

Cutaneous Forms of Sporotrichosis: Report of Two Cases

¹Dr. Sonali T. Dhote, Pathology Resident

²Dr. Anjali Patrikar, Associate Professor

³Dr. Archana Joshi, Associate Professor

⁴Dr.Sadhana Mahore, Professor and HOD

^{1,2,3,4}Department of Pathology, NKP salve Institute of Medical Sciences and Research Centre, Digdoh Hills, Nagpur
440019, Maharashtra

Correspondence Author: Dr. Sonali T. Dhote, Pathology Resident, Department of pathology, NKP salve Institute of Medical sciences and Research centre, Digdoh Hills, Nagpur 440019, Maharashtra

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Sporotrichosis is most common subcutaneous mycosis. It is caused by the fungus *Sporothrix schenckii* which is dimorphic fungus, the infection is usually acquired by traumatic inoculation. We described two cases of cutaneous forms of sporotrichosis with no identifiable source of infection. This dimorphic fungus occurs in both temperate and tropical areas. It occurs sporadically in many countries.

Keywords: sporothrix schenckii, cutaneous form of sporotrichosis.

Introduction

Sporotrichosis, known as rose gardener's disease^[1], is a subacute or chronic mycosis caused in most cases by traumatic inoculation of the saprophytic dimorphic fungus *sporothrix schenckii*^[2]. Only one species of sporothrix was classically described however phenetic and genetic studies have identified additional sporothrix species. Knowledge of the geographic distribution, virulence, clinical presentation, and response to antifungal therapy of this newly identified species is an area of active research^[1]

The characteristic infection involves suppurating subcutaneous nodules that progress proximally along lymphatic channel (lymphocutaneous sporotrichosis). Primary pulmonary infection (pulmonary sporotrichosis) is rare, as is direct inoculation into tendon, bursae, or joints. Osteoarticular sporotrichosis is caused by direct inoculation or hematogenous seeding. In rare cases, disseminated *S.schenckii* infection (disseminated sporotrichosis) occurs, characterized by disseminated cutaneous lesion and involvement of multiple visceral organ; this occurs most commonly in person with AIDS.^[1]

The traditional form of transmission, however, is traumatic inoculation of the fungus into skin, through contact with contaminated soil, plants or organic substrate.^[2] The most common presentation is cutaneous and the disease has been classified into three different clinical forms: cutaneous-lymphatic, fixed and disseminated^[2]

In adult the most common location is the upper limb, while in children it tends to occur more commonly on the face^[2]

Case reports

Case1-

65 yrs old male patient came with complaints of lesion over bilateral lower limb since 1 yr. It started as small papule which grew slowly with history of burning sensation. There is no history of trauma prior to onset of this lesion.

On clinical examination he was thin built. All system were normal and he had no lymphadenopathy. He had multiple well circumscribed hyperkeratotic dome shaped plaque with verrucous and crusted surface present over bilateral ankles with palms and soles (figure 1,2)



Figure 1: Hyperkeratotic dome shaped plaque over bilateral ankles



Figure 2: Hyperkeratotic plaque over left palm

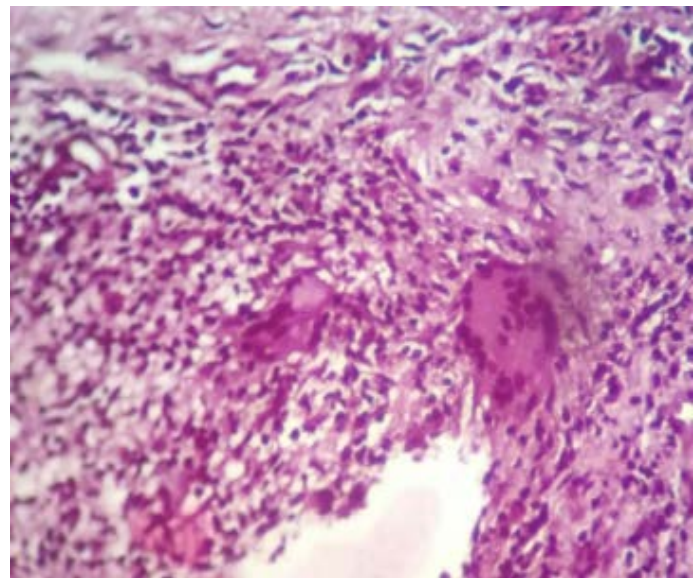


Figure 3 : histopathology showing granuloma formation with round spores (H &E,10x)

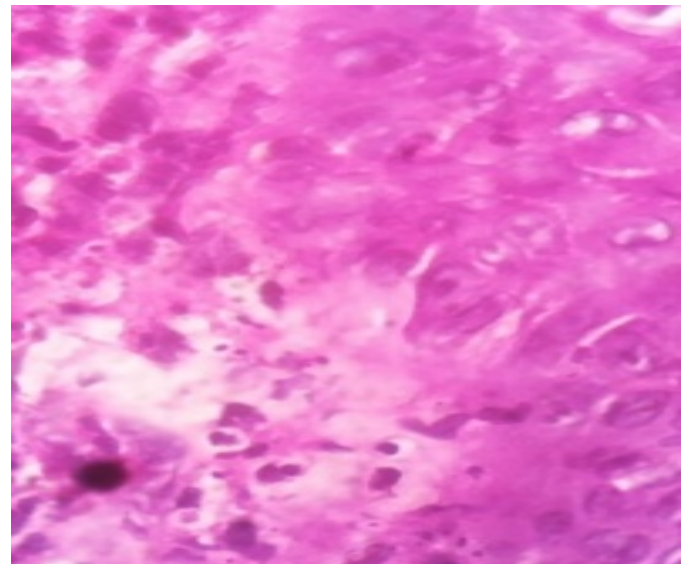


Figure 4 :Histopathology showing Many densely stained round spores and budding forms(H&E,40x)

His chest x ray was normal. A complete blood count, urea and electrolyte and liver function were normal.

Skin biopsy was sent for histopathological examination. Histopathological report – Sections revealed skin composed of squamous epithelium with hyperkeratosis, parakeratosis and elongated rete ridges along with crusting. Focal areas show microabscess formation. Subepithelial tissue shows acute suppurative inflammation showing plenty of neutrophils and few eosinophils and lymphocytic infiltration. Many densely stained round

spores are seen with granuloma formation. Deeper tissue does not show spores. (fig 3,4)

PAS stain shows few fungal spores.

A diagnosis of Sporotrichosis (cutaneous form) was given.

Case2

40 yrs old female came with complaints of swelling over foot since 2 yrs. There was history of trauma by cow leg. Ulcerated lesion with multiple discharging sinuses present over dorsum of right foot. No history of burning sensation over the lesion.

On clinical examination she was moderately built. All systems were normal and she had no lymphadenopathy. She had single well-circumscribed polypoidal fungating mass with ulceration and multiple discharging sinuses. (fig 5)

Suspected clinical diagnosis was actinomycosis right foot.



Figure A:



Figure B:

Figure 5 : Figure A and B : Fungating mass with ulceration and multiple discharging sinuses

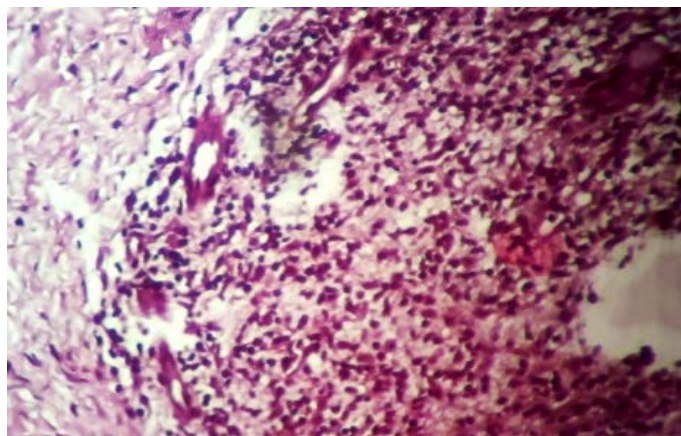


Figure 6: Histopathology showed dense chronic inflammatory infiltrate with neutrophils and fungal spores (H&E, 40x)

Her chest x-ray was normal. A complete blood count, urea and electrolyte and liver function were normal.

A biopsy was sent for histopathological examination. Histopathological report – sections revealed stratified squamous epithelium with acanthosis. The dermis showed perivascular focal dense collection of chronic inflammatory infiltrate, composed of lymphocytes, plasma cells and eosinophils. Deeper dermis showed granuloma formation composed of epithelioid cells, multinucleated giant cells and peripheral mantle of lymphocytes. Many congested blood vessels are also seen. Few yeast forms of fungus were seen. (fig 6,7)

PAS stain was negative for actinomycotic colonies or filamentous fungi.

A diagnosis of sporotrichosis (cutaneous form) was given.

Discussion

Sporotrichosis can be diagnosed through a correlation of clinical, epidemiological, and laboratory data. Laboratory analysis for the determination of sporotrichosis includes direct examination of specimens such as tissue biopsy specimens or pus from lesions. In case of disseminated infections, other specimens, such as sputum, urine, blood, and cerebrospinal and synovial fluids, can be analyzed, depending on the affected organs.^[3]

Sporotrichosis occurs more frequently in workers who deal with contaminated soil and plants. The groups at most risk are gardeners, florists, horticulture and farmers.^[2] Zoonotic transmission has been described in isolated cases or in small outbreaks. The lesions are usually restricted to the skin, subcutaneous cellular tissue, and adjacent lymphatic vessels. Eventually, this fungus can disseminate to other organs, and alternatively, on rare occasions, inhalation of conidia may lead to a systemic disease. Several factors, such as inoculum load, immune status of the host, virulence of the inoculated strain, and depth of traumatic inoculation, influence the different clinical forms of sporotrichosis. The gold standard for sporotrichosis detection is culture; however, serological, histopathological, and molecular approaches have been recently adopted as auxiliary tools for the diagnosis of this mycotic infection.^[3]

Clinical forms

Cutaneous forms

The fixed form is represented by a single lesion or a few lesions at the inoculation site, which is often ulcerated with erythematous edges. The morphology can also be vegetative, verrucous, plaque infiltrated, or tuberous, without lymphatic involvement. Some cases may spontaneously regress.^[4]

A disseminated cutaneous form is characterized by multiple skin lesions at noncontiguous sites without extracutaneous involvement. Lesions of the fixed and lymphocutaneous forms may coexist in the same patient.^[3]

Extracutaneous form

Primary pulmonary sporotrichosis, resulting from inhalation of the fungus, is usually associated with chronic obstructive pulmonary disease, alcoholism, chronic use of corticosteroids and, immunosuppressive diseases.^[5] The clinical presentation is similar to that of tuberculosis, and the diagnosis is often delayed due to the rarity of pulmonary involvement. Radiological patterns include

cavitary disease, tracheobronchial lymph nodes enlargement, and nodular lesions.^[6]

Histopathological examination

Usually reveals a diffuse, granulomatous, chronic inflammatory infiltrate involving the entire dermis. It may show pseudoepitheliomatous hyperplasia, microabscess and fibrosis.^[7] Cutaneous sporotrichosis shows hyperkeratosis, parakeratosis.^[3] Besides intact polymorphonuclear cells, granulomas in sporotrichosis usually contain cellular debris, caseous material, giant and epithelioid cell lymphocytes, plasmacytes, and fibroblasts as well as *S. schenckii* yeast cells within phagocytic cells or in the extracellular medium.^[3]

If spores are present, they appear as round to oval bodies 4 to 6 µm, that stain with PAS stain more strongly at the periphery than in the centre. Single or occasionally multiple buds are seen.^[7]

S. schenckii may be seen in tissue with the routinely used hematoxylin and eosin (H&E) stain, other special stains such as Gomori methenamine silver (GMS) or periodic acid-Schiff (PAS) stain can be employed to enhance fungal detection. Fontana-Masson staining is negative. Atypical *S. schenckii* cells can appear spherical and surrounded by a PAS-positive capsule, resembling *Cryptococcus* cells.^[3]

Some histopathological alterations, such as presence or predominance of epithelioid granulomas, presence of foreign body granulomas, predominance of lymphocytes, presence or predominance of caseous necrosis, and predominance of fibrinoid necrosis and fibrosis, are related to the lack of observation of the fungus in tissue sections.^[8]

In exceptional cases asteroid bodies, the Splendore-Hoeppli reaction can be seen.^[7]

Direct Examination

Direct examination of specimens is usually conducted with 10% potassium hydroxide in order to observe parasitic budding yeast cells. These yeasts are small (2 to 6 µm in diameter) and scarce and consequently are difficult to detect upon direct examination of specimens collected from humans. When the Gram stain is used on the clinical material, yeast cells appear positively stained, sometimes within giant cells or polymorphonuclear lymphocytes. For the detection of *S. schenckii*, some authors recommend Giemsa stain after 10 to 15 dilutions of pus in physiological solution. These staining procedures also lack sensitivity.^[3]

Culture

A whitish colony with membranous aspect and surrounded by blackened halo grew on Sabouraud's agar^[7]

Positive cultures provide the strongest evidence for sporotrichosis, allowing diagnosis of almost all cases of cutaneous disease. Nevertheless, culture diagnosis has significant limitations, mainly in some manifestations of the disease such as *S. schenckii* induced arthritis, where the collection of material for culture is difficult.^[3]

Sporotrichin Skin Test

The cutaneous sporotrichin skin test detects delayed hypersensitivity, i.e., the cellular immune response, and can be a useful diagnostic tool, but its major usefulness is in epidemiological investigations. This reaction is usually positive in about 90% of confirmed sporotrichosis cases but can also indicate previous infection with the fungus.^[9]

The sporotrichin skin test has been successfully applied to confirm the diagnosis of bulbar conjunctival sporotrichosis after the pathological examination revealed yeast-like cells.^[3]

Differential Diagnosis

In the lymphocutaneous form, other disorders that present nodular lymphangitis should be investigated,^[9,10] particularly mycobacteriosis. Nocardiosis (caused mainly

by *Nocardia brasiliensis*), chromoblastomycosis, cryptococcosis, blastomycosis, and cat scratch disease also can be differential diagnoses. Sporotrichosis can also mimic cutaneous bacterial infections, sarcoidosis, lupus vulgaris, tuberculosis, and scrofuloderma, among others. These conditions should be differentiated by history, areas of endemicity, and lab tests.^[3]

Treatment

For fixed cutaneous and lymphocutaneous sporotrichosis, itraconazole 200mg daily for 3-6 months is recommended^[7]. Terbinafin administered at a daily dose of 250mg to 500mg is also effective for cutaneous sporotrichosis. Potassium iodide is also used^[7]

Prevention

Wearing gloves was protective. It is also advisable to wear heavy boots to prevent puncture wounds.^[3]

Conclusion

Sporotrichosis is usually a chronic disease with great polymorphism. It may take several clinical forms. A high clinical suspicion and a detailed history are essential for diagnosis. The lesional polymorphism of sporotrichosis makes the diagnosis on clinical ground difficult.^[7] Early diagnosis and treatment is essential to avoid further complications.

References

- Higuera N; Sporotrichosis.[Available from:<http://emedicine.medscape.com/article/228723-overview#showall>]
- Reis BD¹, Cobucci FO¹, Zaccaron LH¹, D'Acri AM¹, Lima RB¹, Martins CJ; Sporotrichosis in an unusual location--Case report. An Bras Dermatol. 2015 May-Jun;90(3 Suppl 1):84-7
- Barros et al; *Sporothrix schenckii* and Sporotrichosis.clinical microbiology reviews, Oct. 2011, p. 633–654
- Almeida, H. L., Jr, C. B. Lettnin, J. L. Barbosa, and M. C. Dias. 2009.Spontaneous resolution of zoonotic

sporotrichosis during pregnancy. *Rev. Inst. Med. Trop. Sao Paulo* 51:237–238.

5. . Ramirez, J., R. P. Byrd, Jr., and T. M. Roy. 1998. Chronic cavitary pulmonary sporotrichosis: efficacy of oral itraconazole. *J. Ky. Med. Assoc.* 96:103–105.

6. . Palomino, J., O. Saeed, P. Daroca, and J. Lasky. 2009. A 47-year-old man with cough, dyspnea, and an abnormal chest radiograph. *Chest* 135:872–875.

7. Motswaledi H, Nkosi L, Moloabi C, Ngobeni K, Nmutavhanani D, et al. (2011) Sporotrichosis: A Case Report and Literature Review. *J Clin Exp Dermatol Res* 2:132. doi: 10.4172/2155-9554.1000132

8. Quintella, L. P., S. R. Lambert Passos, A. C. Francesconi do Vale, et al. 2011. Histopathology of cutaneous sporotrichosis in Rio de Janeiro: a series of 119 consecutive cases. *J. Cutan. Pathol.* 38:25–32.

9. DiNubile, M. J. 2008. Nodular lymphangitis: a distinctive clinical entity with finite etiologies. *Curr. Infect. Dis. Rep.* 10:404–410.

10. Tobin, E. H., and W. W. Jih. 2001. Sporotrichoid lymphocutaneous infections: etiology, diagnosis and therapy. *Am. Fam. Physician* 63:326–332.