

Spinal cord herniation following cervical meningioma excision: a rare clinical entity and review of literature

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Abstract

Background Spinal cord herniation following surgery is an extremely uncommon clinical condition with very few reports in published literature. This condition usually occurs as a spontaneous idiopathic phenomenon often in the thoracic spine or following a scenario of post traumatic spinal cord/nerve root injury. Rarely has it been reported following spinal cord tumor surgery.

Purpose To document a case of cervical spinal cord herniation as a late onset complication following spinal cord tumor surgery with an atypical presentation of monoparesis.

Design Case report.

Methods We describe the clinical presentation, operative procedure, post operative outcome and review of literature of this rare clinical condition.

Results A 57-year-old man presented with right upper limb monoparesis due to a spinal cord herniation 6 years after a cervical intradural meningioma excision. The patients underwent surgery to reduce the herniation and duroplasty with subsequent complete resolution of symptoms.

Conclusions Spinal cord herniation must be considered as differential diagnosis in scenarios of spinal cord tumor excision presenting with late onset neurological deficit. These cases may present as paraparesis, Brown-sequard syndrome and rarely as in our case as monoparesis.

Keywords Spinal cord herniation · Cervical meningioma · Pseudomeningocele · Monoparesis · Post-operative

Introduction

Spinal cord herniation is a rare clinical entity and was first reported in 1974 by Wortzman et al.; subsequent published literature appears in the form of isolated case reports. Spinal cord herniation can occur spontaneously, following spinal cord/nerve root injury and due to iatrogenic injury. We present a case of cervical spinal cord herniation five and half years following cervical meningioma excision with an atypical clinical presentation of monoparesis and to the best of our knowledge this appears to be the fourth published case of spinal cord herniation following spinal cord tumor excision surgery since it was first described over four decades ago.

Case report

A 50-year-old man was first seen in December 2008 with gradually progressive weakness in the right upper limb and associated gait disturbance over the previous 6 months. Clinical examination revealed brisk deep tendon reflexes, hand myelopathy signs, an extensor plantar response and normal sensory examination. Motor power in the right upper limb was grade four out of five. Subsequent MRI (Fig. 1) revealed an intradural, extramedullary tumor at the C2–3 level. He underwent C2–4 laminectomy and durotomy to expose the tumor tissue located in the right lateral spinal canal attached to the lateral dural surface. Tumor excision was performed and dural margins were cauterised.

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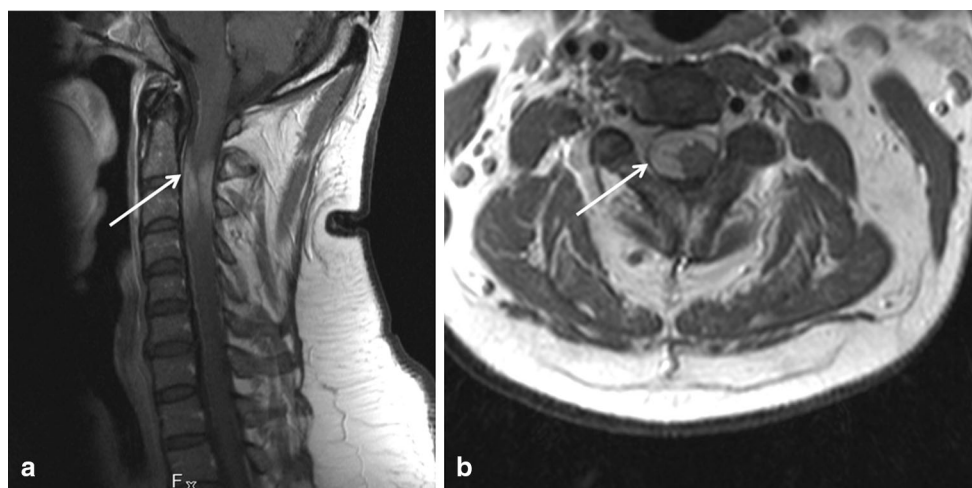


Fig. 1 Pre-operative MRI of the case at first presentation. **a** Post-contrast sagittal image show moderate uniform enhancement of the lesion (*arrow*) with presence of a 'dural tail' sign. **b** Axial section of

the post contrast image showing a large intradural extramedullary lesion (*arrow*) compressing the cord

Dura was closed in a watertight manner with no graft augmentation, followed by wound closure in layers.

Post-operatively the patient had uneventful recovery and was discharged on sixth post operative day. The post-operative MRI showed complete tumor clearance and histopathological analysis confirmed presence of cervical meningioma. Neurological deficit improved on subsequent follow-up and patient was otherwise well and asymptomatic.

He presented again in July 2014, five and half years following the first surgery with progressive right arm radiating pain and weakness in the right arm for 6 months. Motor power in the right arm in C8, T₁ myotomes was grade three out of five and four out of five in the rest of the myotomes. There was no objective weakness in the left arm and both lower limbs. Clinical examination showed loss of dexterity in the right hand with clumsiness of gait. There was an upper motor neuron lesion with brisk reflexes and an extensor plantar response. There was no sensory, bowel or bladder disturbance noted. MRI images (Fig. 2) showed a pseudomeningocele sac with herniated spinal cord at the C3 level causing cervical cord compression with no evidence of tumor reoccurrence.

Surgical procedure

Patient underwent a posterior cervical approach along previous scar. The pseudomeningocele sac was dissected and excised. The herniated cervical cord was isolated and dissected free from surrounding adhesions from the dural margins. The herniated portion was reduced after release of adhesions and durotomy defect was closed using synthetic dural graft (G-Patch). Wound was closed in layers and

patient had an uneventful post operative recovery. The neurological deficit improved on subsequent follow up and the symptoms abated.

Discussion

Spontaneous idiopathic spinal cord herniation has been described with clinical presentation varying from paraparesis to paraplegia and often with Brown-Sequard syndrome [1–4]. Monoparesis is atypical clinical presentation of a spinal cord herniation [3, 4]. Wortzman first described this entity 40 years ago in a case report of a spinal cord incarceration in the dorsal aspect of T₇ resulting in paraplegia in 1974 [1]. Based on review of literature spinal cord herniation may be classified as idiopathic, post-traumatic and iatrogenic [2, 3]. Idiopathic spinal cord herniation occurs spontaneously and is commonly seen in the thoracic spine and is speculated to arise from a ventral defect in the dural sheath; however, exact pathogenesis is still elusive [1–4]. Spinal cord herniation following spinal cord tumor excision remains a rare occurrence with three previous reports in literature.

Spinal cord herniation has also been reported following traumatic nerve root injury [5–8]. Spinal cord herniation into pseudomeningocele following brachial plexus avulsion injury with development of late onset fresh neurological deficit has been previously described by various authors [6–8]. Ijiri described a rare case of cord herniation following nerve root injury in the conus medullaris region [5]. There have been occasional reports of spinal cord herniation following other post-traumatic scenarios such as stab injury, vertebral body fracture, iatrogenic durotomy for spinal cord injury and implant related dural injury [3, 9–11].

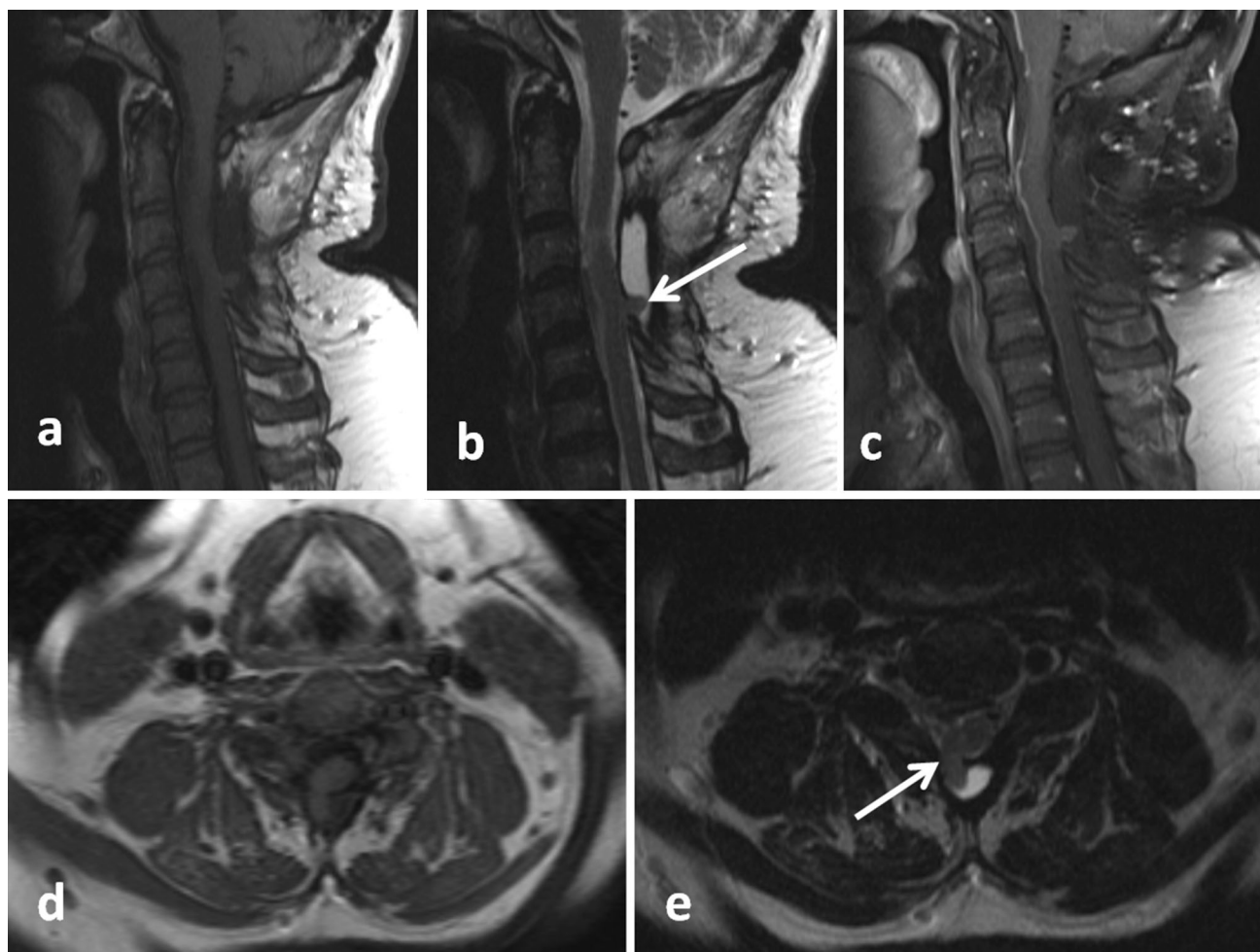


Fig. 2 Pre-operative MRI images at the second presentation. **a** T₁ weighted image of sagittal section of the cervical spine showing a spinal cord herniation at the C4 level along the dorsal aspect. **b** The T₂ weighted sagittal section image shows a pseudomeningocele sac with cord herniation along the dorsal aspect (*arrow*). **c** The post

contrast sagittal section shows no evidence of recurrence of the tumor. **d** Axial T₁ weighted images show a spinal cord herniation on the right posterolateral aspect through the dorsal dura into the pseudomeningocele sac. **e** T₂ weighted image of the axial section with herniated spinal cord (*arrow*)

Belan et al. described spinal cord herniation following foramen magnum decompression which was performed 7 years prior to presentation for an Arnold Chiari malformation [12]. Kawsar and colleagues described a case of thoracic spinal cord herniation through anterior dural defect following en bloc vertebrectomy for a Giant cell tumor presenting with late onset progressive paraparesis [13].

Pathogenesis of spinal cord herniation is unclear and many theories implicating a variety of factors including abnormal CSF flow dynamics, adhesions between the margins of the dural defect and the cord and alignment of the spine have been suggested previously [3, 4, 14]. Kumar suggested that the cause of the cord herniation was a dural defect either congenital in origin resulting in an arachnoid cyst or an iatrogenic dural defect resulting in a pseudomeningocele. Based on the sagittal alignment of the

spine and position of the spinal cord in the dural sac herniation develop in the concavity of the curvature. As a result, dorsal herniations are commonly seen in the cervical spine and ventral herniation are seen in the thoracic spine [2]. Other postulated mechanisms include adhesions developing between the dural defect and the spinal cord. These adhesions lock normal CSF flow into the arachnoid cyst or pseudomeningocele sac. The pulsatile CSF then progressively pushes the spinal cord into the dorsal or ventral defect resulting in spinal cord herniation [3, 14].

Pseudomeningocele formation following durotomy or duroplasty is a rare but well-known complication; however, spinal cord herniation into such a pseudomeningocele sac is a rare phenomenon. In this case report following the excision of the meningioma primary dural closure was performed using non absorbable sutures. Dura was cauterised over the area of the tumor. Whether this could result

in a late onset spinal cord herniation through the dural defect is a matter of speculation and cannot be conclusively proved. Spinal cord herniation following spinal cord tumor surgery was first reported by Hosno in a case of cervical cord herniation at the C2–3 level presenting with paraparesis, 14 years following surgery for excision of a cervical neurofibroma [14]. Similar cervical cord herniation following tumor excision surgery has been documented by Moriyama et al. 10 years following a cervical tumor excision in a case which also presented with paraparesis [15].

Our patient presented with monoparesis which appears to be a rare presenting feature of spinal cord herniation. Although there was no clinically demonstrable weakness in both lower limbs an objective assessment using motor evoked potential may have provided an insight into any subclinical neurological dysfunction in the lower limbs [16]. A similar clinical presentation of monoparesis was documented by Elwahas et al. in a case of cord herniation following cervical neurofibroma excision [17]. Additionally all cases of spinal cord herniation following tumor excision surgery have shown a long asymptomatic interval between the index surgery and development of symptomatic cervical cord herniation.

Conclusion

Spinal cord herniation, though a rare entity, must be considered in the clinical evaluation of late onset neurological deficit in cases with previous spinal cord tumor excision surgery. Though paraparesis and Brown sequard syndrome appear to be the common form of presenting neurological deficit, atypical pattern of neurological deficit such as monoparesis can also be seen.

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Compliance with ethical standards

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

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