



Case Report

Acute cervical epidural abscess with concurrent calcium pyrophosphate deposition after cervical spinal surgery: A case report

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ABSTRACT

Background: Spinal epidural abscess (SEA) is a rare condition that may result in catastrophic outcomes. On the other hand, calcium pyrophosphate (CPP) deposition disease (CPPD) causes inflammatory arthritis. Spinal involvement of a crystal-induced inflammation caused by CPPD is also common. Surgery is a common risk factor for both SEA and CPPD; however, the postoperative acute onset of SEA complicated with CPPD is extremely rare.

Case Description: A man in his 70s presented to our hospital, complaining of right upper limb weakness, loss of dexterity, and gait disturbance. The diagnosis of cervical spondylotic myelopathy was made, and he performed laminectomy at C3, C4, and C5 levels. Four days after the laminectomy, he suffered from acute neck pain, weakness, and hypoesthesia in his arms and legs. Magnetic resonance imaging revealed a mass occupying the dorsal epidural space of C6 and C7, compressing the cervical spinal cord. Considering the acute symptomatology, an acute spinal epidural hematoma after surgery was suspected; therefore, emergency C6 and C7 laminectomy was performed. Surgical findings indicated that the pressure inside the spinal canal was elevated, and the mass was purulent exudate. Pathological examination showed suppurative inflammation with concomitant deposition of CPP. SEA complicated with CPPD was considered; therefore, antibiotics and non-steroidal anti-inflammatory drugs were administered. The motor weakness and hypoesthesia were improved despite a slight residual deficit in his dexterity.

Conclusion: An acute onset of SEA complicated with CPPD after cervical surgery has rarely been reported. The suppurative inflammation fostered by the crystal-induced inflammation may account for the acute symptomatology.

Keywords: Calcium pyrophosphate deposition disease, Chondrocalcinosis, Epidural abscess, Pseudogout, Surgical site infection

INTRODUCTION

Spinal epidural abscess (SEA) is a rare disease, but the early diagnosis of SEA is mandatory due to its potentially fatal outcome. Since the recent advent of imaging techniques enabled us to detect SEA with high sensitivity, the prevalence of SEA is increasing. However, mortality and the rate of the neurologically poor sequelae of SEA remain high.^[1,3]

On the other hand, calcium pyrophosphate deposition disease (CPPD) causes crystal-induced arthritis mimicking gout. The patients typically presented with the acute onset of mono- or

oligoarticular arthritis and constitutional symptoms in the inflammatory response to calcium pyrophosphate (CPP) crystals.^[12] Although the knee or wrist joints are more susceptible to CPPD, spinal involvement of CPPD has also been reported.^[6,10,11] Spinal CPPD affects the facet joint, intervertebral discs, and the ligamentum flava, in some cases leading to neurological symptoms due to the compression of the spinal cord.^[2,7]

Although a surgical invasion is known to be followed by SEA and CPPD, the acute onset of which after the surgery has been rarely reported.

CASE DESCRIPTION

A man in his 70s presented with a weakness in the right arm, loss of dexterity, and gait disturbance for one month. As the symptoms gradually worsened, the patient came to our department. His medical history included hypertension and hypercholesterolemia. He was an immunocompetent host and did not take any oral antithrombotic medication. Magnetic resonance imaging revealed cervical spinal canal stenosis at the level of C3/4 and C4/5 without ossification of the posterior longitudinal ligament and yellow ligament, and there is no evidence of cervical spinal instability [Figure 1a]; hence, he was performed laminectomy at C3, C4, and C5 level. His symptoms subsided after the surgery; however, four days after the procedure, he complained of acute neck pain, motor deficit, and hypoesthesia in four limbs.

Magnetic resonance imaging showed a mass located in the dorsal epidural space at C6 and C7 levels, which we did not manipulate during the surgery. The mass was well-demarcated, low-intensity in T1-weighted, and homogeneously hyperintensity in T2-weighted image [Figures 1b and c]. Based on imaging characteristics and the acute onset, an acute epidural hematoma after the surgery was suspected.

The emergency C6 and C7 laminectomy was performed. The surgical findings implied elevated pressure inside the spinal canal because a purulent, whitish exudate erupted from the thinned cortical bone [Figure 2a]. Coagulase-negative staphylococci (CNS) were detected in the culture test of the exudate. The pathological examination of the exudate showed the infiltration of neutrophils and the small clusters of weak birefringent, basophilic crystals in hematoxylin and eosin stain, suggesting the SEA of CNS with concurrent crystal-induced inflammation of CPP [Figures 2b and c]. During the postoperative course, he complained of pain in his left knee. Arthrocentesis of the joint capsule of the knee also revealed CPP deposition. He was diagnosed with SEA of CNS complicated with CPPD and treated with antibiotics and non-steroidal anti-inflammatory drugs. His motor weakness and hypoesthesia improved despite a mild residual deficit of dexterity ten weeks after the surgery.

DISCUSSION

SEA is characterized by pus accumulation in the epidural space, causing spinal cord compression and neurological deficit. The diagnosis of SEA is difficult, and the treatment is prone to be delayed.^[14] However, since SEA sometimes leads to catastrophic neurological sequelae, the appropriate and immediate treatment is crucial. The causes of infection can be direct transmission from a local infection, indwelling catheter, surgical invasion, and hematogenous infection; however, in some cases, no obvious origin of infection is proven. Surgical drainage with systemic antibiotics is the widely accepted treatment option.^[1,3,13,14]

On the other hand, CPPD is characterized as the crystal deposition in several joints, causing joint pain and constitutional symptoms. CPPD commonly involves the

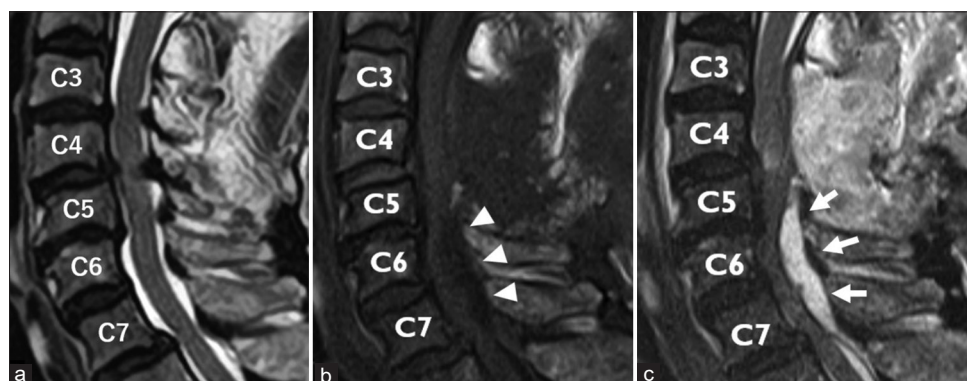


Figure 1: Magnetic resonance image before and after the surgery. (a) Cervical spinal stenosis was found in the T2-weighted image, sagittal section, and the diagnosis of cervical spondylosis was made; therefore, the patient performed the C3 to C5 laminectomy. (b) Four days after the surgery, the mass occupying the dorsal epidural space (arrowhead) was well demarcated and low-intensity in T1-weighted and (c) homogeneously hyper-intensity in T2-weighted image (arrow).

large joints such as knees, wrists, shoulders, and hips, but it also involves the spine.^[2] CPPD is associated with several metabolic diseases, such as calcium metabolic disorders, iron overload, and hypothyroidism. Mechanical stress, including trauma and surgical invasion, is also relevant to the onset of CPPD. According to the reported cases of spinal involvement of CPPD, a surgical procedure is indicative to patients who suffer from neurological symptoms, including a motor deficit, sensory abnormality or bladder, and bowel incontinence. Histopathological findings obtained from the surgical specimen showed the birefringent rhomboid-shaped crystals, indicating CPP deposition.^[4,8-10]

The present patient complained of neck pain and neurological symptoms immediately after the surgery for cervical spondylotic myelopathy, suggesting cervical spinal cord compression. Although a surgical procedure is the common predisposing factor of SEA and CPPD, the

acute onset of 4 days of SEA and CPPD after the preceding surgery has been rarely reported. The interval time between the onset of SEA and the spinal surgery has not been well investigated. Literally reviewed, five cases of CPPD after the spinal surgery were reported, and the interval time after the surgical procedure ranged from 4 weeks to 8 years; however, acute onset within a week has not been reported [Table 1].^[2,5,7,9,11] In addition, the mass occupied the epidural space at the C6 and C7 levels where the surgical manipulation was not performed, which either infection or crystal-induced inflammation could not explain. Whether the diagnosis was SEA or CPPD, the acute postoperative interval of 4 days before the onset of neurological symptoms and the level of the mass formation was atypical. Considering the clinical course, the suppurative inflammation fostered by the crystal-induced inflammation may account for the acute symptomatology of this patient.

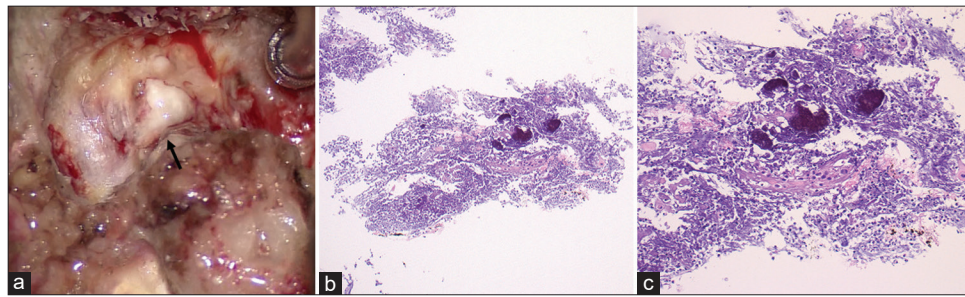


Figure 2: (a) Intraoperative finding. After the laminectomy of C7, a purulent, whitish exudate (arrow) erupted from the thinned cortical bone; Hematoxylin and eosin stain showed the infiltration of neutrophils and the small clusters of weak birefringent, basophilic crystals compatible with calcium pyrophosphate deposition. (b) Low- ($\times 40$), and (c) high-magnification ($\times 100$).

Table 1: Reported cases of calcium pyrophosphate deposition disease after the surgical procedure.

Authors/year	Age/sex	Symptoms	Level	Surgical history	Interval after the previous surgery	Treatment
Bridges K <i>et al.</i> 2017 ^[2]	66 / Female	Mild back spasm, fever, and chills	C7/T1, T1/2 and T9-12	Laminectomy(L2-5), bilateral foraminotomy, TLIF with PLF(L4/5)	5 weeks	Antibiotics, followed by biopsy and IL-1 inhibitor
Jaffee S, <i>et al.</i> 2022 ^[5]	75 / Female	Radiculopathy, paresthesia	L3/4, L4/5	Laminectomy(L4/5)	9 months	laminectomy, foraminotomy (L3/4)
Lam HY <i>et al.</i> 2007 ^[7]	72 / Female	Buttock pain, lower limb numbness	L4/5	L5 hemilaminectomy	2 years	Removal
Moon AS, <i>et al.</i> 2020 ^[9]	66 / Male	Back pain, radicular pain	T12-L1	LIF (L1/2) with PLF (L1-3), sacro-iliac fusion, ACDF(C5-7)	8 years	hemilaminectomy, partial facetectomy (T12-L1)
Ogawa Y <i>et al.</i> 2012 ^[11]	72 / Male	Lower back pain	L4/5	ALIF(L5/S1), TLIF+PLF (L4/5)	4 weeks	Conservative therapy without antibiotics

LIF, lumbar interbody fusion; PLF, posterolateral fusion; ACDF, anterior cervical decompression and fusion; TLIF, transforaminal lumbar interbody fusion; IL-1, interleukin-1; ALIF, anterior lumbar interbody fusion

CONCLUSION

A case of acute epidural abscess suspected of epidural hematoma after cervical laminectomy was reported. The acute onset of SEA complicated with CPPD after cervical spinal surgery has rarely been reported. The crystal-induced inflammation of the CPP could amplify the pyogenic inflammation.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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