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Sporotrichosis as an unusual case of osteomyelitis: A case report and review of the literature



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1. Introduction

Sporotrichosis is a fungal infection due to *Sporothrix schenckii* which is found worldwide in decaying vegetation, plants including rose bushes and sphagnum moss, and the soil [1]. After cutaneous inoculation, typically due to trauma associated with outdoor work, the fungus may cause cutaneous and/or lymphocutaneous infection. Rarely infection involves the joints and/or bones as a result of local inoculation or hematogenous spread. The infection is typically chronic and indolent in nature which may result in delayed diagnosis. We report a case of osteoarticular sporotrichosis, and provide a comprehensive review of the literature to describe the risk factors, diagnosis, and treatment of this unusual infection.

2. Case

A 39-year-old male presented on day 0 to our facility with left knee pain and swelling with progressive difficulty ambulating over the preceding 180 days. His history was significant for alcohol abuse and homelessness whereby he resided in a local park. There was no history of diabetes, immunosuppression, or intravenous drug use. Physical examination on presentation revealed a

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ABSTRACT

Sporotrichosis is an infection of worldwide distribution caused by the dimorphic fungus, *Sporothrix schenckii*. Acquisition typically occurs via cutaneous inoculation with development of a localized cutaneous and/or lymphocutaneous infection. We present a rare case of osteoarticular sporotrichosis in a 39-year-old man and review the literature noting only 20 published cases since 1980. Recommendations on the diagnosis and management of this unusual infection are provided.

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> moderate left knee effusion with limited passive and active range of motion to 15 degrees, and a skin abrasion on the overlying skin. The remainder of the examination was unremarkable, and there was no other skin lesions or lymphadenopathy. Laboratory evaluation showed a white cell count of 8200 cells/mm³, erythrocyte sedimentation rate (ESR) of 53 mm/h, and a C-reactive protein (CRP) of 28.5 mg/L. An HIV test and a drug screen were negative. Kidney and liver testing was within normal limits, and a hemoglobin A1c was 5.9%.

> The patient had presented previously to the Emergency Department (ED) at our facility on day – 140 complaining of left knee pain and an ultrasound was done which found a mild to moderate joint effusion. An arthrocentesis was performed which showed a synovial white cell count of 7470 cells/mm³ with 87% neutrophils. Laboratory evaluation during this visit showed a white blood cell count of 8700 cells/mm³, erythrocyte sedimentation rate (ESR) of 50 mm/h, and a C-reactive protein (CRP) of 15.2 mg/L. The patient was diagnosed with a reactive joint effusion and discharged home from ED with pain medications and followup with an orthopedic surgeon. Sporothrix schenckii was identified from the arthrocentesis culture on day -126. When this result returned, an attempt was made to contact patient to return to ED for reassessment and initiation of antifungal therapy, however due to patient's homeless status and lack of contact information, he could not be reached.

> During the current visit (day 0), the patient again presented with ongoing left knee pain. MRI of the left knee on day +2 demonstrated large complex joint effusions and bone marrow

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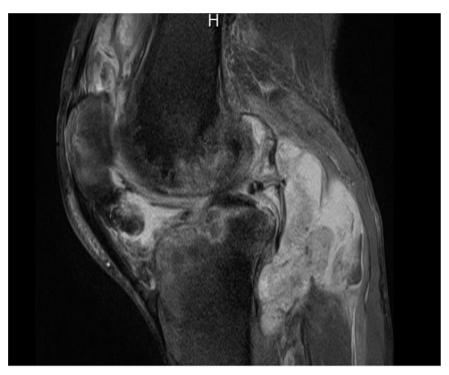


Fig. 1. MRI demonstrating large complex joint effusions and bone marrow edema within the femoral condyles and tibial plateaus.

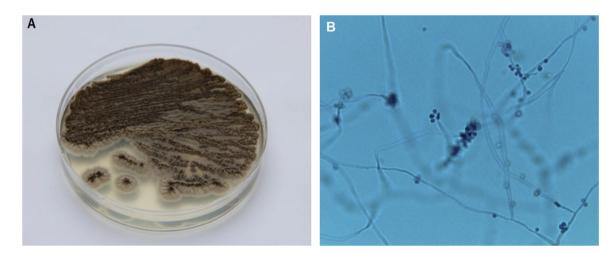


Fig. 2. (A) Growth of filamentous colonies on Sabouraud dextrose agar plate incubated at 30 °C. Colonies at first often appear white to creamy, but then turn brown to black after a few days of incubation; the figure represents growth after 21 days of incubation. Colonies are typically wrinkled in appearance and over time form heaping, mountain-like colonies. (B) Microscopic image of hyphae which are septate and approximately $1-2 \mu m$ in diameter. Conidia are oval-shaped and classically occur in a flower or bouquet-like arrangement (arrow). Colonies incubated at 30 °C; lactophenol cotton blue stain, $500 \times magnification$. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

edema within the femoral condyles and tibial plateaus consistent with osteomyelitis (Fig. 1). On day +3, the patient underwent surgical debridement and synovectomy. Bacterial and acid-fast bacilli (AFB) cultures collected during surgery were negative from all specimens. On day +14, growth on the fungal cultures (five specimens) showed *Sporothrix schenckii* (Fig. 2A and B). The identification of this fungus was based on growth of its mold form on a Sabouraud dextrose agar plate incubated at room temperature (30 °C) showing black-pigmented filamentous colonies, and microscopic slide examination showing lateral conidiophores with clusters of pyriform conidia appearing as flowers or bouquets. Identification was confirmed after transition to the yeast form after plating mycelia on rich culture media (e.g., brain heart infusion agar) at 37 °C. On day +3, the patient began treatment with

oral itraconazole 200 mg twice daily based on the culture from the previous ED visit that had grown *Sporothrix schenckii*. His knee function progressively improved, and he is receiving a planned 12-month antifungal treatment course with good clinical response.

3. Discussion

Sporotrichosis is a disease of worldwide distribution most commonly found in tropical and temperate regions. The first case was described in 1898 by Schenck who demonstrated the pathogenicity of this fungus [2]. The disease was subsequently named *S. schenckii*, with recent data showing that different genetic lineages exist in varying geographical regions [1]. Classically,

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