

CASE REPORT

Bilateral Thumb Lymphocutaneous Sporotrichosis

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ABSTRACT

Sporotrichosis is a fungal infection caused by *Sporothrix*, a thermally dimorphic fungus that affects humans and animals. It is most commonly associated with cutaneous and subcutaneous tissue exposed to fungus-containing soil, moss, or organic material. Sporotrichosis is challenging to diagnose because it can mimic various other dermatological diseases. Thus, as demonstrated in this case, the importance of making a diagnosis based on history and a thorough physical examination, as misdiagnosis can delay proper treatment. A 53-year-old Malay woman presented with bilateral thumb swelling that had been treated with two courses of antibiotics but had failed, resulting in disease progression. Clinical suspicion of lymphocutaneous sporotrichosis was made based on history, physical examination, and investigations finding. The patient was started on an empirical course of Itraconazole 200mg twice daily, and the lesions responded well.

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INTRODUCTION

Sporotrichosis is a subacute to chronic mycosis infection caused by the dimorphic fungus of the genus *Sporothrix*. Even though most cases are reported in the tropics and subtropics, the actual prevalence in Malaysia is unknown. However, it was reported that 73.7% of sporotrichosis infections in Southeast Asia occurred in Malaysia (1).

Lymphocutaneous sporotrichosis is the most common form seen in clinical practice. Infection can occur as a result of trauma to the skin or mucous membrane from activities such as landscaping, rose gardening, and other activities. Hence the disease was also named "rose gardener's disease." There have also been reported cases of zoonotic transmission involving cats. Most reported cases showed a primary lesion on the extremities that can progress along the regional lymphatic channels upwards or downwards, depending on the anatomical site, after a few weeks (2).

CASE REPORT

Madam N, a 53-year-old lady with no known medical illness, presented with painless bilateral thumb nodules for three weeks. The nodules started with a small pea size which progressively increased. Initially, it appeared at

her right thumb's pulp but progressed to the left thumb. It was associated with redness, and the nodules became ulcerated after that. She also notices the presence of multiple small painless nodules arranged in a line at both of her forearms which developed within two weeks of illness.

She has a history of being scratched by a sick stray cat on her right hand before the lesion developed. Since she is working as a cook, the lesion has affected her work performance. She received two courses of oral antibiotics, but the lesion was not improving. Otherwise, there was no itchiness at the lesion site. She denied any fever, shortness of breath or hemoptysis.

On examination, she appeared comfortable. Her blood pressure was 120/84mmHg, her pulse rate was 78 beats per minute, and her temperature was 37°C. There were non-purulent ulcerated nodules with raised edges and surrounding erythema on the pulp of both thumbs (Figure 1). Multiple non-tender nodules were observed arranged linearly over her bilateral forearm. The ulcerated nodule hampered the ranges of motion of the right thumb's interphalangeal joint (Figure 2). Other systemic examinations were unremarkable. Tissue culture and sensitivity for bacteria, including mycobacterium and fungus, revealed no growth on day five. However, based on the history and physical examination, she was clinically diagnosed with lymphocutaneous sporotrichosis. She was started with an empirical course of oral Itraconazole 200mg twice daily for three months, and the lesions responded well with the resolution of ulcers.

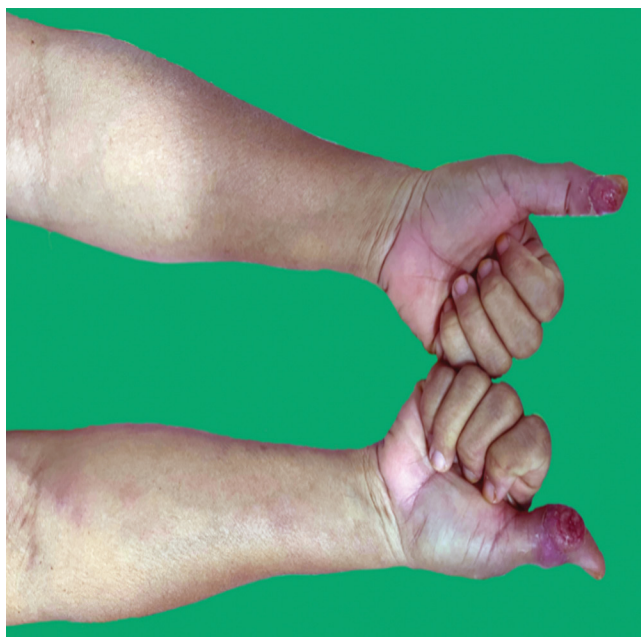


Figure 1: Lymphocutaneous sporotrichosis. Nodules appear along the lymphatics proximal to the initial inoculation injury site

DISCUSSION

Sporotrichosis is a mycotic infection caused by fungi of the *Sporothrix* genus that has a worldwide distribution, particularly in regions with tropical or subtropical climates. The classical transmission route in humans is from traumatic inoculation of contaminated material such as soil, decaying wood, and hay into the skin. Direct transmission to humans can also occur from animals such as cats, dogs, and horses through biting and scratching. In Malaysia, cats are the most popular domestic pets, which explains the aetiology because most of our population touches pets without proper protection (3).

There are several types of sporotrichosis, including lymphocutaneous, pulmonary, and disseminated sporotrichosis. Lymphocutaneous sporotrichosis is the most common clinical subtype, accounting for 70-80% of cases. The vast majority of cases reported had a primary lesion on one of their extremities (4). The disease can spread to various organs and systems in individuals with weakened immune systems. Following inoculation, the incubation period may range from a few days to a few months. Initially, a papular-nodular lesion appears at the infected site, progressively increasing in size and may ulcerate, as illustrated in this case. A string of similar nodules may form along the proximal lymphatics channels without causing systemic symptoms (4).

Clinical suspicions are essential for early diagnosis, as cutaneous lesions need to be differentiated from other differential diagnoses such as blastomycosis, cutaneous leishmaniasis, and atypical mycobacterial infection. The presence of fever, cough, night sweats, and verrucous skin lesions with irregular borders characterize the



Figure 2: The ulcerated nodule of the right thumb's interphalangeal joint.

clinical features of blastomycosis, which were absent in this patient. Another possible differential could be cutaneous leishmaniasis, which manifested as a painless, non-pruritus erythematous papule that progressed to a plaque or ulcer. However, this infection is prevalent among people with poverty and malnutrition. Atypical mycobacterial infection caused by trauma, surgical procedures, or indwelling medical equipment may mimic sporotrichosis. Nevertheless, it is most commonly associated with digit erythema and granuloma formation, although it can also progress to nodular lymphangitis of the hands and forearms (3).

The gold standard for diagnosing sporotrichosis is the isolation and the identification of the *Sporothrix* species through tissue culture from clinical samples such as skin lesions, as in this case. However, a high index of suspicion is required for a clinician to consider the diagnosis before sending samples for fungal culture. Moreover, adequate fungal culture may take up to two to four weeks before the result can be reported as negative (2). As illustrated in this case, no organism growth was isolated after day five of the culture. However, this doesn't mean the treatment should be delayed since the presenting history and clinical features support the provisional diagnosis of lymphocutaneous sporotrichosis.

Since spontaneous resolution is extremely rare, most patients will require treatment. Treatment can be initiated if there is a high clinical suspicion of sporotrichosis. The treatment for sporotrichosis is determined by the clinical form of the disease, the host's immunological status, and the *Sporothrix* species. The low cost, ease of administration, safety profile, and site of infection must all be considered when deciding on a therapy. Itraconazole is recommended as the first-line drug of

choice due to its effectiveness, safety, and convenience. It is highly effective for localized subtypes of the lesion. According to Malaysian national antimicrobial guidelines, oral antifungal agents derived from the azoles group such as Itraconazole should be the first-line drug of choice and administered at 200mg twice daily until all the lesions have resolved (5). This typically required a duration of three up to six months. Oral terbinafine or fluconazole can be considered for patients who cannot tolerate Itraconazole (4).

Based on this case, a history of being scratched by a sick cat and the pattern of skin manifestation make lymphocutaneous sporotrichosis the most likely diagnosis. She received oral Itraconazole 200 mg twice daily and was treated for three months, during which the lesion was then completely resolved. The oral antifungal duration can be extended up to one year if the infection spreads to the lungs or becomes disseminated (2).

Clinicians should consider lymphocutaneous sporotrichosis as one of the most likely diagnoses if patients with a history of environmental exposure to the causative agents present with papular-nodular painless skin lesions that progress to non-healing ulcers. The appearance of the papular-nodular lesion following the lymphatic channels further supports the clinical diagnosis. This infection can lead to cellulitis, unhealed scars, and sepsis if left untreated. As a preventive measure to avoid recurrence, we advise patients to be careful with stray and sick cats.

CONCLUSION

In conclusion, comprehensive history, proper physical

examination and a high index of suspicion of an experienced physician are important. Correct diagnosis is vital to ensure proper patient treatment and to prevent further complications or suffering for the patient.

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