Upper thoracic spinal cord herniation after traumatic nerve root avulsion

Case report and review of the literature

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✓ Transdural herniation of the spinal cord, a rare but well-documented entity, has been reported sporadically for more than 25 years as a possible cause for various neurological signs and symptoms ranging from isolated sensory or motor findings to myelopathy and Brown-Séquard syndrome. The authors report, to the best of their knowledge, the first case of upper thoracic spinal cord herniation occurring after traumatic nerve root avulsion.

KEY WORDS • Brown-Séquard syndrome • nerve root avulsion • pseudomeningocele • spinal cord herniation • spinal cord tethering

SPINAL cord herniation is an uncommon entity that can occur spontaneously or after surgery or trauma. Idiopathic (spontaneous) spinal cord herniation is usually seen anteriorly and commonly in the thoracic spine. It most commonly affects middle-age women and manifests with Brown-Séquard syndrome and myelopathy without radiological evidence of compressive lesions.

Postsurgical or traumatic spinal cord herniation, on the other hand, can be found anywhere in the spinal column and can present clinically, in an acute or delayed fashion, with a much broader spectrum of signs and symptoms.^{13,16} We describe the case of a 41-year-old man with thoracic spinal cord herniation who presented 19 years after sustaining a traumatic brachial plexus nerve root avulsion. The pathogenesis, clinical and imaging spectra, treatment alternatives, and outcome are discussed.

Case Report

History. This 41-year-old man was involved in a motorcycle accident in 1982, 19 years prior to his presentation. At that time, he sustained a significant head injury, which rendered him comatose and required intensive care unit admission. During that time, he also suffered marked right brachial plexus and right lower leg injuries. The brachial plexus injury resulted in a flail arm; elements of the brachial plexus from C-5 to T-1 were involved, with the

lower nerve roots known to be avulsed. The patient's right arm was amputated 18 months later after no improvement was exhibited. Subsequently, he developed phantom pain syndrome, which was managed nonsurgically. His lower right leg required multiple reconstructive procedures and 5 years later, with a shorter leg, he was finally able to walk independently.

Two years prior to his presentation, he started to complain of new right leg weakness and a tendency to fall, which was particularly noticeable while climbing stairs. No neck, back, or leg pain was noted and he denied any sensory, bowel, or bladder dysfunction.

Examination. On examination, a right Horner syndrome was present. Amputation of his right arm at mid-level was apparent. There was marked wasting and weakness of the intrinsic muscles of his left hand. The bulk and strength of the remaining muscles of that extremity as well as the tendon reflexes were normal. He exhibited wasting of the right thigh and calf muscles, with shortening of the leg. There was mild weakness (Grade 4/5) of the right hip and knee flexors and right tibialis anterior, and Grade 3/5 of the right extensor hallucis longus. An upgoing toe and hyperreflexia of the knee and ankle jerks were also present ipsilaterally. Normal strength, tone, and bulk of left lower-extremity muscles were noted. The sensory examination to pinprick, joint position, and vibration was normal in both lower extremities and left arm.

The patient underwent MR imaging and CT myelography (Figs. 1–3). The MR imaging studies revealed a lateral pseudomeningocele arising from the right T1–2 inter-

Abbreviations used in this paper: CSF = cerebrospinal fluid; CT = computerized tomography; MR = magnetic resonance.

FIG. 1. A: Axial postmyelography CT scan demonstrating extravasation of contrast material into pseudomeningocele and prolapse of the spinal cord (arrow). B: Axial fast-spin echo T₂-weighted MR image revealing tethering of the spinal cord and pseudomeningocele (arrow).

vertebral foramen, resting on the chest apex, with spinal cord atrophy, herniation, and tethering. Intramedullary hyperintensity was also present on fast-spin echo T₂-weighted sequences. Postmyelography CT scanning demonstrated extravasation of contrast into the cystic cavity, which consisted of intra- and extraspinal portions.

Operation. Because of the patient's progressive symptoms surgery was deemed necessary. This was performed via a posterior approach. We conducted T-1 and T-2 laminectomies. Intraoperatively attenuation and outpouching of the spinal cord were seen through a 6-mm-diameter dural defect (Fig. 4). Clear communication of CSF was identified between the spinal subarachnoid space and intra- and extraspinal components of the pseudomeningocele. Arachnoidal bands tethered the spinal cord to the dural ring. Meticulous dissection and lysis of such bands allowed reduction of the herniated cord (Fig. 4). Primary closure was supplemented using a dural patch (Duragen; Integra Neurosciences, Plainsboro, NJ) to seal the track between the subarachnoid space and the pseudomeningocele (Fig. 4), as well as to prevent spinal cord reherniation.

Postoperative Course. Six weeks postoperatively, he exhibited a subjective improvement of the right leg weakness and experienced less difficulty when climbing stairs. Continuing improvement of his myelopathy was evident at the 6-month follow-up examination.

Discussion

Spinal cord herniation is an entity that rarely occurs,

with little more than 50 cases reported and approximately 30 surgically proven cases.^{16,21,28} Spinal cord herniation can be classified as spontaneous, posttraumatic, and iatrogenic.^{1,16,21,28} Although spontaneous herniation is now being recognized more frequently on MR imaging, posttraumatic herniation remains a rare condition with only a small number of reports in the literature.^{6-9,14,15,19,22,25} In some of these posttraumatic cases, the presumed cause has been based on a history of remote and sometimes trivial trauma, which, in our opinion, raises the possibility that these cases are not related to injury but are more likely idiopathic (spontaneous) in occurrence. The usual clinical presentation of spontaneous spinal cord herniation is that of myelopathy and Brown-Séquard syndrome affecting middle-age patients. There is a female/male ratio of approximately 2:1.^{4,12,13,16-18,25,28} Cases of isolated motor and sensorimotor deficits, impotence, and bowel and bladder dysfunction affecting younger patients, on the other hand, have been observed after traumatic injury or surgery.^{25,28} Spontaneous spinal cord herniation is often thoracic in location and the spinal cord is displaced ventrally.^{12,17,20,23} Posttraumatic and iatrogenic herniation occurs throughout the spine, with cases of cervical posterolateral herniation developing after C1-2 wiring failure and direct posterior conus medullaris prolapse after L-1 burst fracture.²⁸

In cases of traumatic spinal cord herniation, patients usually present after a postinjury interval of several years, rarely within few days of the initial injury. For example, in 1997 Lee, et al.,¹⁴ reported on a case of delayed paraplegia associated with spinal cord herniation 2 days after a stab

FIG. 2. Coronal postmyelography CT scan (A) and fast-spin echo T₂-weighted MR image (B) revealing an intra- and extraspinal pseudomeningocele and dural hiatus with spinal cord herniation (arrow).

FIG. 3. Sagittal postmyelography CT scan (A) and fast-spin echo T₂-weighted MR image (B) demonstrating an intraspinal cyst with the herniated spinal cord (arrow).

injury to the spine, whereas in 1996 Urbach, et al.,²⁵ described a case of thoracic spinal cord herniation 38 years after the patient suffered blunt trauma to the spine (Table 1).

In 1995 Kumar, et al.,¹³ proposed that a dural defect located at the concavity of the spinal curvature, development of arachnoidal adhesions, and CSF pulsation-induced anterior-posterior movement of the spinal cord are pivotal factors leading to the herniation of the cord out of its dural sleeve into a preexisting cyst.¹³ Some authors have theorized that a herniated, calcified disc as well as intradural disc herniation can erode the dura mater; such theories that have been proposed to explain the dural hiatus in spontaneous spinal cord herniation, which seems to be of degenerative rather than congenital origin because of the characteristic time of presentation.^{10,16,17,29}

Radiologically, spinal cord kinking and neural tissue prolapse are the hallmarks of this disorder.^{10,18,24,28} Spinal cord atrophy, intramedullary hyperintensity, syrinx formation, tethering, vertebral body nuclear trail sign, and scalloping of vertebral body have been also observed.^{2,16,28} Postmyelography CT scanning and phase-contrast MR imaging are helpful in demonstrating the normal CSF flow pattern dorsally and excluding the possibility of a compressive posterior arachnoidal cyst.⁵ Intraoperatively, herniation of the spinal cord and nerve roots through a dural defect or duplicated dura have been reported.^{1,12,17,18,20}

In the present case, one can speculate that traumatic avulsion of the T-1 nerve root caused a dural “hiatus” at the origin of the nerve root sleeve, through which CSF extravasated and created the observed complex pseudomeningocele; here CSF pulsations ultimately culminated in the herniated spinal cord. Arachnoidal adhesions likely developed over time, leading to tethering of the cord to the dural ring and thereby preventing the cord from returning into the spinal canal.

The rarity of this condition makes it hard to define a treatment algorithm. It is, however, prudent to follow expectant therapy in asymptomatic cases. This also seems reasonable for mildly symptomatic and nonprogressive cases.¹⁶ In more severe and progressive cases, surgical repair is the intervention of choice. This can be achieved via primary closure with or without placement of a dural patch, using muscle, fascia graft, synthetic membrane, or lyophilized dura^{4,17,20,29} or, less commonly, by enlarging the dural opening and untethering the spinal cord alone.²⁷

Despite the retrospective nature of the vast majority of reported cases, outcome appears favorable in most surgically treated patients, with persistent sensory symptoms observed in some cases.^{1,12,17,18,20,26,28} A worse outcome is associated with an inability to reduce fully the herniated spinal cord because of the presence of extensive arachnoidal adhesions limiting untethering of the spinal cord.^{3,11} Increasing awareness of this disorder seems to be the key to establishing early diagnosis and preserving functional status in patients presenting with myelopathy or Brown-Séquard syndrome but in whom there is no evidence of extrinsic spinal cord compression.

Conclusions

Spinal cord herniation following traumatic nerve root avulsion is rare but should be considered in patients presenting with delayed Brown-Séquard syndrome or myelopathy. Appropriate MR imaging allows exclusion of compressive disease such as disc, tumor, or pseudomeningocele, and specific diagnosis. Surgical repair is recommended for symptomatic cases; this intervention is technically feasible and leads to improvement or at least stabilization of the neurological status.

FIG. 4. Intraoperative photographs illustrating the spinal cord herniating through the intradural defect. *Inset*: the dural defect is shown after microlysis of adhesions and reduction of the spinal cord hernia.

TABLE 1
Summary of reported cases of posttraumatic spinal cord herniation*

Authors & Year	Age (yrs), Sex	Spinal Level	Herniation Direction	Spinal Pain	Clinical Deficits	Yrs Since Trauma	Postop Course
Wortzman, et al., 1974	63, M	T-7	anterior	yes	myelopathy	31	worsened, then improved
Sachdev, et al., 1981	44, F	L-1	rt anterior	yes	rt leg sensorimotor	6	worsened, then improved
Borges, et al., 1995	68, M	T2–3	lt anterior	no	Brown–Séquard	48	improved
Urbach, et al., 1996	44, M	T5–6	rt anterior	no	myelopathy	38	improved
Watters, et al., 1998	46, M	T-6	lt anterior	yes	lt leg pyramidal	3	improved, then worsened
	35, M	T-3	posterior	NR	myelopathy	17	unknown
	46, M	L-1	rt posterior	yes	myelopathy	1	unchanged
present case	41, M	T-1	rt lat	no	myelopathy	19	early improvement

* NR = not reported.

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