Case Report

Collagenous Colitis with Spondyloarthropathy Presenting as Fibromyalgia Syndrome

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ABSTRACT. Collagenous colitis is a newly recognized clinicopathologic syndrome that presents with diarrhea and weight loss. In some patients arthropathy may be a concomitant feature. We describe a patient whose initial presentation masqueraded as fibromyalgia with associated bowel symptoms, but who was finally diagnosed as having collagenous colitis and inflammatory spondyloarthropathy. (J Rheumatol 2004;31:1455–6)

Key Indexing Terms:
COLLAGENOUS COLITIS FIBROMYALGIA SERONEGATIVE SPONDYLOARTHROPATHY

Rheumatological syndromes have been associated with a number of gastrointestinal disorders, mostly of an inflammatory nature, with the mechanism of action postulated to be increased bowel permeability to antigenic material[1,2]. Collagenous colitis, a rare and recently described cause of chronic watery diarrhea, characterized by subepithelial collagen deposition and mucosal inflammation[2,3] on colonic biopsy, has rarely been reported to occur with rheumatological disorders[4-7]. We describe a patient who initially presented with a diffuse pain syndrome diagnosed as fibromyalgia (FM), and soon thereafter developed symptoms of collagenous colitis. The final rheumatologic diagnosis was a seronegative spondyloarthopathy.

CASE REPORT

A 48-year-old woman presented with a 2-year history of diffuse body pain that had been diagnosed by her primary care physician as FM. She was treated with non-opioid analgesics as a trial of ibuprofen had been ineffective. Six months after onset of musculoskeletal complaints, she reported severe watery non-bloody diarrhea associated with a 5-kg weight loss over a 3-month period. She underwent a colonoscopy, and the pathology of the colonic biopsy specimen showed the following features compatible with the diagnosis of collagenous colitis: thickened subepithelial collagen layer (thicker than 10 microns), increased intraepithelial lymphocytes, lymphocyte and plasma cell infiltration in the lamina propria, and detachment of surface epithelium above the collagen band (Figure 1). No specific treatment other than antidiarrheal agents was prescribed. Musculoskeletal symptoms persisted and prompted rheumatology consultation.

In addition to diffuse widespread pain, the patient complained of prominent lower and mid-thoracic back pain associated with prolonged morning stiffness, increased pain with immobility and improvement with exercise, as well as considerable fatigue. The report of spinal pain was consistent with inflammatory spinal pain. There were no other symptoms that might have been associated with an inflammatory spondyloarthropathy. Clinical examination revealed diffuse body tenderness but without specific evidence of enthesopathy, arthropathy, or tender points at soft tissue sites. There was marked limitation of spinal mobility at the cervical, thoracic, and lumbar sites. The modified Schober's test was reduced and measured 14 cm of distraction.

Abnormal laboratory tests included: platelet count of 528 × 10^9/l, erythrocyte sedimentation rate by the Wintrobe method 33 mm/h, and C-reactive protein 11 mg/dl. Other laboratory variables including serum chemistry and thyroid function were normal. Tissue typing was negative for MHC HLA-B27 and positive for MHC HLA-B7. Radiographs of the sacroiliac joints showed unilateral suspicious changes, which were confirmed to be erosions compatible with sacroiliitis on computed tomography. Nonsteroidal antiinflammatory drugs (NSAID) gave minimal relief. Treatment was, however, moderately successful with amitriptyline, cyclobenzoprine, and opioid analgesic agents.

DISCUSSION

Seronegative spondyloarthropathy associated with collagenous colitis was the final diagnosis in this patient presenting with diffuse musculoskeletal pain, erroneously labeled as only FM. Features atypical for FM included prominent inflammatory-type spinal pain, prolonged morning stiffness, and limited spinal mobility. Upon further evaluation, we radiographically confirmed the presence of sacroiliitis, consolidating a diagnosis of inflammatory spondyloarthropathy[8]. Prominent spinal pain and stiffness in women who have been labeled with FM should suggest a diagnosis of spondyloarthropathy[9]. This association has only rarely been reported with collagenous colitis[6,7].

Collagenous colitis, a clinicopathologic syndrome first described in 1976, is characterized by chronic watery diarrhea, crampy abdominal pain, normal colonic appearance, and histopathologic colorectal findings of subepithelial collagen deposition, chronic inflammation of the lamina propria, and increased intraepithelial lymphocytes[3,10]. This condition, seen mostly in middle-aged women, has an incidence of about 1.8 cases per 100,000. An association

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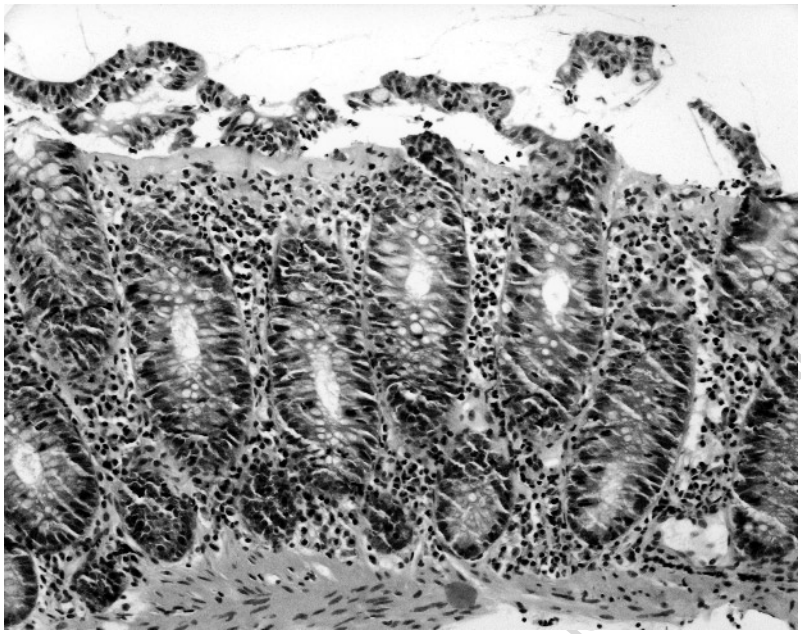

Figure 1. Colonic mucosa with increased chronic inflammation, a thickened subepithelial collagen band, and sloughing of the overlying epithelium (H&E, original magnification 200×).

between the ingestion of NSAID and collagenous colitis has also been suggested, but has not been substantiated[11]. Foreign luminal antigen, possibly bacterial, likely initiates colorectal inflammation leading to immunologic cross-reactivity with an endogenous enterocytic antigen[11]. Treatment is supportive with the use of dietary modifications, antidiarrheal agents, and antiinflammatory drugs. The majority of patients achieve remission with or without treatment.

Collagenous colitis has been associated with a number of autoimmune diseases including thyroiditis, discoid lupus, iritis, Sjögren's syndrome, and myasthenia gravis[4,12]. Since the first report of arthritis and collagenous colitis in 1983[5], there have been additional reports of peripheral inflammatory arthritis: monoarthritis in 2 of 29 patients and polyarthritis in 16 of 163 patients with collagenous colitis[4,12]. Spondyloarthropathy and collagenous colitis have been described in 2 additional patients[6,7]. In view of the small number of patients reported to date, there is limited information on the natural history of this enteropathic arthritis. Both self-limited disease and prolonged destructive disease have been described.

Our patient is of interest as she was initially diagnosed with FM, but both history and examination findings suggested underlying inflammatory spinal process. Collagenous colitis should now be added to the growing list of enteropathic peripheral and axial arthropathies. Whether this reported association is due to an underlying autoimmune disease influencing both the gut and musculoskeletal system, or whether bowel permeability of antigenic material is causative will need to be elucidated. Further studies will need to address the actual frequency and natural history of joint disease in collagenous colitis. This entity, from both the gastrointestinal and the rheumatological perspectives, may be considerably underdiagnosed.

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