

## 症例報告

### RADICULOMYELOPATHY DUE TO TUMORAL CALCIFICATION OF THE LIGAMENTUM FLAVUM OF THE CERVICAL SPINE

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*Abstract* : A case of a 68-year-old woman with radiculomyelopathy caused by unilateral tumoral calcification of the ligamentum flavum of the cervical spine is reported. The patient developed the left shoulder pain and tetraparesis in the previous two years, and the symptoms has deteriorated in spite of conservative therapy. Neurological examination revealed motor weakness, hyperreflexia and hypesthesia in the four extremities. Neuroradiologically, a unilateral nodular opacification was shown in the left C4-C5 level. CT scan revealed nodular calcification, and MRI showed marked compression of the cervical cord by the calcified mass. Removal of the calcification through the left hemilaminectomy was performed using ultrasonic bone curettage. Light microscopic examination showed calcified deposits in the ligamentum flavum without crystals of calcium pyrophosphate dihydrate (CPPD). The patient was discharged from the hospital with improvement of the neurological symptoms

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**Key words** : cervical spine, calcification, ligamentum flavum

#### INTRODUCTION

Ossification of ligamentum flavum was reported usually in the lower thoracic and lumbar region, and rarely seen in the cervical region. Calcification of cervical ligamentum flavum is also relatively rare<sup>1)</sup>. Calcification of the ligamentum flavum and calcium pyrophosphate dihydrate (CPPD) crystal deposition disease<sup>2, 3)</sup> in the cervical region are thought to be the same entity, representing different aspects at different chronological stages<sup>4)</sup>.

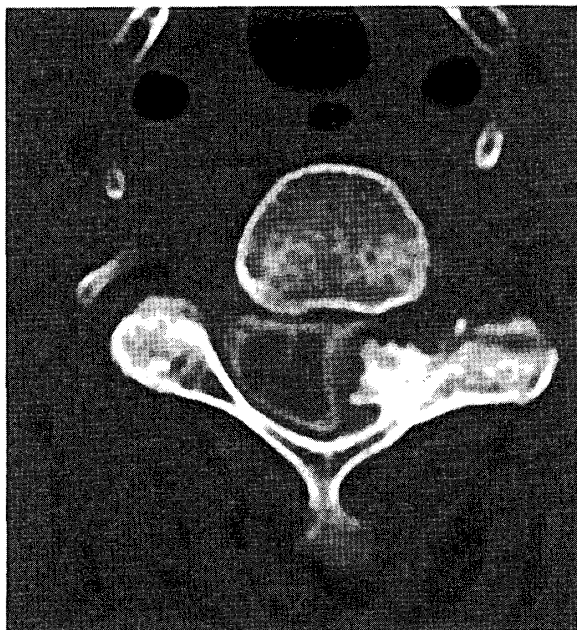
CPPD deposition disease is an inflammatory arthropathy that is defined by the deposition of CPPD crystals in articular and periarticular structures. The deposition of CPPD in hyaline cartilage and fibrocartilage leads to the chondrocalcinosis that is characteristic of the disease. It can occur independently or in association with any of a number of inflammatory or endocrine disorders. This form of crystal-induced arthritis tends to affect the peripheral joints, particularly the knees, ankles, shoulders, wrists, and second and third metacarpophalangeal joints, but involvement of the lumbar spine is not uncommon. However,

cervical spine disease due to CPPD deposition is rare<sup>5-7</sup>.

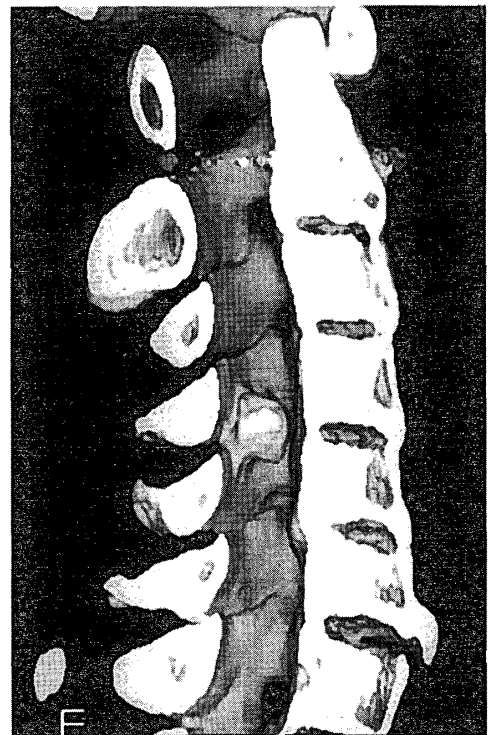
We here report a case with compressive radiculomyelopathy due to tumoral calcification of the ligamentum flavum of the cervical spine. This report presents the clinical and neurological manifestations in a case of calcification of the ligamentum flavum of the cervical spine.

### CASE REPORT

This 68-year-old woman was admitted in June, 2003. She had developed left shoulder pain and tetraparesis in the previous two years, and the symptoms progressed gradually until the time of admission in spite of conservative therapy. She had no history of spinal injury or joint pain. On admission, general physical examination showed no abnormalities. Neurological examination revealed motor weakness, hyperreflexia and hypesthesia in the upper and lower extremities. Plain radiographic examinations of the cervical vertebrae in the lateral projection disclosed a round shadow of calcification between C4 and C5 laminae. Computed tomography (CT) scan, metrizamide CT scan with bone algorithms (Fig.1A) and 3-D CT (Fig. 1B) revealed a unilateral nodular opacification in the C4-C5 level in the left, and metrizamide CT (Fig. 1A) and MRI (Fig. 2) showed marked compression of the cervical cord by the nodule. Bone scintigram showed symmetrical accumulation in C4-C5 level, which showed spondylotic change.



(A)



(B)

Fig. 1. Axial metrizamide computed tomographic (CT) scan with bone algorithms (A) and 3-D CT (B), showing the densely ossified ligamentum flavum markedly compressing the spinal canal in the left.



Fig. 2. Axial T2-weighted MRI scan, showing ovoid, hyperintense, ossified ligamenta flava compressing the spinal cord at the C4-C5 level.

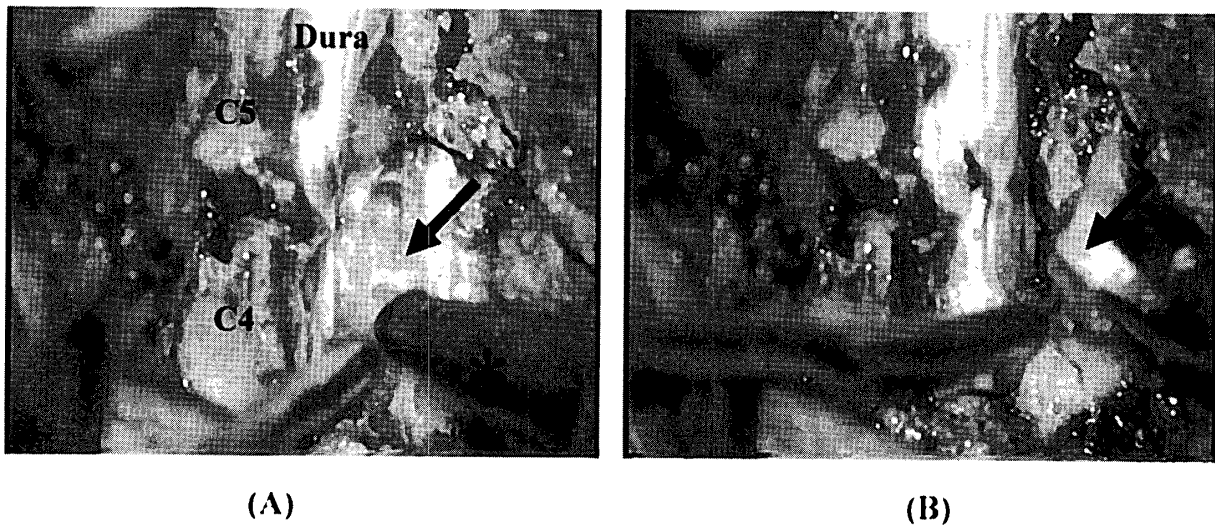


Fig. 3. Operative view. A: Spinal cord is compressed by the ossified ligament (arrow) located at C4-C5 level, and the calcification was removed by ultrasonic bone curette (\*). B: The ossified ligament (arrow) was removed, and the dura recovered to the normal size.



Fig. 4. Postoperative axial CT scan with bone algorithms, showing the disappearance of the ossified ligament.

Left hemilaminectomy was performed from C4 through C5. Calcified nodules were found in the left. Although the nodule adhered to the dura tightly, it was possible to remove it completely using ultrasonic bone curettage (SONOPET UST-2001, Miwatec Co. Ltd., Kawasaki, Kanagawa, Japan) (Figs 3A and 3B). Postoperatively, the patient showed remarkable improvement of neurological signs and symptoms (Fig. 4). Light microscopic examination of the specimen showed calcified deposits in the ligamentum flavum without crystals of CPPD.

#### DISCUSSION

Hypertrophy and ossification are well known disorders of the ligamentum flavum of the thoracic and lumbar vertebrae. However, only a few reports are available describing patients with symptomatic calcification of the ligamentum flavum of the cervical vertebrae<sup>1)</sup>. Iwasaki et al. reported the clinical characteristics of symptomatic calcification of the ligamentum flavum of the cervical vertebrae from the 10 cases in the literature: (1) nine of the 10 patients were women; (2) all patients were over 60 years old; (3) the calcification was frequently observed in the lower cervical vertebrae; (4) associated disc disease was sometimes noted; and (5) concurrent ossification of the posterior longitudinal ligaments appeared only in one male patient<sup>1)</sup>. With the increasing availability of MRI, calcification

and ossification of the ligamentum flavum is being more frequently recognized.

Although the mechanism of the calcification of the ligamentum flavum has not yet been established, the induction by CPPD deposit (as seen in cases of pseudo-gout<sup>2, 3)</sup>) and the degeneration of the elastic fiber of the flavum due to abnormal nutritional conditions are postulated. In every case of calcified and ossified ligamentum flavum, the excised specimen should be examined histologically, which will lead to the recognition of more cases of spinal CPPD.

Muthukumar et al. reported that tumoral CPPD of the ligamentum flavum was rare, and it commonly occurred among middle-age or elderly female patients and presents with progressive myelopathy. CT and MRI are complementary in the diagnosis of this condition. Surgery, if performed early, leads to good improvement. However, long-term follow-up monitoring of these patients is necessary, because surgery provides only symptomatic relief and does not treat the underlying disease<sup>8, 9)</sup>. Sato et al. reported neuroradiological findings of calcium crystal deposition in cervical ligamentum flavum; plain radiography showed oval calcified nodules. On computed myelography, oval calcified nodules were seen in the ligamenta flava of the posterior cervical spinal canal compressing the dural sac. MRI revealed oval corresponding hypointense foci both on T1- and T2-weighted images. In most symptomatic cases, the cervical cord showed hyperintensity at the compression site on T2-weighted images<sup>10)</sup>.

As to the operation for such calcifications, the ossified ligament tenaciously adheres to the dura<sup>9)</sup>. Therefore, the ossified ligament should be drilled to thinning the ligament to a fine sheet, and to allow the thinned ligament to float as islands of bone adhering to the dura. This method relieves the spinal cord compression without causing troublesome postoperative cerebrospinal fluid leaks. This method is similar to the anterior floating method for treatment of cervical myelopathy caused by an ossified posterior ligament. However, concerns exist regarding direct and heat injury to dura and spinal cord from the spinning drill bit. In this case, we used an ultrasonic bone curette<sup>11)</sup>. The ultrasonic surgical equipment comprises a power supply unit, footswitch, and handpiece (HB-13S, length 210 mm). The handpiece tip is 2.8 mm wide, and the amplitude of longitudinal vibration can be 320 microns at an ultrasonic frequency of 25 kHz. Cool-controlled irrigation fluid emerges near the tip, through the sheath. Ultrasonic bone curettage represents safe instrumentation for such surgery without damage to surrounding structures and without incurring the surgeon's psychomotor stress.

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