Case Report

Bilateral Giant Iliopsoas Bursitis Presenting as Refractory Edema of Lower Limbs

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ABSTRACT. A 69-year-old man with rheumatoid arthritis presented with bilateral leg swelling. Magnetic resonance studies revealed bilateral giant iliopsoas bursitis with intrapelvic expansion and compression of pelvic vessels and bladder. The case was refractory to intensive systemic and local medical treatment. (J Rheumatol 2004;31:1452–4)

Key Indexing Terms: ILIOPSOAS BURSITIS LEG SWELLING RHEUMATOID ARTHRITIS

Iliopsoas bursitis is an unusual clinical event that has been described in cases of rheumatoid arthritis (RA), osteoarthritis, microcrystalline arthropathies, infections, avascular necrosis, trauma, pigmented villonodular synovitis, and synovial chondromatosis. In general it is unilateral, and presents as a mass anterior to the hip joint, behind the psoas-iliac muscle, and it can extend through the inguinal arch to the minor pelvis. Clinical manifestations that it commonly causes are pain and palpable mass at the inguinal area. We describe a case of bilateral giant bursitis in a patient with RA that extended to the abdominal cavity and manifested as refractory edema in lower limbs.

CASE REPORT

A 69-year-old man presented with swollen legs of 2 years’ duration. He had a history of seropositive RA since 1989, and in spite of treatment with several disease modifying antirheumatic drugs (DMARD) during this period (gold salts, sulfasalazine, methotrexate, cyclosporine combined with methotrexate, and leflunomide) he never had satisfactory clinical response. He was admitted in 1996 because of massive popliteal cyst in the right leg, which dissected calf muscles to the ankle and was complicated by deep venous thrombosis.

In May 2000, he noted bilateral diffuse swelling of his lower limbs. His internist attributed the edema to postphlebitic syndrome in the right leg and bilateral varicosities. The swelling worsened in March 2002, and he was treated with support stockings, leg elevation, and diuretics, with no improvement. He began to experience diffuse pain in both legs, but never complained of groin pain. He had no erythema or warmth of his lower extremities.

After 6 months he came to us with massive edema in both legs, mainly in the right side. Treatment for RA in the last 18 months included leflunomide 20 mg and prednisone 7.5 mg daily. Examination was remarkable for pitting edema of both legs. He had synovitis in small joints of hands and wrists, consistent with RA. There were no palpable masses in the pelvic or inguinal region, and hip movements were complete and painless.

A lumbar roentgenogram was normal. Hip radiograph disclosed joint space narrowing and erosions related with RA. Erythrocyte sedimentation rate was 57 mm/h. Complete blood count and routine biochemistry were normal.

Computed tomography (CT) and ultrasonography of the pelvis and upper legs revealed a fluid collection inside the iliopsoas bursa in the right side (20 × 12 × 11 cm), extending from the internal proximal soft tissues of the thigh to retroperitoneum in the iliac fossa, associated with the iliopsoas muscle. It produced compression in the iliac vessels and bladder. On the left side another similar cyst (18 × 12 × 9 cm) was visible, which also extended to the pelvic cavity and compressed the iliac vessels and bladder.

A magnetic resonance imaging (MRI) study showed bilateral hip effusion and enlarged bursa of the iliopsoas muscle, filled with pannus and synovial fluid, with intrapelvic extension (Figure 1). A communication between the iliopsoas bursa and the hip joint was seen in both sides.

Ultrasound-guided drainage was performed in the right pelvic cyst, yielding more than 1000 ml of straw-colored fluid in the next 10 days. Laboratory analysis revealed a cell count of 2500/mm³ with 65% segmented neutrophils. The protein was 4.9 g/dl and glucose 10 mg/dl. Gram’s stain, tuberculous, fungal and bacterial cultures, crystal analysis, and cytology were all negative.

The patient underwent systemic steroid therapy with 30 mg per day prednisone. The edema of the legs disappeared at Day 12 after drainage. Leflunomide was stopped and treatment with infliximab (3 mg/kg on Days 0, 14, and 45 and every 2 months thereafter) and methotrexate (7.5 mg/wk) was started.

Two months after the initial aspiration he returned to hospital because of marked edema of the right leg. On echography and MRI, the size of the right mass was seen to be the pre-drainage measurements, and the left mass was clearly decreased in volume. A second intrapelvic aspiration of the right iliopsoas bursa was performed and drainage was maintained for 12 days, yielding 1600 ml fluid overall. At the end, 80 mg of triamcinolone acetonide was injected into the right cystic cavity.

Fifteen days later, the bursal fluid of the right side had reaccumulated, and because of problems of venous stasis and because the patient was very uncomfortable, a complete surgical excision of the right iliopsoas bursa was performed. Histopathology of the resected cyst exhibited changes compatible with hypertrophic rheumatoid synovitis.

Since then the evolution of the case has been satisfactory. Treatment was stable with infliximab (3 mg/kg every 2 mo), methotrexate (7.5 mg/wk), and because of problems of venous stasis and because the patient was very uncomfortable, a complete surgical excision of the right iliopsoas bursa was performed. Histopathology of the resected cyst exhibited changes compatible with hypertrophic rheumatoid synovitis.

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mg/wk), and prednisone (5 mg/day), and there was no further edema in the legs over 8 months of followup.

**DISCUSSION**

The iliopsoas bursa is a $6 \times 3$ cm structure that lies between the anterior surface of the hip joint and the iliopsoas muscle. While the iliopsoas bursa communicates with the joint in 15% of healthy adults, a communication has been documented in most reported cases of bursitis.$^{6,7}$

In patients with RA several cases of swelling of the iliopsoas bursa have been described. It may extend above the inguinal ligament to the point of entering the pelvic cavity.
cavity\textsuperscript{7,8}. In most cases it is asymptomatic or presents with groin pain, snapping sensation, and palpable mass in the inguinal area\textsuperscript{9,10}. Not exceptionally it causes compression on crural vessels and nerves, giving rise to leg edema or entrapment neuropathy in the affected side\textsuperscript{1,6,11}.

Ultrasonography and CT are reliable techniques for diagnosis of this entity, but MRI is the most adequate means of diagnosis, providing an exact anatomic delimitation and revealing the fluid content of the cyst\textsuperscript{2,10}. It is also possible by this means to visualize a communication between the hip joint and the iliopsoas bursal collection\textsuperscript{4,7}.

The interesting aspect of our case is the unusual presentation of this rare condition. We found no report of a case of iliopsoas bursitis with intrapelvic bilateral extension of this size. It was remarkable that there was absence of both local pain and palpable mass. The histological findings in the iliopsoas bursa were diagnostic of chronic synovitis.

Others have reported a good response of psoas bursitis to RA treatment with DMARD and systemic corticosteroids, as well as to local infiltration with long-acting corticosteroids\textsuperscript{2,9,12}. Unfortunately, our patient did not respond to treatment with infliximab at doses of 3 mg/kg/day on Days 0, 14, 45 in combination with methotrexate 7.5 mg/wk, and both local and systemic corticosteroids; this made it necessary to undertake surgical excision at 2.5 months after the biologic therapy was started.

From this case we learn that iliopsoas bursitis in RA can be overlooked until reaching a considerable size, and it can be refractory to local and systemic treatments for RA. One should maintain a high index of suspicion for it when dealing with unexplained persistent lower extremity edema or pelvic pathology in the setting of established RA.

REFERENCES