abstract
Phaeohyphomycosis is a chronic and rare mycotic infection caused by various heterogenous groups of phaeoid or dematiaceous fungi usually involving the skin and subcutaneous tissue manifestating commonly subcutaneous abscesses or cystic swellings. Subcutaneous phaeohyphomycosis are also characterised by papulonodules, verrucous, hyperkeratotic or ulcerated plaques, pyogranuloma, non-healing ulcers or sinuses. The commonly associated genera are. This condition involves the presence of brown-walled hyphal structures in the dermis and epidermis. Here, we are reporting a rare case of phaeohyphomycosis presenting as an cystic lesion on the dorsum of the hand which was surgically excised.

Keywords: chronic, cystic, dematiaceous, mycosis, surgery

Introduction
Phaeohyphomycosis is a chronic infectious condition caused by dematiaceous fungi which usually involve the skin and subcutaneous tissue and occasionally the paranasal sinuses or the central nervous system. Rarely involving the paranasal sinuses, eyes, central nervous system, lymph nodes, and bone [1]. The term 'phaeohyphomycosis' was coined by Ajello *et al*. in 1974 [2]. Phaeohyphomycetes are fungi that produce brown-black structures, particularly spores, at least at some period in their life cycle. These dematiaceous fungi (pigmented) contain melanin in their cell walls and melanin is a known virulence factor in these fungi and acts by scavenging free radicals produced by phagocytic cells in the oxidative process, which would kill most organisms, and also prevents their action on the plasma membrane by binding to hydrolytic enzymes [3]. The fungus is present in host tissues as brownish hyphae, pseudohyphae, yeast cells, or combination of these forms [4]. These organisms are widespread in the environment, being found in soil, wood, and decomposing plant debris. They are distributed worldwide and are more common in tropical and subtropical climates. The major etiologic agents of phaeohyphomycosis are species of Altenaria, Bipolaris, Exophiala, Phialophora, Cladosporrium, Fonsecaea, Curvularia, Chaetomium, Phoma, Exserohilum, and Wangiella [5]. The clinical presentation ranges from solitary cutaneous nodules to deep subcutaneous abscesses [6]. The subcutaneous infections typically occur on the exposed areas of the body, especially the extremities, fingers, wrists, knees, or ankles, resulting from traumatic inoculation [5]. Histopathological examination (HPE) and culture is important in the diagnosis of phaeohyphomycosis.

Case Report
75 year old elderly male, a farmer by occupation, presented with a swelling over the dorsum of the right little finger for the past 5 months. It was spontaneous in onset and gradually progressed to the present size. He complained of pain occasionally over the swelling. There was no ulceration or bleeding from the swelling. There was no history of trauma or thorn prick to the dorsum of the right little finger. There was no history of any co-morbidities. On examination, a swelling of size 5 x 4cm was present over the 4th web space extending onto the dorsum of the little finger. The swelling was not warm or tender, was cystic in consistency but not translucent. It was free from the underlying structures like the extensor tendons and the bones. A clinical diagnosis of a dermoid was made and planned for excision biopsy.

Under axillary block and tourniquet control, an elliptical incision was made and subdermal flaps were raised. Cystic lesion was identified and was filled with pus. The pus was drained and sent for cultures and the cyst was excised in toto. Haemostasis was secured and incision was closed with 4-0 nylon over Segmuller drains. Compression dressing was applied. Post-operative period was uneventful and sutures were removed on the 10th post-op day. Microscopic examination reveals fibrocollagenous cyst walls lined by granulomas composed of epitheloid cells, foreign body and Langhans giant cells. The wall contains aggregates of foamy histiocytes, mixed inflammatory cell infiltrate and necrosis. Pigmented fungal hyphae are seen. On Periodic Acid Schiff stain, branching and septate fungal hyphae morphologically consistent with phaeohyphomycetes were seen.

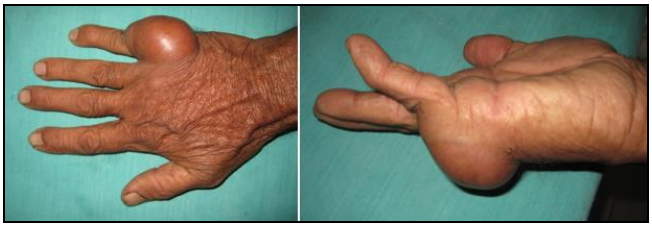

Fig 1: Clinical photograph showing the lesion

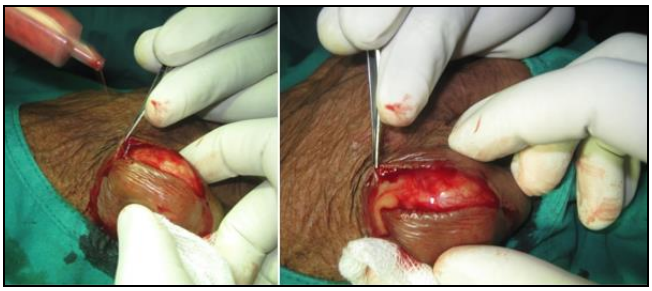

Fig 2: Photograph showing the skin incision with the cyst containing pus

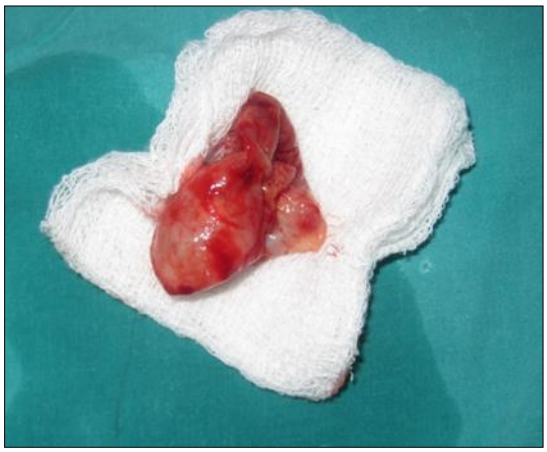

Fig 3: Photograph after excision showing complete cyst wall

Discussion

Dematiaceous fungi are characterized by their dark pigmentation due to the melanin complex in their cell wall. They are found everywhere, but mostly in soil and vegetation. Infection usually occurs through traumatic inoculation of the skin and subcutaneous tissue with contaminated matter, with majority of lesions occurring on the feet and legs of outdoor workers, or inhalation of the etiologic agent [7-10]. The infections caused by dematiaceous (melanized) fungi are grouped into three classes that include phaeohyphomycosis, chromoblastomycois, and eumycotic mycetoma [5]. Phaeohyphomycosis represents a broad range of primary and opportunistic mycoses classified into four clinical forms, which includes superficial or cutaneous, subcutaneous, systemic, and disseminated forms (brain, eye, central nervous system, peritoneum, bones). Subcutaneous infections mostly occur on the exposed parts of the body such as the limbs, fingers, wrist, knees, or ankles and are usually secondary to direct inoculation, often succeeded by trauma. Subcutaneous phaeohyphomycosis occurs in all climates throughout the world. The climate ranges from tropical to temperate in India, and the disease has been reported from the subcontinent [1]. Current prevalence is well documented, but in recent years, the number of cases has increased, especially in immunocompromised patients. Males are commonly affected because of their outdoor occupation and the highest incidence is seen in the third and fifth decades of life [11]. The disease most commonly presents as cysts, commonly seen in the phalanges, or well-demarcated, slow-growing nodules. The most common subcutaneous lesion is the cystic form which is characterised as a firm lesion with well-defined edges and intact skin surface [12]. Differential diagnoses include lipomas, epidermal cysts, bacterial abscesses, fibroids, foreign body granuloma and squamous cell carcinoma [13]. Laboratory diagnosis includes isolation of fungal hyphae on potassium hydroxide mounts and culture on Sabouraud's dextrose agar [9]. Histologically, the lesions show brown-walled septate hyphae or yeast or a combination of both in tissue. The Fontana-Masson stain (specific for melanin), PAS and Gomori methenamine silver stains can be used to confirm the diagnosis [9]. All dematiaceous fungi are similar in morphology and can be differentiated only by culture. Sharma *et al*. reported 23 patients with subcutaneous phaeohyphomycosis from India predominantly involving the extremities and very few cases with dissemination and reviewed the literature [1, 5]. Murayama *et al*., studied and reviewed 54 cases of phaeohyphomycosis due to *E. jeanselmi*, but failed to identify any underlying disease in 31 cases [14]. Local infection may be treated with excision or cryotherapy or in combination with antifungals [1]. Dematiaceous fungi are most susceptible to voriconazole, itraconazole and posaconazole, whereas ketoconazole and fluconazole have limited activity. Amphotericin-B and 5-flucytosine may also be used as treatment modality. Systemic disease is often refractory to therapy [9]. Successful treatment of these lesions includes complete excision. In case of incomplete resection or relapse, oral itraconazole is still considered as the standard treatment for this condition [15]. Systemic disease is often refractory to therapy [9, 16]. Therefore, non-invasive techniques like ultrasound biomicroscopy can be useful in delineating the dermal lesions and monitoring response to therapy. Ultrasound biomicroscopy (UBM) is high frequency ultrasonography performed using a 50-MHz probe (depth of penetration of 4mm). Dermatological ultrasound is usually performed using a 20-MHz probe (depth penetration of 8-10 mm). However, UBM gives a much superior resolution of the dermis and epidermis, compared with lower-frequency probes. It can be used to measure skin thickness, axial and lateral extension of tumors and inflammatory processes [17].

Conclusion

Subcutaneous phaeohyphomycosis should be considered as a differential diagnosis for lipoma, fibroma, epidermal cyst, or foreign body reaction. Surgical excision of the lesion with or without antifungal agents is the mainstay of treatment of subcutaneous phaeohyphomycosis. Our patient is doing well and is under follow-up with no relapse.

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