Case Series

Sporotrichosis from a tertiary care hospital in the Eastern State of India

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ABSTRACT

Sporotrichosis is a form of subcutaneous or systemic fungal infection caused by *Sporothrix schenkii*. This is a thermophilic, dimorphic fungus that occurs worldwide in tropical and temperate areas. We report a case series of sporotrichosis in three patients from a tertiary care center in Assam. The source of infection could be identified in each patient. The patients were successfully treated with oral and topical antifungal agents, which included capsule Itraconazole and supersaturated potassium iodide therapy.

Key words: Fungus, Itraconazole, Saturated solution of potassium iodide, Sporotrichosis

Sporotrichosis is an acute or chronic subcutaneous mycosis caused by the thermophilic, dimorphic saprophytic fungus *Sporothrix schenkii*. The distribution of fungus is worldwide, with focal areas of hyperendemicity except for latitudes 50° further north or south of the equator [1]. It is particularly common in tropical and subtropical areas and temperate zones with warm and humid climates. The most frequent occurrence of the disease is seen in countries like India (along the Sub-Himalayan region), China, Japan, and Central and South America (Mexico, Brazil, Colombia, and Peru). The prevalence in the sub-Himalayan region of India has been reported to be around 12.9% [2-7].

CASE SERIES

Case 1

A 36-year-old Indian male from the Tezpur district of Assam, a soldier by profession, presented with multiple asymptomatic swellings over both arms for months. The lesion started as a small papule, which over a period of time evolved into tender nodules distributed along the lymphatic supply of the upper arm. He gave a positive history of trauma with thorns while he was actively involved in jungle warfare and training activities. There was no history of preexisting cutaneous or systemic illness in the patient or his family members.

On clinical examination, he was found to be an average-built individual, weighing 65 kg with a body mass index (BMI) of 26.7. All systems were normal, and he had bilateral axillary lymphadenopathy with no lymphadenitis. He had multiple well circumscribed and dome-shaped tender nodules over the flexor and extensor aspects of both arms with ulcerated and crusted surfaces (Fig. 1). Differential diagnoses of subcutaneous mycoses, leishmaniasis, Hansen's disease, sarcoidosis, atypical mycobacterial infection, and nocardiosis were taken into consideration.

His chest X-ray and Mantoux test were normal. A full blood count, urea and electrolytes, blood sugar, and liver function tests were normal. The human immunodeficiency virus (HIV) was non-reactive. A smear from the lesion was negative for Leishmania donavani (LD) bodies on staining with Geimsa. A biopsy sample from the lesion was taken and sent for fungal and mycobacterial culture in normal saline. Another biopsy specimen was sent for histopathological examination in formalin. Histopathology showed hyperkeratosis with crusting in the epidermis. The dermis showed a mixed inflammatory infiltrate with neutrophil microabscesses, granulomas, and giant cells. A small, non-branching ball of fungal hyphae as well as an asteroid body located in a microabscess were also identified (Fig. 2). Special fungal stains could not be done in all three cases because of their unavailability in the resource-limited setting. Biopsy material cultured on Lowenstein-Jensen medium did not show any growth after 6–8 weeks of incubation; however, creamy white mold-like growth appeared on Sabouraud dextrose agar (SDA) with chloramphenicol incubated at 30°C after 7 days of incubation. After prolonged incubation of 14 days, the creamy white mold turned black with a wrinkly surface (Fig. 3a). The patient was started on a capsule of itraconazole 100 mg twice daily for a period of three months. The nodules healed well with atrophy and pigmentation.

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Case 2

A 40-year-old Indian male from rural Assam, a gardener by profession, presented with multiple asymptomatic ulcerations and a few pus-filled swellings over the inner aspect of the right leg for 5 months. The lesions started as a small papule, which over a period of time grew slowly to become tender nodules and a few pus-filled plaques distributed along the medial side of the right leg. He gave a positive history of trauma with thorns while gardening. The history of pre-existing dermatological or systemic illness in the patient as well as his family members was negative. He consulted a surgeon, who advised him to get regular dressings. The wound did not heal after taking antibiotics and regular dressings, so he finally reported it to the skin department of the hospital.

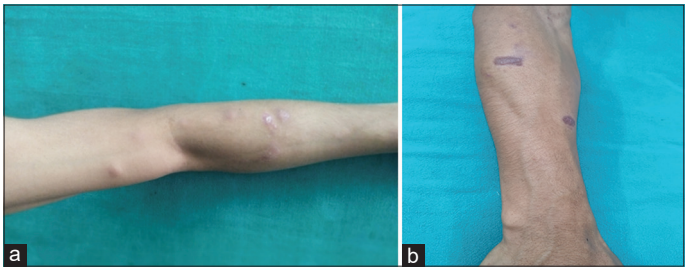

Figure 1: (a) Multiple well circumscribed, dome shaped nodules over flexor and extensor aspect of both arms with ulcerated and crusted surface along the sporotrichoid pattern before treatment; (b) atrophic hyperpigmented plaques post treatment (case 1)

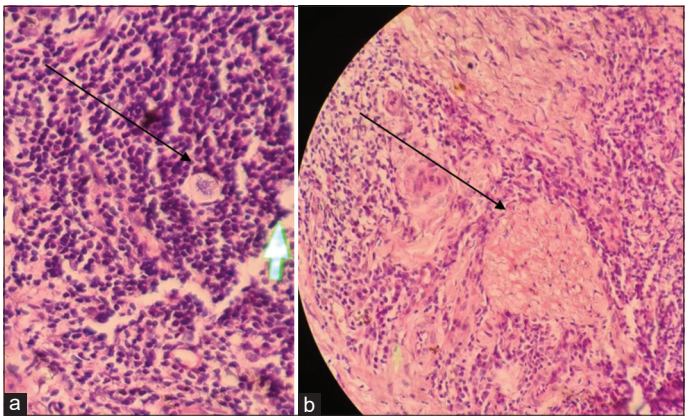

Figure 2: Histopathology showing (a) an asteroid body located in a microabscess; (b) mixed inflammatory infiltrate with neutrophils microabscesses, granulomas and giant cell. A small non-branching ball of fungal hyphae located in a microabscess was also identified (case 1)

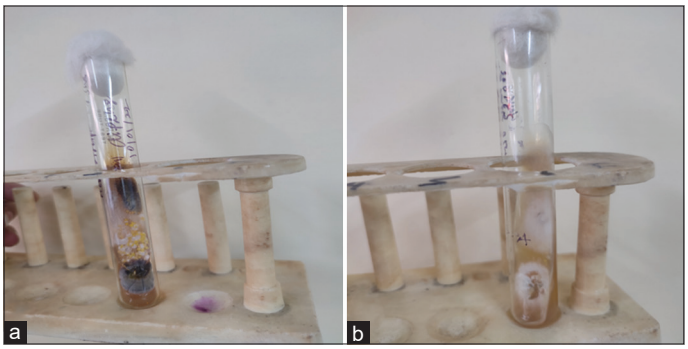

Figure 3: (a) Creamy white mould which turned black wrinkled in appearance on prolonged incubation on Sabouraud dextrose agar (SDA) with chloramphenicol incubated at 30°C after 14 days of incubation (case 1); (b) Biopsy material cultured on SDA with chloramphenicol incubated at 30°C showed positive growth of fungus in form of creamy white colonies after 12 days of incubation (case 2).

On clinical examination, he was found to be a lean-built individual, weighing 45 kg and having a BMI of 23.4. All systems were normal, and he had no lymphadenopathy or lymphadenitis. Laboratory tests and chest radiography were normal, and serology for HIV was negative. He had multiple well-circumscribed, dome-shaped tender nodules and pus-filled plaques over the medial aspect of the right leg with an ulcerated and crusted surface (Fig. 4a). Differential diagnoses of subcutaneous mycoses like mycetoma foot and chromoblastomycosis, leishmaniasis, Hansen's disease, sarcoidosis, atypical mycobacterial infection, and nocardiosis were kept in mind.

The smear from the lesion was negative for LD bodies on staining with Giemsa. A biopsy sample from the lesion was taken and sent for fungal mycobacterial culture and histopathology. Histopathology showed hyperkeratosis with crusting and ulceration in the epidermis. The dermis showed a mixed inflammatory infiltrate with neutrophils, microabscesses, and granulomas. Biopsy material cultured on SDA with chloramphenicol and incubated at 30°C showed positive growth of the fungus after 12 days of incubation (Fig. 3b). Lactophenol cotton blue mount of the culture showed thin, hyaline, septate hyphae arranged in a cigar-like pattern directly arising from the hyphae (Fig. 4b). The patient was started on capsule itraconazole 100 mg twice daily. The patient is presently in mid-way treatment and is responding fairly well.

Case 3

A 32-year-old male patient from Sonitpur, Assam, a gardener by profession, came to the dermatology outpatient department with a few nodules on the left side of his face. He gave a positive history of splinter injuries to his face while cutting grass in the garden 2 months ago. A red nodule developed within 3–4 weeks after the injury and was non-ulcerated and non-scaly. They were not associated with itching, and gradually, similar nodules appeared over the nearby area on the face (Fig. 5a). There was no history of contact with pets, chronic dermatoses, or any other ailment in

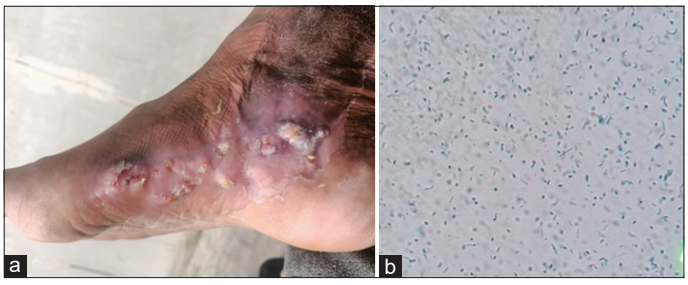

Figure 4: (a) Multiple well circumscribed, dome shaped tender nodules and pus filled plaques over medial an aspect of the right leg with ulcerated and crusted surface; (b) a wet preparation of the colony showed fungal hyphae and spores with typical cigar shaped appearance (case 2)

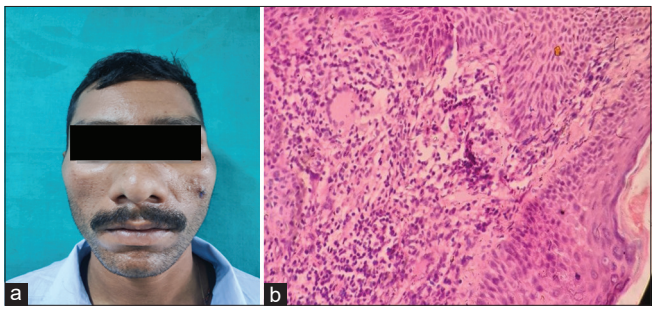

Figure 5: (a) Well-defined ulcerative nodules on the left side of the face. (b) Histopathology from the lesion showed hyperkeratosis, and acanthosis in epidermis. The dermis showed a mixed inflammatory infiltrate with foreign body giant cells, granulomas and microabscesses (case 3).

myself or family members. He was an average-built individual with a BMI of 25. He had consulted a local doctor, who prescribed a tablet of augmentin (1 g) twice daily for 7–10 days with no relief. He was referred to the skin department and evaluated with differentials of granuloma faciale, sarcoidosis, lupus vulgaris, Hansen's disease, leishmaniasis, subcutaneous mycosis, Jessner's lymphocytic infiltrate, and lymphocytoma cutis in mind.

An incisional biopsy was taken from the lesion, which showed hyperkeratosis and acanthosis. The dermis showed a mixed inflammatory infiltrate with foreign body giant cells, granulomas, and microabscesses (Fig. 5b). However, budding yeasts as well as an asteroid body could not be located in a microabscess. Biopsy material was cultured on SDA with chloramphenicol and incubated at 30°C. Culture growth appeared after 7 days, which was cream-white mold. The patient was diagnosed with a case of fixed cutaneous sporotrichosis and was started on five drops of super-saturated potassium iodide solution three times a day, mixed with one cup of milk. The dose was increased daily by one drop three times a day, gradually until 30 drops three times daily were reached. The patient tolerated the saturated solution of potassium iodide (SSKI) quite well, with minor side effects of nausea and the metallic taste of iodine. Thyroid function tests were monitored. The patient was discharged as the lesion started resolving. However, he failed to follow up after treatment.

DISCUSSION

Sporotrichosis infection occurs from subcutaneous inoculation of the spores of fungus from infected wood splinters, haystalks, barbs, splinters, soil, or thorns. The most common professions susceptible to exposure are farmers, gardeners, florists, foresters, nursery workers, horticulturists, soldiers, and mine workers [8]. The infection is common in the age group of 20–50 years, which is the most active year of exposure. The incubation period varies from a few weeks to months, with the average incubation period being 3 weeks [8]. Sporothrix infection can be broadly classified into cutaneous, extracutaneous, or systemic forms. It may manifest as a lymphocutaneous, fixed-cutaneous, disseminated, or systemic infection, or in extracutaneous forms like nasal and ocular. The varied clinical spectra of the disease depend upon the size and depth of the inoculum, the virulence of the fungus, the resistance mechanisms of the host, and the mode of inoculation. The fixed cutaneous form of sporotrichosis occurs as a well-circumscribed lesion at the site of inoculation; in the lymphocutaneous variety, multiple lesions are distributed along the lymphatic distribution. In our first patient, the lesions began as a small papule 4 weeks following the thorn injury. The lesions developed into well-defined tender nodules with superficial ulceration with or without satellite adenopathy. Similar lesions appeared along the lymphatic channel in a typical string-like "sporotrichoid" pattern of distribution [8,9].

Extracutaneous or disseminated sporotrichosis affects internal organs like bones, lungs, or the brain. It is seen in the setting of immunocompromised status in patients with cancer, chemotherapy, diabetes mellitus, organ transplantation, alcoholism, or prolonged corticosteroid therapy. The infection results from hematogenous spread in the body from the primary inoculation site or from direct inhalation of conidia and spores [8,9]. The localized and cutaneous-lymphatic forms are the most commonly reported presentations [10].

Areas exposed to trauma, such as the extremities and the face, are more susceptible to developing the disease. It is seen most commonly in males due to their higher chances of exposure to the external environment. In adults hands, the forearms and arms are the most common locations, while in children, the face is affected first [11,12]. The comparative study of all three cases of sporotrichosis is given in Table 1.

All patients with sporotrichosis invariably require treatment, as the spontaneous resolution of lesions is not reported. A super-SSKI and oral anti-fungal drugs remain the first line of treatment in a resource-limited setting [13]. The exact mechanism of action of SSKI is not clear. However, it is postulated that it gets converted to free iodine by the myeloperoxidase enzyme, which then exerts its effect by inhibiting granuloma formation. SSKI itself is not known to have any fungicidal or static effects. Side effects include metallic taste, gastrointestinal upset, hypo- or hyperthyroidism, flu-like symptoms, acneiform or papulopustular eruptions, pustular psoriasis, iododerma, irritability, and lesional pain and inflammation. It is contraindicated in pregnancy and lactation due to the risk of hypothyroidism or thyromegaly in the neonate [14,15]. Our first patient was managed with capsule itraconazole 100 mg twice daily for a period of three months with no major side effects. The lesions resolved with post-inflammatory hyperpigmentation and atrophy in some of the lesions. Liver function tests were monitored monthly. The patient achieved a complete mycological cure in a period of three months. There has been no recurrence so far. Itraconazole is an azole oral antifungal drug that acts on the fungal enzyme cytochrome P450 lanosterol 14α-demethylase and inhibits cell wall synthesis. This drug has a very high efficacy rate of 90–100% and a MIC of 0.1–1 mg/L. It is easily available with negligibly low rates of recurrence and has replaced amphotericin and SSRI for the treatment of cutaneous and systemic sporotrichosis [16]. Terbinafine is also effective for cutaneous sporotrichosis at a daily dose of 250–500 mg [17]. The role of newer antifungals like posaconazole or ravuconazole remains understudied in patients with sporotrichosis [18].

Table 1: Comparative study of parameters in three cases of sporotrichosis

Parameters	Case 1	Case 2	Case 3
Age	36	40	32
Sex	Male	Male	Male
Site of presentation	Upper limb	Lowe limb	Face
Profession	Soldier	Gardner	Gardner
BMI	26.7	23.4	25
Treatment	Itraconazole 100 mg twice for 3 months	SSKI dissolved in milk	Itraconazole 100 mg twice daily ongoing

SSKI: Saturated solution of potassium iodide

Voriconazole has limited *in vitro* activity against the sporothrix species complex, and echinocandins have not shown any activity against *S. schenckii* [19,20].

CONCLUSION

The rationale of this article is to report cases of sporotrichosis from a known highly endemic zone. Often, these are presented to surgeons, who try to treat them with antibiotics and surgical debridement, thereby complicating the situation. Clinicians should have a high degree of suspicion when they come across a case with a typical string-like pattern of distribution of noduloulcerative lesions. Lesions of sporotrichosis resemble those of cutaneous leishmaniasis, mycobacterial, atypical mycobacterial infection, nocardiosis, Hansen's disease, chromoblastomycosis, and blastomycosis. Prompt and correct diagnosis of sporotrichosis will contribute to timely and effective treatment, reduction of morbidity, and chronicity of the disease.

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