INTERNAL MEDICINE

□ CASE REPORT □

RS3PE Syndrome with Iliopsoas Bursitis Distinguished from an Iliopsoas Abscess Using a CT-guided Puncture

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Abstract

A 55-year-old man was diagnosed with remitting seronegative symmetrical synovitis with pitting edema (RS3PE) syndrome. Contrast-enhanced computed tomography for cancer screening showed a mass with low-density centers with an enhanced rim in the left iliopsoas muscle. We suspected an iliopsoas abscess and performed computed-tomography-guided puncture of the mass. Both Gram staining and the culture of the fluid were negative. We diagnosed the patient with RS3PE syndrome with iliopsoas bursitis and administered low-dose corticosteroids without antibiotics. The symptoms, including left hip pain, quickly disappeared following treatment. Clinicians should be aware that iliopsoas bursitis may resemble an iliopsoas abscess. As a result, it is important to make an accurate differential diagnosis.

Key words: remitting seronegative symmetrical synovitis with pitting edema syndrome, iliopsoas abscess, iliopsoas bursitis, iliopectineal bursitis, polymyalgia rheumatica

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Introduction

Remitting seronegative symmetrical synovitis with pitting edema (RS3PE) syndrome is characterized by symmetrical synovitis with pitting edema on the dorsum manus and the absence of rheumatoid factor (1). Although the sudden onset of polyarthritis associated with prominent distal symptoms is one of the characteristics of RS3PE, proximal symptoms, such as pain in the shoulder, hip, thigh, neck and torso [as in polymyalgia rheumatica (PMR)], are also known to emerge (2). We herein describe a case of RS3PE syndrome with iliopsoas bursitis in which computed tomography (CT)-guided puncture was necessary to distinguish it from an iliopsoas abscess.

Case Report

A 55-year-old man was admitted to our hospital complaining of shoulder pain and hip joint pain that had continued for the previous 2 months. He had occasional episodes of a low-grade fever in the month prior to this admission. His previous physician diagnosed him as having frozen shoulder and prescribed a non-steroidal anti-inflammatory drug. However, his symptoms worsened. For two weeks previous to being admitted he could not toss about in bed. Swelling of the bilateral dorsum manus emerged a week before this admission. The swelling was accompanied by morning stiffness and an impaired grip function.

The patient was previously healthy. His past medical history included lumbar disc herniation and a smoking history of 20 cigarettes a day for 35 years. On the physical exami-

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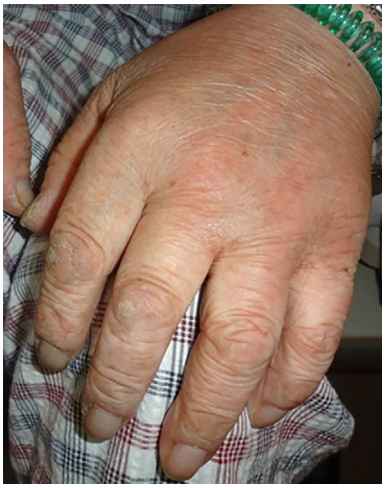

Figure 1. **The patient's dorsum manus was swollen.**

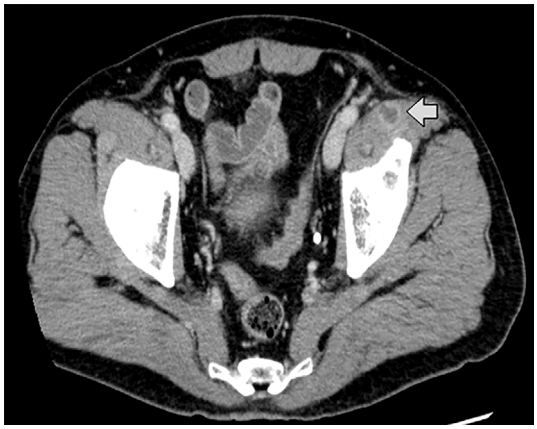

Figure 2. **CT shows a 2 cm mass with enhancement in the left iliopsoas muscle. The mass contained homogenous fluid (arrow).**

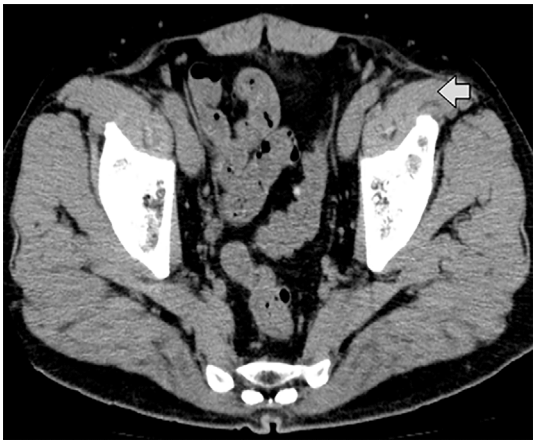

Figure 3. **Three months following admission, the mass with enhancement in the left iliopsoas muscle had disappeared (arrow).**

nation, his body temperature was 37.3℃ and all other vital signs were within the normal limits. His right shoulder and left hip joint showed tenderness. He also had swollen bilateral dorsum manus (Fig. 1), which showed a remarkable pitting edema. The finger joints had a reduced range of motion with reduced grip strength and hand opening ability. Both the metacarpophalangeal and proximal interphalangeal joints showed swelling and tenderness. However, the dorsum pedis showed no edema and there was no tenderness of the temporal arteries. The cardiopulmonary and abdominal findings were normal.

The laboratory data included the following: white blood cell count, 7,000/μL; C-reactive protein (CRP), 3.25 mg/dL (normal range: <0.3 mg/dL); serum creatinine, 0.67 mg/dL; matrix metalloproteinase 3, 194.5 ng/mL (normal range: <121 ng/mL); and rheumatoid factor (RF), anti-cyclic citrullinated peptide antibody, antinuclear antibody, myeloperoxidase antineutrophil cytoplasmic antibody (MPO-ANCA) and proteinase 3 antineutrophil cytoplasmic antibody (PR3-ANCA) negative. The serum vascular endothelial growth factor (VEGF) level was 2,884 pg/mL. A superficial ultrasonography examination showed tendinitis of the extensor of the fingers and caput longum musculi bicipitis brachii. Joint synovitis of both the metacarpophalangeal joints and proximal interphalangeal joints was not prominent. Magnetic resonance imaging (MRI) of the right shoulder showed tendinitis of the caput longum musculi bicipitis brachii and subcoracoid bursitis. We therefore diagnosed the patient to have RS3PE syndrome according to the classification criteria (3). Because RS3PE syndrome has recently been recognized as a potential paraneoplastic syndrome, we searched for malignancy. Contrast-enhanced computed tomography (CECT) showed a 2 cm mass with low-density centers with an enhanced rim in the left iliopsoas muscle which contained homogenous fluid (Fig. 2). CT indicated no fluid in the left hip joint. It was not clear whether the mass with an enhanced rim had a connection to the hip joint. Because these findings suggested an iliopsoas abscess, we performed aspiration of the mass guided by CT in order to drain the mass of fluid and test the fluid; 3 mL of yellowish translucent fluid was collected. The leukocyte count of the fluid was 22,000/mm^3 with 74% of polymorphonuclear leukocytes. Crystals were absent from the fluid. Gram staining of the fluid was negative, and a culture failed to grow any organisms. According to these results, we diagnosed the patient with RS3PE syndrome with iliopsoas bursitis. We initiated a daily 15 mg dose of prednisolone. His right shoulder and hip joint symptoms rapidly improved and the level of CRP declined. Three months later, CT showed that the mass in the left iliopsoas muscle had vanished (Fig. 3).

Discussion

We herein reported a case of RS3PE syndrome that was complicated with iliopsoas bursitis. It was difficult to distinguish iliopsoas bursitis from an iliopsoas abscess and a CT-guided puncture was necessary.

The relationship between RS3PE syndrome and PMR is controversial because both diseases share many features.

Both occur in the elderly with abrupt onsets and have symmetric manifestations and good responses to low-dose glucocorticoid therapy (3). These similarities suggest that these two conditions may belong to the same entity, with RS3PE syndrome being a distinctive clinical pattern of PMR characterized by the development of edema.

We diagnosed the patient with RS3PE syndrome because his symptoms fulfilled the criteria suggested by Olive et al. (3). The criteria include the following: (1) bilateral pitting edema of both hands; (2) a sudden onset of polyarthritis; (3) age >50 years; and (4) seronegativity for RF. Additionally, Cantini et al. suggested that extensor tenosynovitis may be responsible for the edema in the subcutaneous and peritendinous soft tissue of the dorsum in RS3PE syndrome (4). Our patient also had extensor tenosynovitis confirmed by the ultrasonographic findings. The serum VEGF level has been reported to be useful for the diagnosis of patients with RS3PE syndrome (5). Our case also had a high level of VEGF.

The iliopsoas (or iliopectineal) bursa is the largest synovial bursa in the body, and it is present bilaterally in 98% of adults. Interposed between the iliopsoas muscle and the anterior capsule of the hip, this structure averages 6 cm by 3 cm in size (6). There is communication between the iliopsoas bursa and hip joint in 15% of adults (7). Aging and abnormalities of the hip joint lead to further communication between the iliopsoas bursa and hip joint (8). MRI is often useful for evaluating whether there is communication between the iliopsoas bursa and hip joint (9). The differential structures of the iliopsoas bursa include iliopsoas abscess, femoral or inguinal hernia, lymphoma or another neoplastic disease, lymphadenopathy, undescended testicle, and vascular abnormalities, such as femoral vessel aneurysm (10).

Inflammation in the iliopsoas bursa is called iliopsoas bursitis and is caused by bacteria (11) and crystals (12). Additionally, rheumatic diseases, including rheumatoid arthritis (RA) (13) and PMR (14), cause iliopsoas bursitis. In PMR, 10 of 20 patients had iliopsoas bursitis confirmed by MRI findings. To the best of our knowledge, the present case is the first case of RS3PE syndrome complicated with iliopsoas bursitis.

The radiological findings cannot always distinguish infection from non-infectious inflammation. For example, the presence of a gas bubble in the iliopsoas area does not always suggest infectious disease (15). However, our patient had both subcoracoid bursitis and tendinitis of the caput longum musculi bicipitis brachii. If the mass with low-density centers with an enhanced rim was an iliopsoas abscess or bacterial iliopsoas bursitis, subcoracoid bursitis and tendinitis would be assumed to occur via sepsis or bacteremia. Sepsis and bacteremia were unlikely because a general status was sufficiently maintained in the present.

It is reasonable to suspect that these cases of bursitis and tendinitis were caused by systemic diseases such as RS3PE syndrome and PMR. The differentiation of bursitis and tendinitis resembles the differentiation of monoarthritis and polyarthritis. Monoarthritis is thought to be caused by septic arthritis and crystals that induce arthritis, but polyarthritis is thought to be caused by systemic diseases like RA.

In the present case, only the pain in the left hip worsened after the patient's admission, which suggested the presence of an iliopsoas abscess. If a patient presents without bursitis or arthritis except in the mass with low-density centers with an enhanced rim in the iliopsoas muscle, then puncturing is always necessary. Conversely, if the pain does not worsen, the mass with an enhanced rim should be considered to be iliopsoas bursitis accompanied by RS3PE syndrome and the response to corticosteroid therapy must be carefully monitored.

In conclusion, we herein described a case of RS3PE syndrome with iliopsoas bursitis. It is useful to know that in patients with RS3PE syndrome iliopsoas bursitis may resemble an iliopsoas abscess in the imaging findings.

The authors state that they have no Conflict of Interest (COI).

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