Case Report

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A Rare Case of Fungal Osteoarthritis Which was Caused by Phaeohyphomycosis

Pathology section

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ABSTRACT

Phaeohyphomycoses are rare, opportunistic, fungal infections which are usually seen in immunocompromised individuals. These are primarily caused by soil saprophytes, plant pathogens, and contaminants which live in the environment. In humans, cutaneous infections are very common; however, the involvement of lungs, sinuses, eyes, bone, and the central nervous system is rarely seen. To the best of our knowledge, phaeohyphomycosis which causes osteoarthritis has never been reported in the world literature. We are reporting here, a case of fungal osteoarthritis of the right ankle joint, which was caused by phaeohyphomycosis in a 60-year old female diabetic patient. With specific anti-fungal treatment and physiotherapy, she recovered well and after 2 years of follow up, there is no recurrence. We are reporting this case due to its rarity, to study the clinical and the histomorphological features for an early diagnosis and appropriate management of the patient.

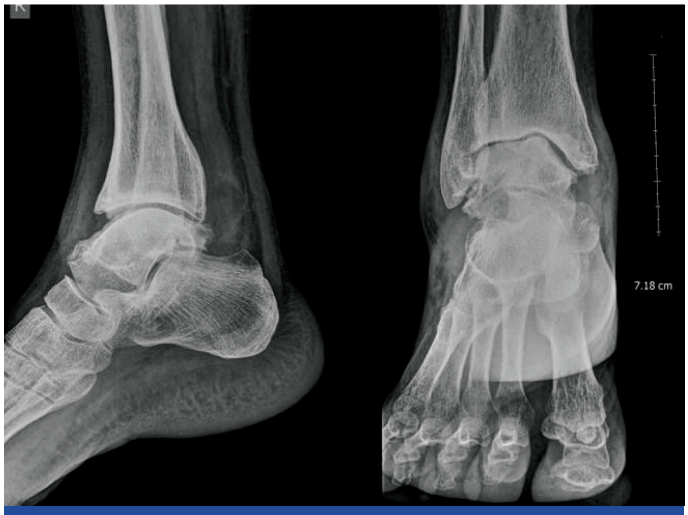
Key Words: Phaeohyphomycosis, Fungal osteoarthritis, Ankle joint

INTRODUCTION

Phaeohyphomycoses are rare fungal infections which are caused by darkly pigmented fungi, which are increasingly being seen in immunocompromised patients [1-5]. More than 100 species which belong to 57 genera are known till date. The human infections are commonly caused by the Bipolaris, Curvularia, Exserohilum, and the Alternaria species [3]. Depending on the immunological status of the patient and the route of infection, they cause sub-cutaneous infections, allergic bronchopulmonary mycosis, chronic allergic sinusitis, keratitis, brain abscess, and disseminated infections [1]. Phaeohyphomycosis which causes osteoarthritis has never been reported till date. We are presenting a case of fungal osteoarthritis of the right ankle joint which was caused by phaeohyphomycosis in a 60-year old diabetic female. With specific anti-fungal treatment and physiotherapy, she recovered well and after 2 years of follow up, there is no recurrence. So, it is important to know the clinical, epidemiological and the histomorphological features of these fungal infections, so that any suspected lesions, especially in immuno-compromised patients, can be evaluated for an early diagnosis and appropriate treatment.

CASE REPORT

A 60-year-old female known diabetic presented with pain and swelling in the right ankle joint of 4 months duration. There was no history of trauma. The local examination revealed tenderness and swelling in the right lateral side of the ankle joint. An X ray revealed erosion of the subtalar joint space, with osteoporosis and osteophyte formation [Table/Fig-1]. The fasting and post-prandial blood sugar levels were 226 mg/dl and 330 mg/dl respectively, whereas the other biochemical and haematological parameters were normal. A synovial biopsy was performed and it revealed cystic cavities with plenty of suppurative granulomas which comprised of epitheloid histiocytes, foreign body giant cells, neutrophils and necrosis. Embedded within these necrotic areas and giant cells, were thick walled irregular branching fungal hyphal

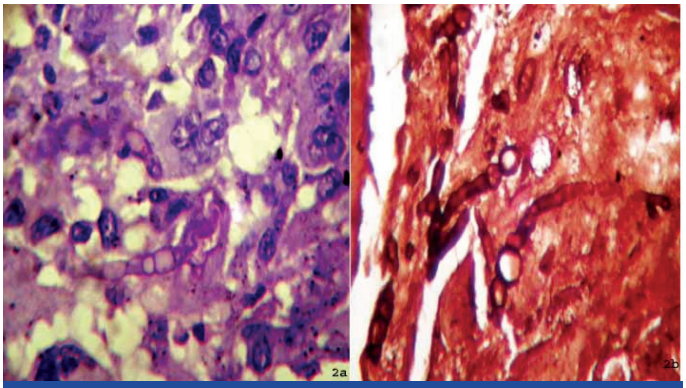


[Table/Fig-1]: Plain X ray of the foot displaying osteoarthritic changes like narrowing of the subtalar joint space with osteophyte formation.

structures with constrictions around their septations, which were highlighted by gomoris methenamine silver (GMS) staining. [Table/Fig-2] The fungal culture showed brownish black colonies with velvety surfaces and grayish hyphae, which confirmed the species as Exophiala jeanselmei. The patient was immediately started with itraconazole and regular physiotherapy. After 10 days of follow up, the swelling and the pain subsided. Now, she is on regular follow up since 2 years, with no evidence of recurrence. A repeat synovial fluid culture showed no growth.

DISCUSSION

Phaeohyphomycosis belongs to a heterogeneous group of fungal infections which was originally described by Ajello and colleagues in 1974 [1]. They proposed the name of the disease as phaeohyphomycosis and listed nine fungi as the aetiologic agents for it. Four years later, they included all the filamentous, dematiaceous and mycelial elements which caused superficial



[Table/Fig-2]: a) Microphotograph showing thick walled irregular branching fungal hyphal structures with constrictions around their septations, epitheloid histiocytes and foreign body giant cells (H&E stain x45X). b) Gomori's methenamine silver stain highlighting the fungal hyphae (GMS stain x45X)

cutaneous, sub-cutaneous and systemic infections [3,6]. More than 100 species and 60 genera have been documented till date [1,6]. These are primarily soil saprophytes, plant pathogens and domestic environment contaminants [5]. Over the past 2 decades, there has been a paradigm shift from Candida infections to these filamentous fungi, especially in immunocompromised individuals [1]. Histopathology shows a single, circumscribed lesion with a central cavity, which is filled with pus which is surrounded by fibrous wall [2,7]. The margins contain suppurative granulomas which are composed of foreign body giant cells, epitheloid histiocytes, plasma cells, and lymphocytes with fungi, which are adjacent to the purulent areas. Despite the dematiaceous nature of the fungi, the brown pigment is not always apparent and the hyphae may appear hyaline on hematoxylin-eosin staining [3]. Skin lesions of traumatic origin, which affect the hands and feet are the most common presentations in healthy people. Subcutaneous or nasal granulomata are relatively frequent, especially in European countries [2]. Systemic phaeohyphomycosis is almost restricted to immunocompromised individuals, with a fatal outcome [5]. Joint infections are secondary to the direct spread in cases of trauma or to systemic spread through inhalation. In our case, since there was no history of trauma or any ulcer around the joint, it was possible that the infection could have spread systemically. The solitary lesions can be treated by surgical excision, and the disseminated disease requires systemic anti-fungal treatment [1,4-7].The propensity of these fungi for dissemination and for resistance to the anti-fungal drugs presents management challenges [1]. Histopathology is diagnostic, whereas studies on the culture and the antibiotic sensitivity of the organisms are required for the species identification and the treatment. The sensitivity of the routine histopathological examination of the skin biopsies for the detection of the fungi is 85% and it can be increased to 100% with the use of GMS staining [1]. The polymerase chain reaction and analysis of the internal transcribed spacer of the ribosomal DNA are powerful tools for a rapid and precise laboratory diagnosis of the invasive fungal infections [5]. Itraconazole is considered as a drug of choice for the treatment of phaeohyphomycosis [1]. Posaconazole, with a similar structure as itraconazole, is another promising agent which can be used against these dematiceous fungi [1].

CONCLUSION

We hereby conclude that for the occurrence of secondary osteoarthritis in an immunocompromised individual; a synovial biopsy and a microbial culture is suggested to rule out the rare possibility of phaeohyphomycosis, for an accurate diagnosis and appropriate management.

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