A Case of Acute Calcium Pyrophosphate Crystal Arthritis in the Lumbar Facet Joint

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Calcium pyrophosphate dihydrate deposition disease most commonly presents with acute arthritis of the peripheral joints. Infrequently, a mass effect of this disease can cause axial symptoms, such as spinal stenosis, radiculopathy, or myelopathy. Herein, we report on the first Korean case of acute arthritis in the lumbar facet joint due to calcium pyrophosphate dihydrate crystal deposition disease. A 73-year-old female presented with acute fever, severe lumbago, and knee arthralgia, 11 days after partial parathyroidectomy. Plain radiographs showed multiple chondrocalcinosis, while a bone scan, computed tomography, and magnetic resonance imaging showed right L5-S1 facet arthritis. In synovial fluid from the facet and knee joints, positively birefringent calcium pyrophosphate dihydrate crystals were observed under polarized light microscopy. Under the diagnosis of acute calcium pyrophosphate dihydrate crystal arthritis (formerly known as ‘pseudogout’) in the facet joint, an intra-articular triamcinolone injection was administered, which resulted in dramatic improvement of the symptoms within 24 hours. (J Rheum Dis 2016;23:125-129)

Key Words. Calcium pyrophosphate dihydrate deposition, Chondrocalcinosis, Zygapophyseal joint, Low back pain

INTRODUCTION

Calcium pyrophosphate dehydrate deposition (CPPD) disease is a crystal-induced arthropathy with several clinical presentations including asymptomatic CPPD, osteoarthritis with CPPD, acute calcium pyrophosphate (CPP) crystal arthritis (formerly called ‘pseudogout’), and chronic CPP crystal inflammatory arthritis, according to the European league against rheumatism (EULAR) recommendations [1]. Aging and osteoarthritis have been known as major risk factors for CPPD disease. But, if CPPD disease develops in individuals younger than 45 years of age, associated metabolic diseases should be considered because hyperparathyroidism, hemochromatosis, and hypomagnesemia are high risk factors for CPPD [1,2]. In addition, familial CPPD disease can occur in a young individual [2].

Acute CPP crystal arthritis presents typically as an acute monoarticular or oligoarticular arthritis and commonly involves large peripheral joints such as the knees, wrists, and ankles [1,2]. It is characterized by an abrupt onset of severe pain and swelling with periarticular erythema and can produce systemic symptoms such as fever, chills, and malaise. Such inflammatory attacks tend to be triggered by intercurrent medical or surgical conditions, and it is important to differentiate between infectious arthritis and acute CPPD disease in clinical practice [3].

Spinal CPPD disease has been reported rarely, and most of the cases present with symptoms related to a mass effect, for instance, foramen magnum syndrome, spinal stenosis, radiculopathy, myelopathy, synovial cyst, or cauda equina syndrome [2]. However, fewer cases of spinal CPPD disease with acute low back pain have been reported [4-11]. We herein report a case of acute CPP crys-
tal arthritis in the lumbar facet joint, which presented with fever and acute severe lumbago.

**CASE REPORT**

A 73-year-old woman with childhood deafness of undetermined cause visited the department of emergency of Seoul National University Bundang Hospital because of pain in the left lower extremity. Over 10 years or more, she had taken oral medications intermittently and received intra-articular injection for the treatment of non-inflammatory pain in both knee joints. She had been diagnosed with chronic kidney disease (stage 4) during an investigation for generalized weakness and anorexia 8 months ago. Two months ago, she visited another hospital due to generalized pain. Because the patient had hypercalcemia and elevated serum level of parathyroid hormone, she was referred to our hospital. Parathyroid scintigraphy and neck ultrasonography showed an increased uptake or a well-defined mass in the lower portion of the left thyroid. Under the diagnosis of symptomatic primary hyperparathyroidism, she underwent a partial parathyroidectomy 8 days prior to admission.

Her initial body temperature was 38.7°C and physical examination revealed limited extension in both hips and swelling without tenderness in the left knee. Laboratory studies showed a white blood cell count of 9,000/mm³ (neutrophils 77.7%, lymphocytes 7.6%), platelet count of 230,000/mm³, hemoglobin concentration at 9.2 g/dL, and C-reactive protein (CRP) level at 2.70 mg/dL. A liver function test showed normal findings with the exception of an elevated level of alkaline phosphatase (419 IU/mL; reference range, 30 to 115 IU/mL). The serum creatinine level was elevated from 2.65 to 6.27 mg/dL and the uric acid level was 10.1 mg/dL. The patient had primary metabolic acidosis and hypocalcemia; the ionized calcium level was 0.71 mmol/L (reference range, 1.05 to 1.35 mmol/L). However, the phosphorous and magnesium levels were found to be within normal limits. Calcific densities were observed in several intervertebral disc and foraminal spaces in spinal radiographs. Additionally, chondrocalcinosis was seen in the symphysis pubis, the left hip joint, and both knee joints (Figure 1A-1C).

Under the impression of hungry bone syndrome and acute renal failure on chronic kidney disease, the patient was admitted to the department of nephrology. She received intravenous and oral calcium, oral sodium bicarbonate, and calcitriol with the correction of hypocalcemia and metabolic acidosis. Empirical antibiotic therapy was initiated because of persistent fever and high CRP levels, but her pain in the left lower extremity did not improve. The patient developed acute severe low back pain on hospital day 3 and a raised CRP level of 21.4 mg/dL was observed on hospital day 7. Blood and urine cultures remained negative. A bone scan did not reveal any insufficiency fractures, but showed increased radiotracer uptake in the right side of the L5/S1 spine (Figure 1D). The patient consulted the department of rheumatology.

![Figure 1. Plain radiographs and bone scan. Radiographs show calcification (white arrows) in the intervertebral and foraminal spaces (A), pubic symphysis (B), and meniscus (C). (D) A technetium bone scan demonstrates increased radiotracer uptake in the right L5-S1 facet joint (black arrowhead).](image-url)
Musculoskeletal examination showed synovitis in the left knee joint and inguinal pain on passive or active extension in both hips. The patient had severe tenderness over the spinous process and the adjacent right paraspinal muscles of the lower L spine. Left knee synovial fluid showed a white blood cell count of 7,600/mm³ with 85% neutrophils and positively birefringent, rhomboid-shaped, intracellular crystals under polarized light microscopy. Spine computed tomography showed calcification of the annulus fibrosus, ligamentum flavum, interspinous ligaments, and facet joint capsules of the lumbar spine. Bone erosion in the facet joint was also identified (Figure 2A-2C). Magnetic resonance imaging of the lumbar spine revealed hypertrophy of the right facet joint of the L5-S1 vertebral segment, subchondral erosion and sclerosis, and a high T2 signal (Figure 2D-2F). Under biplane fluoroscopic guidance, arthrocentesis of the right L5/S1 facet joint was performed and 40 mg of triamcinolone was injected intra-articularly. Synovial fluid from the facet joint was found to contain intracellular CPP crystals on polarized microscopy (Figure 3). A culture of the synovial fluid from the knee and facet joints was negative as well. After a local glucocorticoid injection, the acute lumbar pain and fever were resolved within 24 hours, and the CRP level

Figure 2. Lumbar spine computed tomography scans and magnetic resonance images. (A ~ C) Axial and sagittal computed tomography scans show calcification of the L5-S1 facet joint capsules, ligamentum flavum, and annulus fibrosus (asterisks). (D) Calcification around the facet joint is demonstrated as low signal intensity on a T1-weighted magnetic resonance image (white arrowhead). (E, F) T2-weighted and fat-suppressed imaging reveals facet joint effusion and bone marrow edema (white arrows).

Figure 3. Compensated polarized light microscopy of a synovial fluid sample from the facet joint. Intracellular calcium pyrophosphate dihydrate crystals appear as rhomboids with positive birefringence (× 200). The double headed arrows indicate the axis of the analyzer.
decreased to 1.29 mg/dL after 6 days. The patient had no recurrence of acute CPP crystal arthritis in 4 years of follow-up.

**DISCUSSION**

Axial involvement is an uncommon manifestation of CPPD disease; more cases were reported in the cervical spine than in the thoracic or lumbar spine [2]. Spinal CPPD disease usually presents as a space-occupying lesion with chronic symptoms [2,12]. However, some cases of spinal CPPD disease can present as an acute pain with or without systemic symptoms. In the cervical spine, CPPD disease reportedly produces symptoms of crowned dens syndrome or meningitis-like features [13,14]. On the other hand, there are very few reports of acute CPPD disease in the lumbar spine.

Eight cases of acute CPPD disease presenting acute lumbar pain have been published previously; 6 patients presented with spondylodiscitis [4-9], 1 with facet arthritis [10], and 1 with interspinous bursitis [11]. Fujishiro et al. [10] reported a “pseudogout” attack in the L4/L5 facet joint in a subject with fever and acute lumbar pain. To the best of our knowledge, the present case is the second acute facet joint arthritis case due to CPPD in medical literature and the first in South Korea. Since acute CPP crystal disease could be self-limited or respond well to the non-steroidal anti-inflammatory drugs (NSAIDs) that are generally prescribed for acute back pain, many more cases of acute spinal CPP crystal disease may have gone unrecognized.

As the symptoms and signs of both infectious and acute CPP crystal disease overlap and their treatment and prognosis differ greatly, a correct differential diagnosis is mandatory in clinical practice [3]. The risk factors for CPPD disease may open the door to this differential diagnosis. The features of acute crystal-induced arthropathy in a patient over age 65 years, though not specific, are suggestive of acute CPP crystal arthritis [1]. If he/she has a metabolic disease associated with CPPD disease, acute CPP crystal disease should be considered in differential diagnosis as in our case [2]. The post-operative state, intra-articular hyaluronan, bisphosphonates, or granulocyte colony-stimulating factor is also known to be able to trigger CPP crystal-induced inflammation [2]. Another hint can be the presence of CPPD disease in the peripheral joints. Among the published and current cases with CPP crystal-related acute back pain, 55.6% (5/9 cases) had the past history of or concurrent presence of peripheral acute CPP crystal arthritis [4,7-9]. The presence of radiographic chondrocalcinosis may also indicate acute CPP crystal disease. However, the prevalence of chondrocalcinosis simply increases with age and most subjects with chondrocalcinosis are asymptomatic [1,2]. Thus, a definite diagnosis is based on the presence of intra- or extra-cellular CPP crystals in inflamed body fluid or tissues [1]. Once diagnosed with acute CPP crystal disease, patients show a good therapeutic response to NSAIDs, colchicine, or glucocorticoids [2]. In particular, intra-articular glucocorticoids are very useful in elderly patients with multiple comorbidities.

In conclusion, CPP crystals can cause crystal-induced acute inflammation in various sites of the lumbar spine and acute CPP crystal disease should be included in the differential diagnosis of acute back pain, especially in patients with medical conditions to trigger CPP crystal-induced inflammation and/or peripheral chondrocalcinosis.

**SUMMARY**

Spinal CPPD disease has been uncommonly reported, and most of the cases present with symptoms related to a mass effect in the cervical spine. A few cases of acute CPPD disease involving the lumbar spine have been described. This case report is the first Korean case in which acute CPP crystal arthritis involved the lumbar facet joint. Rheumatologists should be aware of acute CPP crystal arthritis as a rare cause of acute back pain.

**CONFLICT OF INTEREST**

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

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