Sporothrix schenckii infection mimicking sarcoidosis

Sir, Sporothrix schenckii infection (sporotrichosis) is a fungal disease which generally involves the skin, subcutaneous tissues and the surrounding lymph nodes [1, 2]. The disseminated form of sporotrichosis is rare and most often affects the osteoarticular system, with mild, self-limited tenosynovitis and a destructive septic arthritis [1, 3–5]. We describe a man who developed disseminated infection with S. schenckii with cutaneous, pulmonary, ocular and articular involvement, misdiagnosed as sarcoidosis and with a good response to itraconazole treatment.

In September 1998, a 49-yr-old white man was admitted to the rheumatology unit with painful subcutaneous nodules of the arms, lower limbs and abdomen for 6 months. Physical examination showed a well-looking overweight man. His past medical history was unremarkable. He had received a skin injury, inflicted by the branch of a tree. In October 1998, the patient developed arthritis of the wrists and ankles and also blurred vision due to granulomatous anterior uveitis in his right eye. Treatment with oral prednisone 60 mg daily was started, but despite this a new ophthalmological evaluation disclosed granulomatous panuveitis in his right eye and glaucoma in the left eye. Laboratory investigations were normal, apart from leucocytosis of 11.2 × 10³ cells/mm³, thrombocytopenia of 84 × 10³ cells/mm³, an erythrocyte sedimentation rate of 80 mm/h and an antinuclear antibody titre of 1:160 (speckled pattern). Serological tests for hepatitis B and C virus, cytomegalovirus, syphilis, toxoplasmosis and HIV were negative. The tuberculosis test was negative. Chest radiography revealed bilateral interstitial infiltrates in the lower lobes of the lungs. Computed tomography of the chest showed mild enlargement of the mediastinal lymph nodes (<1 cm). Histopathological examination of the skin biopsy specimens showed non-caseous, well-formed granulomas, consistent with sarcoidosis. Staining for acid-fast bacilli, periodic acid-Schiff-positive bacilli and fungi was negative. In November 1998, he developed polyarthritis affecting the small joints of the hands and ankles, daily fever (38–39°C), dyspnea and phthisis bulbi of the right eye, and eye enucleation was performed. The diagnosis of sarcoidosis was based on the clinical and histopathological findings. Treatment with intramuscular methotrexate (15 mg/week) was started and prednisone was tapered to 5 mg daily. One month later, he developed hoarseness and oral examination disclosed granulomatous lesions located in the supraglottic area, nasal septum and floor of the mouth; biopsy showed infiltrates of chronic inflammatory cells and well-formed granulomas. A new skin biopsy was performed and histopathological evaluation showed a non-caseous granulomatous reaction with neutrophils and many fungal structures suggestive of S. schenckii; this was confirmed by positive culture of skin specimens. The diagnosis of disseminated sporotrichosis was made and treatment with oral itraconazole (400 mg daily) was started and maintained for 6 months. Methotrexate and prednisone were tapered off. The skin lesions, hoarseness, fever and articular complaints disappeared. The patient was last seen in January 2000, when he complained of pain of hips and right ankle. The new radiographic evaluation showed diffuse osteopenia and mild reduction of the proximal interphalangeal (PIP) joints and bone erosions of the second metacarpophalangeal (MCP) of the right hand and right ankle. Bone subluxation of the PIP and MCP were noted.

The disseminated form of sporotrichosis is uncommon. Nevertheless, there are reports showing the involvement of bone, periosteum and synovia in almost 80% of cases of disseminated sporotrichosis [1]. These are followed in frequency by pulmonary disease and systemic infection involving the eye and adnexa, sinuses, meninges and gastrointestinal tract. Osteoarticular infection develops in most patients and may occur as destructive arthritis with erosions, periostitis, tendinitis, tenosynovitis and, more rarely, myositis [1]. Arthritis is usually chronic and may involve the small joints of the hands and feet, knees and wrists. Bone lesions can be found in the radius and ulna, femur and ribs [1]. Our patient had a systemic disease that presented initially as a cutaneous disease but evolved with severe articular involvement, leading to joint destruction.

This case report illustrates the clinical problems frequently seen in systemic or articular sporotrichosis. (i) The radiological and histopathological features of Sporothrix infection are not specific [1]. Osteoporosis of contiguous bone surfaces and soft tissue swelling are the most common bone X-ray features. Parasympathetic swelling, subchondral bone erosions and joint-space narrowing occur less often [5]. (ii) It is difficult to distinguish sporotrichosis from other granulomatous diseases, including sarcoidosis [1, 6]. Initially, there appeared to be little doubt that our patient had sarcoidosis, as judged by the combination of mediastinal lymph node enlargement, arthritis, skin lesions and the typical histological findings of a non-caseous sarcoid-like granuloma and with no evidence of acid-fast bacilli or fungi. However, a subsequent positive culture for S. schenckii and the improvement of the patient with itraconazole treatment make the diagnosis of sporotrichosis very likely. (iii) There was a delay in diagnosis. As has previously been reported [5], this could be explained by the scarcity of S. schenckii in the infected tissues. In our case, all previous skin biopsies were negative for fungus, as were bacterial smears on culture. (iv) There was no clear occupational or individual risk for the development of sporotrichosis. Certain occupational groups are at risk of the development of sporotrichosis, including farmers, florists, gardeners, fruit-packers and workers in rural areas who are exposed to frequent skin injuries. Alcoholics and immunosuppressed patients (e.g. those with HIV infection) have also been noted to be predisposed to sporotrichosis [2–5, 7]. The only risk factor found in our patient was a skin injury caused by a tree branch 4 months before the beginning of his cutaneous disease.

The treatment of choice for disseminated sporotrichosis is amphotericin B, but severe side-effects are associated with this drug, such as nephrotoxicity and hyperkalaemia. In addition, parenteral administration is required [8]. Itraconazole, an oral antifungal drug, has been used for the treatment of sporotrichosis, but there is controversy about the results with regard to articular and systemic improvement [9, 10]. Our patient had a good response to treatment with oral itraconazole, and he recovered completely from the skin lesions, tenosynovitis and arthritis. Finally, because cultures for Sporothrix are often negative, we would like to point out the necessity of serial tissue culture for infectious agents in some osteoarticular diseases, particularly sarcoid-like reactions that do not respond to corticosteroid treatment.

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Trimovate (clobetasone butyrate plus antimicrobial agents)) to the groin, with complete recovery.

Terbinafine is very widely used and is the drug of choice in myologically proven onychomycosis. A large postmarketing surveillance programme of 25,000 patients on this drug showed a frequency of cutaneous side-effects in the region of 2.5% [1]. There have been five previous reports of cutaneous lupus in patients receiving terbinafine therapy in the dermatology literature [2–6]. Gupta et al. [7] reported a series of different cutaneous manifestations of the adverse effects of terbinafine, including erythema multiforme, erythroderma, worsening of psoriasis and pityriasis rosea. Contrary to the report of Bonsmann et al. [6], our patient was negative for ANA and anti-histone antibodies. Our patient was anti-Ro (SS-A) positive, and it has previously been noted that this antibody pattern is commonly seen in drug-induced cutaneous lupus [8]. It is notable that there is no reference to cutaneous lupus being a potential adverse effect in the data sheet for terbinafine, and terbinafine is not listed as a potential cause of drug-induced lupus in major rheumatology texts or electronic texts such as UpToDate. We conclude that terbinafine should only be used with caution, if at all, in patients with lupus, especially those who are anti-Ro (SS-A)-positive and then only if nail-clipping mycology is positive.

The authors have declared no conflicts of interest.

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Lower level of synovial fluid interferon-γ in HLA-B27-positive than in HLA-B27-negative patients with Chlamydia trachomatis reactive arthritis

Sir, We read with great interest the study by Bas et al. [1] in which they analysed synovial fluid (SF) cytokine levels in reactive...