Calcium Pyrophosphate Dihydrate Crystal Deposition Disease (Pseudogout) of Lumbar Spine Mimicking Osteomyelitis-Discitis With Epidural Phlegmon

Mark M. Mikhael, MD, Michael A. Chioffe, MD, and Gary S. Shapiro, MD

Abstract

Calcium pyrophosphate dihydrate crystal deposition disease (pseudogout) of the axial spine is rare. To our knowledge, there are few reports of the disease presenting with a presumed diagnosis of infection in the lumbar spine. As reported here, the diagnosis of osteomyelitis-discitis with epidural phlegmon was presumed before intervention.

We present the case of a 60-year-old man with radiographic imaging and worsening clinical presentation at 2 consecutive hospitalizations. Axial magnetic resonance imaging originally showed increased signal intensity at the L5–S1 disk, which suggested an infectious rather than inflammatory process. Aspiration and biopsy at the time were nondiagnostic and showed no evidence of organisms. Two months after conservative treatment, the patient was readmitted with intractable low back pain and radiating bilateral leg pain. Repeat imaging showed increased interval signal in the L5–S1 disk, as well as enhancing soft-tissues that now extended to adjacent levels with extensive erosive changes. After surgical intervention for suspected infection, all cultures and stains for organisms were negative. Final pathology showed granulation tissue with focal inflammatory changes and calcium pyrophosphate crystal deposition.

Although pseudogout is rare, physicians should add the disorder to the differential diagnosis for low back pain with radiculopathy and presumed infection.

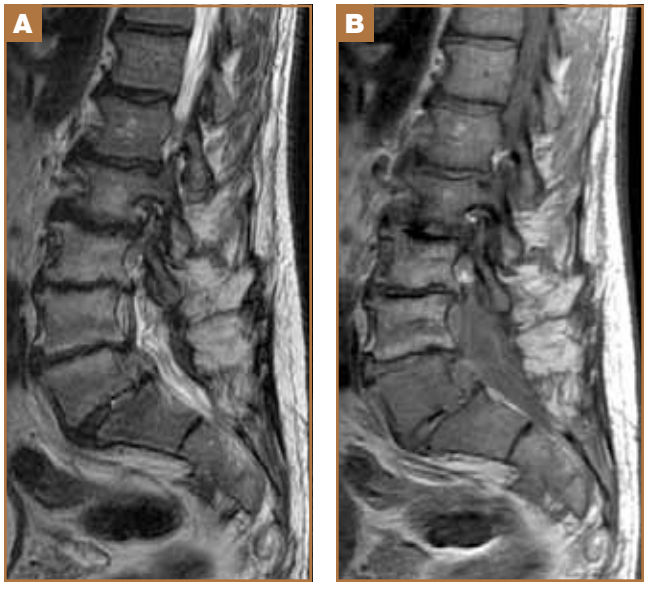

Figure 1. Sagittal T_2-weighted (A) and T_1-weighted (B) magnetic resonance imaging of lumbar spine shows increased signal within L5–S1 disk space. This study was performed at initial crisis and presentation. In this clinical setting, these findings suggest discitis.

Calcium pyrophosphate dihydrate (CPPD) crystal deposition disease, or pseudogout, is a metabolic arthropathy characterized by the clinical features of arthritis and the presence of CPPD crystals.[1] It has been associated with metabolic disorders, rheumatoid arthritis, trauma, and previous spine surgery.[2-4] In the general population, the incidence of CPPD in the spine is much higher with advanced age.[2,5-7] CPPD can be characterized by acute, subacute, or chronic joint inflammation. It commonly involves major joints; symptomatic axial disease is less common.[3,4]

In this article, we describe clinical, laboratory, and radiographic findings that suggested infection in the lumbar spine. The patient had no history of pseudogout. He had worsening pain and radiculopathy, and imaging showed progressive joint destruction. Although we initially believed an infection to be the cause of patient's condition, final pathology confirmed CPPD of the lumbar spine.

Authors' Disclosure Statement: The authors report no actual or potential conflict of interest in relation to this article.

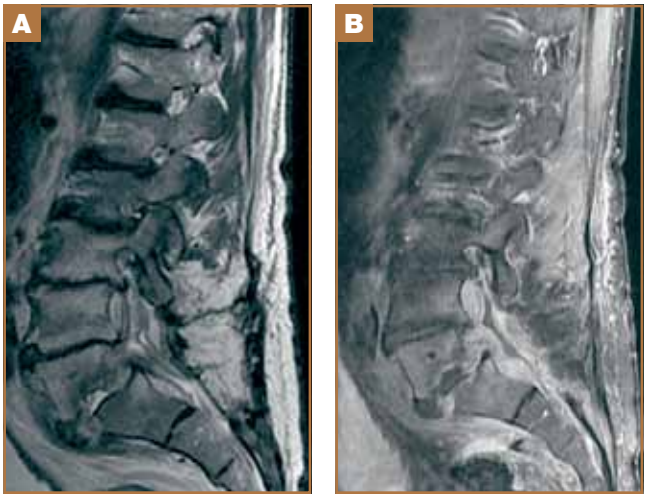

Figure 2. Sagittal magnetic resonance imaging of lumbar spine includes T_2-weighted (A) and postcontrast T_1-weighted (B) sequences performed at second crisis 2 months later. These figures demonstrate interval increase in T_2 signal within L5–S1 intervertebral disk and markedly enhancing soft-tissue in epidural space on T_1-weighted postcontrast sequences.

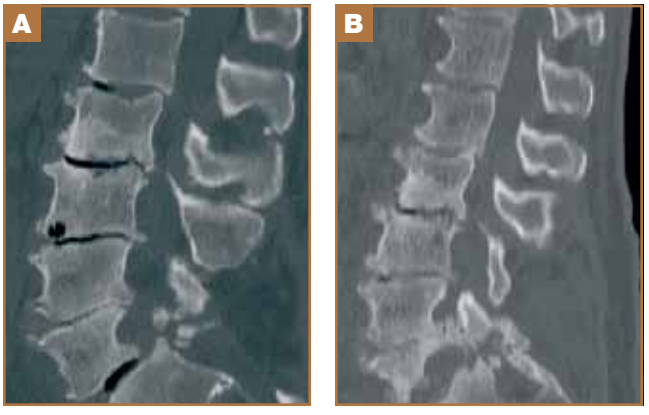

Figure 3. Sagittal computed tomography of lumbar spine at first crisis (A) and second crisis (B) shows progressive erosive changes in L5 vertebral body over 2 months.

The patient provided written informed consent for print and electronic publication of this case report.

Case Report

A 60-year-old man with Parkinson's disease and a remote history of an L4–L5 laminectomy presented with new-onset severe low back pain and bilateral leg pain. Initial examination revealed local tenderness over the lower lumbar spine with intact distal motor and sensation. The patient was afebrile. White blood cell (WBC) count was 11.4 (normal, 4.0-10.0), erythrocyte sedimentation rate (ESR) was 83 mm/h (normal, 0-20 mm/h), and C-reactive protein (CRP) was 66 mg/L (normal, <8.0 mg/L). Magnetic resonance imaging (MRI) showed increased signal within the L5–S1 intervertebral disk and the endplates of the L5 and S1 vertebrae on the T_1- and T_2-weighted sequences (**Figures 1A, 1B**). The diagnosis was presumed discitis, and a fluoroscopy-guided percutaneous biopsy was performed. The patient did not receive antibiotics. Final cultures were negative and pathology was nondiagnostic. Treatment with an epidural steroid injection resolved the pain.

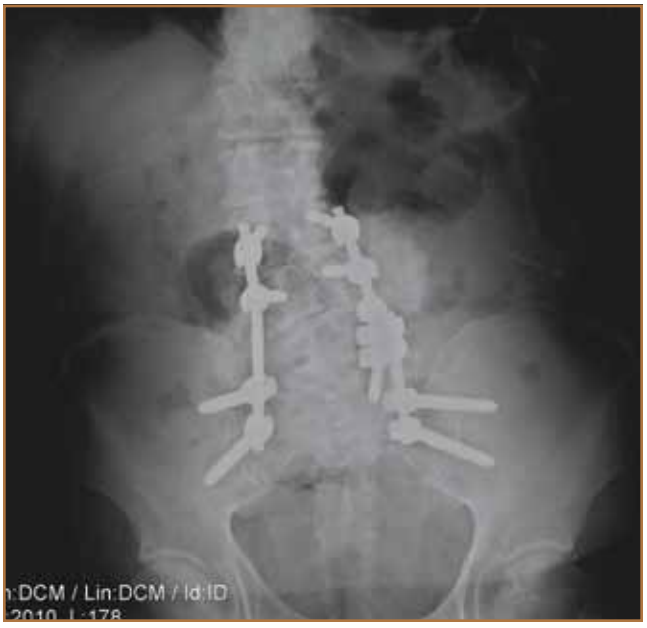

Figure 4. Postoperative anteroposterior radiograph of lumbar spine after spinal decompression and fusion from L2 to pelvis with L5–S1 transforaminal lumbar interbody fusion.

The patient returned 2 months later with worsening bilateral leg pain (right more than left), dorsiflexion weakness, and diminished sensation along the lateral and plantar right foot. WBC count was 8.1, ESR was 94 mm/h, and CRP was 54.7 mg/L. Repeat MRI showed an interval increase in the T_2 signal within the L5–S1 intervertebral disk and markedly enhancing soft-tissue in the epidural space on the T_1 -weighted postcontrast sequences at the L5–S1 level (**Figures 2A, 2B**). Computed tomography (CT) at the time showed progressive erosive changes in the L5 vertebral body when compared to a CT scan from the first crisis (**Figures 3A, 3B**). These findings were highly suggestive of osteomyelitis-discitis with epidural phlegmon.[8-10] Given the nondiagnostic percutaneous-tissue biopsy, and the severity of the current symptoms with radiographic progression of bony destruction, we performed open biopsy, decompression, and stabilization as indicated during surgery. During surgery, there was no evidence of purulence or infection. Intraoperative pathology specimens were sent as sections for immediate evaluation and did not suggest infection.

The patient then had spinal decompression and fusion from L2 to the pelvis with an L5–S1 transforaminal lumbar interbody fusion (TLIF) (**Figure 4**). Given the low suspicion for infection during surgery, a polyetheretherketone cage was used in the reconstruction after debridement of the L5–S1 disk space; use of this device in the setting of known infection has been reported.[11-13] During surgery, chalky white material was noted at the L5–S1 disk space. Cultures and tissue samples did not reveal any organisms. Final pathology indicated CPPD

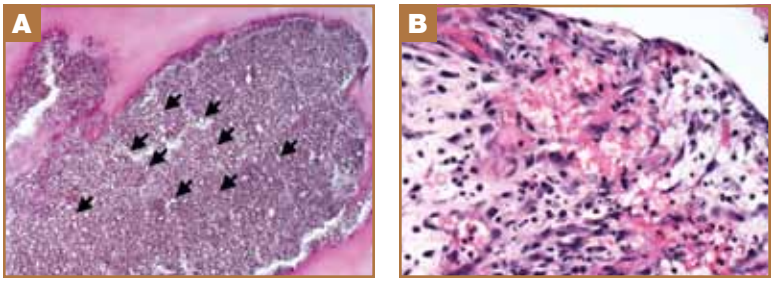

Figure 5. Final pathology specimens prepared on hematoxylin-eosin slides have abundant crystal depositions (arrows) in (A) fibrous tissue and (B) inflammatory granulation tissue.

crystals (pseudogout) with inflammatory granulation tissue (**Figures 5A, 5B**).

Shortly following surgery, the leg pain resolved and motor weakness improved. Fourteen months after surgery, the patient was pain-free, and ambulatory tolerance was improved. The Parkinson's disease, however, had become more advanced.

Discussion

It has been shown that CPPD deposits can cause acute disease from a mass effect, such as axial neck and back pain, myelopathy, radiculopathy, and spinal stenosis.[2-7,14-24] Trauma and previous surgery are associated with deposits in the intervertebral disk tissue, but de novo deposition increases with advanced age.[2,5-7,15]

CPPD in the spine can present with inflammatory symptoms and nonspecific laboratory values that can mimic infection.[3,5] Others have observed that mild leukocytosis and elevated ESR and CRP levels are usually found in acute pseudogout, whereas chronic CPPD arthritis is characterized by elevated ESR and CRP level alone.[25] These findings marked our patient's case. CPPD can also produce severe degenerative disk disease and destructive lesions of the vertebral bodies and disk spaces.[26-29] Tissue biopsy must identify rod or rhomboid crystals that are positively birefringent under polarized light microscopy to confirm the diagnosis of pseudogout.[26-27] If initial tissue biopsy and cultures are nondiagnostic in the setting of suspected infection, perhaps routine polarized light microscopy should also be considered. Further investigation, such as a cost-effectiveness analysis, may be beneficial to support this practice.

In the setting of discitis and osteomyelitis, thorough debridement (eg, with an anterior complete discectomy and corpectomy) is the mainstay of treatment to decrease the incidence of persistent or recurrent infection. In our patient's case, the initial surgical plan included open biopsy and cultures with posterior decompression and stabilization. An anterior debridement would have been done in staged fashion if infection were confirmed. As immediate intraoperative sections did not suggest infection, the index of suspicion was low, and we proceeded with the TLIF pending final pathology and culture results.

Long-term management of pseudogout includes prevention and treatment of acute episodes of inflammation. This includes over-the-counter nonsteroidal anti-inflammatory drugs (NSAIDs) or prescribed medications, such as colchicine or indomethacin. Corticosteroids are often used in patients who do not respond to, or cannot tolerate, NSAIDs.

Symptomatic CPPD has been reported in the axial spine. It most commonly affects the cervical spine, potentially causing myelopathy and cervicomedullary compression.[2-7,14-24] Although symptomatic lumbar deposition is considerably rarer, there have been reports of lumbar stenosis and acute herniated disk syndrome secondary to CPPD deposition.[2-5,14-16,18,20-22] Gouty arthritis of the lumbar spine is likely more common, but also rare.[28,29] Many authors have reported symptomatic gout in the spine, and a few have described gout mimicking infection.[28,29] We found only 1 report of CPPD deposition mimicking infection in the lumbar spine.[5]

Conclusion

Although infection is the most common cause of the symptoms described in this report, we present a rare occurrence of CPPD (pseudogout) mimicking infection in the lumbar spine. Our patient's clinical symptoms, along with imaging studies revealing progressive joint destruction, was initially presumed to be the result of an infection. Final pathology, however, confirmed the ultimate diagnosis of pseudogout. Based on the foregoing, clinicians shoulder consider CPPD in the differential diagnosis for spine infection.

Dr. Mikhael is Reconstructive Spine Surgeon, Illinois Bone and Joint Institute, Glenview; and Clinician Educator, University of Chicago, Illinois. Dr. Chioffe is Resident in Orthopedic Surgery, University of Chicago, Illinois. Dr. Shapiro is Reconstructive Spine Surgeon, Illinois Bone and Joint Institute, Glenview; and Assistant Clinical Professor, University of Chicago, Illinois.

Address correspondence to: Mark M. Mikhael, MD, Illinois Bone and Joint Institute, 2401 Ravine Way, #200, Glenview, IL 60025 (tel, 847-724-4392; fax, 847-832-6713; e-mail, mmikhael@ibji.com).

Am J Orthop. 2013;42(8):E64-E67.

References

1. Kohn NN, Hughes RE, McCarty DJ Jr, Kohn NN, Faires JS. The significance of calcium phosphate crystals in the synovial fluid of arthritic patients: the "pseudogout syndrome". II. Identification of crystals. *Ann Intern Med*. 1962;56:738-745.
2. Mahmud T, Basu D, Dyson PH. Crystal arthropathy of the lumbar spine: a series of six cases and a review of the literature. *J Bone Joint Surg Br*. 2005;87(4):513-517.
3. Salcman M, Khan A, Symonds DA. Calcium pyrophosphate arthropathy of the spine. *Neurosurgery*. 1994;34(5):915-918.
4. Delamarter RB, Sherman JE, Carr J. Lumbar spinal stenosis secondary to calcium pyrophosphate crystal deposition (pseudogout). *Clin Orthop*. 1993;(289):127-130.
5. Bartlett CS 3rd, Casden AM, Abdelwahab IF. Calcium pyrophosphate disease mimicking infection in the lumbar spine. *Orthopedics*. 1999;22(1):79-81.
6. Kawano N, Matsuno T, Miyazawa S, et al. Calcium pyrophosphate dihydrate crystal deposition disease in the cervical ligamentum flavum. *J Neurosurg*. 1988;68(4):613-620.

7. Baty V, Prost B, Jouvet A, Laurent J, Vallée B. Acute spinal cord compression and calcium pyrophosphate deposition disease. Case illustration. *J Neurosurg*. 2003;99(2 suppl):240.
8. Gines R, Bates DJ. Tophaceous lumbar gout mimicking epidural abscess. *Am J Emerg Med*. 1998;16(2):216.
9. Barrett K, Miller ML, Wilson JT. Tophaceous gout of the spine mimicking epidural infection: case report and review of the literature. *Neurosurgery*. 2001;48(5):1170-1173.
10. Steinbach L. Calcium pyrophosphate dehydrate and calcium hydroxyapatite crystal deposition diseases: imaging perspectives. *Radiol Clin North Am*. 2004;42(1):185-205, vii.
11. Pee YH, Park JD, Choi YG, Lee SH. Anterior debridement and fusion followed by posterior pedicle screw fixation in pyogenic spondylodiscitis: autologous iliac bone strut versus cage. *J Neurosurg Spine*. 2008;8(5):405-412.
12. Walter J, Kuhn S, Reichart R, Kalff R, Ewald C. PEEK cages as a potential alternative in the treatment of cervical spondylodiscitis: a preliminary report on a patient series. *Eur Spine J*. 2010;19(6):1004-1009.
13. Brase A, Ringel F, Stüer C, Meyer B, Stoffel M. Debridement and fusion with polyetheretherketone implants in purulent spondylodiscitis: a clinical experience with nine patients. *Acta Neurochir (Wien)*. 2010;152(11):2001-2004.
14. Doita M, Shimomura T, Maeno K, Nishida K, Fujioda H, Kurosaka M. Calcium pyrophosphate dehydrate deposition in the transverse ligament of the atlas: an unusual cause of cervical myelopathy. *Skeletal Radiol*. 2007;36(7):699-702.
15. Siau K, Lee M, Laversuch C. Acute pseudogout of the neck—the crowned dens syndrome: 2 case reports and review of the literature. *Rheumatol Int*. 2011;31(1):85-88.
16. Fujishiro T, Nabeshima Y, Yasui S, Fujita I, Yoshiya S, Fujii H. Pseudogout attack of the lumbar facet joints: a case report. *Spine*. 2002;27(17):396-398.
17. Gadgil AA, Eisenstein SM, Darby A, Cassar Pullicino V. Bilateral symptomatic synovial crystals of the lumbar spine caused by calcium pyrophosphate deposition disease. *Spine.* 2002;27(19):E428-E431.
18. Markiewitz AD, Boumphrey FR, Bauer TW, Bell GR. Calcium pyrophosphate dihydrate crystal deposition disease as a cause of lumbar canal stenosis. *Spine.* 1996;21(4):506-511.
19. Ishida T, Dorfman HD, Bullough PG. Tophaceous pseudogout (tumoral calcium pyrophosphate dihydrate crystal deposition disease). *Hum Pathol*. 1995;26(6):587-593.
20. Lam HY, Cheung KY, Law SW, Fung KY. Crystal arthropathy of the lumbar spine: a report of 4 cases. *J Orthop Surg (Hong Kong)*. 2007;15(1):94-101.
21. Fye KH, Weinstein PR, Donald F. Compressive cervical myelopathy due to calcium pyrophosphate didydrate deposition disease. *Arch Intern Med*. 1999;159(2):189-193.
22. Sadique T, Bradley JG, Jackson AM. Central spinal stenosis due to pseudogout: a case report. *J Bone Joint Surg Br*. 1994;76(4):672-673.
23. Hodge JC, Ghelman B, DiCarlo EF, Cammisa FP Jr. Calcium pyrophosphate deposition within the ligamenta flava at L2, L3, L4, and L5. *Skeletal Radiol*. 1995;24(1):64-66.
24. Berlemann U, Gries NC, Moore RJ, Fraser RD, Vernon-Roberts B. Calcium pyrophosphate dihydrate deposition in degenerate lumbar discs. *Eur Spine J*. 1998;7(1):45-49.
25. Lillicrap M. Crystal arthritis: contemporary approaches to diseases of antiquity. *Clin Med*. 2007;7(1):60-64.
26. Pytel P, Wollmann R, Fessler R, Krausz T, Montag A. Degenerative spine disease: pathologic findings in 985 surgical specimens. *Am J Clin Pathol*. 2006;125(2):193-202.
27. Kishore B, Khare P, Gupta RJ, Gupta C, Khare V. Tumoral calcium pyrophosphate dehydrate crystal deposition disease: a rare diagnosis by fine-needle aspiration. *Diagn Cytopathol*. 2010;38(1):47-50.
28. Tsai C, Chen Y, Hsu HC, Chen HT. Bacteremia coexisting with tophaceous gout of the spine mimicking spondylodiscitis. *Spine.* 2009;34(2):E106-E109.
29. Bonaldi V, Duong H, Starr M, Sarazin L, Richardson J. Tophaceous gout of the lumbar spine mimicking and epidural abscess: MR features. *AJNR Am J Neuroradiol*. 1996;17(10):1949-1952.