

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 quadriparesis 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

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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.^[3,17] 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 quadriparetic 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.

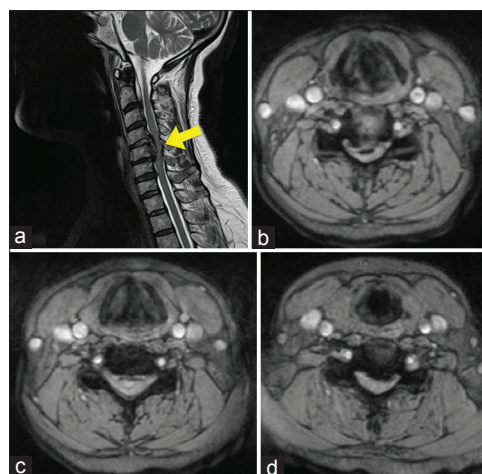


Figure 1: Preoperative T2-weighted sagittal (a) and proton density weighted axial (b-d) MRI images without contrast. The axial images demonstrate the level of spinal canal compromise at the level of the C4-5 disc (b), mid-C5 vertebral body (c) and C5-6 disc (d). An arrow identifies the cervical disc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

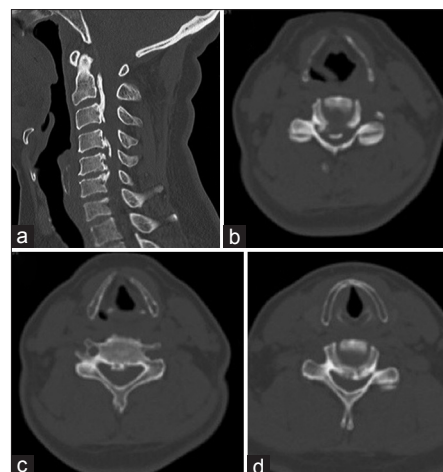


Figure 2: Preoperative sagittal (a) and axial (b-d) CT images. Images b-d correspond to axial MRI images presented in Figure 1. The extent of bony canal compromise secondary to OPLL posterior to the C4-5 disc (b), mid-C5 vertebral body (c) and C5-6 disc (d) is illustrated. Note the double layer sign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

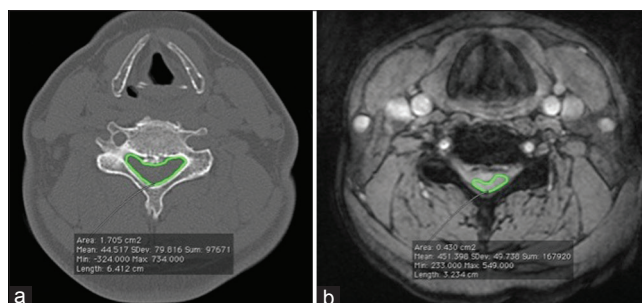


Figure 3: Axial CT (a) and MRI (b) images at the level of maximal canal compromise posterior to the C5 vertebral body. Measurements of the cross-sectional area available for the spinal cord secondary to bony (a) and soft-tissue (b) canal compromise are shown



Figure 4: Postoperative upright anteroposterior (a) and lateral (b) radiographs following anterior cervical decompression with a C5 corpectomy and cage reconstruction with anterior cervical plate stabilization

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.^[2,4,5,7,9,10,12-14,16,18-21] Three reported cases documented imaging findings of OPLL,^[4,9,10] 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.^[19] 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 quadraparesis 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.^[8]

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.^[17] The authors of the three case reports of acute myelopathy with cervical disc and OPLL, as well as Iwamura and Hsiegh, proposed a potential clinical correlation between the two pathologies.^[9,10] 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.^[15] While observation of asymptomatic OPLL remains the standard of care, symptomatic canal stenosis caused by OPLL can require surgery.^[22] 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%.^[15] 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.^[3] 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.^[1] Spinal canal cross-sectional area measurements are shown in Figure 3. Such measurements have been used as indicators of prognosis in surgical decompression for compressive myelopathy, with transverse cross-sectional areas ranging from 0.3 to 0.6 cm² being defined as critical stenosis responsive to surgical decompression, although in a chronic setting.^[6,11] With “boomerang” shaped compression and a minimum spinal cord cross-sectional 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 quadraparesis	72 h	Yes	Yes	No	C4/5	ASF	Complete 14 d
Cheong 2012 ^[4]	43 M	Neck massage. Quadraparesis	4 h	Yes	Yes	Neck massage	C5/6	ASF+L	Partial at 3 yr
Hsieh 2010 ^[9]	61 F	Spinal manipulation therapy for chronic neck pain. Acute Brown-Sequard	2 wk slow, NS acute	Yes (ID)	Yes	Spinal manipulation	C3/4	ASF	Return of function
Liu 2010 ^[13]	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 ^[2]	54 M	Violent neck movement after anesthesia, paraparesis 1 h postoperative	1 h	Yes	No	Extubation	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 ^[10]	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 ^[21]	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 ^[18]	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 ^[5]	25 M	Acute onset Brown-Sequard syndrome	4 d	Yes (ID)	NS	No	C5/6	L	Incomplete resolution
Roda 1982 ^[16]	43 M	Brown-Sequard acute onset after acute chest pain	1 d	Yes	NS	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, ACD: 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.

REFERENCES

1. Chang H, Song KJ, Kim HY, Choi BW. Factors related to the development of myelopathy in patients with cervical ossification of the posterior longitudinal ligament. *J Bone Joint Surg Br* 2012;94:946-9.
2. Chen SH, Hui YL, Yu CM, Niu CC, Lui PW. Paraplegia by acute cervical disc protrusion after lumbar spine surgery. *Chang Gung Med J* 2005;28:254-7.
3. Chen Y, Guo Y, Chen D, Lu X, Wang X, Tian H, et al. Diagnosis and surgery of ossification of posterior longitudinal ligament associated with dural ossification in the cervical spine. *Eur Spine J* 2009;18:1541-7.
4. Cheong HS, Hong BY, Ko YA, Lim SH, Kim JS. Spinal cord injury incurred by neck massage. *Ann Rehabil Med* 2012;36:708-12.
5. Eisenberg RA, Bremer AM, Northup HM. Intradural herniated cervical disk: A case report and review of the literature. *AJNR Am J Neuroradiol* 1986;7:492-4.
6. Fujiwara K, Yonenobu K, Ebara S, Yamashita K, Ono K. The prognosis of surgery for cervical compression myelopathy. An analysis of the factors involved. *J Bone Joint Surg Br* 1989;71:393-8.

7. Goh HK, Li YH. Non-traumatic acute paraplegia caused by cervical disc herniation in a patient with sleep apnoea. *Singapore Med J* 2004;45:235-8.
8. Hardin JG, Halla JT. Cervical spine and radicular pain syndromes. *Curr Opin Rheumatol* 1995;7:136-40.
9. Hsieh JH, Wu CT, Lee ST. Cervical intradural disc herniation after spinal manipulation therapy in a patient with ossification of posterior longitudinal ligament: A case report and review of the literature. *Spine (Phila Pa 1976)* 2010;35:E149-51.
10. Iwamura Y, Onari K, Kondo S, Inasaka R, Horii H. Cervical intradural disc herniation. *Spine (Phila Pa 1976)* 2001;26:698-702.
11. Kadanka Z, Kerkovsky M, Bednarik J, Jarkovsky J. Cross-sectional transverse area and hyperintensities on magnetic resonance imaging in relation to the clinical picture in cervical spondylotic myelopathy. *Spine (Phila Pa 1976)* 2007;32:2573-7.
12. Lee ST, Cheng SY, Lui TN. Intradural rupture of cervical intervertebral disc. *Spine (Phila Pa 1976)* 1989;14:1412-4.
13. Liu C, Huang Y, Cai HX, Fan SW. Nontraumatic acute paraplegia associated with cervical disk herniation. *J Spinal Cord Med* 2010;33:420-4.
14. Lourie H, Shende MC, Stewart DH Jr. The syndrome of central cervical soft disk herniation. *JAMA* 1973;226:302-5.
15. Mazur M, Jost GF, Schmidt MH, Bisson EF. Management of cerebrospinal fluid leaks after anterior decompression for ossification of the posterior longitudinal ligament: A review of the literature. *Neurosurg Focus* 2011;30:E13.
16. Roda JM, Gonzalez C, Blazquez MG, Alvarez MP, Arguello C. Intradural herniated cervical disc. Case report. *J Neurosurg* 1982;57:278-80.
17. Saetia K, Cho D, Lee S, Kim DH, Kim SD. Ossification of the posterior longitudinal ligament: A review. *Neurosurg Focus* 2011;30:E1.
18. Schneider SJ, Grossman RG, Bryan RN. Magnetic resonance imaging of transdural herniation of a cervical disk. *Surg Neurol* 1988;30:216-9.
19. Suzuki T, Abe E, Murai H, Kobayashi T. Nontraumatic acute complete paraplegia resulting from cervical disc herniation: A case report. *Spine (Phila Pa 1976)* 2003;28:E125-8.
20. Tsai HH, Li TY, Chang ST. Nontraumatic acute myelopathy associated with cervical disc herniation during labor. *J Back Musculoskeletal Rehab* 2006;19:97-100.
21. Ueyama T, Tamaki N, Kondoh T, Miyamoto H, Akiyama H, Nagashima T. Non-traumatic acute paraplegia associated with cervical disc herniation: A case report. *Surg Neurol* 1999;52:204-6.
22. Wu JC, Chen YC, Liu L, Huang WC, Chen TJ, Lo SS, et al. Conservatively treated ossification of the posterior longitudinal ligament increases the risk of spinal cord injury: A nationwide cohort study. *J Neurotrauma* 2012;29:462-8.