Acute spontaneous cervical disc herniation causing rapidly progressive myelopathy in a patient with comorbid ossified posterior longitudinal ligament: Case report and literature review

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Abstract

Background: Ossification of the posterior longitudinal ligament (OPLL) and cervical disc herniation are commonly encountered neurosurgical conditions. Here we present an unusual case of nontraumatic rapidly progressive myelopathy due to cervical disc herniation with comorbid OPLL and conduct a literature review focusing on the frequency and management of disc herniations with OPLL.

Case Description: A 52-year-old healthy female presented with a 72-h history of rapid progression of dense quadriplegia with sensory deficits, with a precedent 4-week history of nontraumatic midline neck pain. Clinical examination revealed profound motor deficits below the C5 myotome. Spinal neuroimaging revealed OPLL (computed tomography [CT]) and a cervical disc herniation spanning from C4/5 to C5/6 with significant retrovertebral disease (magnetic resonance imaging [MRI]). Operative management involved an anterior cervical corpectomy and instrumented fusion, with removal of both the sequestered disc material and the locally compressive OPLL. The patient recovered full motor function and independent ambulation with no residual signs or symptoms of myelopathy at the time of discharge.

Conclusion: This unique case of a spontaneous cervical disc herniation in the context of OPLL causing rapidly progressive myelopathy illustrates the complementarity of CT and MRI in diagnosing the underlying cause of a rapidly progressive neurologic deficit in the absence of antecedent trauma. Though the optimal surgical management of such pathology remains uncertain; in this case, the anterior approach was motivated by the significant retrovertebral ventrally compressive sequestrum, and provided for excellent neurologic outcome. This article also reviews the occurrence/management of such acute cervical discs with OPLL.

Key Words: Cervical disc herniation, ossified posterior longitudinal ligament, quadriplegia
INTRODUCTION

Ossification of the posterior longitudinal ligament (OPLL) and cervical disc herniation occur in isolation, but their acute comorbid presentation is highly unusual. Cervical disc herniation, although most often asymptomatic, can contribute to a spectrum of neurological sequelae, including acute radiculopathy, myelopathy, or, rarely, quadriparesis/plegia. Here, we present a review of the literature regarding acute cervical disc herniation and OPLL along with a case study in a patient with acute onset dense quadraparesis. This is highly unusual in the context of OPLL where the osseous reinforcement of the PLL normally protects against neurological compression by an acute cervical disc herniation.

CLINICAL REPORT

A 52-year-old healthy female presented with a rapid, 72-h progression of quadraparesis. She described a prior 4-week history of gradual onset, nontraumatic midline neck pain, requiring narcotic analgesia, without radiculopathy. Immediately prior to admission, she began experiencing a progressive quadriparietic deficit (with sequential numbness of right upper and lower then left lower and upper extremities) without sphincter dysfunction.

Her motor examination revealed weakness in all extremities, most significant in the right lower extremity (grade 1/5 power), diffuse hyperreflexia (positive Hoffman’s sign and up-going Babinski sign bilaterally), and a C7 sensory level to light touch, loss of position/vibration sensation in the lower extremities but intact pin sensation. Notably, the digital rectal examination was unremarkable.

A large, contiguous C4-5 disc extrusion extending from the postero-inferior corner of C4 to the C5-6 disc space was identified on magnetic resonance imaging (MRI) [Figure 1]. Sagittal and axial computed tomography (CT) images identified OPLL surrounding the extruded disc [Figure 2]. Canal compression was measured and computed on axial MR and CT images using imaging software in-house [Figure 3]. Surgery included an anterior C5 corpectomy with removal of both sequestered disc tissue and OPLL. Multiple large fragments of herniated disc were identified posterior to the C5 vertebral body. The OPLL was removed but with an accompanying cerebrospinal fluid (CSF) leak treated with a dural patch, fibrin glue, and a lumbar drain (72 h). The corpectomy defect was reconstructed using a titanium mesh cage filled with autologous local bone, plus an anterior cervical plate. Postoperative upright radiographs indicated good bony alignment and satisfactory instrumentation placement [Figure 4]. She was discharged on her ninth postoperative day with grade 5 motor power in all myotomes and was mobilizing independently.

Figure 1: Preoperative T2-weighted sagittal (a) and proton density weighted axial (b-d) MRI images without contrast. The axial images demonstrate the level of spinal canal compromise at the level of the C4-5 disc (b), mid-C5 vertebral body (c) and C5-6 disc (d). An arrow identifies the cervical disc herniation with extrusion on the sagittal MRI.

Figure 2: Preoperative sagittal (a) and axial (b-d) CT images. Images b-d correspond to axial MRI images presented in Figure 1. The extent of bony canal compromise secondary to OPLL posterior to the C4-5 disc (b), mid-C5 vertebral body (c) and C5-6 disc (d) is illustrated. Note the double layer sign on the axial CT images.

Figure 3: Axial CT (a) and MRI (b) images at the level of maximal canal compromise posterior to the C5 vertebral body. Measurements of the crosssectional area available for the spinal cord secondary to bony (a) and soft-tissue (b) canal compromise are shown.
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DISCUSSION

Literature review of rapidly progressive myelopathy

Acute nontraumatic rapidly progressive myelopathy in the context of cervical disc herniation (with or without comorbid OPLL) is rarely encountered clinically, and a review of cases described in the English literature is summarized in Table 1. Included in this review, a search of PubMed, Web of Science, and gray literature, for cases of acute onset (which we defined as a period of up to 2 weeks) rapidly progressive myelopathy.

Three reported cases documented imaging findings of OPLL, and all three were treated with an anterior surgical decompression, with one case electing for additional posterior instrumentation. The clinical presentation of acute herniation leading to rapid, profound myelopathy often began with minor, focal neck pain, and slow progression of neurologic dysfunction. Only one patient had preexisting myelopathy.

The population of patients with cervical disc herniation and rapidly progressive myelopathy were heterogeneous with respect to age, sex, relative acuity of onset, rapidity of quadriparesis onset, and spinal level of disc herniation. Both the onset and resolution of neurologic dysfunction were variable in the documented cases. Cervical disc herniation stemming from the deterioration of intervertebral discs is believed to be multifactorial, and causes changes to the posterior longitudinal ligament, potentially leading to the development of myelopathy or radiculopathy through direct mechanical pressure and impingement of the spinal cord or nerve roots, respectively.

OPLL is thought to arise from unrestricted osteoblastic overactivity of hypertrophic PLL cells, although the inciting stimulus is uncertain. The typical natural history of OPLL involves asymptomatic progression of the ossification with 71% of patients being free from myelopathy at 30 years and only 17% of patients developing neurologic dysfunction. The authors of the three case reports of acute myelopathy with cervical disc and OPLL, as well as Iwamura and Hsieh, proposed a potential clinical correlation between the two pathologies. Cervical disc herniation is, however, generally not seen in combination with OPLL given the additional barrier force required for herniation.

Surgical management

Surgical intervention is not indicated in asymptomatic patients, with controversy existing over the optimal surgical management of symptomatic OPLL. While observation of asymptomatic OPLL remains the standard of care, symptomatic canal stenosis caused by OPLL can require surgery. Anterior decompression of the ventrally located pathology is often indicated and has shown lower rates of symptom recurrence, but higher rates of postoperative complications. The incidence of CSF leakage in the anterior approach has been estimated to range from 4.3% to 32%. In the present case, the rapidly progressive nature of the myelopathy motivated an anterior surgical approach; whereas slow onset, progressive symptoms in the context of OPLL would motivate long-segment posterior decompression and fusion.

Imaging findings

The surgical risk for CSF leak has been shown to be associated with ossification of the dural membrane as evidenced by the double layer sign on CT, as seen in Figure 2. The extent of OPLL is best visualized with CT imaging; whereas MRI is most useful in characterizing effects on the spinal cord, as displayed in Figures 1-3. Risk factors for the development of myelopathy include age and extent of canal stenosis. Spinal canal crosssectional area measurements are shown in Figure 3. Such measurements have been used as indicators of prognosis in surgical decompression for compressive myelopathy, with transverse crosssectional areas ranging from 0.3 to 0.6 cm² being defined as critical stenosis responsive to surgical decompression, although in a chronic setting. With “boomerang” shaped compression and a minimum spinal cord crosssectional area of 0.43 cm², as seen in the present case, one would expect clinical improvement following surgical decompression based on these previous studies.

CONCLUSION

This case is an example of two comorbid conditions, cervical disc herniation and OPLL, leading to a unique clinical presentation of rapidly progressive myelopathy in the absence of trauma. Surgical decompression was...
Table 1: Literature review of acute rapidly progressive myelopathy in the context of cervical disc herniation with or without comorbid OPLL

| Author/year | Age/sex | Clinical | ww | Cervical disc | OPLL | Trauma? | Level | Intervention | Recovery |
|-------------|---------|----------|----|---------------|------|---------|-------|--------------|----------|
| Present case| 52 F    | Neck pain (4 wk), 72 h quadruparesis | 72 h | Yes | Yes | No | C4/5 | ASF | Complete 14 d |
| Cheong 2012[4] | 43 M    | Neck massage. Quadraparesis | 4 h | Yes | Yes | No | C5/6 | ASF + L | Partial at 3 yr |
| Hsieh 2010[5] | 61 F    | Spinal manipulation therapy for chronic neck pain. Acute Brown-Sequard | 2 wk slow, NS acute | Yes (ID) | Yes | Neck massage | Spinal manipulation | C3/4 | ASF | Return of function |
| Liu 2010[6] | 75 M    | Acute paraplegia when bending forward. Two mo paresthesias prior | 4 h | Yes | No | No | C4/5 | ASF | Partial lower limb strength. Wheelchair bound (15 mo) |
| Tsai 2006[20] | 32 F    | Paraplegia in second stage of labor | 4 h | Yes | No | No | C3/4 | ASF | 4/5 Left, 5/5 Right |
| Chen 2005[21] | 54 M    | Violent neck movement after anesthesia, paraparesis 1 h postoperative | 1 h | Yes | No | No | C6/7 | ASF | Paralysis 3 mo |
| Goh 2004[7] | 57 M    | Progression from weakness to paraparesis | 2 d | Yes | No | No | C4/5 and C5/6 | ASF | NR |
| Suzuki 2003[19] | 29 M    | Slowly progressive myelopathy (5 mo), overnight paraparesis with head turn | <8 h | Yes | No | No | C6/7 | ASF | NR at 14 d or 3 yr |
| Iwamura 2001[18] | 45 M    | Neck pain (10 mo) with 2 wk onset Brown-Sequard | 2 wk | Yes | Yes | No | C6/7 | ASF + OPLL resection | Good. Slight hypesthesia at 7 yr |
| Ueyama 1999[22] | 61 F    | Minor neck pain (2 mo), 1 d onset of acute paraplegia | 1 d acute | Yes | No | No | C5/6 and C6/7 | ASF | Progressive resolution, walking (6 mo) |
| Lee 1989[12] | 53 M    | Sudden arm pain/weakness while lifting an object. Progression to quadraparesis (1 mo) | 1 mo | Yes (ID) | No | No | C5/6 | L | Motor recovery (6 mo) |
| Scheider 1988[16] | 50 F    | Onset of right sided Brown-Sequard while driving | 3 d | Yes (ID) | NS | No | C5/6 | ACD | Minor weakness at 6 mo |
| Eisenberg 1986[21] | 25 M    | Acute onset Brown-Sequard syndrome | 4 d | Yes (ID) | NS | No | C5/6 | L | Incomplete resolution |
| Roda 1982[15] | 43 M    | Brown-Sequard acute onset after acute chest pain | 1 d | Yes | No | No | C6/7 | L | Limited residual symptoms at 31 mo |
| Lourie 1973[14] | 37 M    | 3 d progression of quadraparesis. Neck pain 2 mo prior | 3 d | Yes | No | No | C5/6 | ASF | Recovery at 24 mo |

h: Hour, d: Day, mo: Month, yr: Year, wk: Week, ASF: Anterior spinal fusion, L: Posterior laminectomy, LP: Laminoplasty, ACDF: Anterior cervical discectomy, NS: Not specified, NR: No recovery, ID: Intradural, OPLL: Ossified posterior longitudinal ligament, ASF: Anterior spinal fusion

Successful and lead to complete resolution of symptoms in our patient. The value of complementary types of spinal imaging (CT and MRI) is clear in the context of an acute and rapidly progressive neurological deterioration in the absence of any definable external trauma. Typical imaging findings with OPLL, especially the double layer sign with dural ossification are important in the workup of this condition to determine the cause of neurological deterioration and optimal surgical approach. This case also demonstrates that despite the increased risk of perioperative complications associated with anterior decompression in patients with OPLL, in the setting of comorbid acute cervical disc herniation, this surgical technique is technically feasible and can lead to favorable clinical outcomes.

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