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CASE REPORT

Spontaneous cervical intradural disc herniation presenting with Brown-Séquard and Horner's syndrome: lesson learned from a very unique case

Irene Baudracco[1] · Gordan Grahovac[1] · Vittorio M. Russo[1]

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Abstract
Purpose Cervical spontaneous intradural disc herniation (IDH) is an extremely rare condition. We describe a unique case of a patient presenting with a Brown-Séquard syndrome (BSS) and Horner's syndrome (HS). This study aimed to report an unusual case of spontaneous cervical intradural disc herniation that presented with Horner's and Brown-Séquard syndrome (BSS) and discuss difficulties in preoperative diagnosis and treatment difficulties of intradural cervical disc.
Methods Notes and images review, and analysis of the relevant literature.
Results A 45-year old female presented with acute Horner's syndrome and Brown-Séquard syndrome. The magnetic resonance imaging of cervical spine revealed C4-5 disc extrusion with cord compression. The patient underwent urgent decompression through an anterior cervical corpectomy and fusion. Patient fully recovered 6 months after disease onset.
Conclusion We would like to emphasize that prompt and anterior cervical decompression is the treatment of choice, as it directly address the problem and allows dura repair in spontaneous cervical disc herniation.

Keywords Spontaneous intradural cervical disc herniation · OPLL · Cervical corpectomy · Brown-Séquard syndrome · Horner's syndrome

Introduction

Intradural herniation of intervertebral disc is unusual manifestation and most commonly has been reported at lumbar spine [1]. Cervical intradural disc herniation (IDH) is an extremely rare condition and trauma is the most common aetiology [2].

We describe a case of spontaneous cervical IDH associated with ossification of posterior longitudinal ligament (OPLL) and presenting with Horner's (HS) and Brown-Séquard syndrome (BSS) (Table 1).

Case report

History and neurological examination

A 45 years-old woman presented with an acute onset of severe neck pain radiating to the right upper limb and right-sided hemiparesis, right eye ptosis, miosis and anhidrosis. Neurological examination was consistent with a BSS and HS.

Imaging

The cervical magnetic resonance imaging (MRI) showed central disc osteophyte bar with focal acute right paracentral disc extrusion that has migrated cranially, compressing the right hemicord at the level of C4-5 level.

The Manuscript submitted does not contain information about medical device(s)/drug(s).

✉ Irene Baudracco
Irene.baudracco@gmail.com

[1] Victor Horsley Department of Neurosurgery, The National Hospital for Neurology and Neurosurgery, University College London Hospitals NHS Trust, Queen Square, London WC1N 3BG, UK

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Table 1 Reported cases of spontaneous intradural cervical disc herniation

References	Age	Level	Association with OPLL	Presentation	Surgical approach	Outcome
Marega et al. [4]	41, M	C5-6	Not known	Tetraparesis and Horner's Syndrome	Posterior	Residual symptoms
Schneider et al. [14]	50, F	C5-6	Not known	BSS and bladder dysfunction	ACD	Residual symptoms
Roda et al. [11]	43, M	C6-7	Not known	BSS	Posterior hemilaminectomy	Residual symptoms
Epstein et al. [1]	38, F	C6-7	Not known	C7 radiculopathy	Posterior	Full recovery
Clatterbuck et al. (15)	40, M	C4-5	No	Neck pain, BSS	Anterior	Residual symptoms
		C3-4	No	BSS	Anterior	Full recovery
	32, M	C5-6	No	Neck pain, BSS	Anterior	Full recovery
Iwamura et al. [6]	45, M	C6-7	No	Neck pain, BSS	Anterior *(en bloc)*	Persistent slight hypesthesia
Pan et al. [13]	50, W	C4-5	No, adhesions	Neck pain. Power 4/5 left limbs, gait instability, numbness of upper extremities	ACDF	Full recovery
Pan et al. [13]	58, W	C4-5	No, adhesions	Neck pain. Loss of motor function, sensation and deep tendon relaxes in all four limbs. Hypoesthesia below the T4 dermatome	ACDF	Residual symptoms
Warade and Misra (16)	64, M	C6-7	N/A	Sudden neck pain, BSS	ACDF	Residual symptoms
Wang et al. [2]	52, M	C5-6	Yes	Neck pain, BSS	Two-stage: laminoplasty + anterior corpectomy	Persistent paresthesias
Present case	45, W	C4-5	Yes	Neck pain, BSS and Horner's syndrome	ACDF	Residual weakness

Surgical procedure

The patient underwent urgent spinal cord decompression through an anterior C4 corpectomy. A tear was noted in posterior longitudinal ligament (PLL) on the right side posterior to the OPLL mass. Dural defect was clearly visualized with herniated disc fragment within it. The dura was closed. The cervical spine was reconstructed with Bengal interbody fusion cage filled with autogenous bone (DePuy Synthes, MA, USA) and Skyline cervical plate (DePuy Synthes, MA, USA).

Outcome

The postoperative course was marked with immediate pain relief and gradual recovery of right side hemiparesis. HS was still present after surgery. Clinical examination 6 months after surgery revealed complete resolution of BSS and HS.

Discussion

Cervical IDH is extremely rare clinical entity and due to rarity it remains difficult to definitively diagnose the disease before surgery. The first report of lumbar intradural disc herniation was done in 1942 by Dandy [3] and cervical IDH was done in 1959 by Marega [4]. To the best of our knowledge there have been around thirty cases published so far and only one case was associated with OPLL [2].

The true cause of the IDH is unknown but most commonly is precipitated by trauma on the top of previous degenerative disc disease. Chronic inflammation in the case of OPLL causes scaring and adhesion of the adjacent dura mater to the PLL [5, 6]. The adherent and fragile dura can be easily perforated with extruded disc. In our case, disc extrusion was not associated with previous trauma and intraoperatively we found that the calcified PLL was firmly attached to friable dura.

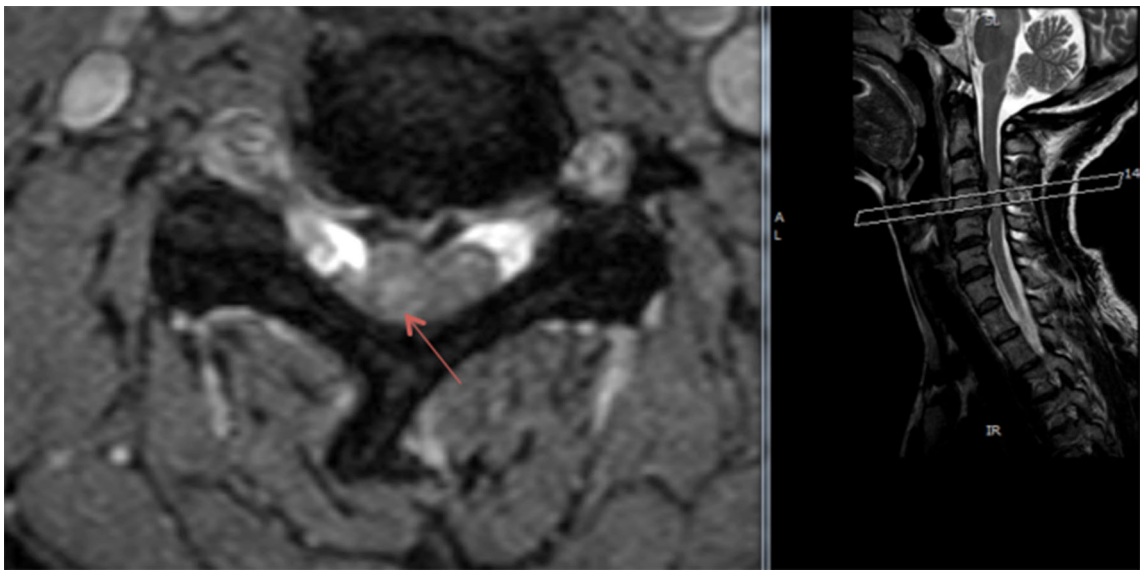

Fig. 1 MRI T2—sagittal and axial views. The *arrow* shows the intradural fragment of the disc prolapse severely compressing the spinal cord

The classical presentation of cervical disc herniation is radiculopathy or myelopathy. Other neurological syndromes such as BSS and HS have been described as first presenting symptoms in patients with IDH [3–11]. Patients with sudden onset hemiplegia are initially assessed to rule out stroke, intracranial space-occupying lesions or other neurological conditions. In our case, the diagnosis of severe cervical spinal cord compression was made incidentally during radiological assessment for a brain stroke. Being HS consistent with posterior circulation stroke [12], the cervical spine was also imaged.

BSS is rarely caused by herniated disc but in case of severe unilateral cord compression due to herniated disc it can be the first clinical presentation of cervical disc herniation [3, 11]. According to Kobayashi et al. the degree of compression, together with the individual distribution of the anterior spinal artery, may account for the level of severity of the sensory deficit [8]. The prognosis of neurological recovery following BSS is slightly worse in case of cervical-IDH compared to extradural cases [13], but good recovery is still achievable in the majority of the cases.

The presence of HS in cases of spontaneous cervical-IDH is exceptionally rare, and it has been reported in only two traumatic cases [1, 14]. The oculosympathetic pathway connecting the hypothalamus and the orbit passes through the ciliospinal center at the level of C8-T2. The direct pressure of the herniated disc on the spinal cord produces compression of the first order neuron of the sympathetic pathway at C4-5 level, interrupting it between the hypothalamus and the orbit [14], as occurred in our case.

Different surgical approaches have been used to address cervical-IDH and variability in outcomes has been reported [1, 7]. In our case, we performed corpectomy that allowed direct visualisation of the intradural disc prolapse with complete decompression of the spinal cord. Primary dural closure was also easily done through the anterior approach. Our patient fully recovered 6 months after procedure (Fig. 1).

Conclusions

Spontaneous cervical-IDH should be considered as part of the differential diagnosis in patients presenting with acute onset of BSS and HS. Prompt surgical intervention allows a good clinical outcome and complete recovery. Anterior approach is the treatment of choice, as it directly addresses the problem.

Compliance with ethical standards

Conflict of interest None of the authors has any potential conflict of interest.

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