Case Report

Subcutaneous pseudomycetoma of the knee caused by *Acremonium species* in a diabetic male: A case report

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Abstract

A subcutaneous infection in the form of pseudomycetoma is rare. *Acremonium species* are rarely considered to be pathogenic in subcutaneous infections due to their ubiquitous nature. We report a case of subcutaneous pseudomycetoma over the dorsolateral aspect of the left knee caused by *Acremonium species* in a 55-year-old diabetic male who was treated successfully with oral itraconazole.

Keywords: *Acremonium*, mycetoma, pseudomycetoma

Introduction

Acremonium species are filamentous molds and ubiquitous soil saprophytes found as contaminants in biological specimens or may rarely be pathogenic in susceptible hosts.[1] Clinical manifestations may range from superficial or localized infections like cutaneous infections, subcutaneous mycetomas, keratitis, onychomycosis, to invasive infections like fungemia, CNS, bone and joint infections, and endocarditis.[2] Only a few cases of human *Acremonium* skin and soft-tissue infections have been reported from India.[3,4] Here, we report an intriguing case of subcutaneous infection caused by *Acremonium species* in a patient with diabetes mellitus without the characteristic features of a mycetoma.

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Case History

A 55-year-old male chauffeur presented with swelling over the lateral aspect of the left knee for 2 months. The swelling gradually increased over 1 week when the patient was evaluated elsewhere. He denied any history of trauma. He was a known diabetic and hypertensive on oral medications.

An initial ultrasound revealed a left knee abscess. The patient's glycosylated hemoglobin (HbA1c) at the time was 7.6%. Incision and drainage (I and D) was carried out to drain the abscess. Empirical treatment of intravenous piperacillin-tazobactam for 2 days was given postoperatively, and oral cefuroxime and linezolid were advised for a week on discharge. Later, he developed a swelling over the operative site that was mildly painful. He denied any history of fever.

One week later, he presented to a surgeon at our hospital with these complaints. On examination of the left knee, an incision

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and drainage wound was seen over the dorsolateral aspect that had not healed completely.

Magnetic resonance imaging of the knee showed a soft-tissue irregularity involving the lateral aspect in the subcutaneous plane. Oral ibuprofen was advised to reduce the size of the swelling for 5 days. Suturing of the I and D wound was done after the swelling subsided.

Four weeks later, he presented with a painful oblong lump of 5 cm × 3 cm over the dorsolateral aspect of his left knee over the operated site [Figure 1]. It was warm, mildly tender, and fluctuant. The postoperative scar had healed. A repeat magnetic resonance imaging of the left knee showed a T2/FS-PD (Fat Suppressed Proton Density) hyperintense collection of 2.5 × 3.3 cm (8.25 cc in volume) along the dorsolateral aspect of the left knee joint [Figure 2].

Diagnostic aspiration of the collection was done. The aspirate was aseptically transported and processed for microbiological studies, including potassium hydroxide (KOH) mount. The KOH mount revealed branching, septate, and hyaline fungal hyphae. Gram staining and cultures were negative. A repeat aspiration was done to rule out contamination. It was concurrent with the previous findings.

Based on the microscopic findings, a working diagnosis of subcutaneous fungal infection was made. The patient was empirically started on oral itraconazole 200 mg twice daily. Culture on the Sabouraud's dextrose agar yielded compact, glabrous, off-white fungal colonies with pale yellow reverse after 6-7 days of incubation at 25°C and 37°C. The lactophenol cotton blue mount from the primary tube and slide culture revealed thin, delicate, hyaline, septate hyphae with unbranched, erect, tapering phialides with no conspicuous collarette. The conidia were oblong (2-3 um × 4-8 um) and one-celled. The characteristic clustering of the conidia at the tip of phialides with no macroconidia helped identify the *Acremonium species* [Figure 3]. Based on the fungal culture report, oral itraconazole was continued for 6 weeks.

At the end of 6 weeks, the swelling had decreased, and he was advised to continue oral itraconazole for 2 more weeks until the subsequent follow-up [Figure 4].

Discussion

Here, we report a case of pseudomycetoma by *Acremonium species*, presented as a swelling on the dorsolateral aspect of the left knee, which had gradually increased. It was mildly painful and did not hinder his daily work. The exact mode of transmission remains unclear as the patient denied any history of local trauma at the site such as from insect bite, thorn prick, or superficial puncture from wooden splinters. However, possible inoculation, either due to an open wound or iatrogenic inoculation during surgical procedures prior to diagnostic aspiration cannot be ruled out. Furthermore, this infection may have been favored by delayed wound healing due to uncontrolled diabetes. The swelling subsided significantly after proper glycemic control and treatment with an antifungal drug.

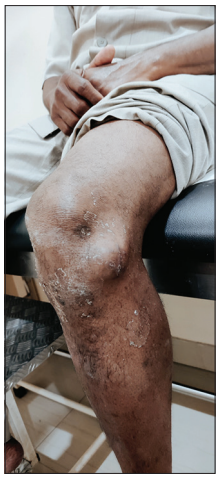

Figure 1: Left knee showing swelling over the dorsolateral aspect

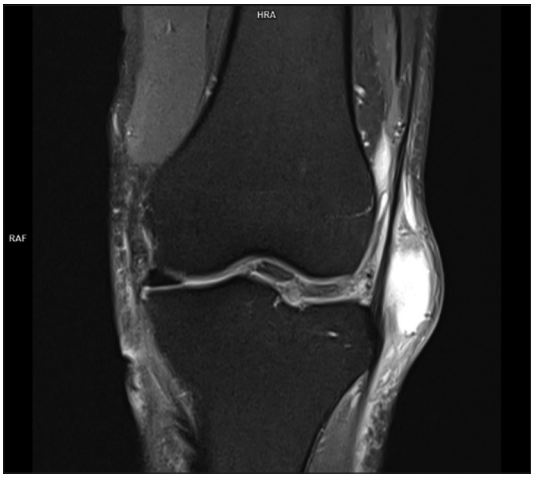

Figure 2: MRI of left knee

Figure 3: Lactophenol cotton blue mount (400X) showing oblong (2-3 um × 4-8 um) and one-celled conidia in clusters over the tip of erect, tapering phialides with no conspicuous collarette

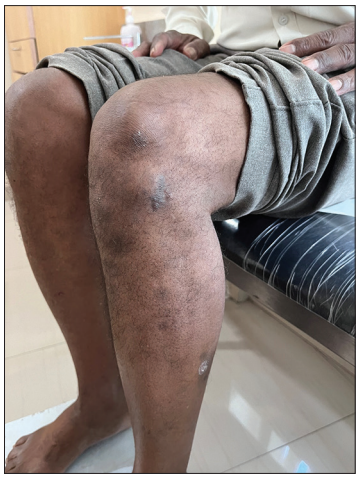

Figure 4: Left knee (follow-up after 6 weeks, showing significant reduction in the size of swelling)

Acremonium is an ascomycete belonging to Hypocreales, characterized by solitary, erect, hyaline, awl-shaped phialides producing single-celled, globose to cylindrical conidia, mostly in slimy heads (clusters).[1] Recently, based on molecular tools and phylogenetic analysis, the taxonomy has undergone significant changes, with several *Acremonium species* shifted to the *Sarocladium*.[5,6]

Acremonium may be confused with *Fusarium* by an untrained eye on microscopy. However, differences can be identified by growth rate and morphology (*Fusarium spp* have a faster growth and characteristic fluffy colony appearance).[1]

With recent taxonomical changes in the *Acremonium*, its distinction from *Sarocladium*is essential and can be made based on morphology (*Acremonium* has conidiophores which are mainly unbranched or poorly basitonously branched, the conidia are more variable in shape [subglobose, obovate, and ellipsoidal] and adelophialides which are usually absent).[5]

In tropical and subtropical countries, mycetomas present as subcutaneous swellings with sinus tracts, edema, and a granular discharge. Lower extremities are commonly involved, with feet being the most prevalent site due to bare feet walking. In an epidemiological study from North India on eumycetomas (fungal mycetomas), 30 cases were identified retrospectively over 13 years. Seven cases were diagnosed with *Acremonium* (three) and *Sarocladium* (four) species, respectively.[7]

This case is of note as our patient denied any history of trauma except the initial surgical procedures that he went through, and the presentation was rather uncharacteristic of mycetoma, with no sinus tract or granular discharge. It was diagnosed as pseudomycetoma caused by *Acremonium species* based on morphological features.

A significant gap exists in the management strategy of these infections due to discordance between *in vitro* studies and clinical cure rates. In immunocompromised hosts, *Acremonium species* are known to cause disseminated infection with fatal outcomes. Amphotericin B and voriconazole were used in most invasive infections in immunosuppressed hosts with variable outcomes.[8,9] Azoles like itraconazole[10,11] and posaconazole[12] achieved good clinical responses in some cases. Higher Minimal inhibitory concentration (MICs) were observed for most agents, including amphotericin B, posaconazole, and voriconazole.[13] In our case, the patient was treated on oral itraconazole 200 mg twice daily with good clinical outcomes.

Conclusions

A subcutaneous infection caused by Acremonium species is rare and does not present commonly as pseudomycetoma. This case highlights the fact that clinical practitioners should consider pseudomycetomas as an important differential in subcutaneous infections with indolent presentations. Direct inoculation is the most likely form of acquisition in immunocompetent hosts. Oral itraconazole can achieve good clinical results and is well tolerated in such patients. A higher degree of suspicion and microbiological expertise is crucial in diagnosing such infections as these can be easily dismissed as contaminants.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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