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CASE REPORT

REACTIVE ARTHRITIS ACCOMPANYING SPOROTRICHOSIS POST CAT SCRATCH: A MALAYSIAN CASE REPORT.

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Abstract

Sporotrichosis is a fungal infection caused by *Sporothrix schenckii*, a dimorphic fungus. Transmission of this infection is usually through contact with infected soil, decaying vegetation and plants as well as zoonotic inoculation such as cat scratches. We present an uncommon case of lymphocutaneous sporotrichosis accompanied by reactive arthritis in a 51-year-old woman due to a cat scratch in Malaysia.

Introduction

Sporothrix schenckii is a dimorphic fungus ubiquitous worldwide and is responsible for sporotrichosis.[1] Pathogenic acquisition of sporotrichosis in immunocompetent patients occurs mainly via cutaneous inoculation with subsequent development of a fixed cutaneous and/or lymphocutaneous infection. Osteo-articular involvement rarely occurs in sporotrichosis with only a handful of cases reported in the literature.[2] Almost all reported cases in the literature originated from Brazil or the United States of America. We report a case of reactive arthritis with sporotrichosis following a cat scratch in Malaysia.

Case report

A 51-year-old Malay lady from Ipoh, Malaysia presented with atraumatic progressive bilateral painful ankle swelling for over two weeks. Other joints were not affected and she denied respiratory, urinary or gastrointestinal disturbances. She has a past history of type 2 diabetes (well controlled on oral hypoglycaemic agents only).

Three weeks prior to this presentation, she suffered an abrasion over the anterior aspect of her right forearm following a cat scratch. She was then treated as bartonellosis with a course of oral ciprofloxacin without improvement. The inoculated site ulcerated and papulo-nodular lesions developed along the lymphatic track. Subsequently, these lesions were surgically resected and itraconazole 100 mg twice daily was commenced (empirically treating for lympho-cutaneous sprorotrichosis) with minimal skin improvement and subsequent development of articular symptoms.

On examination, there was evidence of bilateral ankle swelling most evident anterior to the medial and lateral malleoli as depicted. There were no overlying skin changes. There was evidence of diffuse ankle synovial thickening but no focal tenderness identified upon palpation. The ankle range of movement was mildly restricted particularly upon dorsiflexion. [Figure 1b]. Peripheral and axial joint examination were normal. There was no evidence of dactylitis. There were surgical scars over the right anterior forearm (inoculation site) and above the cubital fossa with interconnecting cutaneous tracking [Figure 1a]. A small subcutaneous nodule was palpable proximal to the track. These cutaneous lesions conform to a linear 'nodular lymphangiitic' pattern which is classically described with sporotrichosis. Otherwise, there were no other skin changes, nail abnormalities or palpable peripheral lymphadenopathies.

Laboratory evaluation demonstrated a leukocyte count of 7.4 x 109/L, haemoglobin of 10.3 g/dL, platelet count of 438 x 109/L, erythrocyte sedimentation rate of 40 mm in the first hour and C-reactive protein of 9.98 mg/L. Rheumatoid factor measurement was low positive at 33.2 IU/mL (normal range: < 15) and the anti-citrulinated cyclic peptide antibody was negative. Renal profile, liver function tests, serum calcium, phosphate and creatine kinase were normal. Blood cultures were negative for fungal and bacterial growth. Unfortunately, her previous tissue samples post skin debridement were not cultured. She declined further excision of the remaining nodule for aetiological confirmation. *Sporothrix* serology and molecular diagnostics were unavailable in our laboratory facility.

Acknowledging these limitations, in view of her cutaneous nodular lesions conforming to a classical 'sporotrichoid' configuration, she was commenced on itraconazole 200 mg twice daily, empirically treating her for reactive arthritis accompanying sporotrichosis. We re-assessed her at the end of a four week course of intensified itraconazole therapy.

Upon follow-up, the surgical scars had healed with hyper-pigmented scarring. The subcutaneous nodule and cutaneous tracking had completely resolved [Figure 2]. There was complete clinical resolution of synovitis in both ankle joints [Figure 2].

Given the good response to intensified itraconazole therapy, we have continued her therapy to complete a 12 months course, as per Infectious Diseases Society of America (IDSA) guidelines.[3]

Discussion

Osteo-articular involvement is the commonest manifestation of extra-cutaneous sporotrichosis.[4] Despite this, a systematic review of the English literature undertaken by Lederer *et al* discovered only 20 cases published between the years 1980 and 2015.[2] Although sporotrichosis has been reported in Malaysia,[5] to our knowledge, this is the first case report of reactive arthritis accompanying sporotrichosis from this country.

The diagnostic probability for sporotrichosis in this lady is high as she suffered a cat scratch with resultant 'nodular lymphagiitic' cutaneous lesions. Cat scratch is the commonest mode of inoculation of *S. schenckii* in Malaysia.[5] Subsequent extra-cutaneous symptoms may indicate infective dissemination. Empirical anti-fungal therapy should be commenced in these situations if fungal cultures could not be obtained to halt further disease progression and prevent chronicity.

Here, the diagnosis of reactive arthritis accompanying sporotrichosis was retrospectively supported by clinical improvement upon intervention with appropriate therapy. Differential diagnoses for 'nodular lymphangitis' include Nocardiosis, Leishmaniasis and *Mycobacterium marinum*. Tissue cultures would distinguish these infections. Where cultures are unavailable, these conditions may be distinguished by virtue of deduction as illustrated below [Table 1].

Despite fungal culture being the diagnostic gold standard, the rate of positive culture for *S. schenckii* displays a significant inter-study variability (32 to 81.2%).[6-7] Pre-treatment with antimicrobials, such as in our case may reduce the yield of fungal retrieval.

Other investigative surrogates such as the sporotrichin skin test and serological methods are sensitive but is non-specific for active disease.8 Molecular methods for detection of pathogenic *Sporothrix* species is well developed. [9] but is not widely available in Malaysia.

Osteoarticular sporotrichosis is recognized in immunocompetent patients as elucidated in Lederer's systematic review.[2] However, the pathogenesis remains obscure. A typical tissue histology of sporotrichosis display mixed tissue granulomatous and pyogenic reactions which suggest impairment of phagocytic function in clearing fungal antigen. We postulate the pathogenesis of osteoarticular sporotrichosis in immunocompetent patients to be specific impairment in phagocytic function (may be hereditary as in chronic granulomatous disease) or immunological cross reactivity akin to the id reaction in dermatophytosis. Identifying the specific pathogenesis requires expensive laboratory investigations which makes it less practical in a third world country.

Conclusion

Osteoarticular sporotrichosis is a relatively uncommon manifestation of *S. schenckii* tissue infection. We report a case of a 51-year-old lady with reactive arthritis accompanying sporotrichosis diagnosed based on clinical grounds. She had complete clinical resolution of cutaneous and articular disease within 4 weeks of twice daily itraconazole 200 mg therapy. The pathogenesis of dissemination of sporotrichosis among immunocompetent individuals is currently obscure, subject to further research and elucidation.

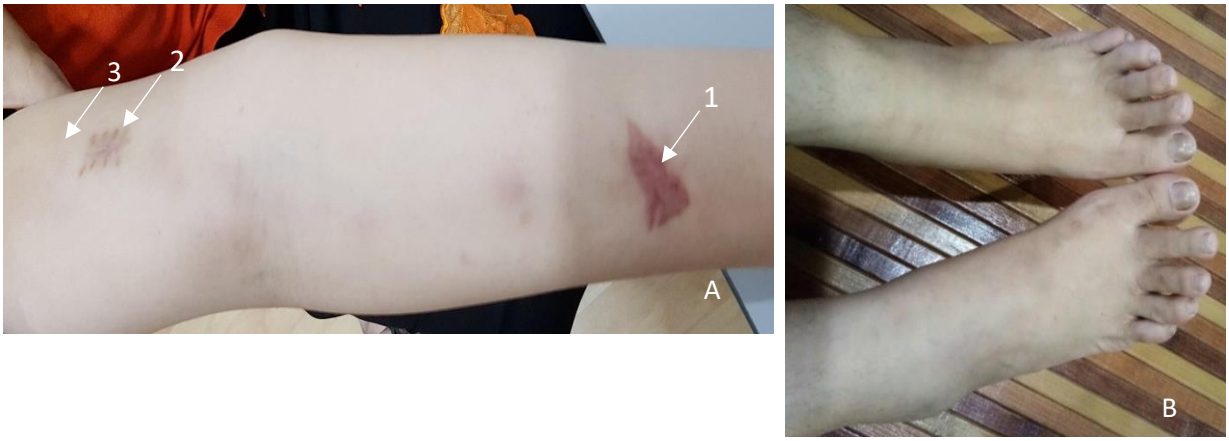

Figure 1. (A) 2 surgical scars post debridement at the inoculation site (arrow 1) and site of papulo-nodular lesions (arrow 2) with interconnecting tracking. A nodule is palpable proximal to the cubital fossa (arrow 3). (B) Bilateral ankle synovitis.

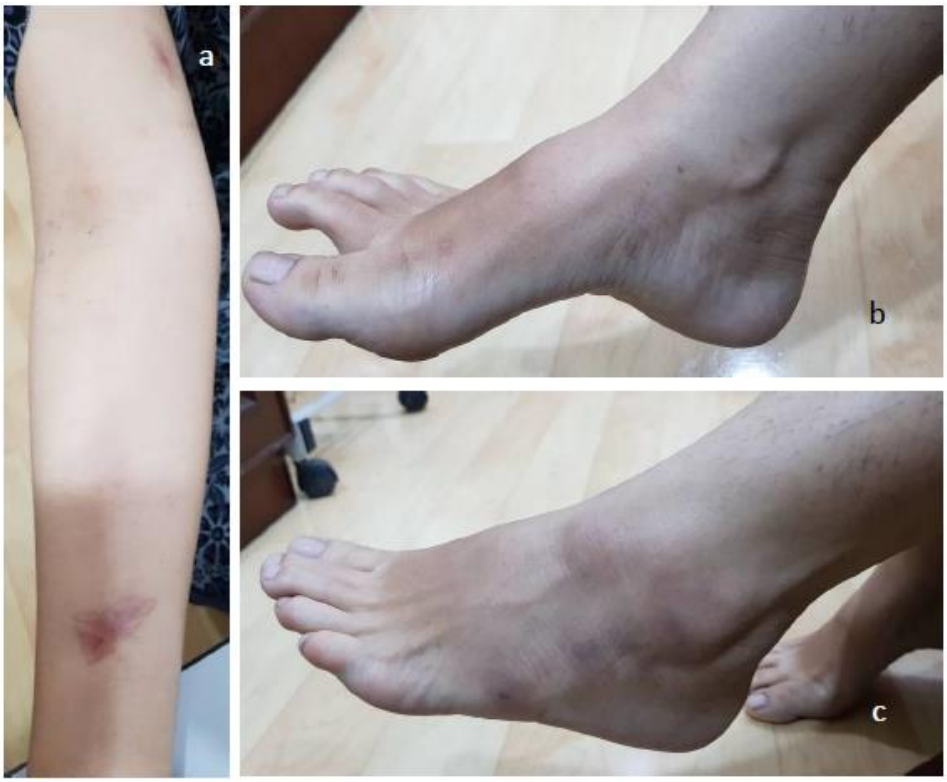

Figure 2. (a) Surgical scars in the maturation phase of secondary healing. There was resolution of tracking and all palpable nodules. Resolution of bilateral ankle swelling (b) Right ankle/foot (c) Left ankle/foot.

Table 1: Differential diagnosis for 'nodular lymphangitis' and their clinical features.

Condition	Clinical features	DGD	Transmission	Antibiotic response	Antifungal response
Sporotrichosis	Cutaneous or lymphocutaneous disease	No	Inoculation (soil, zoonotic, phyto-transmission)	No	Yes
Nocardiosis	Pulmonary or severe disseminated systemic disease	No	Inhalation or traumatic inoculation with nocardia containing soil	Yes	No
Leishmaniasis (cutaneous)	Mucocutaneous or lymphocutaneous disease	Yes	Zoonotic (bite of the female phlebotomine sandfly)	No	No
Mycobacterium marinum	Cutaneous or lymphocutaneous disease	No	Contact with infected water	Partial	No

DGD: *distinct geographical distribution*

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