Case Report

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# Calcium Pyrophosphate Dihydrate Crystal Deposition Disease Involving the Ligamentum Flavum of the Cervical Spine with Intense Enhancement on MRI: A Case Report

## 자기공명영상에서 뚜렷한 조영증강을 보이는 경추 황색인대의 칼슘수산화인회석 결정침착질환: 증례 보고

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Calcium pyrophosphate dihydrate (CPPD) crystal deposition disease is characterized by chondrocalcinosis, which mainly affects the knees, wrists, pelvis, and rarely, the spine. According to previous reports, CPPD crystal deposits display heterogeneous enhancement on MRI. When combined with inflammation of the surrounding soft tissue, strong enhancement by CPPD crystal deposition may appear similar to imaging features of other conditions such as infectious spondylitis. In these conditions, CT plays an important role in differential diagnosis. Here, we present a case of CPPD crystal deposition disease in the ligamentum flavum of the cervical spine that showed intense enhancement on MRI.

**Index terms** Calcium Pyrophosphate; Chondrocalcinosis; Ligamentum Flavum; Magnetic Resonance Imaging

## INTRODUCTION

Calcium pyrophosphate dihydrate (CPPD) crystal deposition disease is characterized by chondrocalcinosis, which affects mainly the knee, wrist, pelvis and rarely spine (1).

It may be associated with para-articular calcifications, joint space narrowing and subchondral cyst formation (1). This disease can involve spine including intervertebral disks, ligamentum flavum, interspinous and supraspinous ligaments. In particular, there were a few reports in which CPPD crystal deposition disease involved ligamentum flavum of cervical spine with heterogenous enhancement on MRI. But as far as we know, CPPD crystal deposits has not been strongly enhanced. We present a case of CPPD crystal deposition disease involving ligamentum flavum of cervical spine that showed intense enhancement on MRI.

## CASE REPORT

A 90-year-old man presented to the emergency room for neck pain that began 4 days ago. Physical examination revealed posterior cervical tenderness and mild limited range of motion in all directions without neurologic deficit. On the day of admission, body temperature was measured at 38.5 degree Celsius and complete blood cell count showed mild leukocytosis of 11000/$mm^3$ (normal range, 4000–10000/$mm^3$). Serum C-reactive protein (CRP) was 11.97 mg/dL (normal range, 0–0.3 mg/dL). Cerebrospinal fluid examination was within normal limits. He did not have history of cervical spine trauma or infection.

He was admitted to an internal medicine department for evaluating infectious diseases, and underwent several tests to determine the cause of infection, but CT of chest and abdomen didn't show any signs of infection. Blood culture and urine culture were also negative.

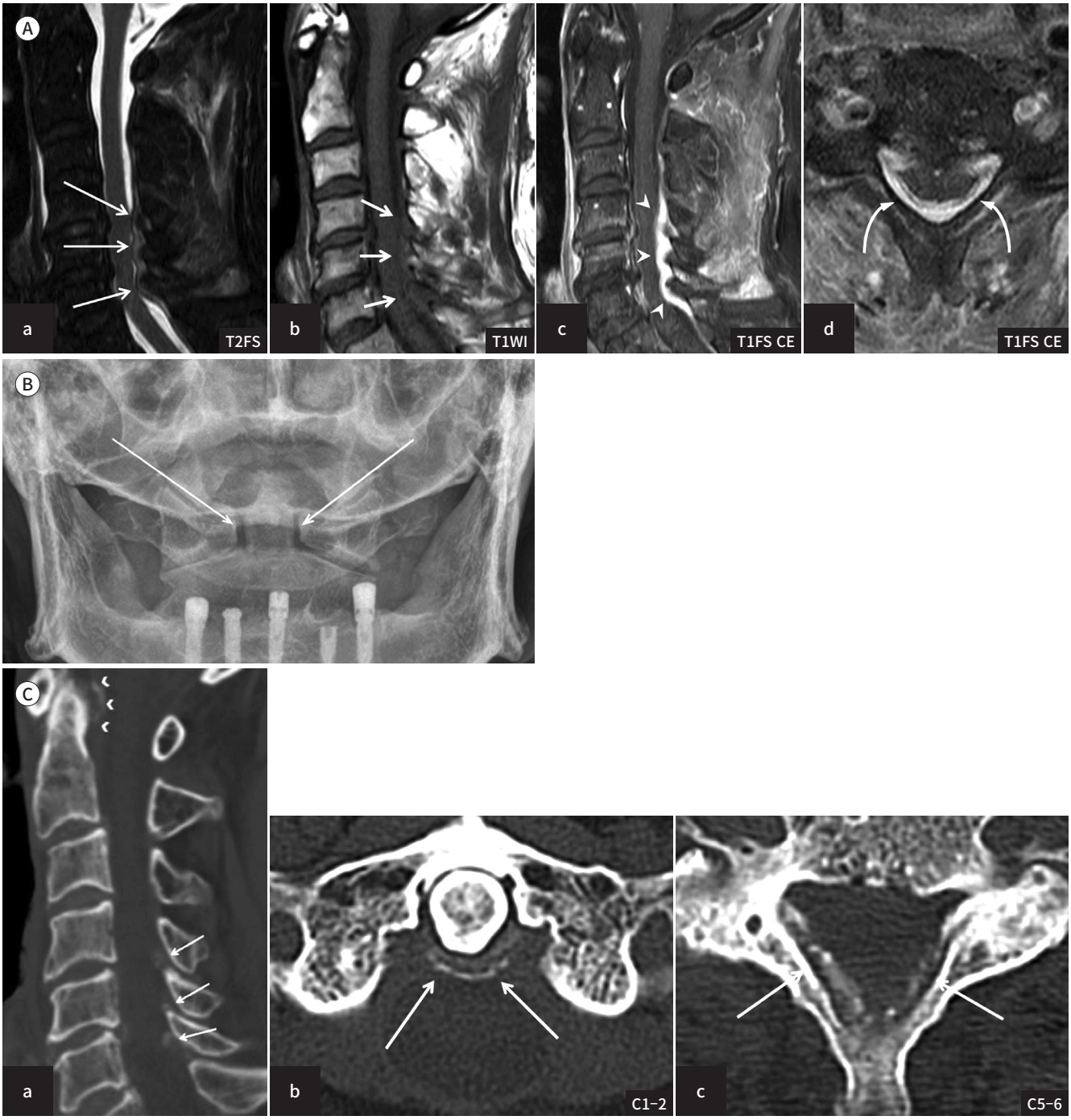
On MR imaging of cervical spine, the lesion appeared as a hypointensities on both pre-contrast T2 (Fig. 1A-a) and T1 (Fig. 1A-b) weighted images. And contrast-enhanced T1-weighted images with fat suppression (Fig. 1A-c, d) showed intense enhancement at epidural space of C4–C6 level. There was soft tissue edema around paraspinal and interspinous spaces. Regarding MR and laboratory findings, infectious spondylodiscitis was suspected. However, cervical spine radiographs with odontoid view (Fig. 1B) demonstrated radio-opaque lesion beside the odontoid process. CT was performed one day later. CT imaging of cervical spine (Fig. 1C) revealed curvilinear calcifications in the peri-odontoid soft tissue and diffuse punctate calcifications in all intervertebral disks with several nodular calcifications of ligamentum flavum at C4–C6 level. Therefore, it was thought that inflammation of the spine by CPPD crystal deposition was more suspected than infectious spondylodiscitis.

Nonsteroidal anti-inflammatory drug was administrated for 4 days, which led to resolve symptoms and normalize serum CRP. Antibiotic drugs were not used. Based on the clinical and laboratory findings, the final diagnosis was CPPD crystal deposition disease of cervical spine with active inflammation.

## DISCUSSION

A patient may demonstrate several different clinical patterns during the course of CPPD disease; pseudogout, pseudo-rheumatoid arthritis, pseudo-osteoarthritis (with acute exacerbations), pseudo-osteoarthrosis (without acute exacerbations), pseudo-neuropathic osteoarthropathy, and miscellaneous pattern (2). The most common pattern is pseudo-osteoarthritis, which is caracterised by chronic progressive arthritis with superimposed acute inflammatory epi-

Fig. 1. Calcium pyrophosphate dihydrate crystal deposition disease involving cervical spine in a 90-year-old man, presenting with neck pain.
A. Sagittal T2- (a) and T1WIs (b) show low signal intensity lesion in posterior epidural space. Signal intensity indicated by long and short arrows in T2- and T1WIs, respectively. Sagittal (c) and axial (d) FS post-contrast T1WIs demonstrate intense enhancem ent of the lesion (arrowheads and curved arrows).
B. An open-mouth odontoid radiograph shows a linear radiopaque lesion around the odontoid process (arrows).
C. Sagittal CT scan (a) shows crown-shaped calcifications surrounding the odontoid process (arrowheads) and diffuse punctate calcifications in all cervical intervertebral discs with several nodular calcified deposits at the ligamentum flavum (arrows). Axial CT scan at the C1–2 level (b) at the C1–2 level shows curvilinear calcified deposits of the transverse ligament on the atlas in a double band (arrows). Axial scan CT scan at the C5–6 level (c) shows linear calcification along the ligamentum flavum (arrows).
CE = contrast-enhanced, FS = fat suppressed, T1WI = T1-weighted image



sodes in large joints such as the knee and hip (2). Although osteoarthritis and CPPD are distinct disorders, it has been suggested that destruction of the articular cartilage caused by the crystals may predispose a patient to developing secondary osteoarthritis (3).

The definite diagnosis of CPPD disease is the identification of CPPD crystals in synovial fluid or biopsy. According to Ryan and McCarty's criteria for CPPD (4), presence of typical calcification on radiographs can be diagnosed as probable CPPD disease. CPPD crystal deposits have been observed in the spine, but less commonly noted (5, 6). The intervertebral disk, posterior longitudinal ligament, facet joints and ligamentum flavum are the sites where crystal is most commonly deposited in cervical spine (7). This case could be classified as probable CPPD disease. Several nodular calcified deposits were observed along ligamentum flavum at C4–C6 with intense enhancement on post-contrast T1 weighted MRI. CPPD crystal deposition disease ranges from an acute inflammatory process mimicking a gout-attack to progressive chronic arthropathy (8). Our case revealed that if acute inflammation is induced by CPPD crystal deposits in the spine, it may appear clinically or radiologically similar to infectious spondylodiscitis. As it has been known in previous studies (6), CT is superior to MRI in detection of CPPD crystal deposition in our case, thus CT can be critical in a differential diagnosis.

The strong enhancement in spinal epidural space at C4–C6 and the paraspinal inflammation in our case required differentiation from infectious spondylodiscitis. It was not easy to distinguish infectious spondylodiscitis because MRI features were not always typical. But, infectious spondylodiscitis usually occurred in the lumbar spine, destroying vertebral body endplates and invading intervertebral disk spaces (9). Tuberculous spondylodiscitis was common in the thoracic spine, might forming paraspinal abscess and showing skip lesions (9). So MRI signal change of the adjacent vertebral body and paraspinal fluid collection would be helpful in differential diagnosis.

Homogenous epidural enhancement might be seen after continuous epidural anesthesia (10). In this condition, there was no signal change in vertebral body, endplate and no paraspinal fluid collection. It was posterior epidural space lesion as same as CPPD crystal deposits in ligamentum flavum (10). However, calcification deposits were not observed.

In conclusion, we described that CPPD crystal deposition disease of spine could be mistaken for other disease entities such as infectious spondylodiscitis due to acute inflammatory response induced by CPPD crystal deposition. An awareness of CPPD crystal deposition disease and its radiological findings could be helpful to avoid diagnostic errors and unnecessary procedures. It would be important to use CT in a timely manner.

### Author Contributions

Conceptualization, L.J., L.S.; data curation, L.J., L.S.; investigation, L.J.; methodology, L.S., B.J.; project administration, L.S., B.J.; resources, all authors; software, L.J.; supervision, L.S., B.J.; visualization, L.J.; writing—original draft, L.J.; and writing—review & editing, L.S., B.J.

### Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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# 자기공명영상에서 뚜렷한 조영증강을 보이는 경추 황색인대의 칼슘수산화인회석 결정침착질환: 증례 보고

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칼슘수산화인회석(calcium pyrophosphate dehydrate; 이하 CPPD) 결정침착질환은 연골석회증을 특징으로 하는 질환으로, 주로 무릎, 손목, 골반, 드물게는 척추를 침범한다. 이전 보고에 따르면, CPPD 결정침착은 MRI에서 불균질하게 조영증강이 될 수 있다. 주변 연부 조직의 염증과 동반된 CPPD 결정침착의 뚜렷한 조영증강은 감염성 척추염 등 다른 질환과 유사한 영상 소견을 보일 수 있다. 이 경우 CT는 감별진단에 중요한 역할을 한다. 저자들은 MRI에서 황색인대에 저명한 조영증강을 보이는 CPPD 결정침착 증례를 보고하고자 한다.

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