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CASE REPORT

Full endoscopic surgery for calcium pyrophosphate deposition disease (CPPD) in the cervical ligamentum flavum: report of two cervical myelopathy cases

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Abstract

Calcium pyrophosphate deposition disease (CPPD), known as pseudogout, is characterized by the accumulation of calcium pyrophosphate crystals in musculoskeletal structures, primarily joints. While CPPD commonly affects various joints, involvement in the cervical spine leading to myelopathy is rare. Surgical intervention becomes necessary when conservative measures fail, but reports on full endoscopic surgeries are extremely rare. We present two successful cases where full endoscopic systems were used for CPPD removal in the cervical spine. The surgical technique involved a full endoscopic approach, adapting the previously reported technique for unilateral laminotomy bilateral decompression. Full-endoscopic removal of cervical CPPD inducing myelopathy were successfully removed with good clinical and radiologic outcomes. The scarcity of endoscopic cases for cervical ligamentum flavum CPPD is attributed to the condition's rarity. However, our successful cases advocate for endoscopic surgery as a potential primary treatment option for CPPD-induced cervical myelopathy, especially in elderly patients or those with previous cervical operation histories. This experience encourages the consideration of endoscopic surgery for managing cervical ligamentum flavum CPPD as a viable alternative.

Keywords Calcium pyrophosphate deposition disease · Cervical spine · Endoscopic spine surgery · Cervical myelopathy

Introduction

Calcium pyrophosphate deposition disease (CPPD), also known as pseudogout, is a condition characterized by the accumulation of calcium pyrophosphate crystals within the musculoskeletal structures such as joints [17]. It can manifest with a wide range of clinical symptoms, from asymptomatic cartilage calcification to various conditions such as inflammatory pathologies reminiscent of pseudogout [13]. CPPD can manifest in various locations, including joint cartilage surfaces, joint capsules, ligaments, and intervertebral discs [4], and while the predilection for numerous joints has been extensively cataloged in the literature, involvement of the cervical spine remains an infrequent entity. Within the realm of cervical manifestations, the emergence of myelopathy secondary to CPPD crystal deposition is an even rarer clinical scenario. While initial management often involves conservative treatments such as anti-inflammatory medications and physical therapy, progressive neurological deterioration due to mechanical compression from deposits in the spine often necessitates surgical intervention, and several previous reports have demonstrated the need for surgical resolution in cases of CPPD deposition in the cervical spine as well [1].

Recently, spine surgery using endoscopy has gained preference among medical professionals and patients due to similar or better outcomes compared to open surgery [9]. The use of this minimally invasive technique is already widely accepted, and its utilization is continuously expanding. More and more indications that were not considered good candidates for endoscopic surgeries are being explored recently [2]. However, due to the scarcity of CPPD-induced cervical myelopathy, literature reporting surgeries using endoscopy for cervical CPPD patients is extremely rare. In this report, we aim to present two cases where full endoscopic systems

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were successfully used to remove CPPD deposits in the cervical spine.

Case report

Case 1

An 82-year-old female patient presented with cervicalgia and right upper and lower limb motor weakness that had been occurring for the past three weeks. The patient had a medical history of bilateral total knee replacement surgery performed seven years ago. Additionally, she had a history of angina, dyslipidemia, hypertension, and diabetes and was taking medications for these conditions. Neurological examination revealed a weakness of grade 4 in the right thumb extensor and grade 4 in the right hip flexor, along with the presence of upper motor neuron signs including Hoffmann's sign in the right hand. Hypoesthesia was also observed across the front of the right hand.

On X-ray imaging, a posterior mass at the C5/6 level was identified, and the CT scans clearly revealed a posterior calcified mass at both the C4/5 and C5/6 levels, beneath the right lamina. Magnetic resonance image (MRI) of the cervical spine presented the calcified mass at the C4/5 and C5/6 levels, narrowing the spinal canal and compressing the spinal cord with significant high signal intensity. Together with the clinical manifestation it was indicative of accompanied compressive myelopathy (Fig. 1). Cord compression was more prominent at the C4/5 level than at the C5/6 level. The diagnosis was believed to be CPPD of the cervical ligamentum flavum inducing myelopathy.

A surgical resection of CPPD was planned. Given the patient's advanced age and the goal of minimizing subsequent injury of normal cervical structures, a full endoscopic approach was chosen to access the adjacent two segments with a single incision. The ligamentum flavum including CPPD was totally resected and the central canal was successfully decompressed. Post-operation CT and MRI confirmed the successful removal of the canal compromising mass, with full decompression of the spinal cord (Fig. 2).

Histopathology report confirmed the diagnosis of CPPD within the removed ligamentum flavum. The patient experienced significant pain relief compared to before surgery and showed considerable improvement in gait and hand mobility.

Case 2

A 74-year-old female patient presented with bilateral hand numbness and clumsiness that had been occurring for the past three months. She had a history of multi-segment posterior cervical decompression, without knowing the detailed diagnosis.

The MRI scan presented bilateral thickening of the ligamentum flavum with resultant spinal canal stenosis and cord

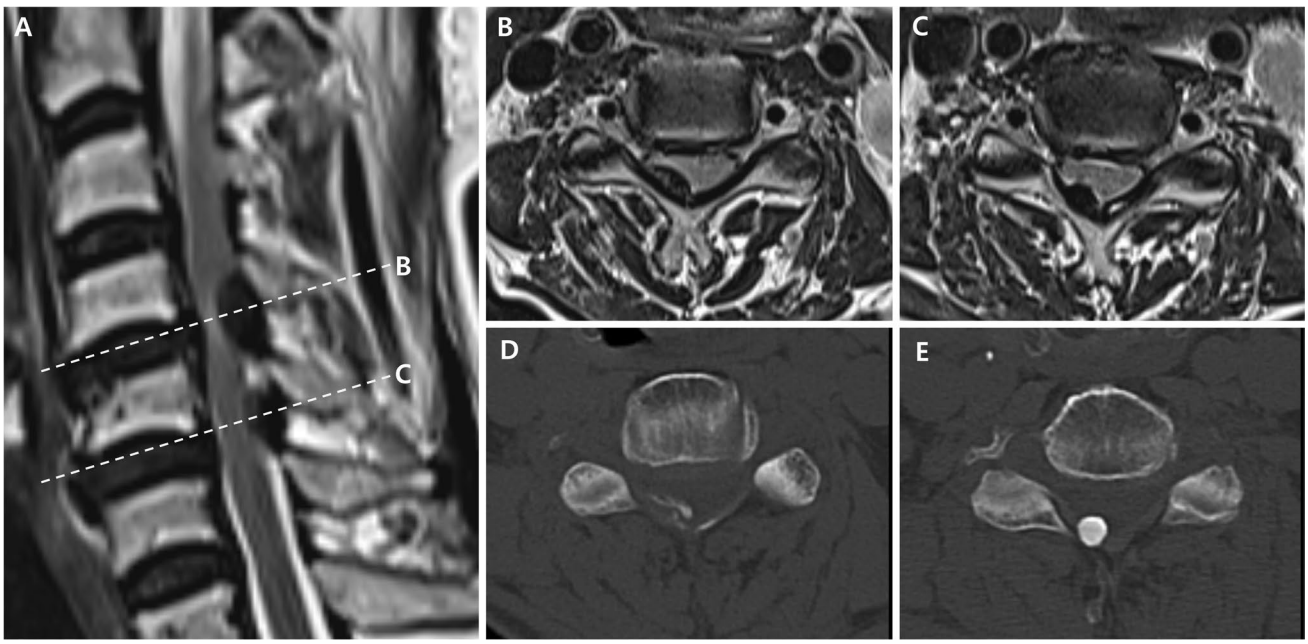

Fig. 1 Pre-operative images of Case 1. **A**: Sagittal plane of MRI scan, indicating compressive myelopathy at C4-5 and C5-6 levels, with more severe cord signal change at C4-5. **B**: Axial plane of MRI scan at C4-5 level. **C**: Axial plane of MRI scan at C5-6 level. **D**: Axial plane of CT scan at C4-5 level. **E**: Axial plane of CT scan at C5-6 level, with a clearer boundary of the mass observed at C5-6 compared to C4-5

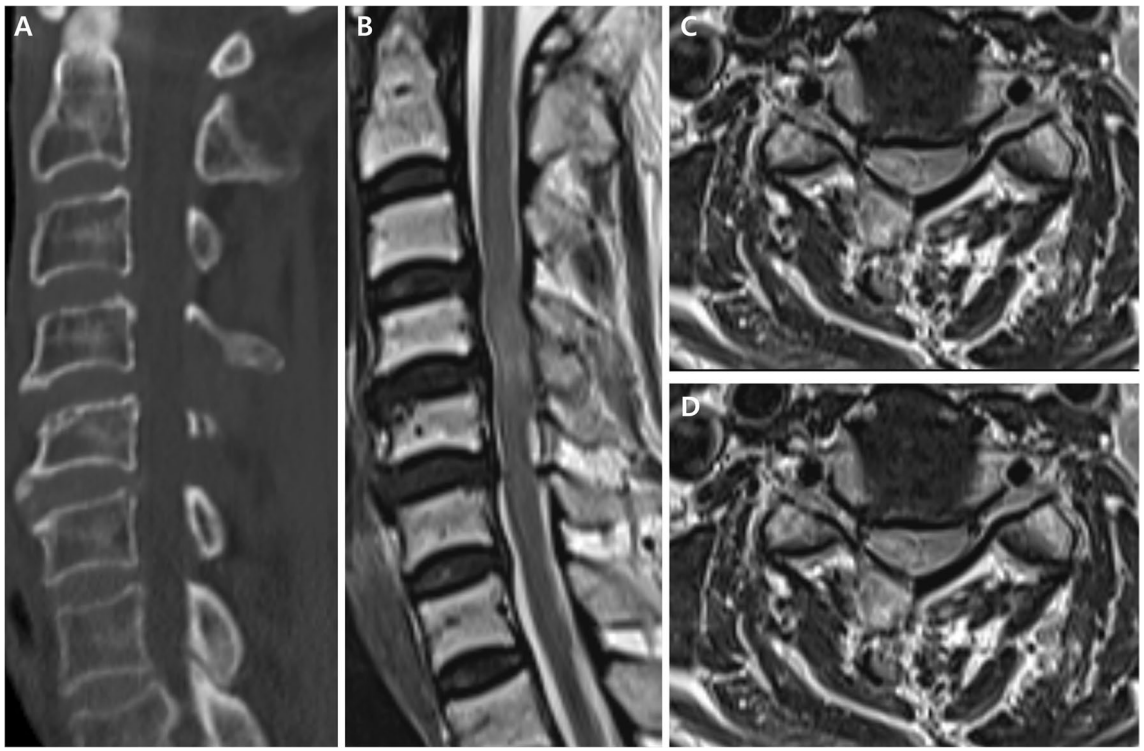

Fig. 2 Post-operative images of Case 1. **A**: Sagittal plane of CT scan, confirming complete removal of the mass. **B**: Sagittal plane of MRI scan. **C**: Axial plane of MRI scan at C4-5 level. **D**: Axial plane of MRI scan at C5-6 level, indicating sufficient cord decompression after surgery

signal change, and the following CT scan showed highly dense CPPD compromising the spinal canal (Fig. 3).

As there was significant clinically and radiologically confirmed myelopathy induced by CPPD in the ligamentum flavum, a posterior decompression surgery was planned. Due to the expected presence of scar tissue formation and adhesions from the previous surgery, limitations were anticipated in muscle dissection and other procedures. Therefore, an endoscopic approach was chosen. Total resection of the ligamentum flavum was performed and post-op CT and MRI

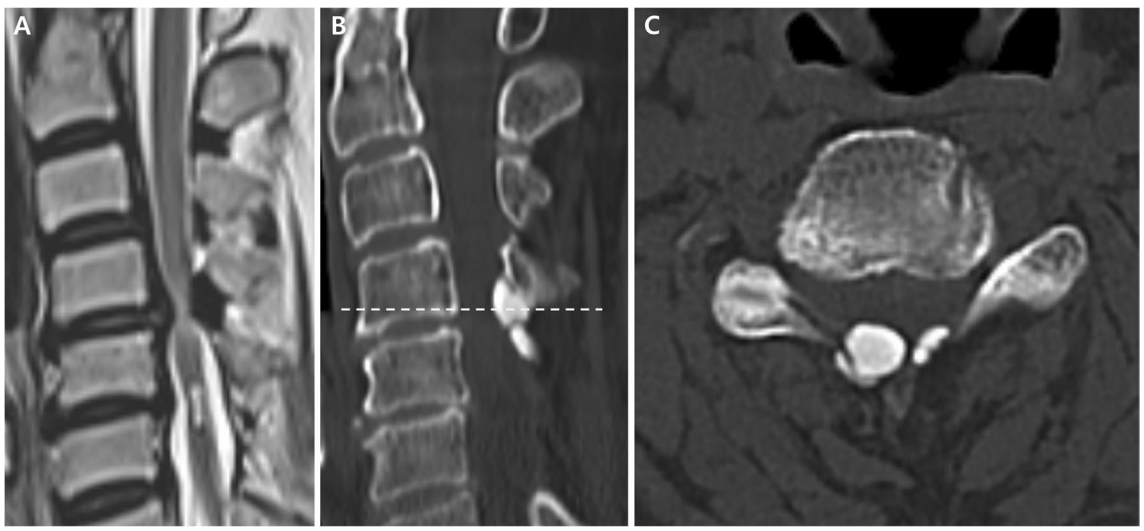

Fig. 3 Pre-operative images of Case 2. **A**: Sagittal plane of MRI scan, displaying the posterior mass at C4-5 with clear cord signal change. **B**: Sagittal plane of CT scan, revealing the previously operated area at C5 and C6 and the posterior mass above it. **C**: Axial plane of CT scan at C4-5 level, showing a well-defined high-density mass on both sides

confirmed the successful removal of the mass, resulting in spinal cord decompression (Fig. 4).

The patient was discharged two days after surgery. She exhibited improved hand numbness and better hand mobility compared to before the surgery.

Surgical technique

The surgical technique for full endoscopic CPPD removal for myelopathy follows the previously reported technique for full endoscopic unilateral laminotomy bilateral decompression (ULBD).

First, under general anesthesia, the patient is positioned face down on a radiolucent table. During the procedure, the use of a Mayfield head-fixator is optional. Slightly flexing the patient's neck helps widening the interlaminar space, minimizing the size of the following laminotomy and aiding in effective lesion removal. With anteroposterior fluoroscopic imaging guided by a C-arm fluoroscope, the medial border of the pedicle is checked, and a line connecting cranial and caudal medial pedicle line is drawn (Fig. 5). Subsequently, using lateral fluoroscopic imaging, a virtual line is drawn between the spinous processes above and below the desired surgical level, which will be the trajectory for surgery. Usually this imaginary line lands on the facet joint of the affected level. The incision line is made less than 1 cm in length and intersects the two drawn lines at their intersection point. After skin incision, serial dilation is performed, followed by insertion of the working tube (30-degree beveled), with caution throughout to prevent unexpected contact with the spinal cord, necessitating guidance from lateral fluoroscopic images. Upon confirming the positioning of the working channel at the V-point where the upper and lower lamina meet on both lateral and anteroposterior fluoroscopic images, the endoscope (15-degree viewing optical angle, 7.3 mm outer diameter, 4.7 mm working channel and 150 mm working length; iLESSYS Pro, Joimax, Karlsruhe, Germany) is introduced. Afterward, the soft tissue is removed using a bipolar cautery, punches and graspers to expose the V-point.

The next step involves performing a precise laminotomy (Fig. 6). Using a diamond burr, the caudal half of the cranial lamina and the cranial half of the caudal lamina are drilled carefully until both ends of the ligamentum flavum are exposed. A Karrison punch is used to carefully cut the lamina where the ligamentum flavum is attached. In extremely narrow spinal canals, paper thin drilling of the lamina to minimize the use of Kerrison punches can be effective. After completing this process, the ligamentum flavum which embeds the CPPD within it is completely detached from the lamina and then removed. Unilateral or bilateral removal of the CPPD and ligamentum flavum is performed based on the bilaterality of the disease. In cases of bilateral lesions, contralateral decompression is required. Before this process, ensuring sufficient space by undercutting the spinous process is crucial to prevent compression of the spinal cord by the instruments. Undercutting of the spinous process is carried out by widening the ipsilateral hemi-laminotomy medially using a diamond burr. After ensuring enough space to remove the contralateral ligamentum flavum and lesion without compressing the spinal cord with instruments, they are removed. Once it is confirmed that the lesion causing spinal cord compression has been completely removed, the surgery is concluded. Meticulous hemostasis is required throughout the procedure, and the insertion of a drain is optional. Wound is sutured and glued

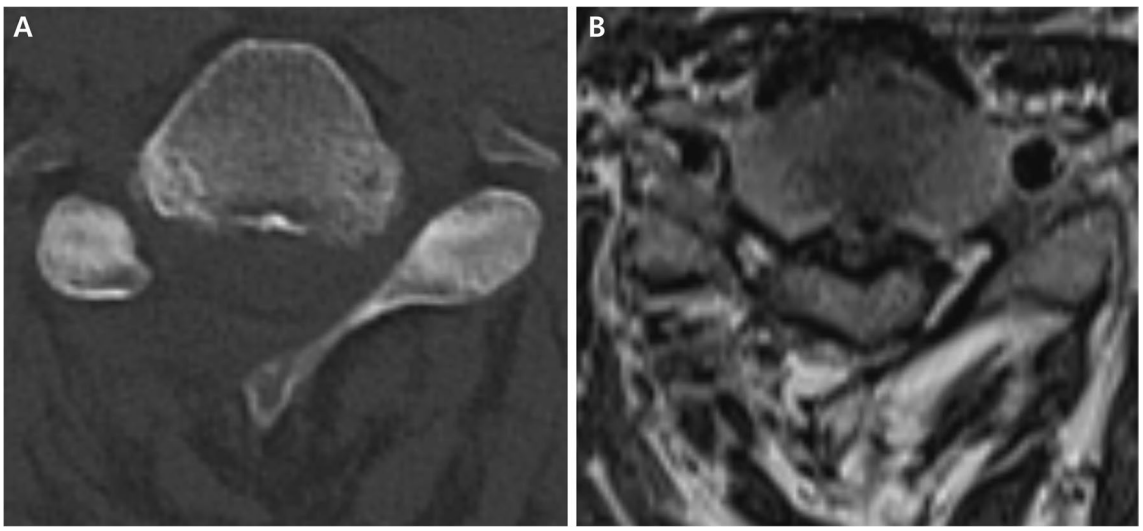

Fig. 4 Post-operative images of Case 2. **A**: Axial plane of CT scan, confirming the removal of nodular masses bilaterally. **B**: Axial plane of MRI scan, showing sufficient cord decompression

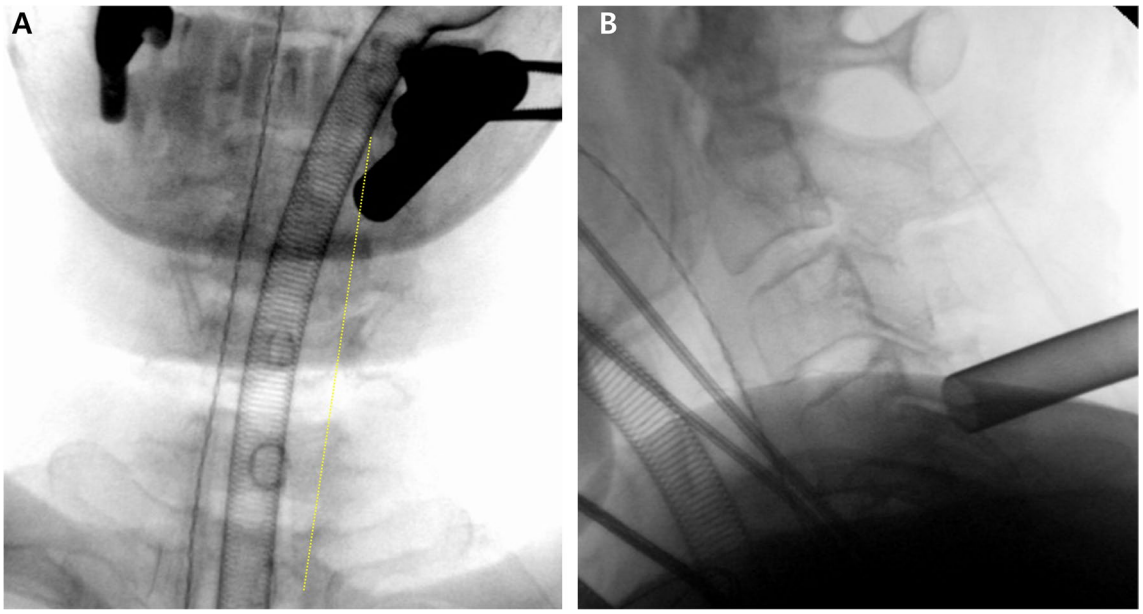

Fig. 5 Fluoroscopic images during surgery. **A**: Anteroposterior (AP) X-ray shows the tip of the working channel is placed on medial pedicle line (yellow dotted line). **B**: Lateral X-ray shows the approach trajectory

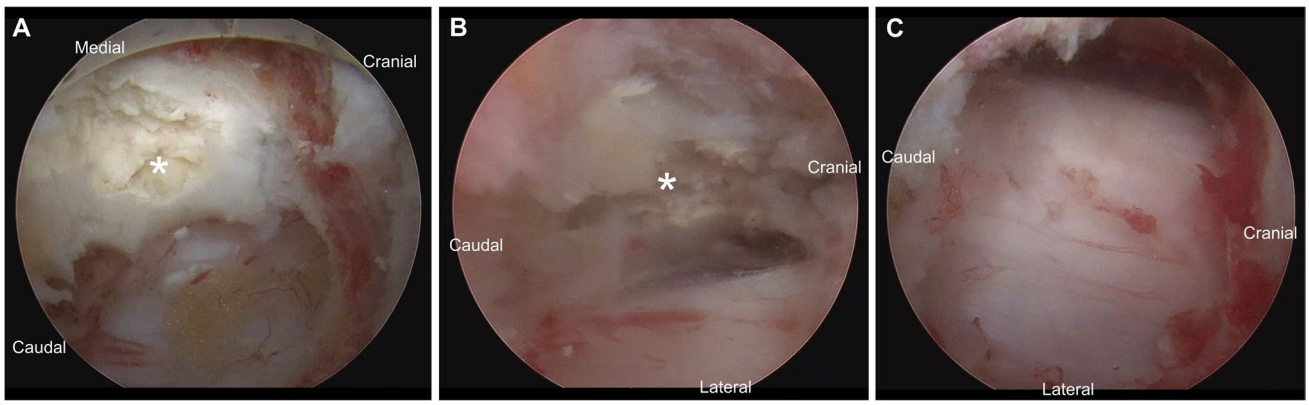

Fig. 6 Intra-operative image after laminectomy. **A**: Clearly visible calcium pyrophosphate deposition (CPPD) within the ligamentum flavum (white asterisk). **B**: Contralateral CPPD (white asterisk). **C**: Bilateral full decompression of the spinal cord presented after removal of the ligamentum flavum and CPPD embedded within it

with topical skin adhesives (Fig. 7). The overall summarized video of a representative case is provided as Video 1.

Discussion

CPPD is characterized by the deposition of calcium pyrophosphate crystals in joints and connective tissues, causing inflammation and joint damage [17]. While most cases of CPPD are idiopathic, associations have been reported with aging, osteoarthritis, gout, hyperparathyroidism, hemochromatosis, and hypomagnesemia [13]. CPPD commonly occurs in large joints, but it can also be found in the intervertebral discs or ligaments of the spine.[3]In the cervical spine, CPPD deposition in the ligamentum flavum subsequently affects the spinal canal and therefore can manifest with clinical symptoms such as pain or myelopathy symptoms [12]. CPPD deposition in the ligamentum flavum of the cervical spine, like in other locations, may be managed with NSAIDs or steroids on a symptomatic basis unless significant neurological deficits are accompanied [4, 13]. However, when mechanical compression occurs, leading to progressive myelopathy similar to other spinal disorders, surgical intervention becomes necessary [1, 4, 12].

Cervical posterior laminectomy is a surgical treatment option for CPPD deposition in the ligamentum flavum of the

Fig. 7 Wound after posterior cervical full-endoscopic surgery

cervical spine, particularly when there is no severe spinal stenosis or excessive neck motion requiring stabilization [1, 4, 12]. Previous case reports have highlighted the successful surgical treatment of progressive myelopathy due to cervical CPPD [1, 4, 12]. However, reported cases of surgical intervention using endoscopy for CPPD deposition in the ligamentum flavum are extremely rare. Endoscopic spine surgery, introduced in the 1990s, has evolved and become preferable due to advantages such as shorter surgery times, minimal tissue damage, good visualization of the surgical field, minimal bleeding, reduced surgical wounds, and shorter hospital stays compared to traditional open surgery [3, 6, 9]. While there are specific points that need to be addressed regarding complication management [8–10], the utilization of it in spinal pathologies is expanding significantly. Endoscopic surgery was once considered relatively contraindicated for complex scenarios, including revisional surgeries and calcified discs, recent studies demonstrate its applicability even in challenging cases, expanding its scope to fusion surgery and some variant procedures [7, 16]. In the current scenario, as the application of endoscopy is expanding, the scarcity of endoscopic surgical cases for CPPD deposition in the cervical ligamentum flavum can be attributed to the rarity of the condition. We believe that our report on the full-endoscopic treatment of this rare pathology will add value to the field by addressing another expanded indication for full-endoscopic cervical surgery.

CPPD deposition in the cervical ligamentum flavum may resemble OLF, but it demonstrates distinct radiological and intraoperative differences. Lu et al. reported that CPPD deposition in the cervical ligamentum flavum differs from OLF, presenting as nodular and not extending to the posterior facet joints, with no continuity with the lamina [12]. Additionally, Takahashi et al. reported that CPPD deposition in the cervical ligamentum flavum does not adhere to the lamina or dura mater [14]. These observations were consistent in our cases, where chalky calcium crystal particles were visibly identified within the ligamentum flavum. Despite the differences in characteristics, our two cases suggest that they can be successfully treated through full-endoscopic posterior decompression, using the same technique as posterior cervical ULBD. This, in turn, may reduce the hesitation among surgeons in choosing endoscopy as a surgical option for CPPD deposition.

In our cases, endoscopic surgery was chosen based on the patient's condition and circumstances. As the patients were both elderly patients, selecting the least invasive available technique was why we did posterior full-endoscopic decompression. Another major reason was that the second patient had a previous surgery on the posterior cervical spine. As significant epidural adhesion was expected, trying a full-endoscopic surgery removed the necessity of getting through epidural scars and directly led us to the affected laminae. Open surgery poses the risk of incidental durotomy during the opening procedure[5, 15], however under full-endoscopic surgery this opening step can be skipped as the working channel directly lands on the target site, and this is a great advantage reducing the need of work at the previous op scar tissue adjacent to the spinal cord.

Drawing from other reports from different groups, ossified ligamentum flavum can be easily and safely removed [11], and in our cases we report that CPPD removal could be successfully achieved by the similar technique. Although the patients were both elderly and had specific circumstances such as previous operation scars, full endoscopic CPPD removals were uneventful and both cases showed neurological symptom improvement postoperatively. Our cases highlight endoscopic surgery as a potential surgical option for cervical ligamentum flavum CPPD deposition. We hope that our experience encourages the consideration of endoscopic surgery as a viable option for the surgical treatment of cervical ligamentum flavum CPPD deposition.

Conclusion

The scarcity of endoscopic surgical cases for cervical ligamentum flavum CPPD can be attributed to the condition's rarity. Despite concerns about complications, our cases

demonstrate successful endoscopic surgeries for CPPD removal, suggesting endoscopic surgery as a potential alternative option for this condition. It can be especially more beneficial for elderly patients or those with previous posterior cervical operation histories. This experience encourages considering endoscopic surgery for cervical ligamentum flavum CPPD as a viable primary surgical treatment.

Supplementary Information The online version contains supplementary material available at https://doi.org/10.1007/s00701-024-06080-4.

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