

Retroperitoneal Fibrosis Secondary to Spondylodiscitis After Infection with *Prevotella*

CARINE SALLIOT, FRÉDÉRIC LAVIE, ALEXANDRE AZRIA, DIDIER CLERC, ANNE MIQUEL, and XAVIER MARIETTE

ABSTRACT. We report a case of severe retroperitoneal fibrosis (RPF) secondary to lumbar spondylodiscitis caused by infection with *Prevotella* resolving after antibiotic therapy. Infection is an unusual cause of RPF, and infection in such cases with this anaerobic bacterium has never been described. (J Rheumatol 2005;32:957–8)

Key Indexing Terms:

RETROPERITONEAL FIBROSIS

SPONDYLODISCITIS

PREVOTELLA

Retroperitoneal fibrosis (RPF) is a rare disease (prevalence less than 1 in 10,000) characterized by chronic inflammation of the retroperitoneum, leading to fibrosis and severe dysfunction of the surrounding organs including compression of ureters and adjacent blood and lymphatic vessels¹.

The disease is idiopathic in more than 70% of cases. Identifiable causes include use of drugs (certain beta blockers, ergotamine, methysergide, bromocryptine), advanced malignant disease (in the breast, stomach, prostate gland, or lymphoma), radiotherapy, retroperitoneal surgery, vascular prosthesis, and some systemic diseases such as vasculitis. Infectious causes have been described in only a few cases¹. We describe a patient with RPF and spondylodiscitis caused by infection with *Prevotella*.

CASE REPORT

A 62-year-old white man presented with a 4-week history of inflammatory low back pain with intermittent sciatica on the left side and asthenia. He had had psoriasis for 10 years and a long history of non-specific lumbar pain. On examination, he was afebrile, had no weight gain or loss, and had lumbar stiffness (Schöber test result = 10 + 2 cm). Laboratory tests revealed inflammation: erythrocyte sedimentation rate (ESR) of 96 mm/h, C-reactive protein (CRP) of 23 mg/l and hemoglobin level of 10.9 g/dl. The leukocyte count was $12 \times 10^9/l$, and prostatic-specific antigen results were normal, with no evidence of monoclonal immunoglobulins. HLA-B27 and rheumatoid factor were not detected. Radiographs of the lumbosacral spine and sacroiliac joints were normal, but magnetic resonance imaging (MRI) of the spine showed erosive spondylitis of the L5 and S1, without discitis. Inflammatory signals from fat saturation sequences revealed an anterior paravertebral mass.

Biopsies of the L5 and the mass revealed inflammatory tissue with cellular infiltrate made of lymphocytes and macrophages associated with some fibrosis. There were no tumor cells. Cultures for bacteria (including *Mycobacterium*) were negative.

From the Departments of Rheumatology and Radiology, Hospital Bicêtre, Paris XI University, Le Kremlin-Bicêtre, France.

C. Salliot, MD, Rheumatologist; F. Lavie, MD, Rheumatologist; A. Azria, MD, Rheumatologist; D. Clerc, MD, Rheumatologist; A. Miquel, MD, Radiologist; X. Mariette, MD, PhD, Rheumatologist.

Address reprint requests to Pr. X. Mariette, Rheumatology Unit, Hospital Bicêtre, 78 rue du Général Leclerc, 94275 Le Kremlin-Bicêtre, France.

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bacterium) were negative. The diagnosis of spondyloarthropathy was suggested because of the history of psoriasis, chronic lumbar pain, and spondylitis, and the inflammatory patterns of the biopsy, and negative cultures. The patient was treated with a nonsteroidal antiinflammatory drug (NSAID).

Three months later, the patient was readmitted to the hospital because of moderate fever and increased back pain and stiffness despite the NSAID treatment. The inflammation had increased (ESR 125 mm/h, CRP 122 mg/l). Blood and urine tests gave negative results.

Repeat MRI showed an increase of inflammatory vertebral lesions and an extension of the retractile mass towards the retroperitoneum, iliopsoas muscles, iliac vessels, and ureters (Figure 1A). Intravenous urography revealed severe hydronephrosis on the left side caused by external stenosis. A second biopsy of the paravertebral mass showed dense fibrotic tissue with a few inflammatory cells. These morphologic and histologic abnormalities were characteristic of RPF. The cultures were again negative.

No infectious or iatrogenic cause was found, including evidence of tumor on biopsies or thoracic and abdominal computed topography, and we made a diagnosis of idiopathic RPF or RPF associated with spondyloarthropathy as described in the literature.

The patient was treated with oral prednisone (1 mg/kg/day), and a double J stent was inserted in the left ureter.

After an initial clinical and biological improvement (decreased inflammation and fever), the patient was again admitted 1 month later with severe fever, shivers, weight loss and pain in the left lumbocruical area. Examination revealed a weakness of the left psoas and quadriceps muscles. The inflammation had reappeared (ESR 96 mm/h, CRP 232 mg/l). Lumbar MRI revealed L5-S1 spondylodiscitis with multiple abscesses within the fibrosis and psoas muscles, especially on the left side (Figure 1B). Blood and urine cultures again gave negative results. Puncture of the left psoas abscess brought pus, which was positive for *Prevotella spp* (the species could not be identified; it might have been either *P. fragilis* or *P. bivia*). Fibrocolonoscopy and echocardiography results were normal.

The patient was treated with ceftriaxone and metronidazol administered intravenously for 3 weeks, followed by oral amoxicillin-clavulanic acid for 3 months. The left psoas abscess was drained percutaneously, and corticosteroid therapy was gradually reduced. The patient quickly experienced a decrease of pain and disappearance of fever, neurological deficit, and inflammation (CRP 6 mg/l after 15 days' treatment). Further, 2 months after the patient began antibiotic therapy, the abscesses had disappeared, as revealed on MRI, and RPF symptoms were reduced (Figure 1C).

DISCUSSION

The final diagnosis in this case was RPF caused by spondylodiscitis due to infection with *Prevotella*. This anaerobic gram-negative bacillus is a saprophyte of the human mouth and colon responsible for endogenous infections.

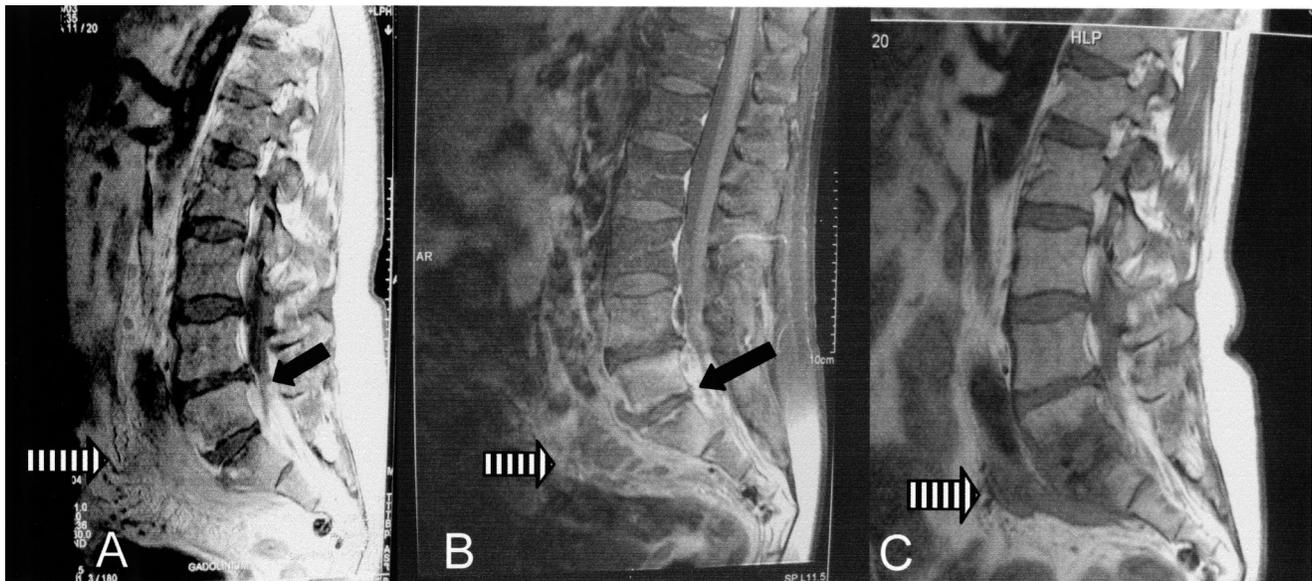


Figure 1. Lumbar magnetic resonance imaging. A. T1 sequence with gadolinium, L5-S1 spondylitis (black arrow) and retroperitoneal fibrosis (black and white arrow). B. T1 sequence, L5-S1 spondylitis (black arrow) with abscesses inside fibrous mass (black and white arrow). C. T1 sequence, with abscesses resolved and fibrosis (black and white arrow) reduced after antibiotic treatment.

We did not find an iatrogenic route of entry for this bacterium: the urine cultures after the JJ stent insertion were negative and moreover *Prevotella* is not an organism found in the urinary tract. The 2 initial negative biopsy results might be explained by the anaerobic pattern, fragility, and slow growth of this bacterium and its need for special medium not used in routine investigations. During biopsy culture, exposure to oxygen probably killed the anaerobic bacteria. Conversely, culture of pus was made in anaerobic conditions that enabled identification.

Investigation of the origin of infection in our case revealed dental treatment a few days before the onset of back pain that could have been a route of entry for *Prevotella*.

The initial improvement of symptoms with prednisone could be explained by the antiinflammatory action of this treatment. Total regression of the RPF with antibiotic treatment is a major point in favor of the infectious origin of this disease.

We found only 4 cases of RPF caused by infection in the literature. Two cases were RPF secondary to Pott's disease² and abdominal ganglionic tuberculosis³, and 2 were due to actinomycosis⁴⁻⁵. Like *Prevotella*, *Actinomyces* is anaerobic and belongs to the commensal flora of the mouth and colon⁶. The first case of RPF with actinomycosis involved a patient with a pelvic fibrotic mass and microabscesses typical of *Actinomyces israeli* infection⁴; the second involved a young woman with a bad dental state who had a retroperitoneal fibrotic mass with skin fistulizations⁵. These 2 patients had hydronephrosis. Both conditions resolved with surgery and 4 months of penicillin treatment.

We initially suspected an association between spondyloarthropathy and RPF, which can show extra-articular fibrotic manifestations of ankylosing spondylitis (AS). This association seems to be exceptional: 17 cases were reported

between 1961 and 2002⁷⁻¹¹, predominantly in men with a mean age of 43 years. In 80% of cases, patients had AS. The 2 diagnoses are often made simultaneously (56%). Until now, the relation between the diseases remains speculative.

As illustrated by our case, caution is necessary before linking RPF to spondyloarthropathy. Treating RPF with antibiotics could be effective in cases of exceptional infectious origin.

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