**BRIEF REPORTS**

**Long-Term Follow-Up of Multifocal Osteoarticular Sporotrichosis Treated with Itraconazole**

Sporotrichosis is a granulomatous disease caused by the dimorphic fungus *Sporothrix schenckii*. Disseminated disease may involve bones, joints, lungs, and meninges and is rarely seen in immunocompetent hosts. Amphotericin B has been the treatment of choice for deep-seated sporotrichosis, although recent reports describing cases with <24 months of follow-up suggest that itraconazole therapy may be more effective [1]. Relapse of multifocal osteoarticular disease has been reported following treatment with alternate antifungal agents 36 months after treatment was discontinued [2]; this finding indicates that longer follow-up is required to establish efficacy. We describe two patients with multifocal osteoarticular sporotrichosis who each received a 24-month course of therapy with itraconazole and have not relapsed 41 and 68 months after discontinuing treatment.

**Case 1.** A 51-year-old male developed itching of both arms in July 1986 after working in his garden. The following week he developed swelling of the upper extremities. The titer of antinuclear antibody (ANA) was 1:320. Microscopic examination of biopsy specimens from the forearm demonstrated necrobiosis with granulomas. His condition improved with symptomatic treatment, followed by prednisone therapy. In November 1986, he was referred for evaluation of recurrent symptoms.

Physical examination demonstrated livedo reticularis, swelling of the wrists and hands, and a tender, edematous left forearm. Roentgenograms of the hands and wrists revealed soft-tissue swelling. He subsequently developed vasospasm in his fingers and tender, red, fluctuant lesions at many sites on the wrists, left forearm, and calves. He denied gardening and exposures to other infectious agents.

Physical examination demonstrated limited range of motion of the wrists and thickened skin over the right prepatellar area and distal left forearm. The WBC count was 4,400/mm³ with 7% eosinophils, and the titer of ANA was 1:320 (table 1). Culture of biopsy specimens from the forearm yielded bacteria, yet despite treatment with steroids and antibiotics, his condition worsened. Further debridement was performed, and cultures of the removed tissue yielded *S. schenckii* and serology was positive at 1:320 dilution. Following therapy with amphotericin B and ketoconazole, pain recurred in his right knee, and an osteolytic lesion was found in his femur. This area was biopsied, and cultures yielded *S. schenckii*. He was treated with itraconazole (200 mg/d) for 24 months. In the ensuing 41 months, he remained free of symptoms.

*S. schenckii* is generally considered to be of low virulence; consequently, localized disease is most common in immunocompetent hosts. Disseminated disease may affect the lungs, bones, eyes, and meninges, but articular disease is most common. Single or multiple joints (typically the knees, hands, and wrists) may be involved [3]. Until recently, intravenous amphotericin B had been considered the treatment of choice for deep-seated sporotrichosis [4], and such therapy is successful in 50%-70% of cases. Alternate treatments include the azoles. However, the results with ketoconazole in the treatment of disseminated sporotrichosis have been disappointing [5].

While fluconazole has a role in treatment of cutaneous disease, it is less effective for treatment of osteoarticular disease [4]. Itraconazole better in vitro and in vivo activity against *S. schenckii* in animal models than does ketoconazole or fluconazole [6]. In recent years, case reports have documented the effectiveness of itraconazole in the treatment of *S. schenckii*. In the sole nonrandomized trial [7], patients were treated for various durations, and of 35 patients, 25 responded to itraconazole therapy. In that study, 13 patients with osteoarticular disease received 200–400 mg/d of itraconazole for 5–18 months, and 4 (31%) relapsed after a mean duration of 5.8 months of therapy.

The two patients described herein are of interest for several reasons. Both had high ANA titers for which there was no other explanation than infection with *S. schenckii*, and both patients responded transiently to therapy with corticosteroids. In addition, our cases represent the longest reported follow-up of multifocal osteoarticular sporotrichosis treated with itraconazole. Findings after follow-up periods of 41 months and 68 months demonstrated that itraconazole therapy has resulted in a sustained cure of multifocal osteoarticular sporotrichosis. These results suggest that doses of 200 mg/d for 24 months are sufficient to effect a long-term cure of multifocal osteoarticular disease.

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Table 1. Clinical features of two patients with osteoarticular sporotrichosis treated with itraconazole.

<table>
<thead>
<tr>
<th>Patient no.</th>
<th>Age (y)/sex</th>
<th>Symptoms at presentation</th>
<th>Time from onset of symptoms to diagnosis</th>
<th>Laboratory test (titer or cell count)</th>
<th>Previous ineffective treatment (dosage)</th>
<th>Itraconazole regimen</th>
<th>Outcome (length of follow-up)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>51/M</td>
<td>Bilateral arm pain</td>
<td>6 mo</td>
<td>ANA (1:640); sporothrix serology (1:640)</td>
<td>Amphotericin B (2,176 mg); ketoconazole (400 mg/d for 4 mo)</td>
<td>200 mg/d for 23 mo</td>
<td>Cure (68 mo)</td>
</tr>
<tr>
<td>2</td>
<td>49/M</td>
<td>Tender lesions on forearms and legs</td>
<td>12 mo</td>
<td>ANA (1:320); sporothrix serology (1:320); WBC count (4,400/mm³ with 7% eosinophils)</td>
<td>Amphotericin B (3 g); ketoconazole (400 mg/d for 43 mo)</td>
<td>200 mg/d for 24 mo</td>
<td>Cure (41 mo)</td>
</tr>
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NOTE. ANA = antinuclear antibody.

References

An Unusual Case of Hydatid Disease: Localization to the Gluteus Muscle

Echinococcus granulosus is a species of tapeworm that is endemic in certain parts of the world, particularly in many rural zones of South America [1]. Infection with this organism occurs when an intermediate host (cattle or humans) ingests the released eggs, which are contained in the feces of the definitive host (dogs or wolves). The larva is then liberated, penetrates through the intestinal epithelium, and finally is transported to target organs where the parasite develops, forming a hydatid cyst. These cysts mainly occur in the liver and lungs (65% and 25% of cases, respectively); other sites such as the kidneys, spleen, and bones are less frequently involved. Muscular involvement with hydatid cysts represents only 4% of cases, and when this disease occurs in unusual sites, it may mimic other pathological processes [2-4]. We describe an unusual case of human hydatid disease that developed in the gluteus muscle.

A 49-year-old woman who lived in a rural area of Argentina was admitted to the University Hospital in Cordoba complaining of an abscessed tumor-like mass that was located in the upper zone of her right gluteus muscle. She had had no other symptoms until 2 weeks before admission, when she noted that the mass had become warm and painful. The patient mentioned that she had received intramuscular medication in the same muscle several days before admission, and we suspected that there might be a relationship between the needle puncture and development of the abscess. Therefore, the abscess was drained, yielding 100 mL of colorless transparent liquid. Microscopic examination of the liquid revealed the presence of scolices and hooks.

Ultrasonographic examination of the zone showed a 19 × 49-mm abscess with liquid density that was surrounded by fibrotic areas. Findings on abdominal ultrasonograms and chest radiographs were normal. The leukocyte count was 7,400/mm³ with 6% eosinophils, and all tests for hydatid disease were positive. On the basis of these results, resection of the cyst was planned. The patient received oral albendazole (10 mg/kg) for 4 weeks before the cyst was removed. After surgery, she recovered uneventfully, and she continued to receive oral albendazole for 1 month at which time she was seen at a follow-up visit.

Hydatid disease that involves unusual sites is seen in patients in many countries throughout the world; however, such disease may be overlooked if clinicians fail to consider the possibility of parasitic involvement at unusual sites. Hydatid cysts that are located in the gluteus muscle represent a rare presentation of E. granulosus infection, which may be confused with other lesions such as tumors or may mimic other pathological processes. The present report serves to remind physicians and other public health workers that it is important to initially make the correct