CASE REPORT

Sporotrichosis atypical presentation as a soft tissue tumour

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Abstract

Sporotrichosis is a mycosis caused by a saprophytic dimorphic fungus named Sporothrix schenckii. Infections occur following traumatic inoculation of fungus from plants and infected cat bites and scratches. We report a case of a farmer who presented with a solitary subcutaneous nodule initially diagnosed as a soft tissue tumour. A history of agricultural activity and feline contact should draw the clinician’s attention to sporotrichosis, as the diagnosis can be easily missed in atypical cases. The diagnosis, microbiology and management of the case are discussed.

Keywords: sporotrichosis, mycoses, Sporothrix schenckii

INTRODUCTION

Sporotrichosis is a form of chronic subcutaneous infection caused by Sporothrix schenckii. The disease was first diagnosed by Benjamin Schenck in 1896 following its discovery in a patient with refractory chronic subcutaneous abscess.1 Many cases are reported worldwide, predominantly in Central and South America, Mexico, Eastern Europe, India and Japan. The fungus is frequently found in soil, decaying vegetable debris and timbers, making agriculture-related recreational activities and forestry jobs as unique factors in acquiring the disease. The most famous epidemic was between 1941 and 1943 at Witwatersrand, South Africa involving thousands of gold-miners.1 Sporadic cases involving avid gardeners and nursery workers have been reported.2 Domestic cats are considered important carriers for sporotrichosis to humans.3 We describe an elderly farmer who presented with a solitary subcutaneous mass, which was initially thought to be a soft tissue tumour.

CASE REPORT

This patient was a 70-year-old Malay farmer, who was seen at the Orthopaedic Clinic, Universiti Kebangsaan Malaysia Medical Centre (UKMMC) with a one-month history of progressive painless nodule on the left leg. There was no history of trauma, fever, weight loss, or loss of appetite. Systemic review was otherwise unremarkable. He owned an oil palm plantation in Johor. Physical examination revealed a solitary mass measuring 6 x 4 x 3cm over the lateral aspect of the left leg, approximately 5 cm above the ankle. It was non-tender, firm and fixed to the surrounding tissue. There was no lymphocutaneous dissemination or regional lymphadenopathy.

Laboratory investigations showed mild neutrophilia and raised erythrocyte sedimentation rate (48mm/hour). A plain radiograph of the affected limb showed a soft tissue shadow. Ultrasound assessment revealed a heterogeneous echogenic mass with cystic changes. Based on these imaging findings, he was diagnosed to have a soft tissue tumour and aspiration biopsy was performed.

Pathology

A fine-needle aspiration cytology (FNAC) smear exhibited few polymorphonuclear neutrophils and abundant macrophages and multinucleated giant cells, suggestive of a chronic granulomatous inflammation. This FNAC result suggested the possibility of an infectious process. The nodule was excised for histopathological examination, mycobacterial and fungal culture.
Macroscopically, the mass measured 5 cm in diameter. It was encapsulated, multinodular and filled with yellow-brown jelly-like material. Histological sections showed granulomata and microabscesses. Grocott stain showed abundant “cigar-shaped bodies” (Figure 1A). Periodic acid-Schiff stain showed numerous oval yeast cells (Figure 1B). Culture of the nodule on Sabouraud dextrose agar at room temperature grew a white filamentous mould identified as *Sporothrix schenckii* after 3 days of incubation. The mould subsequently darkened as it matured (Figure 2A). Microscopical examination demonstrated thin, septate hyaline hyphae with slender conidiophores rising at right angles. Rosette-like clusters of conidia were seen at the tip of the conidiophores. Some conidia are arranged singly on the hyphae (Figure 2B). A dimorphic conversion test was done by subculturing the filamentous moulds on brain-heart infusion agar supplemented with blood and incubated at 37°C. Yeast colonies grew on day-7 of incubation. Microscopic examination demonstrated budding yeast cells.

We also assessed the patient’s risk factors for subcutaneous sporotrichosis. He did a lot...
of shearing in the palm-oil plantation. Besides, he also had regular contacts with eight domestic cats. However, he denied having any recent cat bite or scratch. Likewise, he could not recall any obvious trauma while at work. Given the fact that the wound healed uneventfully, he was not prescribed any anti-fungal agent upon discharge. The patient remained asymptomatic, without signs of recurrence or lymphadenopathy six months post-operatively. He still did the shearing, but wearing proper walking boots and attire to prevent further *Sporothrix schenckii* infection. He was also advised to have his cats examined and treated by the veterinarian, as necessary.

**DISCUSSION**

Sporotrichosis is classified into 4 clinical categories: (i) lymphocutaneous, (ii) fixed cutaneous, (iii) multifocal or disseminated, and (iv) extra-cutaneous. The most common clinical presentation is the lymphocutaneous variant, representing over 75% of all cases. This variant is characterized by the emergence of indurated papule, which progresses to nodule formation and ulceration. Further nodules appear in the lymphatic course contiguous to the initial lesion and finally produce cutaneous fistulae. In this particular patient, he had a fixed solitary lesion in subcutaneous tissue plane, with no lymphatic dissemination and regional lymphadenopathy. His condition had been overlooked initially due to the atypical presentation mimicking soft tissue tumour. On-line literature search did not find similar reported cases. The unexpected findings in cytology smear; coupled with raised ESR had shifted our clinical diagnosis from benign soft tissue tumour to an infection-related condition. Close liaison between the histopathologist, medical microbiologist and orthopaedic surgeon is vital in achieving correct aetiological diagnosis in a timely manner. As illustrated in this case, definitive diagnosis was accomplished by histopathological examination and mycology culture of the biopsied nodule. Culture remains as the most specific method in diagnosing sporotrichosis. This method is cheap, simple to perform and readily available in most clinical laboratories. Although it requires days to weeks for isolation, it allows genus-species identification that guides clinician in selecting the appropriate antifungal agent. *In-vitro* fungal dimorphism test and demonstration of characteristic conidia arrangement on microscopy finally confirmed the diagnosis. To date, there is no reliable serological test for the diagnosis of sporotrichosis. Several antigens have been identified; however, further studies are needed to elucidate the usefulness of these antigens.

Most cases of sporotrichosis require long-term itraconazole therapy. If left untreated, the lesions tend to become chronic. This patient was fortunate, as he recovered post-operatively without any antifungal treatment. *En-bloc* removal of the encapsulated mass in intact condition might be the explanation. Application of local heat through infrared radiation can be a useful adjuvant in this patient. Daily infrared heat at 43°C for two to three months ensures further elimination of residual fungal infection. *Sporothrix schenckii* infection does not confer complete immunity. Hence, patient’s education on sporotrichosis is essential, with special emphasis on the mode of transmission and preventive measures.

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**REFERENCES**