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Musculoskeletal Sporotrichosis

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Abstract. Sporotrichosis is a chronic, indolent, fungal infection that rarely involves the musculoskeletal system. The etiologic agent, *Sporothrix schenckii*, is ubiquitous in nature and has been isolated from soil, timber, decaying vegetation, and a variety of foliage. The organism gains entrance to the body through trauma to the skin or, in rare instances, by inhalation. The vast majority of infections in humans is characterized by nodular or ulcerated lesions of the cutaneous tissues and adjacent lymphatics. Osteoarticular involvement may occur either by contiguous spread from a cutaneous focus, through direct inoculation of tissue by the organism, or by hematogenous dissemination. The rarity of musculoskeletal sporotrichosis often causes a delay in diagnosis which leads to inappropriate therapy and permanent deformity in some patients. Three cases which show a spectrum of bone and joint involvement are presented.

Key words: Infection – Fungal – Arthritis – Osteomyelitis – Bursitis

The first documented case of sporotrichosis was reported from the Johns Hopkins Hospital in 1898 by Schenck [19]. Between 1906 and 1912 de Beurmann reported 210 cases of sporotrichosis and, for the first time, described extracutaneous infection in 15 patients [5]. There have been periodic reports of sporotrichal epidemics; one of the most famous of these epidemics was in South Africa where in a two year period, over 3,000 gold miners were infected by the fungus which was growing on timbers in the mines [21]. Sporotrichosis is the most common subcutaneous and deep mycosis in Mexico where most patients give a history of gathering grass for use as packing material or for basket weaving. In the United States, Canada, and other temperate countries, infection in man is usually associated with gardening [17], construction work, and mining. The disease is generally considered an occupational hazard of rose growers [24] and those workers who are in contact with gardening soil or sphagnum moss [3]. Infections have been traced to inoculation of the fungus by a variety of foreign objects as well as by the bites of wild or domestic animals, birds, and insects [23]. The disease has also been transmitted via contaminated dressings from suppurating lesions, and there is one report in which the disease was transmitted by direct contact from the cheek of a mother to the cheek of her child [22].

In the majority of instances, sporotrichosis infection is limited to the skin and regional lymphatics. Extracutaneous sporotrichosis, either focal or disseminated, is rare with only five instances occurring in 3,300 cases reported in the South Africa epidemic [14]. In a review of the world literature from 1898 to 1967 Wilson et al. found only 30 documented cases of disseminated sporotrichosis. Two patients suffered from diabetes mellitus and two had sarcoidosis. Bone, periosteum, and synovium were involved in 24 patients [23].

Gladstone's review of the American and English literature between 1924 and 1970 revealed only 24 cases of osseous sporotrichosis. Fourteen patients had disseminated skin lesions. The tibia, fibula, and small tubular bones of the hand and feet were most frequently infected (17 patients) [8].

Altner reported six cases of sporotrichosis of bones and joints and found an additional 24 cases in his review of the English literature from 1898

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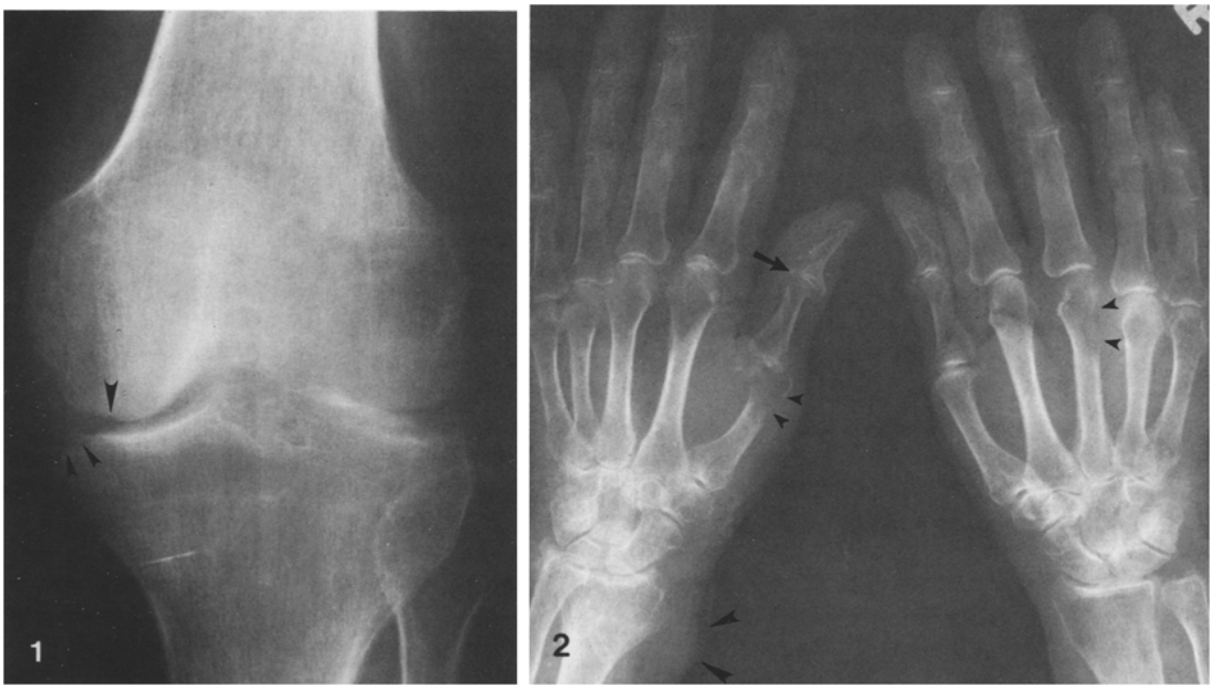

Fig. 1. Examination of the knee shows diffuse osteopenia and severe joint space narrowing. Subchondral and periarticular erosions are present (*arrowheads*)

Fig. 2. Posteroanterior view of both hands shows a soft tissue nodule adjacent to the distal left radius (*large arrowheads*) and destruction of the distal aspect of the left first and right third metacarpals (*small arrowheads*) with associated soft tissue swelling. There are erosions involving the left first interphalangeal joint (*arrow*)

to 1970 [1]. Two of the six reported cases had disseminated infections. A review of the English language literature from 1898 to 1979 by Bayer et al. revealed only 44 documented cases of sporotrichal arthritis [2]. In 21 cases, the infection was confined to the site initially involved. Ten patients developed disease in contiguous cutaneous or subcutaneous tissues with resultant abscesses, fistulae, and/or draining sinuses. Dissemination of the infection, presumably by hematogenous spread, occurred in 14 patients who had distant involvement most commonly in other joints, skin, subcutaneous tissue, or muscle.

Disseminated sporotrichosis is felt to represent an opportunistic infection. In support of this Bayer et al. found that 23 of 26 patients with detailed clinical information cited had associated diseases [2]. The most common underlying disorders were alcoholism, myeloproliferative diseases, underlying malignancy, and corticosteroid dependence.

Case Reports

Case 1

A 56-year-old white male construction worker first presented to a local physician in March 1982 with pain and swelling in his left knee, and he was initially diagnosed as having tendinitis. The knee pain continued and on the second visit to his physician he was told that he had a Baker cyst. Surgery was performed in November 1982, at which time a soft tissue mass was removed. Culture and histologic evaluation of the specimen yielded no diagnosis. The knee pain worsened and he became unable to walk. A second surgical procedure and synovial biopsy were performed in February 1983. Again no specific diagnosis was made from either the culture or the pathology. The patient was referred to Barnes Hospital for further evaluation.

On admission the laboratory data were unremarkable except for a slightly elevated erythrocyte sedimentation rate. Physical examination revealed a slightly warm left knee which was painful upon valgus stress. A joint effusion was present. Admission radiographs of the knee confirmed a joint effusion and showed diffuse osteopenia and generalized severe joint space narrowing. Small subchondral and periarticular erosions were present (Fig. 1). Arthrocentesis yielded serosanguinous fluid which was culture positive for *Sporothrix schenkii*. The patient was begun on systemic amphotericin B with subsequent decrease in his joint pain.

Case 2

A 77-year-old white woman presented in January 1955 with a two month history of arthralgias in the hands followed by the development of soft tissue nodules and widespread ulcerating skin lesions. Approximately three weeks after they appeared, the lesions became fluctuant, ulcerated, and drained serosanguinous fluid. The patient also had a three week history

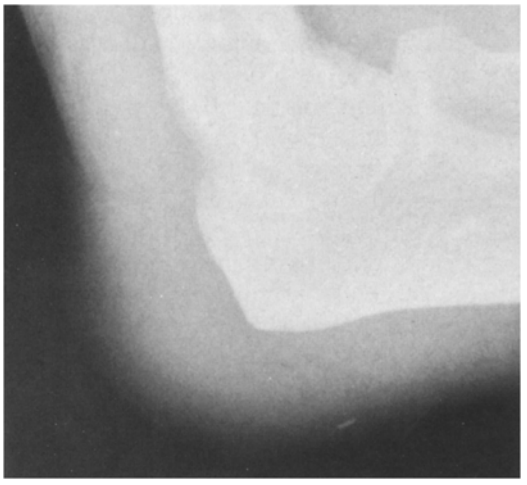

Fig. 3. Lateral view of the right elbow demonstrates marked enlargement of the olecranon bursa. No joint effusion or bony abnormality is evident

of night sweats, chills, and dry cough. She had been treated with oral penicillin and topical sulfa powder without any improvement. A skin biopsy was performed and it yielded no diagnosis.

On admission to Barnes Hospital the physical examination showed several types of cutaneous lesions. A 3×3 cm raised, reddish purple, fluctuant lesion was present on the dorsum of her right hand. Encrusted, raised lesions having violaceous borders and measuring 1–6 cm in diameter were present over the upper extremities. Weeping, ulcerated lesions measuring approximately 1–2.5 cm in diameter were present over her lower extremities and buttocks. Laboratory data included white blood cell count of 14,500 and a negative tuberculin skin test. Her chest radiograph was normal. Radiographs of the hands demonstrated nodular soft tissue swelling adjacent to the distal left radius, destruction of the left first and right third metacarpal heads, and erosions of the left first interphalangeal joint (Fig. 2).

Approximately 5 cc of fluid were aspirated from a nodule on the dorsum of the right hand; culture of this fluid grew *Sporothrix schenckii*. Biopsies of several lesions on the right arm showed acute and chronic inflammation with cultures positive for *Sporothrix schenckii*. Because of the positive cultures, a Schiff stain was performed on the biopsy specimen, but this failed to show any fungal organisms. After being discharged on potassium iodide oral solution (SSKI) the patient improved clinically. A radiograph of the right hand three months later showed early healing of the osseous lesions.

Case 3

A 71-year-old white male engineer enjoyed gardening as a hobby. During the summer of 1982 he developed a small, red nodule on the volar aspect of his right forearm. During the subsequent two weeks this nodule increased in size while several more developed in the right anticubital fossa and over the olecranon. He was seen by a dermatologist in September 1982, at which time a clinical diagnosis of sporotrichosis was made, and treatment with SSKI (40 drops three times a day) was begun. Although his cutaneous lesions improved clinically, the patient developed episodes of numbness and tingling in his right ring and little fingers. On physical examination in January 1983, the olecranon bursa was swollen and culture of a bursal aspirate was positive for *Sporothrix schenckii*.

Physical examination on admission to Barnes Hospital revealed a 4×4.5 cm fluctuant erythematous mass involving the right olecranon bursa, and a separate 3.5×5 cm soft tissue mass in the right anticubital fossa. These did not have warmth, tenderness, or ulceration. The admission laboratory data were unremarkable. Radiographs showed enlargement of the olecranon bursa without evidence of arthritis, osteomyelitis, or soft tissue calcification (Fig. 3). A biopsy of the olecranon bursa showed granulomatous inflammation and fungal organisms. Cultures of the specimen grew *Sporothrix schenckii*.

The patient was begun on a course of amphotericin B. Because his serum creatinine became elevated, the dosage was lowered, and his bursitis failed to show clinical improvement. Subsequently, an excision of the right olecranon bursa was done. Histologic evaluation showed extensive fibrosis and chronic inflammation. There were several areas of central fibrinoid necrosis with pallisading borders, consistent with a caseating granulomatous infection. Special stains were positive for *Sporothrix schenckii*. The amphotericin B was discontinued and the patient was discharged on SSKI (40 drops daily).

Discussion

Sporotrichosis infection has a spectrum of clinical presentation which depends on the site of entry of the organism and the host's response to the inoculum [19]. Three main clinical types are discussed.

Cutaneous Lymphatic Sporotrichosis

The most common form of sporotrichosis follows subcutaneous inoculation from a puncture wound due to a thorn or splinter. The two types of cutaneous infection comprise up to 75% of all reported cases [9]. The first type is a small, nontender, movable subcutaneous nodule which develops at the site of injury 1–12 weeks after inoculation. The nodule eventually ulcerates and closely resembles a chancre. The other, less frequent lesion begins as a small ulcer at the site of injury. The difference in the clinical presentation of these two lesions is probably due to the site of organism inoculation, one in the subcutaneous tissue, and the other in the epidermis [18].

No matter which form the primary lesion takes, if untreated, sporotrichosis infection runs a chronic course, characterized by involvement of the lymphatic system draining the primary lesion. The lymph nodes become enlarged and eventually suppurate; the interconnecting lymphatics become indurated and cord-like. Seropurulent discharge may drain from both the primary lesion and the involved nodes. Within weeks of the development of the primary lesion, multiple, hard, subcutaneous nodules develop along the draining lymphatic channels. Although untreated infections spread to

distant lymph nodes and more subcutaneous nodules develop, there is less tendency for these lesions to undergo necrosis [9]. Similarly, while some cases may heal spontaneously, untreated sporotrichosis usually persists for years without systemic dissemination.

Cutaneous Nonlymphatic Sporotrichosis

Cutaneous lesions may remain localized without involvement of lymphatics [6], and these most commonly occur on the face, trunk, neck, and arm. Their variable appearance including ulcerative, verrucose, papular, acneform, or erythematoid plaques may minic many other cutaneous diseases. The exact reason for this "fixed" form of sporotrichosis is unknown, but it has been postulated that isolated cutaneous lesions occur in people who have been sensitized to *Sporothrix schenckii*, without prior clinically apparent disease [9]. Another theory is that the strain of organism found in the cutaneous nonlymphatic form has a lower temperature tolerance than those strains which cause the lymphocutaneous and disseminated infections [12].

Extracutaneous Sporotrichosis

Extracutaneous involvement is rare, but has been reported in the central nervous, genitourinary, gastrointestinal, pulmonary, and musculoskeletal systems. The skeletal system is the most commonly affected extracutaneous site. Bone and joint involvement was found in 85% of patients in a review of 30 cases of extracutaneous infection [23].

Extracutaneous sporotrichosis may be divided into localized, multifocal, or disseminated forms [9]. Localized disease is limited to a particular anatomic site with or without skin involvement. This type of infection may result from direct inoculation, or may occur in the absence of previous trauma or cutaneous disease. Multifocal or systemic sporotrichosis refers to a widely disseminated disease involving multiple organ systems and, in most cases, multiple skin sites [9]. In disseminated infections, there is probably hematogenous spread from primary cutaneous lesions or from involved suppurating lymph nodes. In those cases of disseminated disease not preceded by skin lesions or penetrating trauma, it seems probable that the lung serves as the portal of entry [16]. Because of the low virulence of the *Sporothrix* organism, disseminated sporotrichosis most likely represents an opportunistic infection. Lynch et al. reported 11 deaths in a review of 37 patients who had systemic sporotrichosis [16]. Although many of the deaths were not due specifically to the sporotrichosis infection, the multiple organ involvement was considered a bad prognostic sign.

In most extracutaneous infections, routine laboratory data are normal with the possible exception of a slightly elevated erythrocyte sedimentation rate or white blood cell count. In those patients with disseminated infection, the laboratory data may reflect an underlying disease. Among our cases, only the patient with multifocal sporotrichosis (Case 2) demonstrated leukocytosis.

Sporotrichal Arthritis. The diagnosis of sporotrichal arthritis is often delayed, and there seems to be two major reasons for the delay. First, *Sporothrix schenckii* is an uncommon cause of arthritis and, second, sporotrichal arthritis often occurs in the absence of the clinically familiar cutaneous lesions. In one reported series, the average time from joint symptoms to diagnosis was 25 months [4]. In Bayer's review the range of time before diagnosis was two months to eight years [2].

Sporotrichal arthritis, a disease seen almost exclusively in adult males, has a documented age range of 10 to 77 years [2]. Over 60% of patients in Bayer's series had agricultural occupations, or were brick masons or miners. Knee, wrist and hand, ankle, and elbow joints were the most frequently involved, while the hips and shoulders were usually spared [1, 2, 23].

In the majority of patients, the presenting signs and symptoms are those of an inflammatory synovitis, i.e., pain, warmth, swelling, and restricted motion of the affected joint. In a review of 44 patients, 84% of them presented clinically with an isolated synovitis [2]. In the remaining cases, cutaneous or pulmonary sporotrichosis preceded or occurred simultaneously with the synovitis. Few patients were constitutionally ill or had a low grade fever.

As in our first case, the radiographic appearance of sporotrichal arthritis is typically characterized by joint space narrowing and diffuse osteoporosis. Joint effusion may be present. Juxta-articular erosions and adjacent bony destruction are significant but nonspecific features and may result in intra-articular joint bodies [24].

Sporotrichal arthritis must be differentiated from rheumatoid arthritis and other fungal or granulomatous infections. According to Winter and Pearson the frequent involvement of the small joints of the hands and feet is helpful in differentiating sporotrichosis from other types of infectious arthritis [24]. In some cases diffuse joint space narrowing and periarticular erosions may

mimic rheumatoid arthritis; however, monoarticular involvement, particularly of a large joint, is more in favor of infection than rheumatoid arthritis. The indolent nature of the sporotrichal infection excludes the usual pyogenic organisms but is not unlike the clinical course of other granulomatous infections. Sporotrichal arthritis should be included in the differential diagnosis of a patient with chronic monoarticular or pauciarticular arthritis [4].

Sporotrichal Osteomyelitis. Osseous sporotrichosis is a chronic infection of bone which may appear as an isolated lesion or as part of a disseminated infection. The tibia, fibula, femur, humerus, and short tubular bones of the hands and feet are most commonly involved [8]. In a review of 22 cases by Gladstone and Littman, 68% of the patients had localized swelling, 41% had local sinus tract formation, and 77% had concomitant arthritis. Sixty-three percent of the 22 patients had distant skin lesions [8].

As in our second case, the usual roentgenographic finding in osseous sporotrichosis is destruction of bone without reactive sclerosis. The bone destruction may extend into adjacent joints. Periosteal reaction is usually absent. Unlike tuberculosis, osteopenia may not be a feature. These radiographic changes are not specific for sporotrichosis and may be seen in other fungal infections, tuberculosis, and neoplasms. In most cases of sporotrichal osteomyelitis, there is some roentgenographic evidence of osseous healing within four to five months following the onset of therapy. Clinical improvement usually precedes radiologic healing [8].

Bursal Sporotrichosis. Bursae are subcutaneous, synovial-lined spaces that provide smooth motion between contiguous muscles, tendons, ligaments, and bones. Several of the 140 bursae in the human body are superficial in location and thus are likely to be traumatized [10]. The olecranon bursa and popliteal fossa are common sites of involvement [7, 13, 23]. Because the normal olecranon bursa is a self-contained potential space that does not communicate with the elbow joint, olecranon bursitis is usually not associated with elbow arthritis (Case 3). On the other hand, a popliteal bursitis will be associated with septic arthritis of the knee because of the normal anatomic communication between the two (Case 1). Patients with bursitis present with painful, focal soft tissue swelling and, if a mass is large enough, there is limited range of motion of the adjacent joint. Hyperdistention of the bursa may result in skin necrosis or a draining sinus [10].

As in our third case, the radiologic picture of bursitis is that of focal soft tissue swelling at the site of a bursa. In most instances an adjacent joint would be spared. Bursal calcification is usually not a feature of sporotrichosis and, in fact, has been described in only one patient [23]. In the absence of typical cutaneous lesions, the differential diagnosis includes gout, synovial arthritis, septic bursitis, and hemorrhage [7, 10, 13].

Diagnosis and Treatment

The diagnosis of sporotrichal arthritis, osteomyelitis, or bursitis is usually made following culture of the organism. Microscopic evaluation of pus or a synovial biopsy rarely produces a laboratory diagnosis due to the paucity of organisms [6]. In those cases where an asteroid body is seen the diagnosis of sporotrichosis can be made histologically. Unfortunately, the cigar-shaped asteroid body, an eosinophilic mass that surrounds the organism in tissue and represents an antigen-antibody complex, is rarely found in cases from the United States, Mexico, or Central America [15, 18].

Because biopsy of a lesion can hinder the healing process, it is recommended that material be aspirated from a fluctuant lesion, joint, or bursa and cultured. *Sporothrix schenckii* grows well on glucose-neopeptone (Sabaroud) agar at 25°–30° C. The culture is usually positive within a few days; however, specimens should not be discarded for four weeks since some cultures grow slowly [6].

Serology and skin tests have not been diagnostic by themselves because the frequency of delayed hypersensitivity has been reported to be as high as 50% in certain geographic areas [20]. For this reason a positive skin test is not helpful in diagnosis unless the frequency of the reactions in the local population is known to be low. In all areas, a negative skin test should exclude the diagnosis of sporotrichosis [5].

Therapy for extracutaneous sporotrichosis is not as simple or as successful as the treatment of cutaneous infections which respond well to oral iodides [8]. Intravenous amphotericin B is the drug of choice for disseminated disease. In some cases amphotericin B has also been directly injected into an involved joint space or bursa with some therapeutic success [23]. Treatment with oral potassium iodide alone or in conjunction with surgery is less successful than amphotericin B in extracutaneous infections.

Summary

Sporotrichosis in humans is characteristically a focal granulomatous infection which is easily recognized when it has a lymphocutaneous presentation. Because sporotrichosis is an uncommon cause of arthritis, osteomyelitis, and bursitis, considerable delay in diagnosis of the causitive infectious agent is frequent. This delay may result in inappropriate therapy and persistent morbidity. Monoarticular arthritis and superficial bursitis in patients who, by occupation or hobby, are in contact with soil or vegetation should raise the possibility of sporotrichal infection. Diagnosis is easily established by culture of fluid aspirate or tissue. Treatment with amphotericin B is effective in most cases.

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