Case Report

A Rare Case of a Subcutaneous Phaeomycotic Cyst with a Brief Review of Literature

Madhavan Manoharan[1], Natarajan Shanmugam[2], Saveetha Veeriyan[3]

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[1] *Department of Pathology, Saveetha Medical College, Thandalam, Kanchipuram District, India, 605102*

[2] *Department of Orthopaedics, Saveetha Medical College, Thandalam, Kanchipuram District, India, 605102*

[3] *Department of Radiology, Saveetha Medical College, Thandalam, Kanchipuram District, India, 605102*

Abstract

Phaeohyphomycosis consists of a heterogeneous group of fungal infections caused by more than 80 genera and species. Subcutaneous infection usually follows traumatic implantation of a fungus by a wooden splinter that the fungus inh abits as a saprophyte. The growth of the fungus forms verrucous plaques or a painless subcutaneous abscess. We report a subcutaneous cyst (phaeomycotic cyst) in the leg of a 60-year-old woman that developed after a thorn prick at that site. With the provisional diagnosis of an epidermoid cyst, she was treated with a simple excision of the cyst. However, histopathological examination of the cyst revealed the typical features of fungus, and a definitive diagnosis of a phaeomycotic cyst was made. As the infective aetiology was not considered clinically, the specimen was not sent for microbiological culture, and hence the exact species was not identified. As the lesion was localised, simple excision was sufficient treatment, and no recurrence was observed during 12 months of follow-up.

Keywords: *clinical microbiology, cyst, dermatomycosis, diagnosis, female, leg dermatoses*

Introduction

Phaeohyphomycosis is a heterogeneous group of fungal infections caused by a variety of naturally pigmented fungi. The aetiological agents of this mycosis, which include more than 80 genera and species, are common saprophytes found in soil, wood, and decaying vegetable matter (1,2). Phaeohyphomycosis is rare, although it is more common in immunocompromised individuals (3). Phaeohyphomycosis affects either superficial tissues, such as the skin, cornea, and subcutaneous tissue, or deep tissues, such as the brain, and cases of phaeohyphomycosis are classified accordingly (2). Subcutaneous infection is usually caused by *Exophiala* and *Phialophora* species (4). The infection typically follows traumatic implantation of the fungi by a wooden splinter or a thorn prick and manifests as a cystic lesion. Herein, we report a typical case of a subcutaneous phaeomycotic cyst occurring in an elderly woman.

Case Report

A 60-year-old woman presented with painless swelling over the anterior aspect of the right leg for the past one and half years. It started as a small swelling and gradually reached the present size. The patient, a labourer, was pricked by a thorn while clearing wild bushes two years ago. Swelling was noticed a few months after the incident. On examination, her general condition was good. She was afebrile. Other systemic examination results were within normal limits. Local examination revealed a firm, fluctuant, mobile swelling measuring about 3×3 cm situated over the anterior aspect of the right leg. It was not painful or tender. It was not attached to the underlying bone. An X-ray of the right leg showed a well-circumscribed soft tissue nodule in the subcutaneous plane in the anterior aspect of the shin (Figure 1). The underlying bone appeared normal, with no abnormal lytic or sclerotic lesions or cortical breaks. The chest roentgenogram was normal. Her haemoglobin level was 9.0 gm/dL, sugar 82 mg/dL, urea 34 mg/dL, and creatinine 1.0 mg/dL. The urinalysis results were normal.

Operative findings

The soft tissue mass was easily separated from the surrounding tissue and was removed in toto. When the excised cyst was cut open in the surgical theatre, it expelled purulent material (Figure 2). The cyst was immersed in 10% formalin and sent for histopathological examination.

Histopathological examination

Microscopy revealed a fibrocollagenous cyst wall lined by granulomas, composed of foreign-body giant cells, epithelioid cells, xanthoma cells, and lymphoplasma cells. Some of the giant cells contained fungal hyphae. The hyphae were pigmented (Figure 3) and septate. Constrictions were present at a few septations, forming a structure similar to a bamboo stem (Figure 4). With these histological characteristics, the diagnosis of a phaeomycotic cyst was made. As the specimen was preserved in formalin, fungal culture to identify the species could not be performed.

Follow-up

The suture was removed after 2 weeks, and the wound healed well. There was no recurrence observed during the 12 months of follow-up.

Figure 1: X-ray showing a well-delineated soft tissue mass anterior to the tibia.

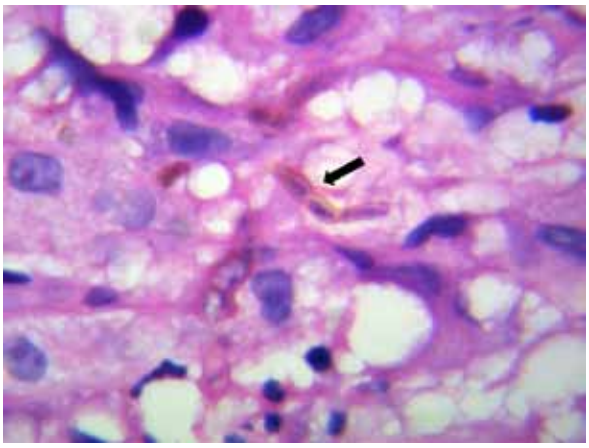

Figure 3: Haematoxylin and eosin staining of the cyst wall (100× magnification). Hyphae are brown in colour (arrow).

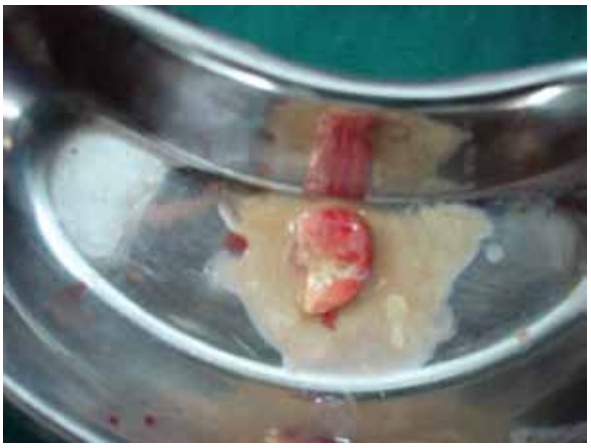

Figure 2: The cyst contained purulent material.

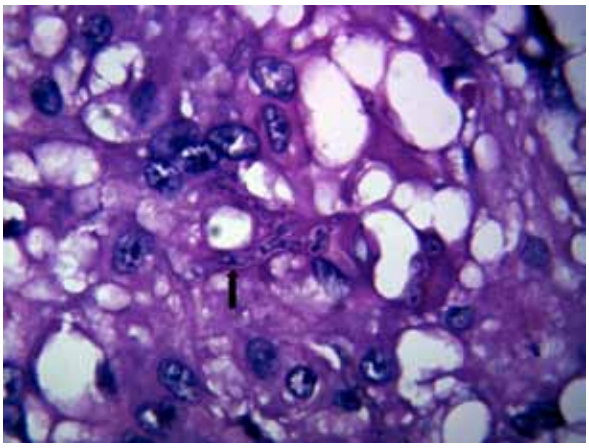

Figure 4: Haematoxylin and eosin staining of the cyst wall (100× magnification). Hyphae showed constriction at the septation, similar to bamboo stem (arrow).

Discussion

Phaeohyphomycosis infections occur worldwide in both animals and humans. These infections are more common in immunodeficient or debilitated hosts and rarely affect healthy individuals (3). Phaeohyphomycosis has been clinically divided into superficial (cutaneous and corneal), subcutaneous, and systemic phaeohyphomycosis by McGinnis (2).

Subcutaneous phaeohyphomycosis is usually caused by *Exophiala* and *Phialophora* species (4). Occasional cases caused by *Fonsecaea* species have also been reported (5). Subcutaneous phaeohyphomycosis usually results in a painless subcutaneous abscess or in verrucous plaques on the hand, arm, face, or neck. Although phaeohyphomycosis has distinct clinical features, it is occasionally confused with chromoblastomycosis (Table 1).

Typically, phaeohyphomycosis follows traumatic implantation of the fungus by a wooden splinter, as in our case. Lymphangitis and regional lymphadenopathy are unusual. Hence, infective aetiologies are usually not considered. Our case was clinically suspected to be an epidermoid cyst.

The host reaction to phaeohyphomycosis is similar regardless of the aetiological agent and the anatomic site of involvement. The lesion is usually situated in the dermis and the subcutaneous plane and is characterised by cyst formation with dense collagenous connective tissue and central suppurative necrosis. The overlying epidermis is usually normal. The wall contains compact aggregates of epithelioid histiocytes and numerous giant cells. Pigmented moniliform fungal elements are usually present inside the giant cells or extracellularly in the necrotic debris. Fungi may vary in their degree of pigmentation and may also appear as infrequently branching hyphae measuring 2–6 μm wide. The fungi are closely septate and constricted at their prominent septations (3). Our case exhibited the typical features of phaeohyphomycosis.

In many cases, identification of the exact species is not attempted, as phaeohyphomycosis is not suspected clinically. Similarly, in our case, an infective aetiology was not suspected, and after excision in the operation theatre, the cyst was fixed in formalin. When culture and identification of species is not pursued, the cyst can be histologically differentiated from black grain eumycotic mycetoma (Table 2) and chromoblastomycosis (Table 3).

Regarding the management of subcutaneous phaeohyphomycosis, excision of the localised lesion is usually curative (6). In our case, after the local excision, there was no recurrence found during the 12 months of follow-up.

Conclusion

Subcutaneous phaeohyphomycosis is a rare fungal infection caused by a wide variety of dematiaceous fungi. A high index of suspicion for infective aetiologies is needed to make a proper clinical diagnosis. For a localised lesion, simple excision is usually curative. When phaeohyphomycosis is not suspected and the identification of species by culture is not pursued, routine histopathological examination is sufficient to arrive at a diagnosis of phaeohyphomycosis.

Table 1: Clinical differences between chromoblastomycosis and phaeohyphomycosis

Chromoblastomycosis	Phaeohyphomycosis
• Starts as a papule that usually develops into a nodule and progresses to multiple nodules involving a large portion of the body	• Usually forms a single subcutaneous cyst and rarely multiple cysts
• Skin surface is scaly, cracked, or verrucous	• Skin surface is usually smooth and uninvolved
• Body reactions against the fungus are ineffective, and the infection spreads	• Body reactions produce an effective fibrocollagenous tissue capsule around the infection and arrest its spread
• Systemic involvement is rare	• Cerebral involvement develops from respiratory infection
• Excision of the early lesion alone is curative; advanced lesions require prolonged antifungal treatment	• Local excision is curative; only systemic infection requires antifungal treatment

Table 2: Histological differences between black grain eumycotic mycetoma and phaeohyphomycosis

Black grain eumycotic mycetoma	Phaeohyphomycosis
• Forms distinct granules	• Never forms tissue granules
• Composed of interwoven mycelial aggregates	• Composed of scattered individual polymorphous fungal elements
• Organised	• Unorganised
• Nearly always extracellular	• Often intracellular

Table 3: Histological differences between chromoblastomycosis and phaeohyphomycosis

Chromoblastomycosis	Phaeohyphomycosis
• Brown, spherical to polyhedral thick-walled muriform cells	• Moniliform cells
• 5–12 µm	• 2–6 µm
• Muriform cells reproduce by septation in 1 or 2 planes	• Moniliform cells
• Do not produce chains of cells	• Do produce chains of cells
• Epidermis is hyperplastic	• Epidermis is normal

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Authors' Contributions

Conception and design, drafting of the article: MM
Provision of patients: NS, SV
Final approval of the article: MM, NS, SV

Correspondence

Associate Professor Dr Manoharan Madhavan
MBBS (University of Madras), MD Path (Pondicherry University)
Saveetha Medical College
Thandalam, Kanchipuram District
India, 605102
Tel: +91-90-9403 3599
Fax: +91-44-2681 1171
Email: madhavan6@gmail.com

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