

## Contrast Enhancing Cervical Spondylotic Myelopathy- An Unusual Presentation: Case Report and Review of Literature

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### Abstract

We present an unusual case of cervical spondylotic myelopathy (CSM) with dual enhancement pattern on MRI. A 54-year-old female presented with neck pain, brachialgia and acute onset tetra-paresis with bowel and bladder involvement. MRI showed disc prolapse at C5-6 and C6-7 with extensive cord edema. Contrast MRI showed intramedullary enhancement along with leptomeningeal enhancement. CSF analysis was equivocal. In view of this clinical and radiological picture, diagnosis of intramedullary tubercular granuloma with coexisting cervical spondylosis was sought. So, she was started on anti-tubercular treatment along with steroids with which patient improved dramatically. Weakness, however, reappeared after which two-level anterior corpectomies (C6, C7) was done. By 12 months patient recovered significantly clinically and radiologically. If the diagnosis of CSM cannot be definitively made because of atypical contrast enhancement on MRI, surgical intervention can be staged after ruling out other medical causes of contrast enhancement. A decompression is initially performed and a spinal cord biopsy is planned as a second stage if the patient shows no clinic-radiological improvement.

**Keywords:** Cervical Spondylotic Myelopathy; MRI; Leptomeningeal; Contrast Enhancement

### Abbreviations

MRI: Magnetic Resonance Imaging; CSM; Cervical Spondylotic Myelopathy; CSF: Cerebro Spinal Fluid; CT: Computed Tomography; TIRM: Turbo Inversion Recovery Magnitude

### Introduction

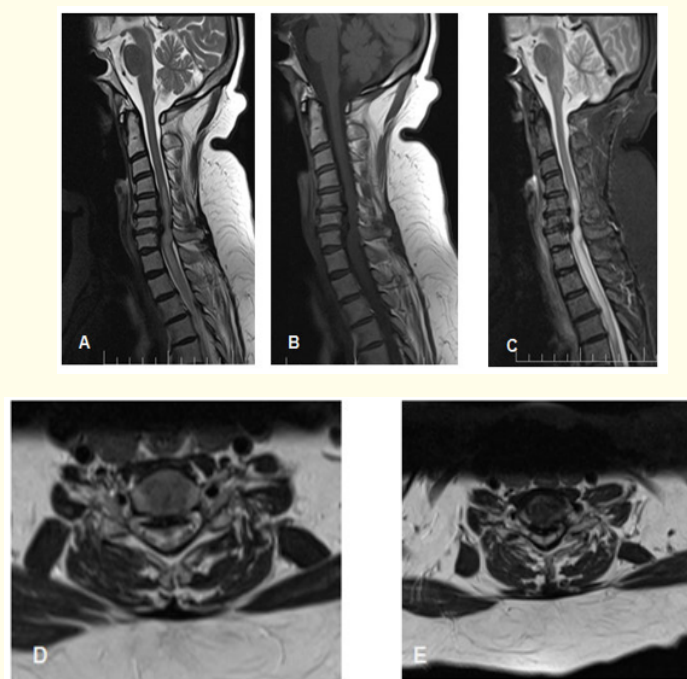
Among all the causes of cervical myelopathy, cervical spondylotic myelopathy (CSM) is the commonest cause for non-traumatic cervical myelopathy [1]. CSM is usually due to static compression of the spinal cord with or without coexisting dynamic compression. The diagnosis of CSM is confirmed by Magnetic Resonance Imaging (MRI) which also helps in differentiating and recognizing the coexistence of other compressive or non-compressive pathologies [2]. Even though it's uncommon for patients with CSM to be worked up with contrast-enhanced MRI, it is usually indicated to exclude other suspected potential compressive or non-compressive causes of cervical myelopathy (infections, inflammations, tumors and vascular malformations). However few studies demonstrated intramedullary contrast enhancement in CSM on sagittal and axial MRI with an incidence of around 7.3% [3,4]. To our knowledge, none of the published literature has documented leptomeningeal contrast enhancement along with intramedullary disc like enhancement in CSM. The purpose of this study is to report a case of CSM with peculiar clinic-radiological presentation with dual enhancement pattern (leptomeningeal and intramedullary) on MRI.

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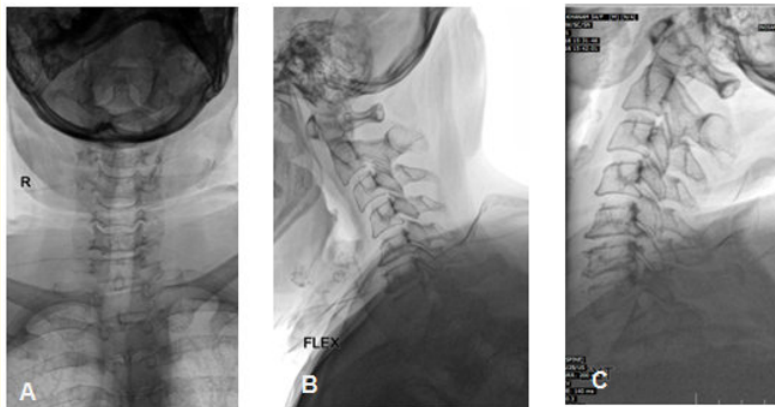
## Case Report

### History

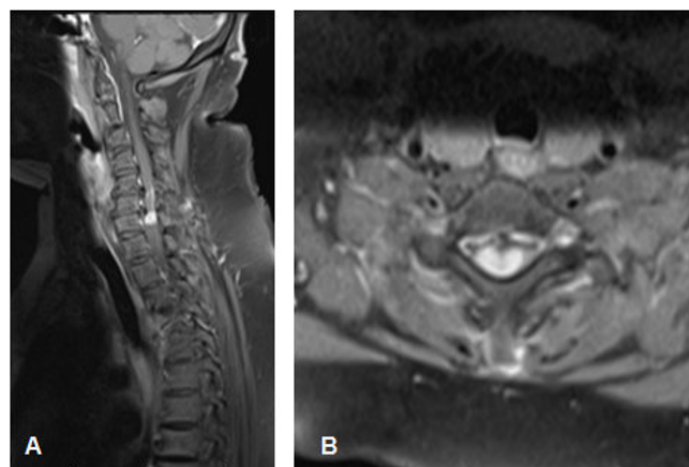
A 54-year-old diabetic female developed insidious onset and gradually worsening axial neck pain with bilateral upper limb radiculopathy for the last 2 months and gradually worsening weakness of all four limbs for the last 1 month. She had difficulty in performing fine motor tasks using upper limbs like buttoning and spoon feeding. She also had gait instability which progressed and finally, she was unable to walk due to her associated lower limb weakness. Since the last 1 week, she developed urinary incontinence with loss of bladder sensation and constipation. The history of adequate trauma, fever and constitutional symptoms was negative. Her higher mental functions and cranial nerve examination were normal. She had a normal tone in both upper limbs with grade 1 spasticity in both lower limbs and weakness in bilateral hand Intrinsic and bilateral lower limbs with positive Babinski and exaggerated deep tendon reflexes in both lower limbs and with a negative Hoffman's sign. Fine touch and pinprick sensation was reduced bilaterally below C8. Vibration and proprioception were affected in both upper limbs and lower limbs, with a positive Romberg's sign. She was clinically diagnosed to be having CSM and was subjected to MRI to confirm the diagnosis. The MRI showed anterior cord compression at C5-6 and C6-7 disc levels with hyperintensity extending from behind C4 vertebral body to behind T4 vertebral body on T2 and TIRM MR sequences (Figure 1). Cervical spine radiographs showed no instability (Figure 2). In view of disproportionate cord edema to the amount of compression, the patient was subjected to contrast MRI which showed intramedullary contrast enhancement behind C7 vertebral body along with cephalad leptomeningeal enhancement (Figure 3). The diagnostic possibilities sought were cervical spondylosis with coexisting tuberculous granuloma or inflammatory myelitis or intramedullary tumor with leptomeningeal metastasis. Total leucocyte count, differential leucocyte count and serum angiotensin converting enzyme levels were normal but with mildly elevated C reactive protein and a negative serum anti-nuclear antibodies. CSF analysis showed relative lymphocytosis with increased protein. Culture analysis for pyogenic organisms and gene expert (Tuberculosis Polymerase Chain reaction) were negative. CSF adenosine deaminase was normal and oligoclonal bands were absent. High-resolution CT Chest and MRI brain were normal. In view of this equivocal picture, she was started empirically on anti-tubercular treatment along with pulse intravenous steroid for 3 days after which her motor power improved substantially in both the lower limbs that she became ambulant. However, she again deteriorated neurologically in spite of ongoing anti-tubercular treatment under cover of a tapered dose of oral steroids.



**Figure 1:** (A to E) showing MRI T2, T1, TIRM sagittal images, C5-6 axial and C6-7 axial images respectively, showing significant compression at C5-6 and C6-7 disc levels and extensive cord edema.



**Figure 2:** (A to C) showing pre-op AP, lateral flexion and extension views respectively.



**Figure 3:** (A and B) showing contrast MRI sagittal and axial views respectively with intramedullary and leptomeningeal enhancement with no central grey matter sparing.

**Surgery and Postoperative course**

In view of this confusing clinical and radiological picture, the patient was advised surgical decompression along with spinal cord biopsy. However, patient rejected spinal cord biopsy in view of the associated risk of neurological deterioration. So, she was operated by anterior decompression where a two-level corpectomy (C6 and C7) was performed and anterior stabilization and fusion were done using harms cage (Figure 4). Post surgery patient was subjected to aggressive rehabilitation and antitubercular treatment was stopped and the patient became independent ambulator by 3 months and recovered bladder sensation by 12months but with persisting urge incontinence. Serial MRI examinations were done post-operatively at 2 weeks, 3 months and at 12 months period which showed a gradual decrease in the edema and complete disappearance of contrast enhancement in the final MRI at 12 months postop (Figure 5-7). However, 12-month postop MRI showed focal myelomalacia changes at C6 -C7 level (Figure 8). The modified Japanese orthopedic association score had increased from 7/17 preoperatively to 14/17 at final follow-up.

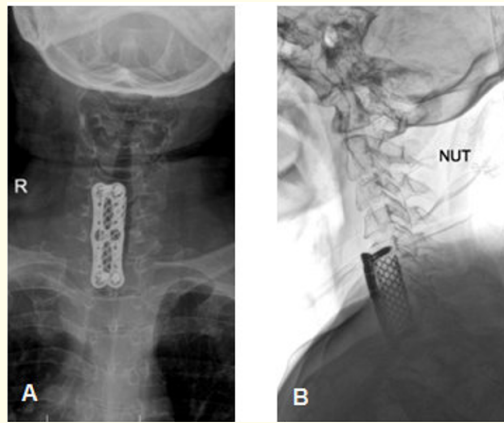


Figure 4: (A and B) showing postop AP and lateral radiographs.

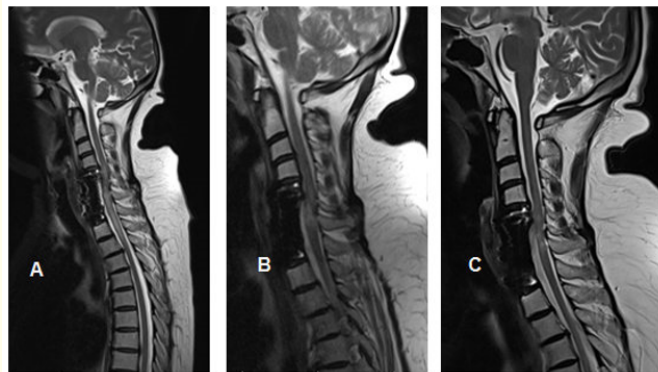


Figure 5: (A, B and C) showing 2-week, 3 month and 12month postop T2 sagittal MRI showing gradual decrease in the length of T2 hyper intensity.

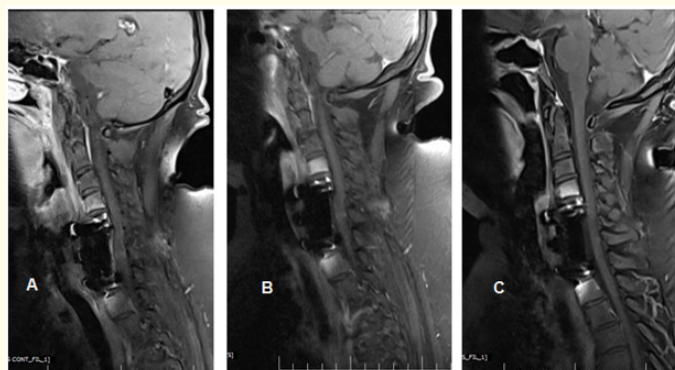
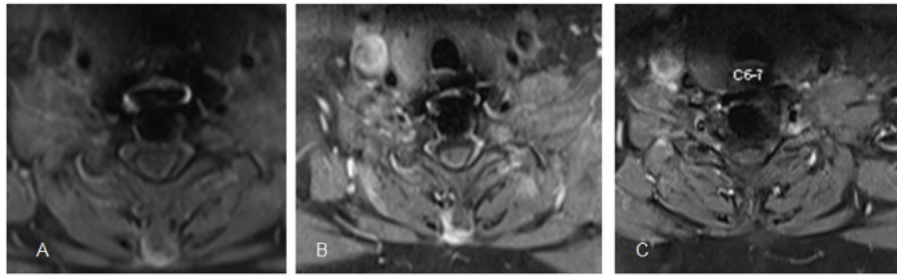


Figure 6: (A, B and C) showing 2 week, 3 month and 12month postop T1 contrast sagittal MRI showing gradual disappearance of enhancement.



**Figure 7:** (A, B and C) showing 2 week, 3 month and 12month postop T1 contrast axial MRI showing gradual disappearance of enhancement.



**Figure 8:** (A and B) showing 12 month postop T2 and T1 sagittal MRI with myelomalacia, recognized by patchy cord hypo intensities on T1 sagittal MRI.

## Discussion

Cervical myelopathy, a common pathological scenario, is a clinical diagnosis confirmed with imaging, predominantly MRI [5]. This can be due to a non-compressive or a compressive pathology. However, occasionally both can simultaneously coexist, posing a diagnostic dilemma. The predominant non-compressive causes of cervical myelopathy are transverse myelitis (para infectious, post infectious or idiopathic), demyelinating disorders (Multiple Sclerosis, Neuromyelitis Optica), systemic disorders (Systemic lupus erythematosus, Sjogren's syndrome, Scleroderma, Behcet disease, and Sarcoidosis), sub-acute combined degeneration and paraneoplastic myelitis [6]. The compressive pathologies can be extradural or intradural. The common extradural compressive etiologies are degenerative, infective and trauma (compression from bone or disc fragments or epidural hematoma). The common intradural compressive etiologies are tumors and cystic lesions (intra or extramedullary), syringomyelia, arterio-venous malformations and infections (tuberculous granulomas or myelitis) [6].

The incidence of contrast enhancement of spinal cord and meninges on MRI varies between different studies. However, the diagnostic significance and mechanism of contrast enhancement of these structures are related to the integrity of blood-spinal cord barrier in normal tissue and its breakdown in different pathologies [7]. Several *in-vitro* and *in-vivo* studies demonstrated that the normal blood-spinal cord barrier function to maintain a controlled homeostatic environment is disturbed by an injury to the spinal cord [8,9]. The usual causes of intra-medullary contrast enhancement on MRI are infections, inflammatory pathologies, demyelinating diseases and intramedullary spinal cord tumors [7]. Traumatic spinal cord injury can also be associated with such contrast enhancement [10,11]. As demonstrated by Flanagan, et al. CSM is sometimes found associated with specific patterns of intramedullary contrast enhancement on MRI, like a pancake like enhancement just below the site of maximal stenosis with sparing of central grey matter [3]. The enhancement pattern in our case though located just below the maximal stenosis in sagittal MRI, was not typically pancake type in appearance and had no central grey



matter sparing. However, it also showed cephalad focal, linear and uniform leptomeningeal enhancement. The usual causes of intra-dural extra-medullary contrast enhancement are infections, inflammatory pathologies, and intradural extramedullary tumors [7,12]. So, we thought of the possible intramedullary causes with associated leptomeningeal enhancement (dual enhancement) i.e. infections [13], inflammatory pathologies [12] and intramedullary tumors with coexisting leptomeningeal metastasis [14], as differential diagnosis and proceeded accordingly. The common multisystem inflammatory pathology associated with such dual enhancement is Sarcoidosis [15]. The incidence of neuro-sarcoidosis is 15%, with spinal involvement ranging from 6 to 8%. However, the incidence of isolated spinal neuro-sarcoidosis is less than 0.5% [16]. This patient had no systemic signs of sarcoidosis and the serum angiotensin-converting enzyme level was also normal. Intramedullary tumors with coexisting leptomeningeal metastasis can present with such dual enhancement pattern. But they are usually associated with thick and nodular leptomeningeal enhancement pattern [7]. However, at times there could be a coexistence of both CSM and an intramedullary tumor which could not be ruled out in our case [17]. Tuberculosis, particularly in endemic regions, is also known the cause of myelitis, which along with coexisting meningitis can produce the dual enhancement pattern. Pulse intravenous steroids constitute a part of medical therapy for tubercular myelitis along with anti-tubercular treatment [13,18].

In view of these differential diagnoses, and tuberculosis being endemic, we have empirically started our patient with pulse intravenous steroids and anti-tubercular treatment for which patient showed a drastic improvement initially followed by a deterioration. So, the patient was planned for posterior laminectomy and instrumented fusion along with spinal cord biopsy. In view of the risks associated with the spinal cord biopsy patient refused for the biopsy and after which an anterior decompression in the form of two-level corpectomy and fusion was performed [19]. Post-surgery, after aggressive rehabilitation patient became independent ambulator by three months and regained bladder sensation by twelve months, at the final follow-up. Though the preoperative axial contrast MRI did not show central grey matter sparing as mentioned by Flanagan, *et al.* the 2wk and 3 month postop MRI showed the pattern of central grey matter sparing. Also, the final MRI at 12 months showed the complete regression of contrast enhancement.

Though many case reports and case series [20-25] documenting contrast enhancement in CSM have been published before, none of them documented any evidence of leptomeningeal enhancement in their cases. The contrast enhancement in our case can also be explained by the leaky blood-spinal cord barrier which got healed once the spinal cord compression was addressed. Keeping this in mind doing a primary intramedullary spinal cord biopsy in such cases might be an aggressive approach.

## Conclusion

If intramedullary contrast enhancement with leptomeningeal enhancement is encountered in a case of CSM, all other causes of such dual enhancement pattern must be ruled out before planning any surgical intervention. If the diagnosis is still not clear, the surgical intervention can be staged, performing an initial decompression and planning a spinal cord biopsy as a second stage, if the patient shows no clinical and radiological improvement. Recognition of such contrast enhancement patterns in CSM is important to reduce the incidence of inappropriate interventions or delay in consideration of a potentially beneficial decompressive surgery.

## Highlights

- CSM is rarely associated with intramedullary contrast enhancement.
- Association of leptomeningeal enhancement is not reported in CSM.
- Our case elucidates dual enhancing CSM that disappeared with adequate decompression.
- So, if there is an atypical enhancement in CSM, surgical procedure can be staged.

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