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Case Report

Case of Polymyalgia Rheumatica Misdiagnosed as Infectious Spondylitis

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A 60-year-old woman visited the authors' clinic with low back pain and arthralgia. Her symptoms had occurred 6 months previously, and she was treated with an epidural injection and a balloon dilatation procedure based on the assumption of spinal stenosis, but both treatments were ineffective. Her low back pain was aggravated, accompanied by fever and chills over a period of 4 months. As a result, she visited another referral hospital and was diagnosed with infective spondylitis associated with the invasive procedure. Her symptoms improved with antibiotics, but they recurred. When she visited our clinic, she still had continuous low back pain and febrile senses. Magnetic resonance imaging of her lumbar spine revealed interspinous bursitis, and 18 F-fluorodeoxyglucose positron emission tomography showed multifocal synovial inflammation. She was diagnosed with polymyalgia rheumatica and treatment was started on prednisolone and celecoxib. Her symptoms improved dramatically and the inflammatory markers normalized. **(J Rheum Dis 2018;25:140-143)**

Key Words. Polymyalgia rheumatica, Back pain, Arthralgia, Positron emission tomography computed tomography

INTRODUCTION

Polymyalgia rheumatica (PMR) is a chronic inflammatory disease that affects people older than 50 years of age. PMR is more commonly reported in Caucasians and females, and is relatively uncommon among Asian, African-American, and Latino populations [1]. In Korea, PMR is a somewhat rare disease, and the annual incidence rate of PMR during 5 years was estimated to be 2.06 per 100,000 individuals over 50 years of age [2]. The clinical symptoms of PMR are aching and morning stiffness of the shoulders and/or pelvic girdle. The muscles supporting the neck can also be involved, but myalgia of the torso is less common. These symptoms are usually symmetric [3]. The cause of PMR is still unknown, and even the anatomical site of the inflammation is unclear. Both environmental and genetic factors appear to play a role [3]. Diagnosis of PMR is based on clinical features and established criteria but there is no pathognomic test [4]. Because of its inflammatory features and localized musculoskeletal pain, patients with PMR tend to be diagnosed as having an infection. In this report, we describe a case of PMR presented with low back pain with fever, misdiagnosed as infective spondylitis.

CASE REPORT

A 60-year old woman visited our clinic for low back pain accompanied by mild pain in both knees and the neck. Her symptoms first occurred 6 months before visiting our hospital. At first she had visited a private clinic and had undergone magnetic resonance imaging (MRI) of the lumbar spine as part of her medical evaluation. She had been treated with epidural injection and balloon dilata-

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tion procedure based on a diagnosis of spinal stenosis. However, this treatment was ineffective and her symptoms became aggravated and persisted, accompanied by fever and chills over a period of 4 months.

She had visited another referral hospital and was diagnosed as having infectious spondylitis that was a result of the invasive procedure for her low back pain and mild fever. Although culture results of soft tissue around the spine were negative, she had been suspected as having an infection because of her low-grade fever, high levels of inflammatory markers and history of local injection. Hence, she was treated with empiric antibiotics and non-steroidal anti-inflammatory drugs (NSAID) for 1 month. Her symptoms gradually improved but recurred after the cessation of those treatments.

At that point, she was referred to our hospital. When we first saw her, she could not sit up in her bed because of low back pain. Our physical examination found there was tenderness in the low back, both shoulders, and in the buttocks, knees, and ankles. The movement of affected joints was limited by pain. She had a fever of 38.0°C, while other vital signs were stable: blood pressure was 100/60 mmHg, heart rate 72 beats per minute, respiratory rate 18 breaths per minute. A blood test revealed increased levels of inflammatory markers of C-reactive protein (CRP) with 3.4 mg/dL, and ESR with 114 mm/hour. However, her white cell count was in the normal range (7,200/mm^3) and her procalcitonin was normal (< 0.05 ng/mL). The results from two blood cultures were also negative.

To rule out other inflammatory rheumatologic conditions, we performed laboratory tests. Although anti-nuclear antibody was positive (1:160 with nucleolar pattern), all of the anti-extractable nuclear antigens were negative. For the tests for rheumatoid arthritis (RA), rheumatoid factor (RF) and anti-cyclic citrullinated peptides (anti-CCP) antibody were negative. Joint fluid aspirated from her knees also revealed no inflammatory conditions; white blood cell count was 390/mm^3 (neutrophil dominant), test for crystals was negative and blood culture was negative for bacteria and tuberculosis.

After evaluation with a simple X-ray to check her chest and spine, we performed an enhanced CT scan on her chest and abdomen to rule out any hidden malignancy. They revealed that there was no evidence of malignancy or infection in the internal organs.

Her MRI showed diffuse enhancement along capsule and adjacent soft tissue at the bilateral facet joint of L1-L5 (Figure 1A), interspinous bursitis at T12-L5 on T1 weighted image (Figure 1B) and broad-based disc protrusion in L3-5. There was no evidence of infection such as vertebral body destruction, abscess, enhanced vertebra etc. Those findings were considered to be consistent with PMR rather than infectious spondylitis or other types of inflammatory arthritis. For the additional information for PMR and malignancy, we performed 18 F-fluorodeoxyglucose (FDG) positron emission tomography integrated with computed tomography (PET/CT). Multiple inflammatory bursitis in pubic symphysis (Figure 2A) and both ischiogluteal, and trochanteric bursa (Figure 2B) were revealed. In addition, increased FDG uptakes were noted at the C6-7, T12-L5 level inter-spinous ligament (Figure 2C) and arthritis on facet joints of L2-3 and L3-4 was revealed (Figure 2D).

Her symptoms, including low back pain and mild fever, dramatically improved after administration of 10 mg of prednisolone (PDS) and 400 mg of celecoxib per day was started on her second day in our hospital. After 4 days of treatment, CRP was normalized and ESR decreased to 78

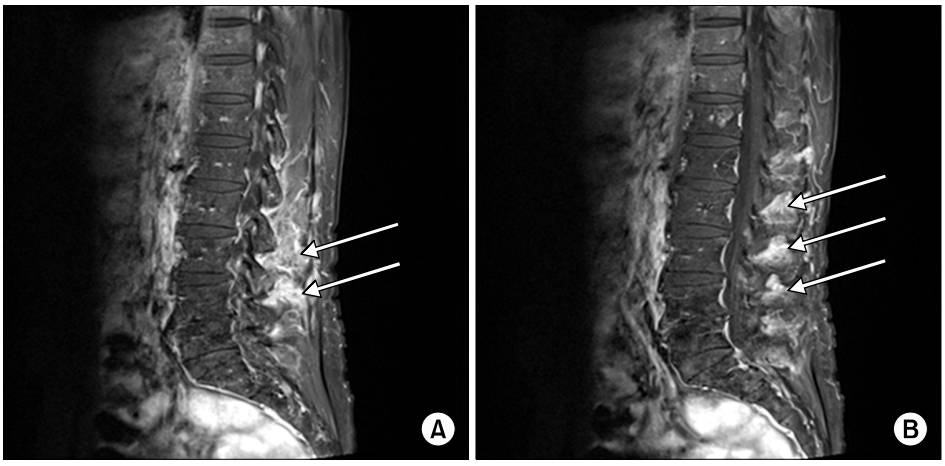

Figure 1. L-spine magnetic resonance imaging. (A) Diffuse enhancement along capsule and adjacent soft tissue at bilateral facet joint of L1-L5, (B) interspinous bursitis at T12-L5 on T1 weighted image.

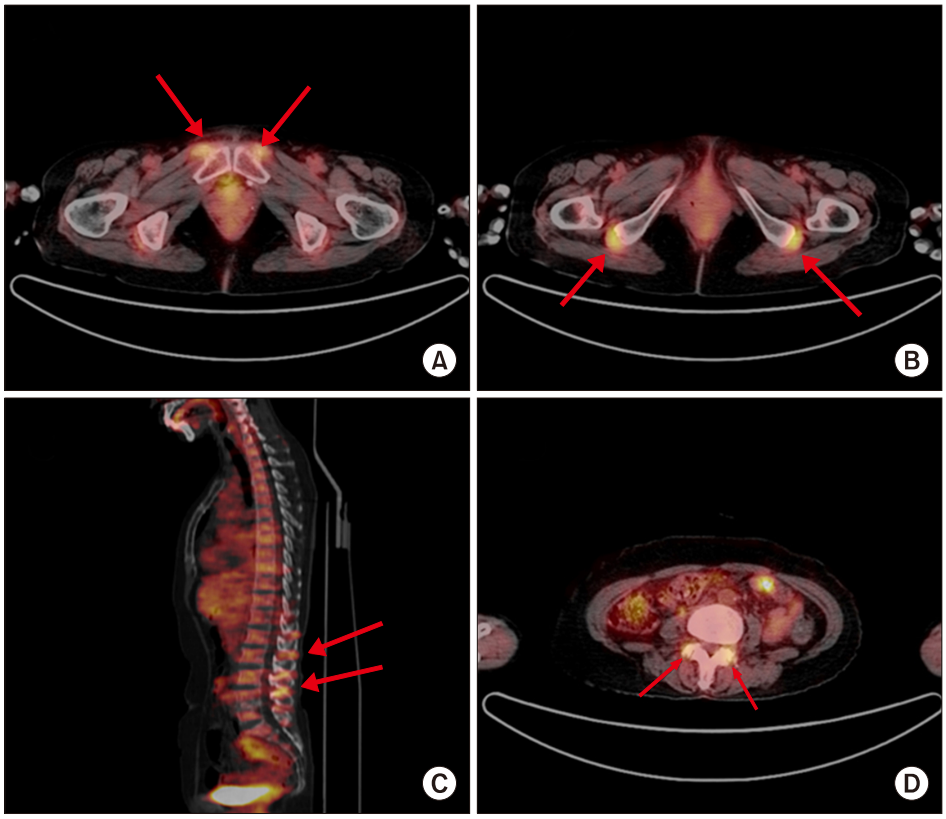

Figure 2. Whole body 18-fluorodeoxyglucose (FDG) positron emission tomography integrated with computed tomography scan. (A) Multiple inflammatory bursitis in pubic symphysis and (B) both ischiogluteal, trochanteric bursa were revealed. In addition, increased FDG uptakes were noted at the (C) C6-7, T12-L5 level inter-spinous ligament and arthritis (D) on facet joints of L2-3 and L3-4 was revealed.

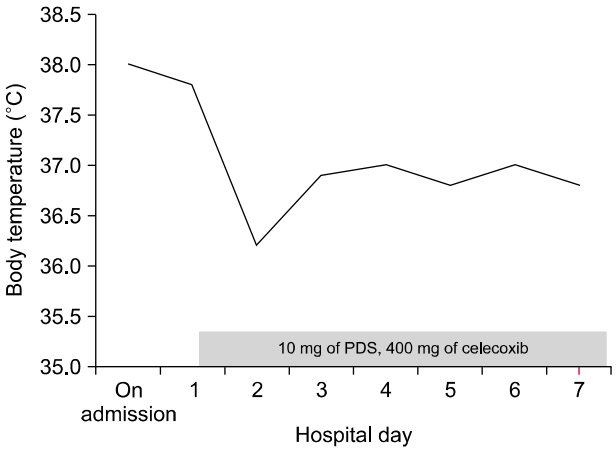

Figure 3. Clinical course of patient. PDS: prednisolone.

mm/hour. After 4 months, her symptoms were improved and there were no additional flare ups, so daily PDS was reduced to 2.5 mg (Figure 3).

DISCUSSION

Because of the absence of a precise pathognomonic test for PMR, many clinicians can misdiagnose it as other inflammatory conditions. In our case, despite several clinical clues suggesting PMR, the history of epidural injection and fever made it more difficult to diagnose her as suffering from PMR. Her partial response to NSAIDs and antibiotics led clinicians to misdiagnose her condition as an infection. However, our detailed medical history review and a careful interpretation of imaging studies make it possible to reach a correct diagnosis.

The diagnosis of PMR is made primarily on clinical grounds. The classification criteria have been proposed by the European League Against Rheumatism and the American College of Rheumatology (EULAR/ACR) as a research tool to identify patients with PMR [4]. However, there remains a need for additional tests to provide diagnostic information, particularly where the diagnosis is not clear. Notably, there is considerable overlap between PMR and seronegative RA in older adults who present with symmetric synovitis.

Our case displayed marked symptoms of PMR according to the EULAR/ACR criteria, but it takes 6 months to be diagnosed as PMR: ≥50 years, both hip and shoulder pain in addition to low back pain and an abnormal CRP or ESR, morning stiffness ≥45 minutes, pain or limited range of motion at the hip, absence of RF or ACPA, and symptom improvement after low dose glucocorticoid

treatment. This shows the difficulties in the early suspicion and diagnosis of PMR.

Although imaging techniques are not the gold standard for the diagnosis of PMR, some modalities such as ultrasonography (US), MRI, and PET/CT can be used to detect underlying inflammation in patients with PMR [5]. Of these approaches, ultrasound is the first choice for the diagnosis of PMR due to its wide availability and recent improvements in the technology. In PMR patients, US can demonstrate synovitis, particularly in proximal joints and subacromial-subdeltoid bursitis. However, since US is considered to be an operator-dependent technology with poor repeatability, a more objective imaging technique has been required.

PET scanning can identify bursitis as well as underlying vascular involvement [6]. A recent study reported that interspinous bursitis is common in patients with PMR, but is not associated with spinal pain. In addition, the most frequent site of spinal pain in patients with PMR was the cervical portion rather than the lumbar spine [7]. Lesions in the spinous processes are more frequently detected with FDG-PET/CT than with MRI. In one previous study, 71% of patients with PMR showed increased FDG uptake in the vertebral spinous processes, while MRI detected only 20% of the corresponding lesions [8]. Salvarani et al. [9] suggested that inflammation of the lumbar bursae may be responsible for the low back pain reported by patients with PMR. This finding was also revealed in this case, in which the patient suffered from low back pain and there was multiple inflammatory bursitis at several C, L-spine level and arthritis on facet joints of L-spine revealed by PET/CT scans. Hence, the FDG-PET/CT scan is thought to be useful in diagnosing PMR by revealing significant uptake in articular and extra-articular sites such as arterial involvement [5,10,11].

In this case, we learned how low back pain can be treated with an injection, and how the subsequent elevation of the inflammatory markers makes diagnosis difficult. The lack of a pathognomonic test to identify PMR can also create confusion. However, it is reasonable to suspect PMR in elderly patients with low back pain and shoulder pain. In addition to careful history taking, some imaging studies such as FDG-PET/CT scan might be helpful in diagnosing PMR in its early stage.

SUMMARY

When patients suffer from low back pain accompanied with the symmetric involvement of other large joints, PMR should be included in the differential diagnosis. In addition to careful history taking, FDG-PET/CT scan can be helpful in diagnosing PMR in the early stage.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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