Fixed Cutaneous Sporotrichosis in an Adolescent Boy: A Case Report

William R. Howe, BS; Capt Oliver J. Wisco, USAF, MC; Col Christopher Sartori, USAF, MC

We present a case of an adolescent boy with a 3-week history of 3 painless purulent erythematous ulcers with raised hyperkeratotic borders associated with regional lymphadenopathy on the medial aspect of the right antecubital fossa. There were no known initiating factors and no other significant associated signs or symptoms. The patient initially was treated with antibiotics, with no improvement. Bacterial and fungal cultures of biopsy specimens demonstrated the presence of Sporothrix schenckii *and confirmed the diagnosis of sporotrichosis. Itraconazole was initiated with an appropriate response. This case demonstrates the importance of understanding the clinical presentation of sporotrichosis in children without a history of the disease.*

Cutis. 2006;78:337-340.

Case Report

A 13-year-old adolescent boy with no significant medical history presented with 3 painless purulent erythematous ulcers of 3 weeks' duration with raised hyperkeratotic borders on the medial aspect of the right antecubital fossa (Figure 1). There also were several fixed lymph nodes with lymphangitic streaks ascending the medial aspect of the arm in the lymphatic drainage region. The patient was afebrile and had no systemic symptoms.

The history was unremarkable, and the patient could not recall any initiating factors and denied any recent insect bites, animal contact, or outdoor exposure. Similar lesions were not found on family members or friends. The patient initially saw his primary care physician who diagnosed the lesions as arthropod bites and started the patient on amoxicillin/clavulanate potassium and ceftriaxone disodium. The patient's condition worsened despite 2 weeks of antibiotic therapy. He was referred to a dermatologist who presumptively diagnosed an atypical *Mycobacterium* infection. The antibiotic was changed to clarithromycin, and a biopsy with tissue culture for bacteria, fungi, and atypical mycobacteria was performed. Microscopic evaluation of the tissue sample demonstrated psoriasiform acanthosis, with an acute and chronic inflammatory infiltrate extending deep into the dermis (Figure 2). The inflammatory infiltrate was predominantly lymphohistiocytic with scattered neutrophils, eosinophils, and plasma cells (Figure 3). Periodic acid-Schiff, Gomori methenamine-silver, and acid-fast bacilli stains of the tissue specimen were negative for fungal organisms. Culture of the tissue biopsy specimen was negative for bacteria and atypical mycobacteria but grew *Sporothrix schenckii* after 3 weeks. Clarithromycin was discontinued and the patient was started on a 6-month course of oral itraconazole (100 mg twice daily). Monthly follow-up examinations demonstrated progressive healing. At the 6-month follow-up examination, the lymphadenopathy had resolved, and the cutaneous ulcers had completely healed with moderate residual scarring in the affected area.

Comment

Sporotrichosis is caused by the fungus *S schenckii*, the most common deep tissue fungus. It can be found worldwide, with most cases occurring in Mexico, Brazil, India, and Australia.[1] The hallmark of pediatric cases reported in the literature is a chronic nontender ulcer with raised borders unresponsive to antibiotics. However, it is often difficult to diagnose sporotrichosis in children because the lesions do not always follow the typical sporotrichoid pattern commonly found in adults, and a history of exposure is not always present.[2]

The differential diagnosis of a purulent ulcer with a raised border and associated lymphadenopathy in the sporotrichoid pattern is broad and includes

Accepted for publication March 3, 2005.

Mr. Howe is from the University of Colorado School of Medicine, Denver. Dr. Wisco is from David Grant Medical Center, Travis AFB, California. Dr. Sartori is from the US Air Force Academy Hospital, USAFA, Colorado.

The views expressed in this article are those of the authors and do not reflect the official policy of the US Air Force or the US Department of Defense.

The authors report no conflict of interest.

Reprints: Col Christopher Sartori, USAF, MC, Department of Dermatology, US Air Force Academy Hospital, 10th Medical Group, 4102 Pinion Dr, USAFA, CO 80840 (e-mail: christopher.sartori@usafa.af.mil).

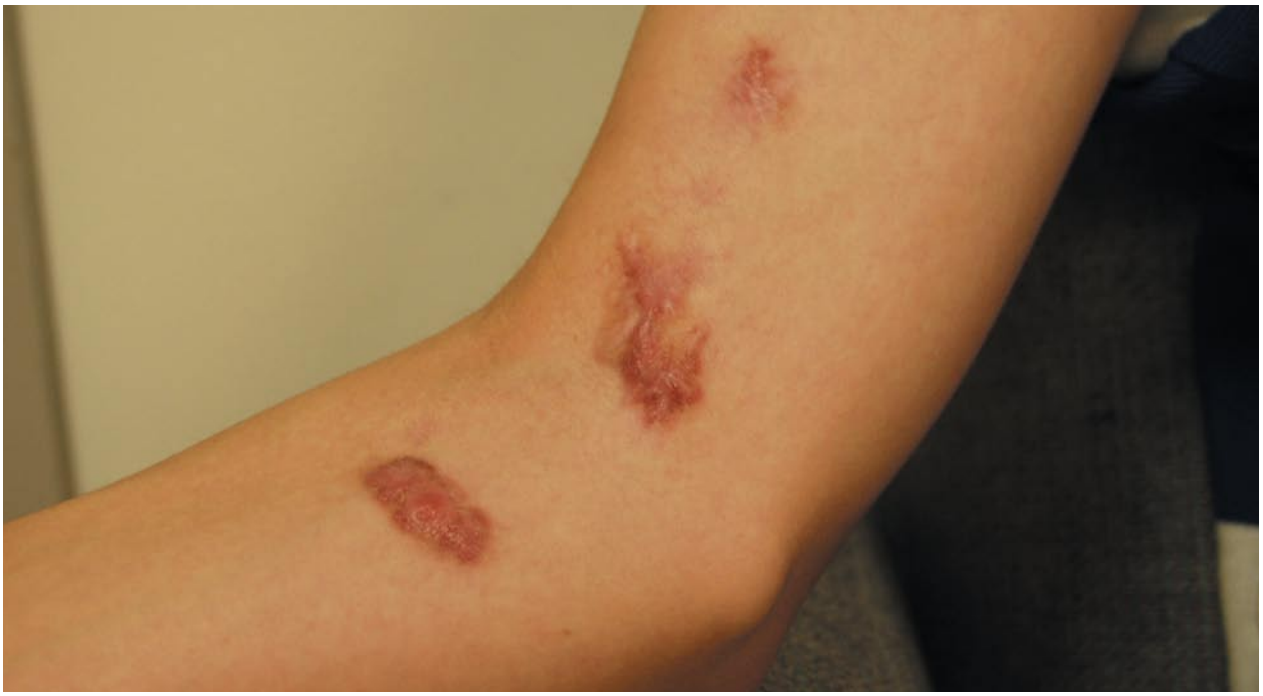

Figure 1. The medial aspect of the patient's right antecubital fossa showing 3 purulent erythematous ulcers with raised hyperkeratotic borders.

sporotrichosis, the atypical *Mycobacterium* species (especially *Mycobacterium marinum*), nocardiosis, bartonellosis (cat scratch disease), tularemia, and leishmaniasis.[2-4] Lupus vulgaris, foreign body granulomas, and pyoderma gangrenosum also should be considered.[5,6]

Sporotrichosis is characterized into 3 main types (disseminated, fixed cutaneous, and lymphocutaneous); however, the fixed cutaneous form is most common in children.[1] A child typically will present with a vague history of nontender isolated erythematous papules, plaques, or nodules that may be pustular, hyperkeratotic, and/or ulcerated. Lymphadenopathy usually is not present. Our patient, however, had significant associated lymphadenopathy. The lesions commonly are found on exposed areas of the skin, such as the face, neck, upper extremities, or lower extremities. Time of inoculation to onset of disease can vary from 3 weeks to 6 months. Sporotrichosis rarely heals spontaneously; thus, treatment is mandatory. A delay in treatment significantly increases the risk of scarring.[1]

The most common type of sporotrichosis is the lymphocutaneous form, which is common in adults and presents as small painless papules that slowly enlarge and spread proximally following the lymphatic channels and forming the typical sporotrichoid pattern.[7] The lesions are associated with regional lymphadenopathy and are very similar to the fixed cutaneous type in their progression, location, timing, and treatment.[8]

The disseminated form of sporotrichosis is an emerging condition in individuals with human immunodeficiency virus.[9,10] Patients with this form present with multiple typical sporotrichoid papules, plaques, and/or nodules, frequently with visceral dissemination.[11] Systemic spread is believed to originate from a pulmonary focus or cutaneous inoculation site. Pulmonary manifestations range from acute bronchitis to end-stage pulmonary fibrosis. Extrapulmonary sites primarily involve the joints, sinuses, and central nervous system.[11]

Cutaneous infection occurs through traumatic implantation from a contaminated plant or animal. Plants commonly implicated in cutaneous infection include rose bushes, tree bark, hay, shrub thorns, sphagnum moss, and wheat grains.[7,12] Animals usually responsible for infection include cats, armadillos, horses, dogs, snakes, rats, and birds.[12] Fish spines are other potential sources of *S schenckii*.[13] Although skin trauma from an animal source usually is required for infection to occur, trauma is not always obligatory. Inoculation can occur through direct contact with an animal's skin lesion, particularly one that contains a large number of organisms.[14]

S schenckii is a dimorphic fungus that exists as yeast in host tissues and as mold at room temperature.[15,16] The virulence factors of *S schenckii* have not been clearly identified, but excreted melanin and extracellular proteinases are implicated. Melanin seems to play a role in destroying monocytes and

macrophages, while the extracellular proteinases bind to fibronectins, laminin, and type II collagen to assist in dissemination.[15,16]

The gold standard for diagnosing sporotrichosis is fungal culture. *Sporothrix* grows on Sabouraud dextrose agar or Mycosel plates at 25°C. Subculture analysis on blood-glucose-cysteine agar or brain-heart infusion broth confirms the presence of *S schenckii*.[4] Microscopic evaluation demonstrates oval or pyriform conidia in a bouquetlike pattern. The yeast-form cigar-shaped organisms are identified using a periodic acid-Schiff or Gomori methenamine-silver stain.[12] Histologic examination of the tissue exhibits epidermal hyperplasia, intraepidermal abscesses, and hyperkeratosis. In 40% of infected patients, granulomas with extracellular asteroid bodies consisting of radiating eosinophilic spicules around a fungal cell can be found within the abscesses and are specific for *S schenckii*.[17] Newer methods for identifying sporotrichosis include immunohistochemical staining, nested polymerase chain reaction assays, and a sporotrichin skin test that detects a delayed hypersensitivity reaction.[8,18,19]

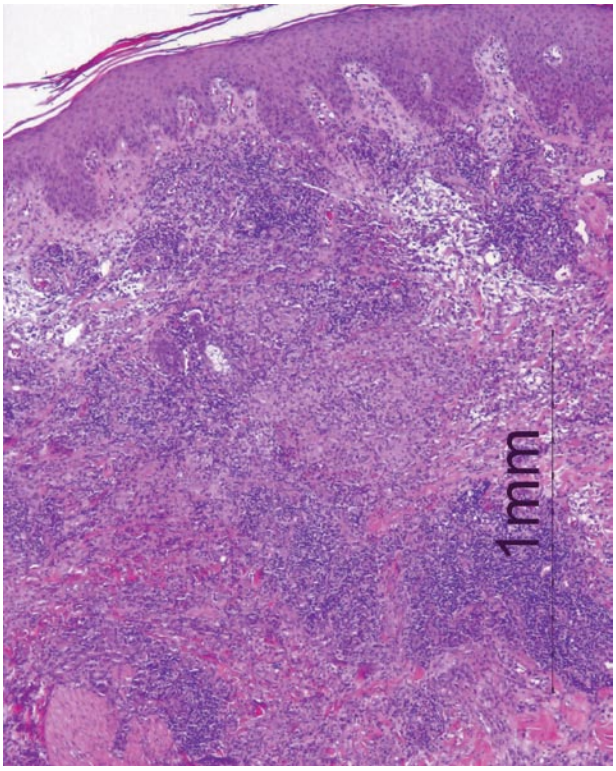

Figure 2. Psoriasiform acanthosis with an acute and chronic inflammatory infiltrate extending deep into the dermis (H&E, original magnification ×10).

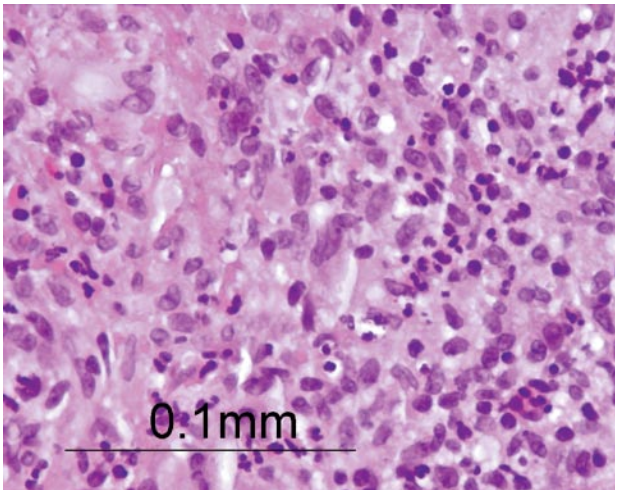

Figure 3. The inflammatory infiltrate was predominantly lymphohistiocytic with scattered neutrophils, eosinophils, and plasma cells (H&E, original magnification ×40).

Since the early 1900s, the main treatment for cutaneous sporotrichosis in endemic areas has been potassium iodide.[20] Potassium iodide is both very effective and inexpensive, but because of its complicated treatment regimen and side effects, the imidazole class of antifungal medications now is usually prescribed. The treatment regimen for potassium iodide progresses from 40 to 50 drops (47 mg/drop) administered 3 times daily for at least 4 weeks. Adverse events include dermatitis, nausea, vomiting, diarrhea, abdominal pain, metallic taste, sore teeth and gums, excessive salivation, headache, hyperkalemia, and disrupted thyroid hormone production.[20] Itraconazole, an imidazole-class antifungal agent, is prescribed at 100 to 200 mg daily for 3 to 6 months. It is the treatment of choice when cost is not a limiting factor. In children, itraconazole is well-tolerated with few adverse effects.[21] Other effective medications include terbinafine hydrochloride (250 mg twice daily for 18 weeks) and fluconazole (400 mg daily for 6 months). Because some of the *S schenckii* strains do not grow in temperatures exceeding 35°C, heat can be provided through handheld heaters, hot baths, and hot compresses. The patient must use the heat source at least one hour daily for several months, making compliance difficult. This method, however, may be particularly useful in patients, such as pregnant women, who cannot safely take the mainstay medications.[22,23]

REFERENCES

1. Welsh O, Schmid-Grendelmeier P, Stingl P, et al. Tropical dermatology. part II. *J Am Acad Dermatol*. 2002;46:748-763.
2. Burch JM. Unsuspected sporotrichosis in childhood. *Pediatr Infect Dis J*. 2001;20:442-445.

3. Kayal JD, McCall CO. Sporotrichoid cutaneous *Mycobacterium avium* complex infection. *J Am Acad Dermatol*. 2002;47:249-250.
4. Queiroz-Telles F, McGinnis MR, Salkin I, et al. Subcutaneous mycoses. *Infect Dis Clin North Am*. 2003; 17:59-85.
5. Byrd DR, El-Azhary RA, Gibson LE, et al. Sporotrichosis masquerading as pyoderma gangrenosum: case report and review of 19 cases of sporotrichosis. *J Eur Acad Dermatol Venereol*. 2001;15:581-584.
6. Khandpur S, Nanda S, Reddy BS. An unusual episode of lupus vulgaris masquerading as sporotrichosis. *Int J Dermatol*. 2001;40:336-339.
7. Tobin EH, Jih WW. Sporotrichoid lymphocutaneous infections: etiology, diagnosis and therapy. *Am Fam Physician*. 2001;63:326-332.
8. Morris-Jones R. Sporothrichosis. *Clin Exp Dermatol*. 2002;27:427-431.
9. Al-Tawfiq JA, Woods KK. Disseminated sporotrichosis and *Sporothrix schenckii* fungemia as the initial presentation of human immunodeficiency virus infection. *Clin Infect Dis*. 1998;26:1403-1406.
10. Moylett EH, Shearer WT. HIV: clinical manifestations. *J Allergy Clin Immunol*. 2002;110:3-16.
11. Ware AJ, Cockerell CJ, Skiest DJ, et al. Disseminated sporotrichosis with extensive cutaneous involvement in a patient with AIDS. *J Am Acad Dermatol*. 1999;40:350-355.
12. De Araujo T, Marques AC, Kerdel F. Sporotrichosis. *Int J Dermatol*. 2001;40:737-742.
13. Haddad VJ. Localized lymphatic sporotrichosis after fish-induced injury. *Med Mycol*. 2002;40:425-427.
14. Reed KD, Moore FM, Geiger GE, et al. Zoonotic transmission of sporotrichosis: case report and review. *Clin Infect Dis*. 1993;16:384-387.
15. Lima OC, Figueiredo CC, Pereira BA, et al. Adhesion of the human pathogen *Sporothrix schenckii* to several extracellular matrix proteins. *Braz J Med Biol Res*. 1999;32: 651-657.
16. Romero-Martinez R, Wheeler M, Guerrero-Plata A, et al. Biosynthesis and functions of melanin in *Sporothrix schenckii*. *Infect Immun*. 2000;68:3696-3703.
17. Rodriguez G, Sarmiento L. The asteroid bodies of sporotrichosis. *Am J Dermatopathol*. 1998;20:246-249.
18. Byrd J, Mehregan DR, Mehregan DA. Utility of antibacillus Calmette-Guerin antibodies as a screen for organisms in sporotrichoid infections. *J Am Acad Dermatol*. 2001;44:261-264.
19. Hu S, Chung WH, Hung SI, et al. Detection of *Sporothrix schenckii* in clinical samples by a nested PCR assay. *J Clin Microbiol*. 2003;41:1414-1418.
20. Sterling JB, Heymann WR. Potassium iodide in dermatology: 19th century drug for the 21st century—uses, pharmacology, adverse effects, and contraindications. *J Am Acad Dermatol*. 2000;43:691-697.
21. Gupta AK, Cooper EA, Ginter G. Efficacy and safety of itraconazole use in children. *Dermatol Clin*. 2003;21:521-535.
22. Hay RJ. Therapeutic potential of terbinafine in subcutaneous and systemic mycoses. *Br J Dermatol*. 1999;141:36-40.
23. Kauffman CA, Hajjeh R, Chapman SW. Practice guidelines for the management of patients with sporotrichosis. For the Mycoses Study Group. Infectious Disease Society of America. *Clin Infect Dis*. 2000;30:684-687.