

Cutaneous Pseudallescheria Boydii Infection- Two Rare Case Reports

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Abstract: *Pseudallescheria boydii*, is a saprophytic fungi of increasing clinical importance which belongs to the genus *Scedosporium*. It is a rare cause of infection in humans. The prevalence of infection is more among immunocompromised patients and it is rarely seen among immunocompetent individuals. It causes broad spectrum of diseases including subcutaneous and soft tissue infection, pneumonia, brain abscess, endocarditis, sinusitis and disseminated infection. Here we report two cases of *Pseudallescheria boydii* in immunocompetent patients presented as subcutaneous swelling. Case 1: A 30 year old female presented with swelling over left elbow, which was clinically diagnosed as lipoma. Fine needle aspiration yielded pus like material. It was surgically debrided and tissue was sent for histopathological examination. Case 2: A 70 year old female presented with swelling over dorsum of right foot. On fine needle aspiration pus was aspirated. In both the cases on cytology, fungal elements showing septate hyphae with acute angle branching, intercalary and lateral conidia were seen in an inflammatory and necrotic background. Diagnosis was confirmed by Gomori methenamine silver stain and culture. On the basis of above findings diagnosis of *Pseudallescheria boydii* was made.

Keywords: Cutaneous mycetoma, Genus *Scedosporium*, *Pseudallescheriasis*, Gomori Methenamine silver stain

1. Introduction

Pseudallescheria boydii is a saprophytic microorganism belonging to the genus *Scedosporium*, which is a rare cause of infection in humans. These fungi are found in soil, polluted water and decaying vegetations.^[1] Clinical types of infections include cutaneous infections (especially mycetoma), keratitis, pneumonia, lung abscess, endophthalmitis, corneal infection and osteomyelitis.^[2,3] In immunocompetent hosts this organism is known to cause localized skin and soft tissue mycetoma especially after trauma.^[4] *Pseudallescheriasis* has been rarely reported in immunocompetent patients and the disease typically occurs in immunocompromised individuals.^[5]

A diagnosis of *Scedosporium* infection can be based on cytology and histopathology with isolation of the fungus in culture, serology and several molecular methods.^[6,7] An overall mortality of 46.9% has been described, but mortality can be as high as 87.5% in cases of disseminated disease.^[8]

Here we are reporting two cases of cutaneous mycetoma in immunocompetent patients.

2. Case Report

Case 1

A 30 year old female presented with complaint of swelling over left elbow since 5 months which was gradually increasing in size. There was no history of pain or trauma to the site. On examination swelling measuring 4×3cm was present over the left elbow which was soft, mobile and nontender. X ray of the elbow was unremarkable. Haematological tests were normal except for normocytic hypochromic anaemia, blood sugar level were within normal limits and serological test for HIV was negative.

Clinically, provisional diagnosis of lipoma was made and patient was sent for FNAC, which yielded pus. Smears were made from aspirated material which were stained with H&E, Wright's stain and Ziehl-Neelsen stain. The smears studied showed fungal elements with irregular septate hyphae, acute angle branching, intercalary and lateral conidia in an inflammatory and necrotic background. Z-N stained smears did not show the presence of acid-fast bacilli. With these findings cytological diagnosis of cutaneous mycetoma by *Pseudallescheria boydii* was made following which it was surgically debrided and tissue was sent for histopathological examination.

Excised specimen was processed and sections were stained with H&E and Gomori Methenamine silver stain. Histologically, section studied showed cyst wall consisting of granulation tissue, dense mixed inflammatory cell infiltrates rich in neutrophils, few foamy macrophages and few foreign body type multinucleated giant cells. On surface of granulation tissue, fungal elements with narrow, irregular septate hyphae having intercalary and lateral conidia were seen. Section showed fungal colonies in Gomori Methenamine silver stain. Part of the excised tissue was sent for culture study, which was inoculated on Sabaroud Dextrose Agar (SDA) media. Initially, it showed grey-white fluffy colonies followed by dark grey colonies. From the reverse, it was pale with brownish black zones which was suggestive of *Pseudallescheria boydii*. Thus, based on macroscopic and microscopic findings, diagnosis of mycetoma by *Pseudallescheria boydii* was made.

Case 2

A 70 year old female presented with swelling over dorsum of right foot since 3 months, which was of 3×3cm and cystic in consistency. There was no history of swelling at other sites. Her routine haematological tests and blood sugars were within normal limits. On fine needle aspiration pus was

aspirated. Smears were made from aspirated material and stained with H& E and Wright's stain. The smear studied showed fungal elements with irregular septate hyphae, acute angle branching, intercalary and lateral conidia in an inflammatory and necrotic background. With the above cytological features, diagnosis of cutaneous mycetoma by *Pseudallescheria boydii* was made.

3. Discussion

Scedosporium infections are caused by opportunistic pathogens. The genus *Scedosporium* includes *S. apiospermum*/P. *Boydii* and *S. prolificans*. The anamorph (asexual state) of *Pseudallescheria boydii* is *Scedosporium apiospermum*.^[1] By convention, the name of the teleomorph, *P. boydii* has priority over the name of the anamorph, *S. apiospermum*.^[6,9]

Because scedosporium infections are caused by opportunistic pathogens, their clinical presentations are dependent on host immune status, which can cause life-threatening infections in immunosuppressed patients.^[10,11] The prevalence of infection is more among immunocompromised patients and it is rarely seen among immunocompetent individuals.^[11] However, neither of our patients were immunosuppressed. The disease in immunocompetent individuals ranges from asymptomatic pulmonary colonization to post-traumatic cutaneous infection and mycetoma.^[10] There is no difference in the virulence between the asexual stage and other stages.^[12] The fungus can cause mycetoma, and also pneumonitis, osteomyelitis, arthritis, meningitis, brain abscess, endocarditis, thyroid abscess and cutaneous and subcutaneous granuloma. In recent years, prevalence of *P. boydii*/ *S. apiospermum* infection is increasing due to widespread use of corticosteroids, immunosuppressants, antineoplastics and broad-spectrum antibiotics.^[12,3] Our patients presented with sub cutaneous swellings.

The most common mode of infection is by trauma, leading on to cutaneous and subcutaneous infection which can progress to mycetoma. In our case although there was no history of trauma, as both the patients were from rural area involved in agricultural activities minor latent trauma or thorn prick can be considered as mode of infection.^[5] Infection can also occur due to inhalation of fungal spores.^[13]

Pseudallescheria boydii/Scedosporium apiospermum infections are difficult to treat due to inherent resistance to available antifungal agents. In order to give appropriate treatment, pathogen must be diagnosed correctly.^[4] A diagnosis of *Scedosporium* infection can be based on cytology and histopathology with isolation of the fungus in culture, serology and several non-culture-based detection using molecular methods such as PCR and DNA sequencing.^[6] On Sabouraud glucose agar it produces floccose colonies that look different from the obverse (upper surface) and from the reverse. On microscopy, septate hyaline hyphae are seen with irregular, acute branching. The hyaline hyphae are approximately 5 µm in diameter but may appear as large swollen cells.^[1,6]

The clinical presentation and even the findings on cytopathology and/or histopathology of *Scedosporium* spp and *Aspergillus* spp., are very similar.^[6,9] Distinguishing *P. boydii* from *Aspergillus* is important for adequate treatment as *P. boydii* usually resistant to amphotericin but sensitive to imidazoles & triazoles in contrast to most of the fungi.^[2,4,14] However *Aspergillus* displays a regular, dichotomous branching pattern in cytology and histopathological sections, while *Scedosporium* present a more irregular branching.^[6] Aside from complete surgical resection (if possible), no optimal treatment option is known for infections caused by all *Scedosporium* species. Voriconazole, possibly in combination with other antifungal drugs (e.g., terbinafine and/or caspofungin), showed high efficacy in vitro and in vivo.^[8]

A systematic diagnostic approach is necessary to identify the etiologic agent in an accurate and timely manner. Starting the treatment as early as possible has a vital importance in immunocompromised patients as delay in diagnosis would most likely delay appropriate therapy, which can lead to potentially fatal consequences, especially in the immunocompromised patient.^[2]

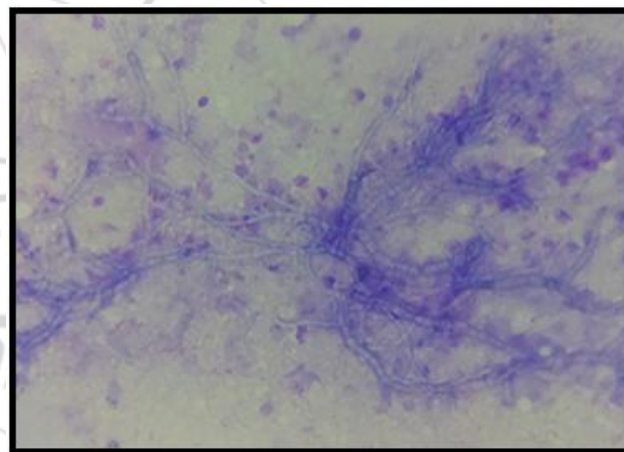


Figure 1: Wright's stain of smear from aspirated material (×40); Showing thin hyaline hyphae, irregular acute angle branching

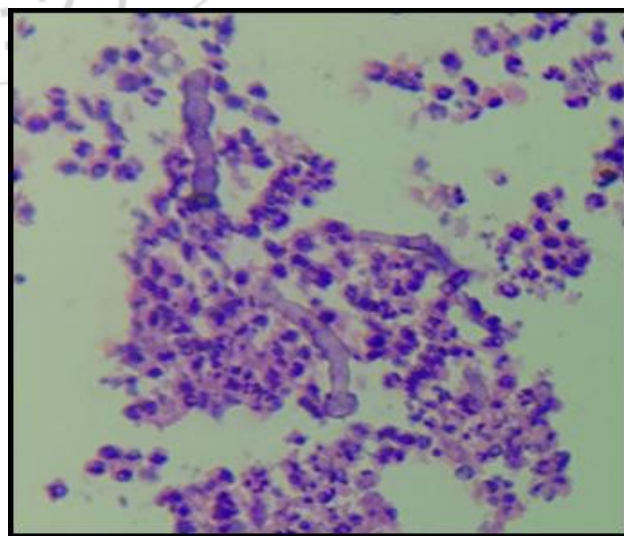


Figure 2: H&E stain (×40) of the excised specimen Showing Mixed inflammatory cell infiltrate,

Predominantly consisting of Neutrophils and Fungal colonies with septate hyphae and terminal Oval conidia

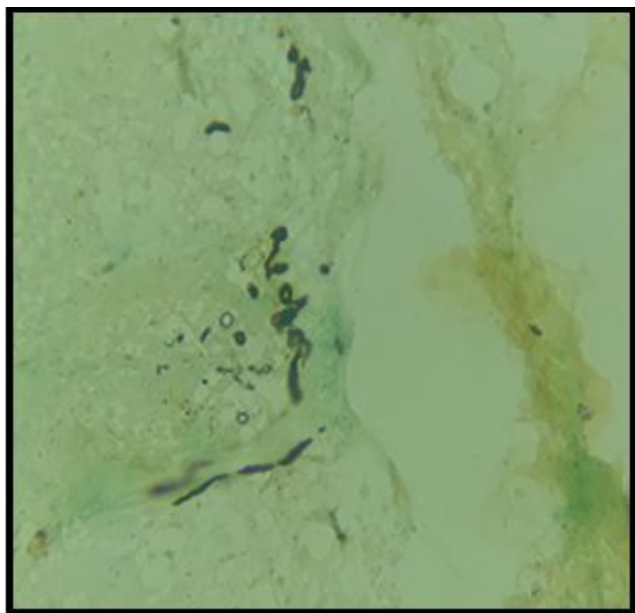


Figure 3: Gomori methanamine silver stain ($\times 40$); Highlighting the fungal colonies

4. Conclusion

It is important to include fungal infection among differential diagnosis in any cutaneous swelling. *Pseudallescheria boydii*/*Scedosporium apiospermum* infections are emerging pathogens among immunocompetent individuals. Careful mycological examination, Isolation, proper identification of the fungal isolates are important steps in the optimal treatment of these infections. Aggressive treatment of locally invasive disease is of value in preventing rapid and fatal dissemination with this organism.

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