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CASE REPORT

Iatrogenic dorsal spinal cord herniation and repair with clip-based expansile duraplasty: a case report

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INTRODUCTION: Myelopathy arising due to dorsal herniation of the spinal cord is a rare phenomenon, particularly so in the thoracic region. Where cases of thoracic dorsal cord herniation have been reported, the aetiology has typically been non-iatrogenic.

CASE PRESENTATION: We report the case of a paediatric oncology patient who presented with neurological deterioration secondary to thoracic dorsal spinal cord herniation, manifesting three months after laminectomy for biopsy of a spinal medulloblastoma lesion. We repaired the dural defect using non-penetrating titanium clips to create a secure expansile duraplasty, resulting in radiologically evident reduction of the cord herniation as well as corresponding clinical improvement.

DISCUSSION: Thoracic dorsal spinal cord herniation is an extremely rare occurrence after spinal surgery. Non-penetrating titanium clips can be used to form a secure expansile duraplasty following reduction of the cord herniation. Successful repair of the dural defect re-anteriorises the cord and can confer neurological benefit.

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INTRODUCTION

Dorsal spinal cord herniation (DSCH) is a rare phenomenon characteristically involving the cervical spinal cord and of iatrogenic aetiology, arising from post-operative dural defects [1–3]. In the thoracic cord, however, reported DSCH cases are typically non-iatrogenic, their occurrence attributed to pre-existing traumatic or structural abnormalities [4, 5]. Only two cases of iatrogenic thoracic DSCH have been reported [4, 6]. Surgical repair of the causative dural defect typically results in clinical improvement or stabilisation [1–6]. We present a case of iatrogenic thoracic DSCH repaired with a clip-based duraplasty technique.

CASE PRESENTATION

A 15-year-old female initially presented to her general practitioner reporting vague symptoms affecting attendance at school: earache, intermittent headache and rotatory vertigo, as well as mild but persistent generalised 'backache'. In the first instance, physiotherapy was offered, with negligible benefit. At three months from symptom onset, there was an acute deterioration in back pain, with prominent localisation of pain to the thoraco-lumbar junction. The patient also reported becoming more unsteady when walking, with occasional 'giving way' of her legs; numbness in the left hand, right leg, and right chest wall; and episodes of constipation. Bladder dysfunction was initially in the form of urinary hesitancy and urgency, though this progressed over ensuing weeks to a requirement for self-catheterisation due to significant post-void bladder residual volumes.

A neurosurgical opinion was sought at a centre different to ours and whole neuraxial magnetic resonance imaging (MRI) was urgently performed. Clinical examination at this stage demonstrated normal limb power. Neuroimaging revealed multiple enhancing intracranial and spinal lesions (*see* Fig. 1), consistent with the mixed clinical picture of multi-level radiculopathy, bladder dysfunction and secondary headache disorder. A multi-disciplinary team (MDT) recommended that a lesion at the T12 level was most amenable to biopsy. The patient accordingly underwent laminectomy at this level, without intra-operative complication. At this time, primary dural closure was performed using sutures and fibrin sealant (Tisseel, Baxter International, Deerfield IL).

The histopathology from the T12 biopsy yielded a neuropathological diagnosis of medulloblastoma, the impression being of cranial primary with multiple spinal 'drop metastases'. The MDT reconvened in light of the histopathological diagnosis, and recommended commencement of a chemoradiotherapy regimen comprising carboplatin, cyclophosphamide and vincristine chemotherapy combined with 41.4-Gray craniospinal megavoltage radiotherapy in 23 fractions. Chemoradiotherapy resulted in significant neurological benefit: gait improved, allowing independent mobilisation, and there was partial resolution of bladder symptoms. Chemoradiotherapy was, however, complicated by diffuse desquamation of skin overlying the cervico-thoraco-lumbo-sacral regions, haemorrhagic cystitis (attributed to cyclophosphamide toxicity, though also a known complication of radiotherapy) and myelosuppression (in

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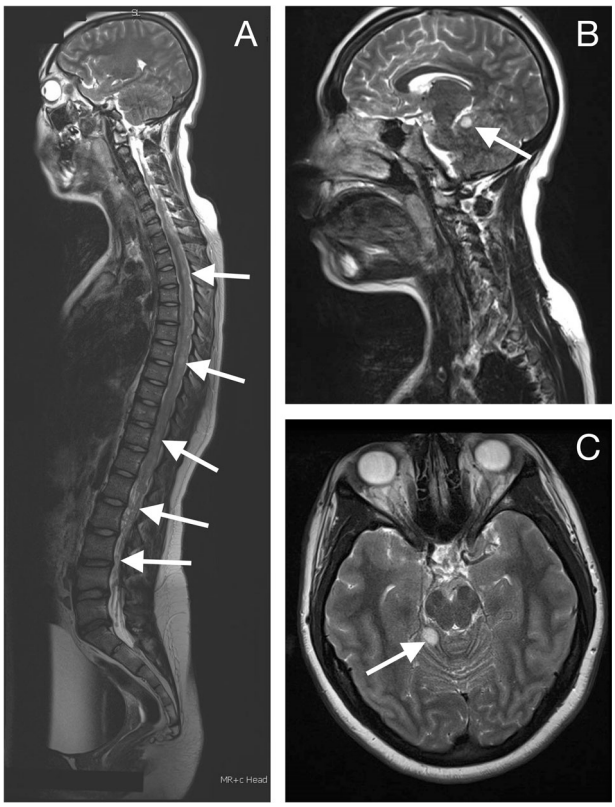

Fig. 1 Magnetic resonance imaging from time of initial clinical presentation. Multiple spinal and cauda equina lesions are demonstrated on a sagittal whole-neuraxis slice (**A**): note also the distended bladder. The cord ends at L1. Cranial lesions are seen on parasagittal (**B**) and axial (**C**) slices. Selected lesions are highlighted by the white arrows.

the form of both thrombocytopaenia and immunosuppressive oesophageal candidiasis).

Three months after initial surgery, the patient presented to our centre with a four-day history of thoracolumbar myelopathy: gait ataxia with loss of independent mobilisation, up-going plantar reflexes and lower limb hyperreflexia; and sensory disturbance below the waist. Examination confirmed globally reduced lower limb power and thoracolumbar dermatomal hypoaesthesia consistent with a T12/L1 sensory level.

Repeat neuroimaging was urgently arranged to determine the cause of the acute neurological deterioration. This demonstrated spinal cord herniation into a significant dorsal dural defect at the previous laminectomy level, with associated pseudomeningocoele and signal change in the T10–T12 spinal cord (Fig. 2A, B).

The patient then underwent surgical reduction of the herniation, performed by the senior authors (A.C. and P.S.). Exploration of the pre-existing laminectomy corridor revealed a section of herniated spinal cord (Fig. 3A). Intraoperatively, extensive arachnoid loculations were found anterior to the herniated cord: careful dissection of these allowed cord release and reduction of the herniated section (Fig. 3B,C). Primary dural repair was deemed unfeasible due to friable dura and the fact that this had already been attempted at the previous operation. The dural defect was instead repaired with an expansile duraplasty formed of a dural patch (Durepair, Medtronic, Fridley MN) secured by non-penetrating titanium clips (AnastoClip, LeMaitre Vascular, Burlington MA) in lieu of conventional stitches (Fig. 3D). The duraplasty was further augmented using a fibrin sealant (Evicel, Ethicon Inc, Bridgewater NJ; Fig. 3E). Neurophysiological monitoring was used throughout. Neuroimaging performed 6 weeks post-operatively (Fig. 2C, D) confirmed successful reduction of the dorsal herniation. Post-operatively, there was significant motor improvement allowing restoration of pre-biopsy mobility, with residual weakness being confined to plantarflexion of the left ankle. Sensation was completely restored. Intermittent self-catheterisation was required for ongoing bladder dysfunction.

Given the remaining disease burden from the primary medulloblastoma diagnosis, the MDT consensus was that the patient continues maintenance chemoradiotherapy post-operatively, in spite of concerns regarding wound healing problems and the onset of cisplatin-related high-frequency sensorineural hearing loss. However, the patient died seven months after the dural repair operation due to significant metastatic disease progression.

DISCUSSION

DSCH is a little-understood and highly uncommon cause of myelopathy previously reported in a mere 18 cases. Of these 18, eleven involve the cervical cord, six involve the thoracic cord and one involves the lumbar cord [1–6].

In the thoracic spinal cord, dural defects through which dorsal herniation occur are characteristically idiopathic or traumatic in aetiology, tending to result from disc herniations or vertebral body defects, but an iatrogenic aetiology—as in our case—has been reported in only two cases [4, 6]. The putative pathophysiology of DSCH implicates chronic inflammatory connective tissue reactions resulting in progressive formation of adhesions between the cord and dural defect, impairing cerebrospinal fluid (CSF) flow [7]. Disrupted local CSF dynamics are believed to give rise to a pulsatile positive pressure gradient, causing dorsal cord erosion through the dura. Inflammatory adhesions may also lead to granulation tissue formation and dural leakage, promoting the formation of intra- or extra-medullary cysts and pseudomeningocoeles that may contribute to a further negative pressure gradient for cord herniation [2, 3].

Abd Elwahab and colleagues have proposed that prevention of iatrogenic DSCH might be accomplished via use of a dural graft rather than primary closure [2], though notably iatrogenic DSCH cases arising spontaneously without initial dural opening have been reported [1, 6].

Since there is local compression of the spinal cord at the herniated segment, the clinical presentation of DSCH typically involves progressive myelopathy or paraparesis, the onset of which is often substantially delayed from original dural insult, by up to 18 years [4]. Surgical repair of the causative dural defects has been performed in all 18 reported symptomatic cases, with no documented re-herniations. Following exploration of affected levels and cord untethering, 16 cases utilised an artificial patch for dural repair: one other case used a muscular patch, whilst primary dural closure was performed in another [4, 6]. Dural closure was performed with sutures in all 16 cases.

In light of existing DSCH literature, our case presents several novel features in predisposition, clinical factors and operative management. The patient was an adolescent female, the first thoracic DSCH case of this demographic. Secondly, the aetiology for thoracic DSCH was iatrogenic, following laminectomy for spinal medulloblastoma biopsy. We propose that recurrent courses of high-dose chemoradiotherapy impaired normal dural healing [8], permitting dorsal cord herniation through a progressively enlarging dural defect. Additionally, factors intrinsic to the malignancy and chemoradiotherapy response—i.e. formation of arachnoid adhesions and loculations giving rise to CSF pressure abnormalities at the local level, as well as the potential for changes in CSF protein load—might have played a contributory role. This further stresses the importance of watertight dural closure in such cases.

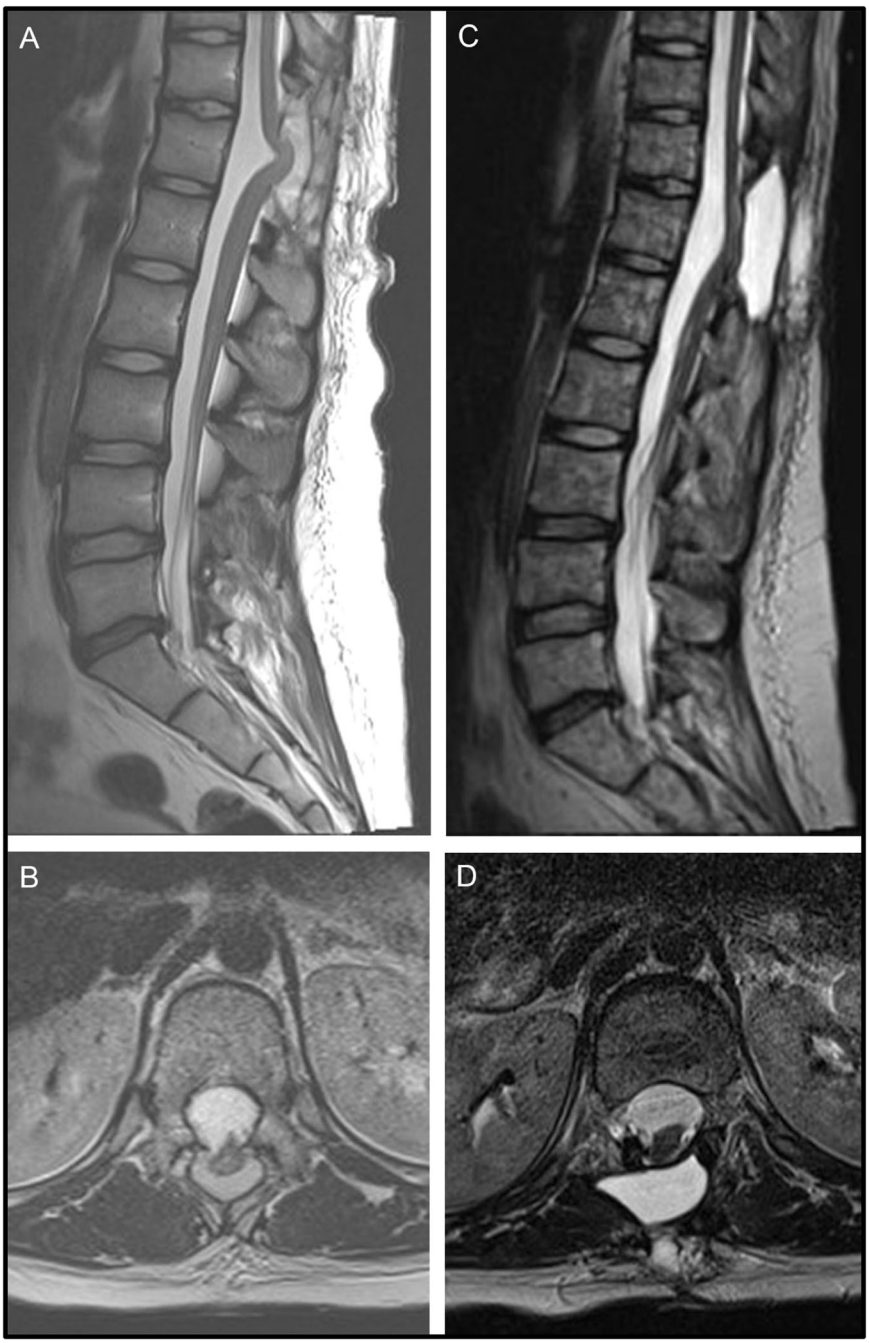

Fig. 2 Pre- and post-operative T2-weighted magnetic resonance imaging. Sagittal (**A**) and axial (**B**) pre-operative images show T12 dorsal dural defect and associated thoracic spinal cord herniation, with a subsequently re-anteriorised cord following dural repair shown in sagittal (**C**) and axial (**D**) views.

Thirdly, the use of non-penetrating titanium clips to affix the dural prosthesis was a departure from the convention of stitch-secured duraplasty for DSCH. Our rationale for selecting expansile duraplasty over primary dural repair was friable dura encountered intraoperatively, the edges of which proved difficult to appose. Non-penetrating clips, originally intended for vascular anastomoses, are designed to be compliant to pulsatile flow, offering maximal watertightness whilst retaining structural integrity. Other advantages over sutures include greater manoeuvrability in confined spaces, shorter repair duration and reduced risk of CSF leak [9]. In our experience, clips are routinely used for primary dural closure after resection of intradural tumours, as well as for repair of inadvertent durotomies. Titanium clips are MRI-compatible and have been shown to give rise only to minimal signal artefact, such that radiological visualisation and assessment of the spinal cord and surrounding structures on MRI is unimpaired [10].

In conclusion, thoracic DSCH is an extremely rare occurrence after spinal surgery. Local CSF flow abnormalities and poorly healed dural repairs are some proposed mechanisms for this uncommon pathology. Non-penetrating titanium clips can be used to form a secure expansile duraplasty following reduction of herniated cord. Successful repair of the dural defect re-anteriorises the cord and can confer neurological benefit.

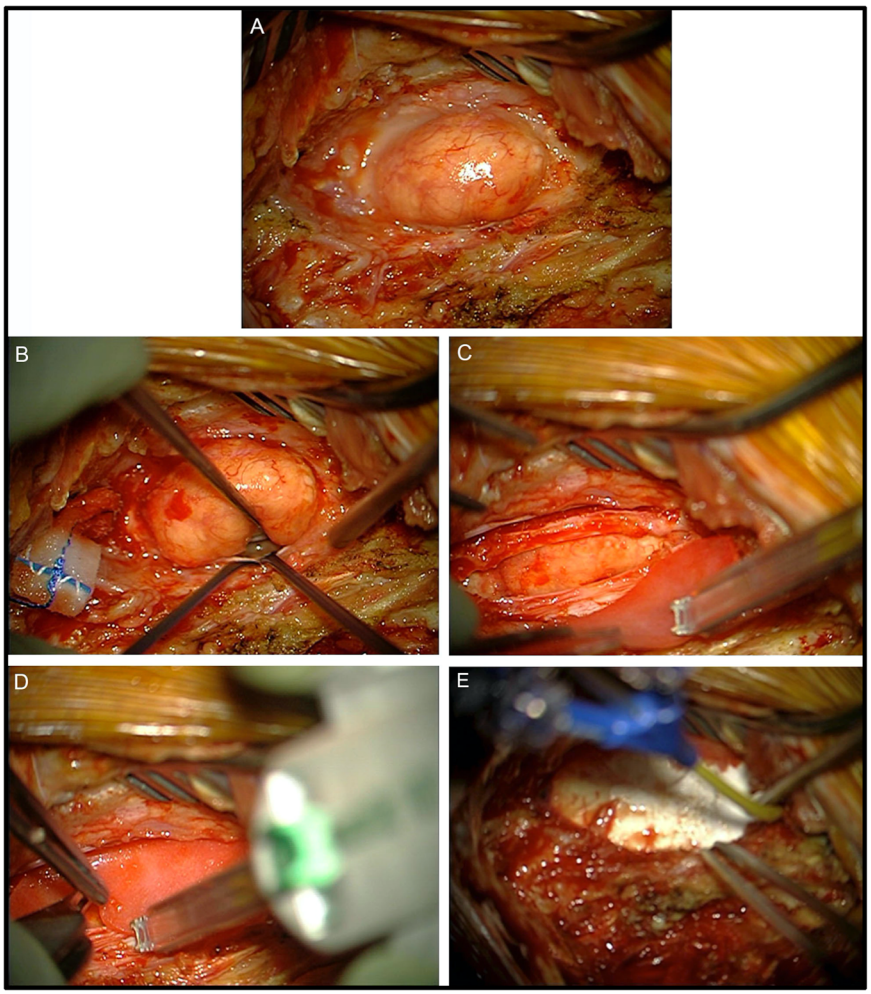

Fig. 3 Intraoperative microscopic images depicting reduction of thoracic cord herniation. The spinal cord was found to be visibly herniated through a large dural defect (**A**). Arachnoid granulations were released via careful dissection (**B**), permitting re-anteriorisation of the cord (**C**). A dural patch was affixed to the surrounding dura using non-penetrating titanium clips (**D**), with fibrin sealant also used to complete dural repair (**E**).

DATA AVAILABILITY

Additional data relating to this case may be available on request.

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COMPETING INTERESTS

The authors report no conflicts of interest and have no personal, financial or institutional interest in any drugs, materials or devices mentioned or described in this article.

ADDITIONAL INFORMATION

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