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UNVEILING ATYPICAL SUBCUTANEOUS PHAEOHYPHOMYCOSIS IN A DIABETIC PATIENT: A CASE REPORT

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ABSTRACT Phaeohyphomycosis is a rare mycotic infection caused by various heterogeneous groups of phaeoid (dematiaceous) fungi that affect the skin and subcutaneous tissues. Common clinical manifestations include subcutaneous abscesses or cystic swellings. We report the case of a 39-year-old male presenting with a subcutaneous phaeohyphomycosis manifesting as swelling over the left elbow. Initial examination suggested a differential diagnosis of abscess or olecranon bursitis. Histopathological analysis revealed granulomatous inflammation, and special staining with Grocott's methenamine silver stain showed broad pigmented hyphae. Culture produced black-colored colonies identified as Exophiala jeanselmi. The patient underwent surgical excision of the lesions. This case underscores the importance of considering fungal infections in atypical presentations of chronic subcutaneous swellings, especially in diabetic patients.

KEYWORDS : phaeohyphomycosis, Exophiala jeanselmei ,itraconazole

INTRODUCTION

Pheohyphomycosis is a rare and heterogeneous group of mycotic infections caused by dematiaceous (phaeoid) fungi. It typically affects the skin, subcutaneous tissue, paranasal sinuses, and occasionally the central nervous system[1]. These fungi are commonly found in soil and can infect subcutaneous tissues, predominantly in the extremities, with potential spread to distant sites, especially in immunocompromised individuals. Pheohyphomycosis is caused by brown-pigmented (phaeoid) or dematiaceous fungi such as Exophiala jeanselmei, Exophiala dermatitidis, Bipolaris, and Alternaria species, affects Indian patients at a younger age.(4) Exophiala dermatitidis is associated with higher mortality rates .

Melanin serves as a significant virulence factor in these fungi by scavenging free radicals generated during phagocytic cell oxidative processes, thereby inhibiting their antimicrobial action on the plasma membrane through binding to hydrolytic enzymes [2]. Clinical manifestations range from solitary cutaneous nodules to deep subcutaneous abscesses [3]. Subcutaneous infections typically manifest on exposed body areas such as extremities, fingers, wrists, knees, or ankles, often resulting from traumatic inoculation [4]. Histopathological examination (HPE) and culture play pivotal roles in phaeohyphomycosis diagnosis. We present this case due to its unusual clinical presentation, featuring multiple cystic swellings on both feet and the right hand in an immunocompetent individual.

Subcutaneous infections commonly develop on exposed areas of the body, such as the extremities (fingers, wrists, knees, ankles), due to traumatic inoculation.[5]Diagnosis of phaeohyphomycosis relies significantly on histopathological examination (HPE) and culture, which are essential for accurate identification and treatment planning. Treatment with newer azoles shows promise, with excision alone or in combination with azoles emerging as effective therapeutic strategies. Clinically, it typically manifests as localized subcutaneous or deep-seated infections, presenting as cysts or abscesses.

Case Report

A 39-year-old male electrician presented with an 8-month history of progressive swelling on the posterior aspect of his left elbow. The patient has a medical history significant for diabetes mellitus for the past five years, managed with oral hypoglycemic agents. There was no history of trauma or tuberculosis exposure, and the patient denied any recent surgical procedures. He reported intermittent yellowish discharge from the swelling but denied fever, chills, or other systemic symptoms.

On physical examination, a firm to soft cystic swelling measuring approximately 6 *5 cm was palpated over the left olecranon, with no evidence of overlying pustules or scars.(Figure 1). The swelling was non-tender to palpation, and there were no discharging sinuses or palpable fluctuance. Regional lymph nodes were within normal limits, and there was no associated lymphadenopathy.

Given these findings, a differential diagnosis of abscess or olecranon bursitis was considered. Routine investigations were conducted, which were generally normal except for elevated blood glucose levels, with an HbA1c of 9%.

During the excisional biopsy, purulent discharge was observed from the biopsy site, prompting the incision and drainage of all lesions. The pus obtained was sent for Gram stain and culture, while the cyst material was sent for histopathological examination (HPE). Gram staining was negative for bacteria, but culture on Sabouraud's dextrose agar identified pigmented fungi. Lactophenol cotton blue staining confirmed the presence of Exophiala jeanselmi species. The Histopathology examination of the cyst material revealed multiple granulomas, and Grocott's methenamine silver (GMS) stain showed broad brownish filaments.(Figure 2 and 3).

Gross examination of the excised tissue showed a nodular, grey-white mass measuring 3x2x2 cm. The cut section revealed multiple cysts containing necrotic material. Therefore, a diagnosis of subcutaneous phaeohyphomycosis was made, and the patient was prescribed a regimen of itraconazole for one week and fluconazole for three weeks each month, over a period of three months.

After three months of therapy, there was marked improvement in the lesion. At the last follow-up session, there were no local signs of recurrence, and the wound had healed completely.

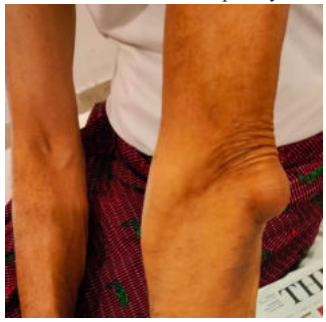

Figure 1:clinical photograph

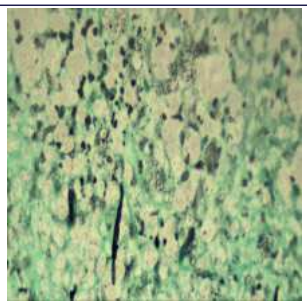

Figure 2:granulomas with pigmented fungal growth -GMS stain

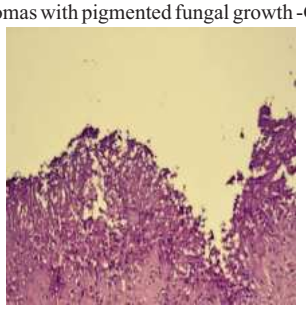

Figure 3:H and E Staining showing granuloma in the dermis composed of epitheloid cells lymphocytes and fibroblasts.

DISCUSSION

Subcutaneous phaeohyphomycosis is a rare infection, but its incidence appears to be rising alongside the growing population of immunocompromised patients[5]. However, our case demonstrates that an immunocompromised state is not a prerequisite for developing phaeohyphomycosis. The infection typically occurs through traumatic inoculation of the skin and subcutaneous tissue [6]with contaminated matter, with most lesions appearing on the feet and legs of outdoor workers, as seen in this patient. The affected age range spans from 3 to 60 years, with males being more commonly affected due to their outdoor occupations. This infection is more prevalent in tropical and subtropical climates. In our case, the patient was a young male who presented with swelling over the elbow

The most common etiological agents of subcutaneous phaeohy phomycosis are E. jeanselmi followed by E. dermatitidis. [4]The genus Exophiala is widely found in the environment and can cause infections in both immunocompromised individuals (such as those with HIV, transplant recipients, chronic debilitating diseases, diabetes, and on immunosuppressive therapy) and, rarely, in immunocompetent persons. E. jeanselmi typically causes mild cutaneous and subcutaneous infections, often presenting as localized and solitary phaeohyphomycotic cysts.[5] Even in severely immunosuppressed patients, Exophiala infections tend to remain localized.

Our case did not exhibit the typical features of phaeohyphomycosis, despite the general host reaction typically being similar regardless of the causative agent or site of infection. Normally, lesions are located in the dermis and subcutaneous tissues, characterized by cyst formation accompanied by dense collagenous connective tissue and central suppurative necrosis. The epidermis above is usually unaffected. Histologically, the lesion typically shows compact aggregates of epithelioid histiocytes and numerous giant cells in the wall, containing pigmented moniliform fungal elements within or outside giant cells amidst necrotic debris. Fungal elements often appear as septate, occasionally branching hyphae measuring 2-6 μm wide, with prominent septations[7].

The typical clinical presentation includes a solitary subcutaneous cyst or abscess that feels firm to fluctuant, usually sparing the overlying skin. Commonly affected sites include the upper and lower limbs, specifically over the fingers, wrists, knees, or ankles, and less frequently on the face, neck, and scalp. All dematiaceous fungi share similar morphology and can usually be differentiated only through culture.

In a study by Murayama et al. [9], which reviewed 54 cases of phaeohyphomycosis caused 1stby E. jeanselmi, 31 cases did not reveal any underlying predisposing disease. Our patient exhibited no underlying immunosuppressive condition predisposing to phaeohyphomycosis, but did have diabetes. Our case report emphasizes the importance of considering fungal infections in the differential diagnosis of subcutaneous soft swellings, which can sometimes be mistaken for conditions like lipoma, fibroma, abscess, or other inflammatory tissue swellings. Treatment typically involves surgical excision of the lesion, often combined with antifungal agents such as itraconazole, ketoconazole, or amphotericin B.

In our case, we utilized both itraconazole and fluconazole for treatment.

CONCLUSION

This case highlights the complexities in diagnosing and treating subcutaneous mycoses, especially in immunocompromised patients like those with diabetes mellitus. Accurate identification of the causative agent, as demonstrated with Exophiala jeanselmi in this patient, is essential for effective management. Treatment with itraconazole and fluconazole over three months led to marked improvement and complete healing of the lesion.

While antifungal therapy and surgical excision are mainstays of treatment, challenges such as drug tolerability and the risk of relapse remain. The development of new antifungal agents and combination therapies holds promise for better outcomes. Antifungal susceptibility testing is crucial for guiding treatment decisions. Continued research and advancements in antifungal therapy are vital for improving management and prognosis of subcutaneous mycoses.

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Conflict of interest
No conflict of interest

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Informed consent
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Authors contributions
All Authors contributed equally to conceptualization, validation, visualization, writing- review and editing

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