Duroplasty in iatrogenic dorsal spinal cord herniation: illustrative case

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BACKGROUND The case report detailed an unusual presentation of an iatrogenic dorsal cord herniation at the level of the thoracic cord after insertion of an epidural catheter 8 months before presentation to the neurosurgical clinic.

OBSERVATIONS Only 13 cases of iatrogenic dorsal cord herniation, most of which occurred after spinal surgery, have been described in the literature. This was the first case of a spinal cord hernia described after the insertion of an epidural catheter. In this case study, the authors described a 38-year-old man who presented with progressive lower limb weakness, sensory deficits, perianal numbness, and urinary/fecal incontinence. He was diagnosed with a spinal cord hernia that reherniated after an initial sandwich duroplasty repair. Definitive repair was made after his re-presentation using an expansile duroplasty.

LESSONS In patients with previous spinal instrumentation who present with neurological symptoms, spinal cord herniation should be considered a likely differential despite its rarity. In this case, a simple duroplasty was insufficient to provide full resolution of symptoms and was associated with recurrence. Perhaps a combination of graft and expansile duroplasty may be used for repair, especially when associated with a tethered cord and in the presence of significant adhesions.

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KEYWORDS spinal cord herniation; spinal duroplasty; spinal cord hernia repair

Spinal cord herniation in and of itself has been noted to be a rare phenomenon and should be suspected in individuals who present with an unusual combination of motor/sensory and autonomic deficits.[1] Dorsal herniation of the cord due to iatrogenic causes is an even more uncommon phenomenon, with only 13 reported cases in the literature to date (Table 1), and these cases have been most commonly due to spinal surgery.

Its association with previous epidural analgesia is even less commonly encountered, and in this case report, we describe the presentation and management of dorsal cord herniation in a patient with complications that may have been secondary to an epidural catheter insertion after a bilateral sequential single lung transplant 5 months before initial presentation.

Posterior cord herniation has been described after trauma, but iatrogenic and idiopathic (due to congenital dural weakness) cases are reported to occur more commonly.[2] This case report is the first description of an iatrogenic herniation occurring after insertion of an epidural catheter.

Illustrative Case

A 38-year-old man presented with a posterior cord herniation at T4 with no history of previous trauma. The history started 5 months before admission to our unit, when he was admitted for a bilateral lung transplant as treatment for his refractory cystic fibrosis. That surgery was mostly uneventful. However, to better manage his pain, an epidural catheter was inserted, the first attempt of which failed.

Eight months after his lung transplant, he presented with reduced sensations in both lower limbs and reports of both urinary and fecal incontinence. He was then referred to the neurosurgical clinic where he was assessed. He described a progressive loss of sensation that started in his right ankle and spread to involve the whole of the right lower limb. Additionally, he reported repeated episodes of both urinary and fecal incontinence. On examination, he had a sensory level at T6, with decreased sensation below his scar on the right side of his trunk. There was also associated dysaesthesia and paraesthesia and loss of temperature on the right side as well as perianal numbness. He was

TABLE 1. A summary of all reported cases of iatrogenic dorsal cord herniation

S/N	Authors	Patient Demographics (yrs, sex)	Initial Presentation	Mechanism of Herniation	Direction of Herniation	Level of Herniation	Time From Initial Insult	Neurological Symptoms	Imaging Findings	Intraoperative Findings
1	Kaliya-Perumal et al., 2019[23]	50 F	Ossification of ligamentum flavum	Removal of OLF, inadvertent dural tear, fibrin sealant	Dorsal	T11-T12	Immediately postoperatively	Deteriorating neurology, nonspecific	Cord herniating out of thecal sac at T11-T12	Hernia irreducible; peri-hernia durotomy, primary closure 6-0 Prolene, & reinforcement w/ fibrin sealant
2	Heller et al., 2017[27]	51 F	Cervical kyphotic deformity w/ cord tethering & adhesions from previous Chiari decompression	No intraoperative dural tear or injury during 3-stage kyphosis correction & stabilization (C5-C7)	Dorsal	C5-C6	4 mos postoperatively	Worsening balance, hand clumsiness, upper limb dysmetria	Extradural herniation of the cervical cord, w/ dural defects on either side of the cord	Adhesiolysis; dura adherent to cord was ellipsed & left attached to cord; expansile AlloDerm patch sutured in place to reestablish thecal sac
3	Watters et al., 1998[28]	33 M	Unstable traumatic odontoid fracture, fixed w/ posterior cervical wiring, which was impacting cord 16 yrs later	Intraoperatively, wire removed w/ dural tear & CSF flow noted	Dorsolateral	C1-C2	2 wks postoperatively (postoperatively from time of wire removal)	Neck pain, w/ return of sensorimotor deficits on lt side	CT myelography: lt dorsolateral herniation, w/ contrast extravasation into the epidural space	Dural tear confirmed; fibrotic tissue formed a subdural membrane; membrane & arachnoid adhesions sectioned; subdural membrane & sutures used to repair dura overlaid w/ gelatin sponge; C1-C3 interlaminar fusion done to restore stability
4	Moriyama et al., 2013[24]	51 M	Excision of intradural extramedullary spinal tumor 10 yrs before presentation	Herniation into pseudomeningocele after cervical cord SOL excision 10 yrs earlier	Dorsolateral	C7	10 yrs	Progressive gait disturbance, spastic paraparesis, urinary symptoms	CT myelogram showed dilatation of the ventral subarachnoid space & DL deviation of the cord into the pseudomeningocele at C7	Cord herniated through dorsal dura, spinal cord tightly adhered around the dural defect; release of adhesions under IOM; primary closure attempted initially but unsuccessful; artificial dura (Gore-Tex) used for subsequent repair
5	Zakaria et al., 2013[5]	57 M	Spinal claudication; intramedullary cyst found at T12-L1; marsupialization of cyst & primary closure of dura performed	Herniation through dural defect postoperatively after cyst marsupialization	Dorsal	T12-L1	8 wks	Worsening back pain radiating down both legs; loss of pinprick sensation up to knees bilat; loss of proprioception & difficulty walking w/ an ataxic gait	Dorsal herniation of the cord through the original dural incision w/ an associated cystic cavity at the level of previous surgery: T12-L1	Posterior approach to expose the thoracolumbar junction; knuckle of herniated cord found under extensive fibrous tissue; cord dissected away from scar tissue, dural defect demarcated, cord reduced, & primary closure done
6	Kwon et al., 2021[29]	64 F	Presented w/ stage IV non-small cell lung cancer & multiple spinal metastases, L1 pathologic fracture, & cord compression	Posterior cord decompression & posterior screw fixation performed from T11 to L3; no dural tear intraoperatively	Dorsal	L1-L2	36 hrs	Abrupt drop in power bilat to 2/5 in both LLs	Ruptured dural sac w/ posterior herniation of the conus	NA; patient declined surgery

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7	Nakashima et al., 2020[9]	55 M	Cervical laminoplasty 8 yrs earlier	Accidental dural tear during laminoplasty	Dorsal	C4-C5	8 yrs	Sudden motor & sensory deficits, gait disturbance, & urinary/fecal symptoms	Posterior displacement of cord through dura at C4-C5 & dilatation of ventral subarachnoid space; CT showed bony defects around the hernia	Lamina & lamina spacer removed; cord repositioned after making 2-mm incisions cranially & caudally (about the defect)
8		60 M	Cervical laminoplasty at C2-C7 due to ossification of posterior longitudinal ligament	Dural tear intraoperatively, not repaired directly, sprayed w/ fibrin glue	Dorsal	C3	6 mos	LL numbness & gait disturbance; UL weakness, & bladder dysfunction	High signal intensity of the cord at C3 & cord herniation at C2-C3	C3-C4 laminectomy & duroplasty; no details provided
9		47F	Durotomy for resection of T11 schwannoma; dura closed primarily using 6-0 prolene; CSF leak noted on postoperative MRI	Dorsal herniation of cord through the dural defect	Dorsal	T11	2 mos	No neurological improvement after initial operation	Dorsal shift of the spinal cord with high-intensity signaling posterior to the cord on initial scans	Cord reduced & defect closed primarily w/ sutures; no other details provided
10	Hosono et al., 1995[25]	45 M	Intradural extramedullary tumor encroaching on the spinal cord at the level of the atlas; dura & arachnoid closed w/ interrupted sutures	Herniation into pseudomeningocele at the level of the atlas	Dorsal	C2-C3	14 yrs	Gait disturbance & clumsiness of rt hand fingers	MRI showed a large cyst posterior to the cord w/ the same intensity as CSF at C2-C3 communicating with the subarachnoid space; cord herniated into the cyst	Adhesiolysis, cyst wall amputation, & the remainder sealed w/ fibrin glue
11	Abd Elwahab & O'Sullivan, 2015[30]	56 M	Incidental finding of an R dumbbell neurofibroma at C2-C3	Herniation into pseudomeningocele at the level of C3 5 yrs postoperatively	Dorsal	C2	5 yrs	Neck pain & progressive weakness of rt UL	Pseudomeningocele w/ cord herniation & entrapment by bone edges	Prone position w/ three-point cranial fixation; incarcerated cord; defect enlarged & adhesiolysis done; cord reduced; dural defect sealed using dural substitute (Neuro-patch) w/ 5-0 Prolene
12	Belen et al., 2009[31]	22 M	Chiari I undergoing FMD & C1 laminectomy w/ subsequent CSF leak	Cord tethering to soft tissues at the level of C2 & eventual herniation into pseudomeningocele	Dorsal	C2	1 yr (diagnosis) 7 yrs (operation)	Unchanged motor deficit & muscle atrophy after initial FMD: 1 yr Worsened hand function in the past 3 mos, dysaesthesias in trunk & limbs: 7 yrs	Spinal cord tethering at C2 & pseudomeningocele at 1 yr 7-yr imaging: posterior herniation of the cord at C1 level	The patient refused operation at 1 yr 7-yr operation: exploration of the posterior fossa in the prone position; soft tissue dissected up to herniated neural tissue; cord noted attached to muscle; freed microsurgically using sharp dissection; pulsation noted; duroplasty done using a Gore-Tex sheet; no other details provided

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S/N	Authors	Patient Demographics (yrs, sex)	Initial Presentation	Mechanism of Herniation	Direction of Herniation	Level of Herniation	Time From Initial Insult	Neurological Symptoms	Imaging Findings	Intraoperative Findings
13	Iencean & Poeata, 2014[32]	51 M	C2-C4 low-grade ependymoma w/ complete tumor removal	Dural defect after surgery	Dorsal	C2-C3	5 yrs	Distal paraesthesias of the limbs & progressive tetraparesis w/ difficulty mobilizing	Posterior spinal herniation through a dural defect at C2-C3	Resection of dural scar around pseudomyelocele, release of the spinal cord, & reconstruction of dura mater

CSF = cerebrospinal fluid; CT = computed tomography; DL = dorsolateral; FMD = foramen magnum decompression; IOM = intraoperative monitoring; LL = lower limb; NA = not applicable; OLF = ossification of the ligamentum flavum; SOL = space occupying lesion; UL = upper limb.


FIG. 1. T1 MRI sequence showing the initial dorsal protrusion of the dorsal cord preoperatively.

noted to have bilaterally brisk tendon reflexes, with clonus in both ankles and bilateral flexor plantar response. However, he had intact motor power in all muscle groups. There was no history of mechanical trauma or previous spinal operations.

A magnetic resonance imaging (MRI) scan (Fig. 1) showed a dorsal cord herniation and posterior breaking of the cord with T2 high-signal intensity of the central and dorsal cord at the level of T4.

Considering the history, presenting signs, and postpresentation MRI scan, the suspicion was that of a spinal cord hernia at the T4 level. The patient was thus admitted a week later for surgical repair of the cord herniation. During the initial operation, the herniated cord was noted to have "mushroomed" through a small 10-mm dorsal dural defect with adhesions to the dura on the outside. Dural and arachnoid adhesions formed a ring around the herniated cord and incarcerated it (Fig. 2). Dissection was started by making two dural midline snip incisions cranial and caudal to the herniated cord. From the cranial opening, careful dissection was performed in the caudad direction until encountering the incarcerating ring and vice versa from the caudal opening. Careful motor evoked potentials-guided adhesiolysis was performed around the incarcerated cord to separate it from the dura. The herniated cord was completely released but never restored to a complete normal morphology, with persistent bleb-like swelling at the location of the herniation. Further dissection would potentially endanger the neural tissue, and eventually, the cord was untethered and returned to its normal position within the thecal sac (Fig. 3), where it was pulsating normally. The dural opening was approximately 25 mm at the end in craniocaudal
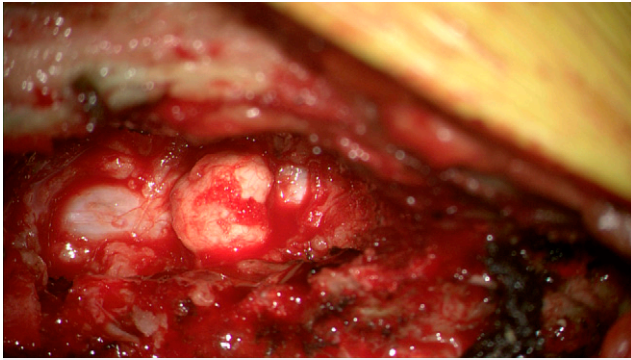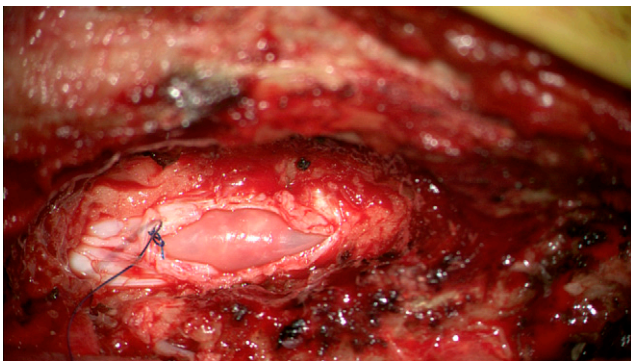
FIG. 2. After access into the skin, obvious incarceration of the cord by dural and arachnoid adhesions is visible, forming a ring around the herniated cord.

FIG. 4. A T2 MRI short T1 inversion recovery sequence showing improved herniation of the cord at clinic follow-up. The inset picture shows the plane at which the larger sagittal slice (the bigger picture) was taken.

diameter, with the reduced cord seen freely pulsating inside. Attempts at primary repair would have ended in compression on the cord, so the dura was left open. In situ synthetic dural substitutes were used as onlay (above) and inlay (below) for this opening (also called the sandwich technique).[3,4] Stitches without tension were used to secure the grafts to avoid misplacement.
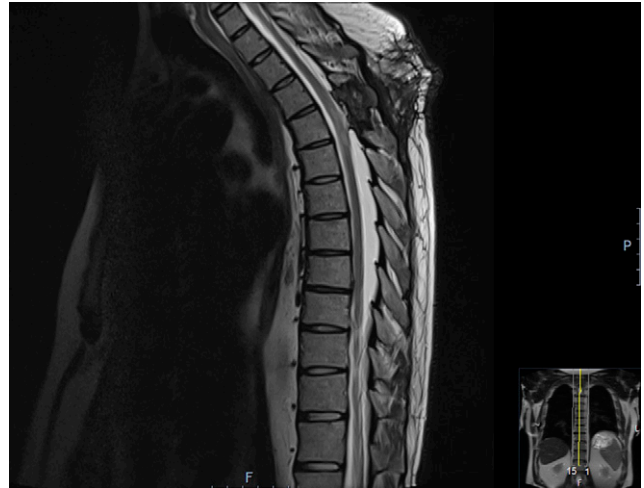
The patient made a good clinical recovery with improved sensations in both lower limbs. He was discharged home a few days later. Unfortunately, 2 weeks after the operation, he presented again with a 48-hour history of sensory deterioration in both lower limbs. Deep sensations were particularly affected. The picture was suggestive of posterior cord syndrome.

Reexploration was decided. Intraoperative findings were as follows: the cord was herniating through the surgical dural opening and pushing the onlay grafts dorsally. A false incarceration ring was created by the edges of the dural opening. It was clear that the cord had retethered although the inlay graft was still in place.

Onlay dural grafts were removed. The dural opening was extended approximately 5 to 10 mm in both cranial and caudal directions. A sleeve of fascia with fat was inserted and sutured in place to the dural edges. This time, dural hitch stitches were applied between the dura and the paraspinal muscles to create a tent-like space behind the cord. Adequate expansile duroplasty was achieved at the end with ample artificial dorsal subdural space. Synthetic dural sealants were used, with no cerebrospinal fluid effluence noted at the end.

FIG. 3. Cord reduced into intrathecal sac before closure using an expansile duroplasty.

After the operation, the patient made excellent progress neurologically as well as with the physiotherapy team and was discharged 18 days after the second operation. He subsequently has made a good recovery. A delayed postoperative MRI scan at follow-up (Fig. 4) shows improved cord edema. Clinically, the patient self-reports a slow improvement in proprioception as well as a return of bladder sensation and a reduction in the frequency of the episodes of urinary incontinence and no new reported episodes of fecal incontinence.

Discussion

Spinal cord herniation is described in the literature as a rare cause of spinal cord dysfunction,[1,5–8] which is often amenable to surgery. It can commonly be classified into spontaneous, iatrogenic, and posttraumatic. Spontaneous and idiopathic presentations have been described more commonly in the literature, and herniation due to a traumatically acquired dural defect is the least reported cause.[1] Iatrogenic dorsal cord herniation has been described even more rarely,[9] with only 13 cases reported in the literature to date (Table 1).

Generally, most herniations have been described in the ventral and/or ventrolateral spinal cord, most commonly at the T4-T5 level.[8] Spontaneous dorsal cord herniation has been described even more rarely, with one case described in a 2-year-old boy secondary to herniation into an extradural arachnoid cyst.[10]

The presentation of spinal cord herniation is reportedly variable, but more than 50% of cases present with Brown-Sequard syndrome, which is suggestive of cord hemisection.[11] Commonly, they also present with spastic paraparesis, a dysfunctional bladder/dysfunctional bowel (up to 71% of patients), lower limb pain, and sometimes low-pressure headaches.[8] When associated with tethering, the presentation may include pain (in the lower back or lower extremities), lower limb weakness, and sensorimotor dysfunction unrelated to dermatomal distribution.

The pathophysiological processes for developing spinal cord herniations are not well understood. However, as much as the entities of tethered cord and tethered cord syndrome have been described differently, with the latter described as a tethering of the filum, their pathophysiological mechanisms share some similarity.[12–16] In

in vivo models, the pathophysiological mechanism of spinal cord malfunction in cord tethering was postulated to be due to metabolic dysfunction in stretched and incarcerated neurons, which results in ischemia because of anterior spinal artery involvement and increased cord susceptibility to hypoxic injury due to distortion of the smaller vessels around the cord.[16,17] Also, traction-induced ischemia has been linked to reduced conduction, leading to both reversible and irreversible nerve damage.[12–15,18] Clinical progression in cord tethering has been postulated to be stretch-related damage to the herniated/tethered fibers in the lateral funiculus involving the spinothalamic and, subsequently, descending tracts.[8,13] The vascular mechanism has also been postulated to explain the lack of improvement in some patients after untethering.[16]

Making a diagnosis of spinal cord herniation is difficult because this entity is classically rare and not the foremost diagnosis on most neurologists'/neurosurgeons' minds, with the median time to diagnosis approximately 17 months and one patient having a 16-year time to diagnosis.[19] This delay in diagnosis typically leads to unnecessary testing, visits to the doctor, imaging, and procedures that may have no benefit to the patient. Tethered cord in itself is notoriously difficult to diagnose, with most cases presenting with a long history of misdiagnosis before being correctly diagnosed.[16]

There seem to be two general types of herniated cord according to the intraoperative morphology. The first type occurs in small defects (<1–2 cm) within a relatively short time of insult.[20] In this type, adhesions are minimal, and herniation is mainly driven by the differential pressure between the subdural and extradural spaces. Good primary closure seems to be sufficient. The second type occurs in larger defects (>1–2 cm) with longer time intervals. Adhesions are the main pathology, and primary repair is difficult and almost impossible.[20] The current mainstay of treatment of this type involves careful microdissection to release all surrounding adhesions, returning the cord to its original position, and effecting closure using one of two techniques: the enlargement of the inner dural defect, first described by Watanabe et al.,[21] which was our initial approach during the first operation, or a dural graft as propounded by Zairi et al.,[22] which was our second approach.[21,22]

We reviewed the literature that reported similar cases with an iatrogenic dorsal cord herniation (Table 1). Special consideration was given to the surgical technique used and its correlation with the presentation.

Primary closure was attempted successfully in three cases.[5,9,23] Those cases had the following factors in common: a short interval between the time of insult and time of presentation (ranging between immediately postoperative to 8 weeks), being in the thoracic region, and having smaller dural defects.

On the other hand, primary closure was performed unsuccessfully in one case reported by Moriyama et al. in 2013.[24] That case had a 10-year interval between the primary cervical operation and the new presentation. The dural defect was larger, with thick surrounding dural adhesions and cord tethering. A different strategy was retried successfully that involved subsequent secondary repair with artificial dura using an expansile duroplasty technique.

Another technique that was used by Nakashima et al. and Hosono et al. involved careful adhesiolysis, amputation of any pseudocysts, reduction of the cord to the thecal sac, and enlargement of the dural opening in both cranial and caudal directions.[9,25] This is done in combination with or without an inlay graft and a dural sealant.

In cases with long-standing pathology with larger defects when thick adhesions are the main driving force of tethering, varying

degrees of bony decompression were done at first to gain safe access to the herniated cord. This was followed by adhesiolysis and dural opening enlargement, as described in the former technique. There is an added use of grafts, either synthetic or autologous. The graft is sutured in place to create an adequate space behind the cord and create expansive duroplasty.

After surgical intervention, a significant improvement in symptoms has been reported, with 78% (with back pain at presentation) and 83% (with leg pain at presentation) of patients reporting improved symptoms.[26]

Observations

For this index patient who presented with an insidious onset of symptoms predominantly suggestive of a posterior cord syndrome, we hypothesize that the precipitating event was the insertion of the epidural catheter after his bilateral lung transplant.

Dorsal cord herniation associated with tethering has rarely been reported in the literature, and unsurprisingly in our index case, it was misdiagnosed repeatedly in the initial workup period, which is commonly encountered with tethered cord.[26]

To remedy his symptoms, which had gotten worse over time, we decided on a surgical approach. Our initial approach was adhesiolysis with enlarging of dural opening and inlay graft, which failed and resulted in retethering. Reoperation was needed with a more extensive expansion duroplasty and closure with an inlay graft with a tenting technique of the dura. That approach was successfully done to reduce the possibility of retethering.

Lessons

Dorsal cord herniation (especially iatrogenic) has been rarely reported in the literature, even less frequently in association with cord tethering. Spinal cord herniation is a rare phenomenon that occurs more commonly as either a spontaneous or idiopathic herniation. The diagnosis of cord herniation possibly with associated tethering should be considered in patients with a bizarre combination of upper motor neuron neurology that cannot be attributed to any other central neurological lesion. Additionally, because of the proposed pathophysiological mechanism of vascular injury in cord tethering, there is a chance of no improvement after detethering of the cord in some patients.

Also, during the management of this case, we attempted a singular technique for closure using graft duroplasty, which resulted in retethering. Therefore, it may be worth considering a combination of both expansion and graft duroplasty for the management of dorsal cord herniation, more so in association with tethering in the presence of significant adhesions. However, more research is required to determine if this is associated with a decreased rate of reoperations and/or recurrence of herniation. The mechanism of cord herniation may have been responsible for his delayed presentation approximately 5 months after his bilateral lung transplant operation.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: all authors. Acquisition of data: Ogbu, Eltoukhy. Drafting the article: Ogbu. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Ogbu. Administrative/technical/material support: Tzerakis. Study supervision: Tzerakis.

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