Unusual Presentation of Tuberculous Rheumatism (Poncet's Disease) with Oral Ulcers and Tuberculid

Tuberculous rheumatism (Poncet’s disease) is characterized by a polyarthritus that occurs during acute tuberculous infection in which no mycobacterial involvement can be found or other known cause of polyarthritus detected [1]. Clinically, arthritis is the most important symptom in patients with this disorder [1–3], and there are only a few reports of Poncet’s disease in the English-language literature that describe extraarticular involvement. The manifestations include erythematous eruption [4], urticaria [5], erythema nodosum [6, 7], papulonecrotic tuberculid [8], and conjunctivitis [4]. To our knowledge, oral ulcers have not been reported previously. We describe a woman with Poncet’s disease who presented with oral ulcers and tuberculid.

A healthy 51-year-old woman presented with a 3-week history of painful oral ulcers, rash, fever, night sweats, and weight loss. Two days before admission, she began to have swelling and pain in both knees and ankles. Indomethacin therapy was provided, without relief of her symptoms. Physical examination revealed fever, numerous oral ulcers (figure 1), and scattered erythematous papules of the distal extremities. She had effusion of both shoulders, elbows, wrists, knees, and ankles (all sites of effusion were slightly warm and tender). There was marked left cervical lymphadenopathy. No other physical abnormality was found.

Laboratory studies revealed an erythrocyte sedimentation rate of 120 mm/h. The remainder of the serum chemistry profile was unremarkable. A urinalysis was normal. A Mantoux test, with 5 units of purified protein derivative, was strongly positive (45 mm induration with central necrosis). Findings on radiographs of the chest, shoulders, elbows, wrists, knees, and ankles were normal. Cultures of blood, urine, and stool were negative for bacteria and fungi. Cultures of sputum, urine, CSF, and synovial fluid were negative for acid-fast bacilli. Serology for HLA-B27 antigen was positive. Results of tests for antistreptolysin O, antinuclear antibodies, rheumatoid factor, cryoglobulins, and antibodies to hepatitis B and C viruses, HIV, herpes simplex virus, varicella-zoster virus, and parvovirus B19 were negative, as were results of tests for antibodies to Brucella, Salmonella, Mycoplasma, Shigella, Campylobacter, Treponema, Yersinia, Chlamydia, and Borrelia species. Lymph-node biopsy specimens showed caseating granulomas containing Mycobacterium tuberculosis. Results of a Tzanck test of the oral ulcers were normal. Histological examination of skin-lesion specimens showed a perivascular mononuclear cell infiltration in the dermis and isolated granulomas without caseum in the deep dermis and subcutaneous tissue. Culture of a skin lesion specimen was negative for Mycobacterium species. Treatment was initiated with isoniazid (300 mg/d), rifampin (600 mg/d), ethambutol (1,200 mg/d), and pyrazinamide (1.5 g/d). Treatment with indomethacin was discontinued. The fever, arthritis, and mucocutaneous manifestations subsided over 3 weeks. Biopsy and viral cultures of the oral ulcers were not done because of the simultaneous evidence for active tuberculous lymphadenitis, skin lesions with histopathological characteristics of tuberculid, and the rapid resolution of the oral ulcers and the other clinical manifestations while the patient was receiving antituberculous therapy. It was believed that all the manifestations of this disorder were related to the M. tuberculosis infection. Complete re-

Figure 1. Round ulcer with a central yellowish necrotic base and a surrounding ring of erythema on the inferolateral aspect of the tongue in a female patient with Poncet’s disease.
mission of the cervical lymphadenitis was achieved after 6 months of treatment. The patient remains well after therapy.

This patient had oral ulcers, skin lesions that met the criteria for tuberculid [9], polyarthritis, and a positive Mantoux test, with a temporal relationship with tuberculous lymphadenitis. M. tuberculosis was not isolated from the skin biopsy or synovial fluid specimens. Rapid resolution of her clinical manifestations was obtained with antituberculous therapy. Therefore, the diagnosis of Poncet’s disease was made. Other causes of oral ulcers were considered, but there was no history of herpes labialis in this case, and clinically our patient had aphthous ulcers on relatively distant regions of the oral mucosa, as opposed to the aphthous ulcers produced by herpes simplex, which usually occur as a few clustered lesions on one part of the oral mucosa. Evidence consistent with a viral infection was not found when a Tzanck test was performed. Otherwise, this patient’s condition did not meet the criteria for Behçet’s disease or other rheumatic disorders.

Although the pathogenesis of this patient’s condition is unknown, we suggest that M. tuberculosis induced a cell-mediated immune reaction with inflammation of the oral mucosa, skin, and synovial structures in a patient predisposed genetically to react to infectious agents (supported by the presence of HLA-B27 antigen). It is possible that this patient had a reactive arthritis, with a sterile-articular inflammation in relation to a distant infection. Skin and mucosal lesions are frequent in such patients [10]. Susceptibility to the disease is linked strongly to the presence of HLA-B27 antigen.

In conclusion, oral ulcers and tuberculid may be an unusual presentation of Poncet’s disease in genetically predisposed patients who have HLA-B27 antigen.

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References

Disseminated Mycobacterium avium Complex Infection Associated with Bifocal Synovitis in a Patient with Dermatomyositis

Infections due to Mycobacterium avium complex (MAC) are usually disseminated and occur in immunocompromised patients, mainly HIV-positive patients. We describe synovitis not associated with arthritis due to MAC in a patient being treated for dermatomyositis.

A 61-year-old male was being treated for dermatomyositis with prednisone (12–60 mg daily) and azathioprine. He was a metallurgist and was not exposed to toxins or to animals. In April 1995, a cold swelling appeared on the dorsum of his right wrist. The condition began to worsen in September 1996, and it was accompanied by right ankle swelling. These symptoms did not improve with local steroid injection, and gradually the extensor tendons of the second, third, and fourth digits ruptured. There was neither fever nor clinical evidence of relapse of dermatomyositis.

Laboratory studies revealed the following values: lymphopenia (lymphocytes, 250/mm³; CD4 cell count, 195/mm³; C-reactive protein level, 120 mg/L; and erythrocyte sedimentation rate, 97 mm/h. The total serum Ig level was 15.9 g/L (normal, 8–15 g/L). Articular fluid aspirated from the right wrist contained 95% impaired polymorphonuclear leukocytes and a protein level of 46 g/L. Direct gram staining was negative, and standard bacteriologic cultures were sterile. Ziehl-Nielsen staining showed acid-fast organisms. The patient was HIV negative, and bone marrow was normal. A chest radiograph showed a diffuse miliary infiltrate. Findings on a right wrist radiograph were normal, but right wrist MRI confirmed tendon rupture and tenosynovitis, without arthritis or osteomyelitis. At this juncture, the diagnosis of tuberculous tenosynovitis was considered, and therapy with isoniazid, rifampin, ethambutol, and pyrazinamide was instituted. Tenosynovectomies were performed for the right second, third, and fourth digits, and a splint was applied.

Four weeks later cultures of synovial fluid, the synovial biopsy specimen, and bone marrow specimens were positive for MAC, results suggestive of disseminated MAC infection with bifocal tenosynovitis. The portal of entry remained questionable. Although the MAC infection was diffuse, it might have been first localized to the right wrist and later generalized. When the wrist swelling appeared in April 1995, the synovitis was believed to be related to dermatomyositis, but it may in fact have been the first sign of MAC infection. Once the MAC infection was ascertained, the treatment regimen was modified to include the following: ethambutol (1,200 mg daily), rifabutin (450 mg daily), and clarithromycin.