Cutaneous sporotrichosis of face: Polymorphism and reactivation after intralesional triamcinolone

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ABSTRACT

Cutaneous sporotrichosis, a subcutaneous mycotic infection caused by the saprophytic, dimorphic fungus Sporothrix schenckii. It commonly presents as lymphocutaneous or fixed cutaneous lesions involving the upper extremities with facial lesions being seen more often in children. The lesions are polymorphic. The therapeutic response to saturated solution of potassium iodide is almost diagnostic. We describe a culture-proven case of cutaneous sporotrichosis of the face mimicking lupus vulgaris initially and basal cell carcinoma later, who did not tolerate potassium iodide and failed to respond to treatment with flucnazole. The patient had reactivation of infection following an infiltration of the scar with triamcinolone acetonide injection. Various other aspects of these unusual phenomena are also discussed.

Key Words: Cicatrical ectropion, Fixed cutaneous sporotrichosis, Lymphocutaneous sporotrichosis

INTRODUCTION

Sporotrichosis is a chronic granulomatous subcutaneous infection caused by a the rapidly growing dimorphic, saprophytic and geophilic fungus; Sporothrix schenckii. It commonly presents as lymphocutaneous or fixed cutaneous lesions depending upon the status of cellular immunity. In both forms, the most common sites involved are the upper extremities and the face. We report here cutaneous sporotrichosis of the face with some unusual features.

CASE REPORT

A 26 year-old male farmer presented with a nonhealing ulcer below the right eye of about 4 years duration. It had started as a small nodule following a roadside injury and an ulcer developed over this after repeated manipulations with a thorn. The lesion rapidly increased in size and became painful following a repeat injury at the same site a year ago.

Cutaneous examination showed a 1 × 1.5 cm, erythematous, tender noduloulcerative lesion over the right infraorbital area. It had brownish-black crusting at places and succulent infiltrated borders. There was no regional lymphadenopathy. Systemic examination and routine laboratory investigations including chest X-ray were normal. Mantoux test was 22 × 20 mm in diameter. Histopathology revealed noncaseating epithelioid cell granulomas, periappendageal lymphocytic infiltrate and occasional Langhans type of multinucleated giant cells in the dermis.

In view of tuberculoid histopathology and reactive Mantoux test, treatment with rifampicin (600 mg), isoniazid (300 mg), pyrazinamide (1500 mg) and ethambutol (800 mg) was started with a provisional diagnosis of lupus vulgaris. A month later, the lesion had developed rolled-out, translucent borders and central brownish black crust mimicking basal cell carcinoma. Four new noduloulcerative lesions were also noticed in the vicinity of the primary lesion [Figure 1]. However, the biopsy material sent earlier for fungal culture grew creamy white colonies of S. schenckii identified by the typical bouquet-like pattern of conidia on
lactophenol cotton blue mounts favoring the diagnosis of sporotrichosis.

Treatment with SSKI five drops thrice a day was initiated. After three days of therapy, the patient complained of flu-like symptoms comprising of malaise, excessive lacrimation, nasal congestion and headache. He had developed lesional inflammation, purulent discharge, tenderness and peri orbital swelling. SSKI was stopped and fluconazole (600 mg/day) was started. Lesional erythema, tenderness and pus discharge subsided in five days. SSKI was reintroduced at a lower dose starting at two drops thrice daily which was increased to five drops thrice daily after a week. However, the patient again developed flu-like symptoms and lesional inflammation as before. The symptoms subsided immediately after the SSKI was stopped and the patient continued only fluconazole. An attempt to reinstitute SSKI five days later at a dose of two drops thrice daily again elicited the side effects of SSKI compelling cessation of SSKI administration altogether. The patient was discharged after the lesional erythema, induration and tenderness subsided and was advised to continue fluconazole for another seven weeks and follow-up regularly.

At the end of 12 weeks, his lesions healed completely and scars had produced ectropion of the lower eyelid [Figure 2]. After four weeks, the scar was infiltrated with triamcinolone acetonide (5 mg/ml) to correct the cicatricial ectropion. The patient returned within three weeks as the scar had become erythematous and indurated with a yellowish crust. With the possibility of reactivation of the infection due to triamcinolone, fluconazole 600 mg/day was restarted. Due to the unsatisfactory response to fluconazole after four weeks, oral itraconazole (100 mg b.i.d) was introduced and the lesions subsided completely after two months of treatment. The patient was advised to continue treatment for one more month and follow-up.

**DISCUSSION**

Sporotrichosis occurs in three clinical subsets: lymphocutaneous sporotrichosis, fixed cutaneous sporotrichosis and disseminated sporotrichosis depending upon the portal of entry of S. schenckii and the initial as well as subsequent immunological status of the patient. Contrary to other clinical forms, the localized form is associated with a high host-resistance wherein the sporotrichin test is positive, spontaneous resolution is not uncommon and response to SSKI is better. These characteristics suggest an immunological spectrum for the disease-fixed cutaneous sporotrichosis with a well-developed immune response at one end, lymphocutaneous and disseminated forms with poorly developed immune response at the other end of the spectrum.[1] Lymphocutaneous sporotrichosis accounts for almost 70% of the cases of sporotrichosis.[1] Involvement of the upper extremities occurs in varying proportions in adults while the face is frequently affected in children and adolescents.[2] The facial lesions are more often of fixed cutaneous form. The initial noduloulcerative fixed sporotrichosis lesion in our patient developed a lymphocutaneous form later probably due to destabilization of the disease following the procedure of biopsy. Such a phenomenon has been observed previously.[3] Lesions of cutaneous sporotrichosis resembling pyoderma gangrenosum,[4] keratoacanthoma,[5] soft tissue sarcoma[6] and facial cellulitis[7] have been described. The primary lesion in our patient had translucent, rolled-out borders with a central black crust resembling basal cell carcinoma. However, histopathology and culture of the fungus was diagnostic.

**Figure 1:** Noduloulcerative plaque with satellite lesions and rolled-out lower edge

**Figure 2:** Cicatricial ectropion
Potassium iodide (SKI) is the drug of first choice because of its consistent results and low cost.\[8\] The frequency of side effects and treatment noncompliance is variable across regions and though it can be as high as 60%; discontinuation of treatment is rarely required.\[2\] Our patient developed flu-like symptoms, general malaise and parotid swelling shortly after starting SKI on three occasions, which warranted its discontinuation. Lesional pain and inflammation was similar to our previously reported case.\[9\] Itraconazole is the treatment of choice for all forms of sporotrichosis particularly when cost does not preclude its use. Fluconazole is less effective than itraconazole and is of value in patients intolerant to itraconazole.\[10\] Fluconazole has been used for cutaneous sporotrichosis in doses from 200 mg/day to 800 mg/day with varying results.\[11\] An initial dose of 600 mg/day was reasonably effective in our patient. Treatment with azoles, as with SKI, also needs to be continued for at least 4-6 weeks after the apparent clinical cure to achieve mycological cure.

Reactivation of the lesion following intralesional triamcinolone acetonide infiltration given for ectropion correction is well-documented. Bickley et al.\[12\] described two patients having fixed cutaneous sporotrichosis lesions resembling other inflammatory skin conditions treated with intralesional corticosteroids. Subsequent skin biopsies from these lesions demonstrated an unusually large number of yeast cells. Furuta et al.\[13\] also observed numerous fungal elements in periodic acid-Schiff-stained histologic sections of a specimen of cutaneous sporotrichosis from a patient being treated with topical corticosteroids. Corticosteroids apparently facilitate fungal proliferation by suppressing local tissue-resistance in such cases. The disease also appears to persist in such cases far longer than the apparent clinical healing of the lesions requiring prolonged high-dose treatment.

REFERENCES