CASE REPORTS

Sporotrichal arthritis

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Abstract

Sporotrichal arthritis is a rare disease entity. Diagnosis is often difficult and delayed. Presentation may be either monoarticular or polyarticular. A case of polyarticular sporotrichal arthritis which exemplifies these problems is reported.

Sporotrichosis is a chronic infection caused by *Sporothrix schenckii* which affects the skin and the lymphatic tissue. Dissemination may cause a pneumonitis, osteomyelitis, and, rarely, arthritis. When the joints are affected it may be difficult to diagnose. This report emphasises the difficulties of establishing such a diagnosis.

Case report

A 49 year old male sanitation worker was seen in the rheumatology clinic at Westchester County Medical Center with a history of progressive massive swelling of the right knee, left ankle, left wrist, and left elbow for six months. It had started in the right knee and within a few weeks affected the other joints. Before his clinic visit he had undergone repeated aspirations of the left olecranon bursa. Details of laboratory tests done or drugs received were not available. He had stiffness in the affected joints and a history of alcohol abuse, but there was no history of back, buttock, or heel pain, urethritis, iritis, or diarrhoea. He had a history of hypertension. His mother had rheumatoid arthritis.

An initial examination showed marked swelling and synovial thickening of the left olecranon bursa, right knee, left wrist, and left ankle. He had tenosynovial swelling of the dorsum of the left wrist with rupture of the fourth and fifth extensor tendons. Systemic examination was unremarkable. Laboratory tests showed a sedimentation rate of 130 mm/h and inflammatory synovial fluid with a white cell count of $57 \times 10^9/l$ (degenerated cells), glucose 1.1 mmol/l, protein 43 g/l, and no crystals. Bacterial cultures were negative. Radiographs initially showed effusions and soft tissue swelling with no significant bony changes. A complete blood count, chemistry profile including uric acid, rheumatoid factor, antinuclear antibody, and serum complement concentrations were normal or negative.

The patient was diagnosed as having an inflammatory asymmetrical oligoarthritis, pos-

sibly due to atypical gout or rheumatoid arthritis. There was no response to various non-steroidal anti-inflammatory drugs or colchicine, however. Local injection of the left olecranon bursa with methylprednisolone was complicated by septic bursitis due to staphylococci coagulase negative, which responded to intravenous antibiotics and drainage. Proliferative synovitis of the affected joints worsened and draining sinuses and fistulae on the right knee and left ankle developed. A closed synovial biopsy specimen from the right knee showed chronic inflammatory cells with epithelioid granuloma negative for acid fast bacillus and fungi, and 'characteristic but not diagnostic of rheumatoid arthritis'. A fungal culture from a right knee aspirate 10 months after the first visit and 16 months after the start of symptoms grew *Sporothrix schenckii*, and a diagnosis of sporotrichal arthritis was made. A subsequent review of tissue specimens showed rare organisms. The patient was treated with repeated courses of intravenous amphotericin B. Progression of radiographic changes was noted (figs 1 and 2). Synovectomy of the right knee and left ankle was performed.

Discussion

Sporotrichosis is a chronic infection caused by the ubiquitous saprophyte *Sporothrix schenckii*. Disease occurs by contact with fomites, cutaneous inoculation, and respiratory inhalation. Generally, however, no history of contact is elicited. It is a disease of adult men with outdoor occupations, such as farmers. The usual manifestation is skin ulceration with lymphatic tissue disease (ulceroglandular sporotrichosis). Dissemination may occur to deeper sites causing sporotrichal pneumonitis, osteomyelitis, or arthritis.¹⁻³ Sporotrichal arthritis is rare and occurs with an incidence of 0.03 to 0.04%.^{1,4} It is seen in patients with underlying conditions such as alcohol abuse, myeloproliferative disorders, malignancy, prolonged steroid treatment, etc.^{2,3} Antecedent trauma is also a contributing factor. Sporotrichal arthritis usually presents as an isolated arthritis with no evidence of skin or pulmonary disease.^{1,5} The course is characteristically indolent and constitutional symptoms are rare. Patients do not seek medical attention early and diagnosis is usually delayed (mean interval of disease onset to diagnosis is about 17 months).^{1,2} A striking

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Figure 1 Right knee. (A) Six months after onset of symptoms—soft tissue swelling and joint effusion with no bone changes. (B) Sixteen months after onset of symptoms—increased soft tissue swelling plus small erosions of medial tibia and femur. (C) Twenty four months after onset of symptoms—increased erosions medially plus small erosions laterally and decrease of lateral compartment joint space. Also note subchondral cyst inferior to tibial spines.

Figure 2 Left ankle. (A) Twenty four months after onset of symptoms—soft tissue swelling plus erosions of medial and lateral malleoli and mild narrowing of tibiotalar joint space. (B) Thirty one months after onset of symptoms—partial obliteration of tibiotalar joint space with increased erosions of tibia and fibula and new erosions of talus. (C) Thirty seven months after onset of symptoms—further obliteration of tibiotalar joint space.

feature of sporotrichal arthritis is that the inflammatory synovitis remains confined to the joints initially affected.¹ Single or multiple joints may be affected—most commonly the knees (66%), and hands and wrists (52%). The ankles and elbows are less often affected.^{2,3} Hands and wrists are usually not affected in other fungal arthritides, such as coccidioidal or candidal mycoses.¹

Sporotrichal tenosynovitis of the radial tendon sheath has been reported.^{6,7} Olecranon bursitis has been described^{8,9} but is rare. Reviews of bursitis do not mention sporotrichosis as a cause.^{10,11} Radiographic changes are usually present on initial examination, reflecting the indolent course and the delay in seeking medical attention. Changes include soft tissue swelling and osteoporosis, juxta-articular erosions, and intraosseous lytic lesions.^{12,13} Osteoporosis is seen in a minority of cases.

Joint subluxations, characteristic features of rheumatoid arthritis, are not seen in sporotrichal arthritis. Laboratory tests are normal except for a raised erythrocyte sedimentation rate.² A sporotrichin skin test is positive in 97.3%, but the antigen is not readily available. A sporotrichal agglutination titre of $\geq 1/80$ is considered diagnostic of active sporotrichosis and is also of prognostic value. Decreasing titres indicate

response to treatment.¹ This test also is not readily available.

Typically, histopathological findings are chronic non-caseating granulomatous synovitis^{1,2}; occasionally, caseating granulomas or chronic non-specific inflammatory synovitis is found. Organisms are difficult to see and are often missed, both on Gram stain of synovial fluid and tissue specimens.¹⁻³ Diagnosis requires synovial fluid cultures or tissue cultures. Repeated cultures and prolonged incubation may be needed. Although there is no mortality directly associated with sporotrichal arthritis, morbidity is high and related to delays in diagnosis. Dissemination occurs in some 30% of patients to distal joints, skin, muscles, and subcutaneous tissues.¹ Abscesses and fistulae formation commonly occur.

Sporotrichal arthritis may be treated with one or more courses of systemic antifungal agents such as amphotericin B intravenously with or without debridement.^{1,10} Synovectomies may be required.

Other antifungal agents, such as ketoconazole, have been tried with little success in sporotrichal arthritis.¹⁴ Potassium iodide used for treating cutaneous sporotrichosis has no effect on sporotrichal arthritis.¹⁵

The exact pathogenesis of sporotrichal

arthritis is not known; direct inoculation occurs only occasionally. Haematogenous spread from an extracutaneous point of entry, such as the lungs, is the most likely mechanism. This case illustrates the frustration and difficulties encountered in the diagnosis and management of sporotrichal arthritis.

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