Cord Splitting Access to Ventral Intradural Cysts of Cervicothoracic Junction and Thoracic Spine

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OBJECTIVE: **Surgical treatment of ventrally located intradural cysts is difficult and controversial. Laminectomy with division of the denticulate ligaments and gentle cord mobilization remains the standard approach but risks further neurologic deterioration secondary to cord manipulation. Our purpose is to evaluate the safety and effectiveness of a midline cord-splitting approach as an alternative for treating ventral thoracic intradural cysts.**

METHODS: **We describe 2 patients who were treated for ventral intradural cysts causing progressive and severe myelopathy. Under general anesthesia and continuous neurophysiologic monitoring, laminectomy, durotomy, and cord splitting through a midline approach gave direct access to both lesions. Cyst drainage was supplemented by a cystopleural shunt in 1 case.**

RESULTS: **Cyst collapse and cord reexpansion were documented in both patients with a magnetic resonance imaging scan 1 week after surgery. In both cases there was a significant neurologic improvement, which was maintained 2 years postoperatively. Intraoperative monitoring recorded no loss of somatosensory or motor potentials during surgery. Follow-up magnetic resonance imaging scans 2 years postoperatively showed no evidence of cyst recurrence, and both patients remained neurologically improved and stable.**

CONCLUSIONS: **We have been able to drain 2 ventral intradural cysts using a cord-splitting technique. This has allowed safe access to purely ventrally located lesions, which were inaccessible dorsally or dorsolaterally. By using this method we have been able to avoid a more invasive ventral transthoracic approach necessitating vertebrectomy and reconstruction and risking serious complications.**

INTRODUCTION

Intradural cysts are uncommon lesions, which can be found at almost any level in the spine.[1-4] Clinical presentation and the rate at which symptoms progress will determine whether surgical treatment is indicated.[5] The size and location of the cyst in relation to the spinal cord are the key factors that need to be taken into account when choosing the best approach.[6,7] Dorsally located lesions can be successfully approached and removed through a conventional laminectomy/hemilaminectomy or laminoplasty.[8-10] The treatment is far more complex in ventrolateral or purely ventral lesions.[11-13] In such cases, a posterior approach may not give adequate cyst exposure, making total removal difficult and increasing the risk of recurrence. In some cases, dividing the denticulate ligaments and rotating the spinal cord have been advocated as a safe adjunct to optimize surgical treatment.[1,13,14] Manipulating a compromised cord in this way can result in aggravating preexisting neurologic deficits.

Over the past few years several authors have reported corpectomy and anterior column reconstruction as a satisfactory method to achieve complete excision of ventral intradural cysts of the cervical spine.[11-13] Depending on the level, the same approach in the thoracic spine may be possible but is a considerable surgical undertaking, with potentially life-threatening complications including cerebrospinal fluid (CSF)-pleural fistula and infection.[15,16]

We present our experience in 2 cases of ventral intradural thoracic cysts, where a conventional posterior or posterolateral

Key words
- Cerebrospinal fluid
- Cyst
- Hydrocephalus
- Neuroenteric
- Spinal cord

Abbreviations and Acronyms
CSF: Cerebrospinal fluid
MEP: Motor evoked potential
MRI: Magnetic resonance imaging
SEP: Somatosensory evoked potential

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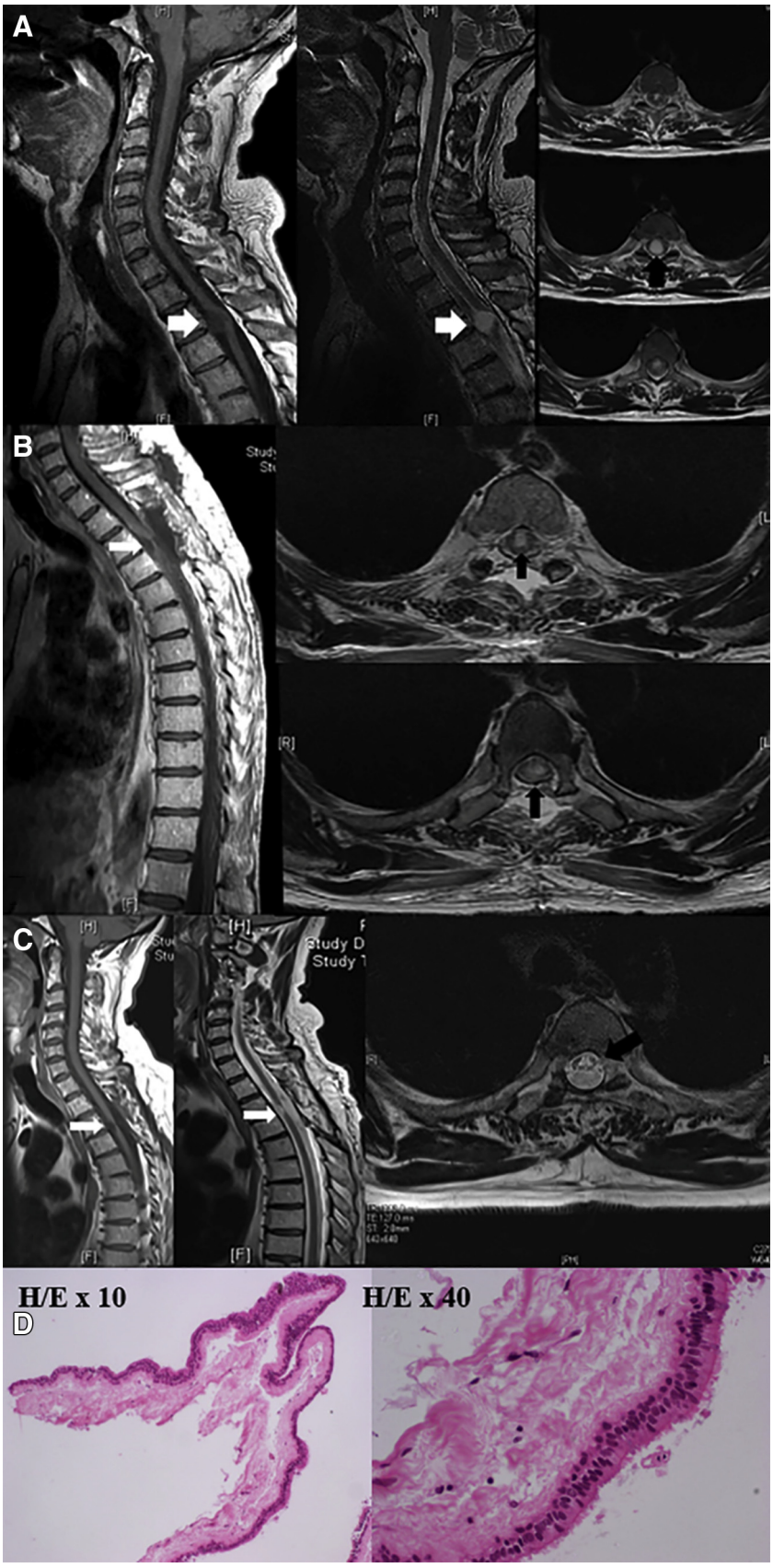
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approach even with denticulate section and gentle cord rotation would have granted neither adequate nor safe access. Both lesions were deeply indenting the spinal cord, severely reducing the anterior-posterior thickness of the cord. We chose a midline approach splitting the cord longitudinally through the median raphe. In the first case the cyst was excised, and in the second case the cyst cavity was shunted into the pleural space.

We discuss the benefits, risks, and technical aspects of our approach and review the literature.

MATERIAL AND METHODS

We reviewed the records of 16 patients with intradural spinal cysts treated over a 2-year period at our institution. Median age was 53 years (9 females, 7 males). In 12 cases the cyst was located dorsally to the thoracic cord, and in 2 cases it was at the conus medullaris. The 2 ventral cysts were cervicothoracic (C7-T4) and upper thoracic cord (T4). A preoperative magnetic resonance imaging (MRI) of the whole spine with contrast was performed in all cases. Clinical presentation included progressive paraparesis (16 patients), loss of sphincter control (5), ataxic gait (8), radicular pain (4), and sensory disturbance. The histology was consistent with 10 arachnoid, 2 neurenteric, 1 ependymal cyst, and 3 cases of arachnoiditis.

Cysts located dorsally to the cord were always approached by a standard laminectomy, which allowed complete and safe removal. In the patients with arachnoiditis (1 posttraumatic, 1 post-irradiation, and 1 following posterior fossa surgery), the laminectomy was widened to the margins of the dura to allow division of the denticulate ligaments and cord adhesions in an attempt to reestablish physiologic CSF flow.

In the remaining 2 cases, a ventral C7-T4 and a ventral T4 cyst were treated as urgent cases due to rapidly progressive paraparesis. We describe these 2 cases in more detail.

The study was approved by the Institutional Review Board, and written informed consent was obtained from all patients/next of kins according to the recommendation of The Declaration of Helsinki for investigations involving human subjects. The research data analysis has no effect on the participants or their medical care.

Illustrative Cases

Case 1. A 64-year-old male was referred with a 12-month history of progressive difficulty in walking, gait ataxia, and symmetrically reduced sensation over the lower limbs. He was seen in the emergency department with a sudden worsening of his symptoms in the days leading up to his admission, with frequent falls caused by his legs suddenly giving way. Neurologic examination revealed reduced power (grade 3/5, Muscle Strength Grading Scale, Royal Medical Research Council of Great Britain) in both legs, diminished reflexes, and equivocal plantars. He had an incomplete sensory level at T4 with no sacral sparing. Anal tone was absent.

An emergency MRI of the whole spine showed a low signal intensity lesion in T1 sequences, high signal in T2, located ventral to the cord at the T4 level and displacing the spinal cord posteriorly. The spinal cord was severely compressed with a thin shell of residual neural tissue surrounding the lesion laterally and dorsally (**Figure 1A**).

Due to the acute clinical deterioration, the patient was scheduled for urgent surgery. Under general anesthesia and continuous neurophysiologic monitoring, he was positioned prone on a Jackson table, with the head fixed in a Mayfield clamp. The T4 vertebral body was located by intraoperative radiographs, and a midline skin incision extending from T2 to T6 was performed, followed by a T3-T5 laminectomy. After opening the dura and retracting it with hitch stitches, a D-wave electrode was inserted subdurally, caudal to the lesion. The cord appeared significantly swollen and almost translucent on its dorsal surface. Two attempts at mobilizing it laterally were unsuccessful. A midline dorsal incision was then made into the spinal cord, and pial stitches were fixed to the dura. The cord was split longitudinally along the median raphe. No change in somatosensory evoked potentials (SEPs) or motor evoked potentials (MEPs) was observed during this phase. The posterior wall of the cyst was encountered, and the exposure through the cord was extended cranially and caudally to identify the superior and inferior borders of the lesion. The cyst was then fenestrated to reduce its volume, its anterior wall was isolated from the ventral dura, and a complete excision was obtained. No change in MEPs was observed during cyst manipulation, and gratifyingly a mild improvement in SEPs and MEPs occurred after the cyst was removed. The postoperative course was uneventful. One week after surgery a postoperative MRI showed partial cord reexpansion with persistence of high T2 signal intensity in the area previously occupied by the cyst (**Figure 1B**). A slight motor improvement was observed within the first 2 postoperative weeks. Histology was consistent with a neuroenteric cyst. Two years after surgery the patient's gait was still ataxic but significantly improved, and he was able to walk with a stick. Residual sensory impairment was minimal, and bladder and bowel function had returned to normal. The 2-year MRI showed no evidence of recurrence with significant cord reexpansion (**Figure 1C**).

Case 2. A 25-year-old female was brought to the emergency department of our hospital due to a 6-month history of progressive gait impairment. Over the 2 weeks before admission her legs had become so weak that she could only lift them from the bed when helped by 2 people and was unable to stand. She also complained that over the preceding 24 hours she was unaware when emptying her bladder and she could only empty her bowels after a

Figure 1. (**A**) Magnetic resonance imaging (MRI) sagittal T1 and T2 images, showing the presence of an intradural cystic lesion located at the T4 level, ventral to the thoracic cord (*white arrows*). On the right side, axial images show the cyst from its cranial to caudal end. At the site of the lesion's largest diameter (*black arrow*), the cyst has pushed the cord laterally and posteriorly, where only a minimal layer of neural tissue seems to remain. (**B**) Postoperative MRI, in sagittal (*white arrow*) and axial (*black arrows*) projections, showing initial cord reexpansion. (**C**) 2 years postoperative MRI, showing full cord reexpansion in the sagittal T1, T2 (*white arrows*), and axial planes (*black arrow*). (**D**) Histology, hematoxylin-eosin coloration, respectively, 10× and 40× magnification pictures. On the left, neurenteric cyst wall demonstrating simple columnar and cuboidal epithelium arranged around a cystic cavity, mimicking respiratory or gastrointestinal epithelium. On the right, at higher magnification, columnar and cuboidal mucin-secreting goblet cells are best appreciated.

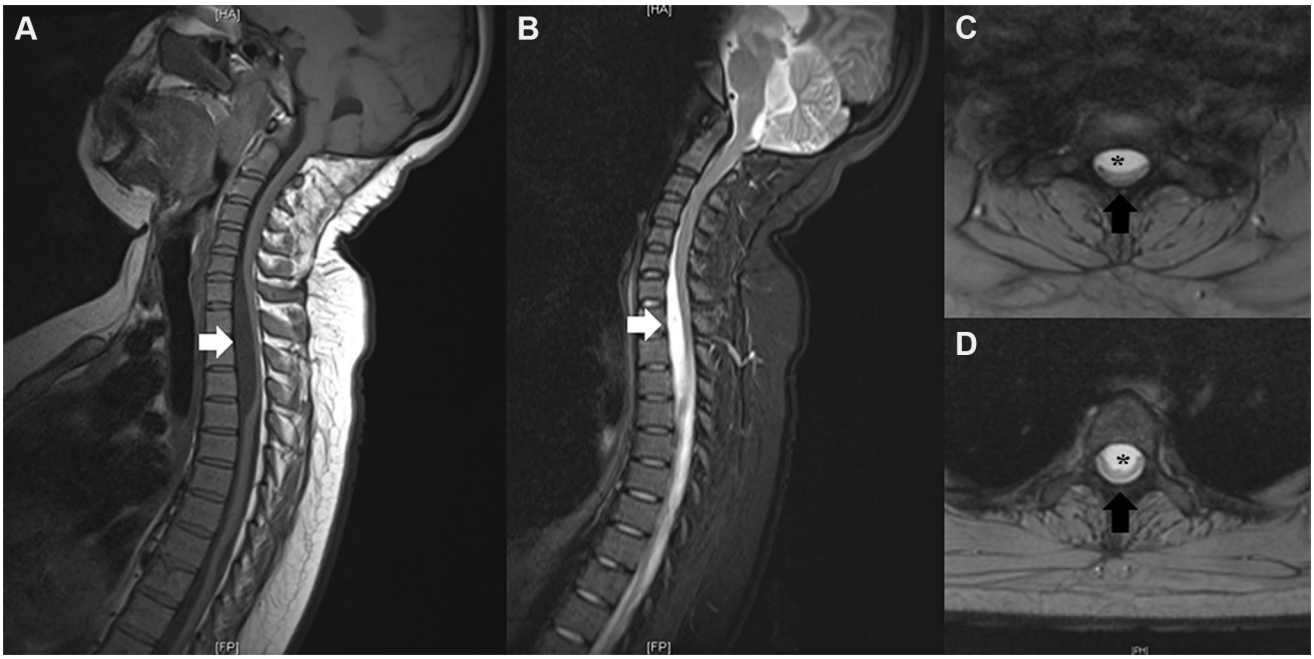

Figure 2. Sagittal T1 (**A**) and T2 (**B**) magnetic resonance images showing a large intradural cystic cavity (*white arrows*) extending from C6 to T4, located ventral to the cord, which is severely pushed backward. Axial T2 images (**C**—**D**) are shown on the right, including the level of larger cyst (*asterisks*) width (**D**). The cord has been molded into a C-shape (*black arrows*).

significant effort. Her previous medical history was interesting. At the age of 3 she had been operated on for the removal of a fourth ventricle ependymoma. She subsequently developed hydrocephalus, and a nonadjustable, medium-pressure ventriculoperitoneal shunt was positioned. At regular follow-up examinations over the years no tumor relapse was observed. Neurologic examination revealed 2/5 power in both legs. Reflexes were absent in the lower limbs and brisk in the upper limbs. There was a sensory level at T2. Sensation around the perineum and perianal area was also diminished, and anal tone was reduced. She underwent an urgent MRI scan of the brain and spine, which demonstrated a large cyst lying ventral to the spinal cord between C7 and T4. The spinal cord was posteriorly displaced, and its anterior-posterior depth significantly reduced (Figure 2). The ventriculoperitoneal shunt function was investigated with an infusion test and was found to be working normally. Due to neuroimaging characteristics, a diagnosis of arachnoid cyst was suggested.

When considering the surgical options, an anterior transthoracic approach was not considered suitable. A posterior approach was selected (Figure 3 and Video,Supplemental Digital Content). Under general anesthesia, the patient was positioned prone on a Jackson table, with the head in a Mayfield clamp with full neurophysiologic monitoring of spinal cord function. The upper and lower borders of the laminectomy were identified by intraoperative radiographs. A skin incision was drawn from C5 to T6, and the laminae were exposed by bilateral subperiosteal dissection. After further x-ray confirmation, a C7-T4, partial C6, and partial T5 laminectomy was performed. The dura was opened in the midline, and dural hitch sutures were inserted. The spinal cord filled the dural opening and was tense (Figure 3A). A D-wave recording electrode was positioned subdurally running caudal to the cyst (Figure 3B). We attempted to access the cyst lateral to the cord, but this proved impossible because the cord was ballooned backwards filling the dural tube (Figure 3C). Pial stitches were inserted. The cord was incised longitudinally in the midline and split down through the median raphe, to gain access to the ventral surface of the intradural space. We did not encounter the anterior spinal artery, and we were careful to ensure that when we opened the ventral pial layer there was no vessel lying deep to the spinal cord. The cyst cavity was opened at the T3 level (Figure 3D). No changes in MEPs and SEPs were observed. The cord splitting was extended rostrally and caudally to improve the access. The ventral wall of the cyst was fenestrated, and the ventral dura was identified (Figure 3E). A cystopleural shunt was then introduced through the split in the cord and fed cranially toward the superior extent of the cyst cavity (Figure 3F). The spinal cord was no longer under tension, and its lateral surface could now be seen, with CSF free flowing around it (Figure 3G). Watertight dural closure was performed, and the proximal end of the shunt was sutured to the dura, to minimize any risk of displacement (Figure 3H). A slight improvement in SEPs and MEPs was noted by the end of the procedure. Three days after surgery the patient was able to stand but not walk. Power was 4/5 in the left leg and 4+/5 in the

Video

Video available at www.sciencedirect.com

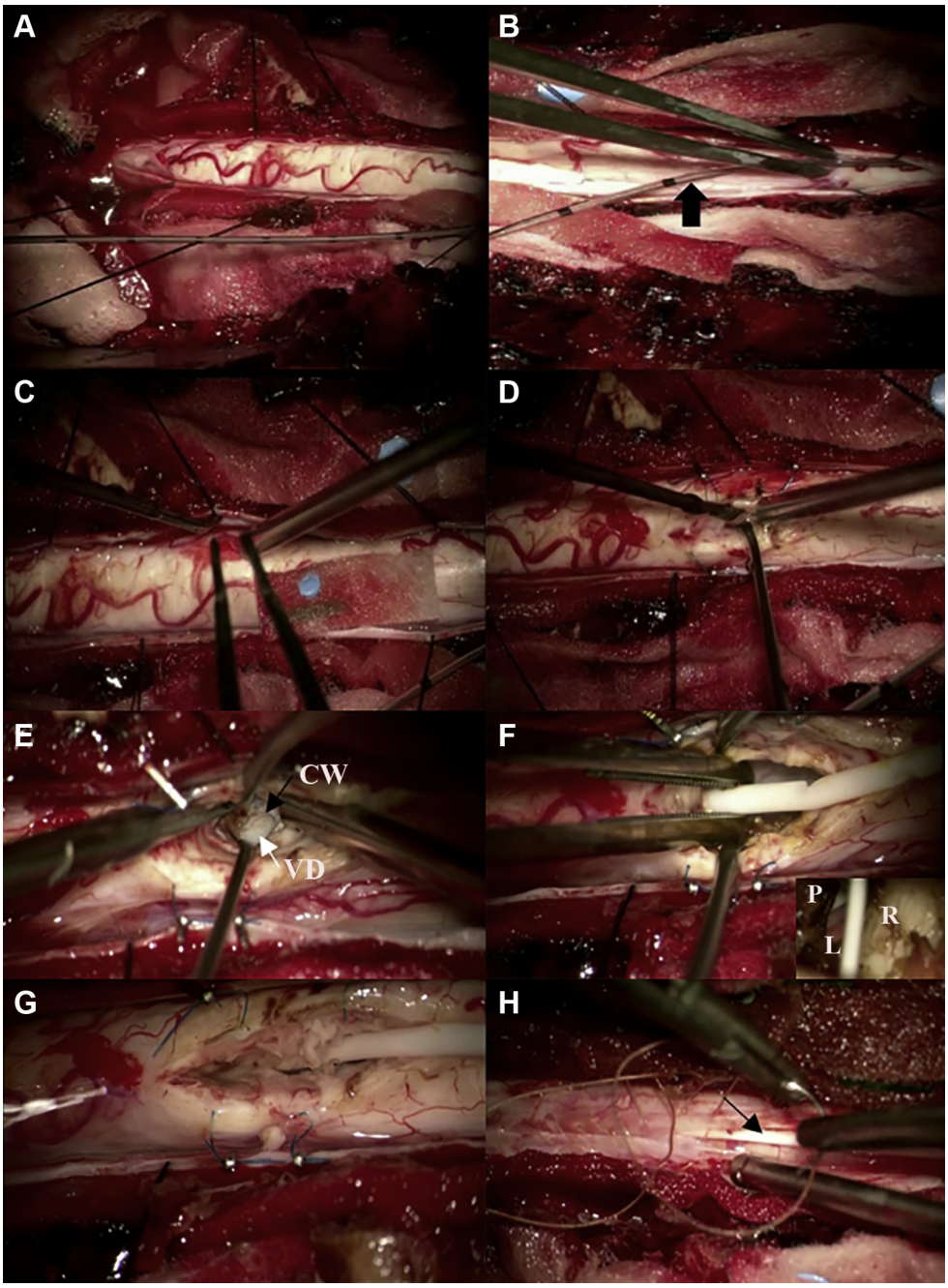

Figure 3. (**A**) After performing a C7-T4, partial C6, and partial T5 laminectomy, the thecal sac has been opened along the midline and dural hitch sutures have been inserted. The spinal cord fills the dural opening and appears tense. (**B**) The D wave electrode (*black arrow*) is carefully inserted subdurally below the caudal end of the cyst cavity. (**C**) The attempt at accessing the cyst lateral to the cord fails because the cord is ballooned backwards filling the dural tube and minimal manipulation causes immediate pial bleeding, requiring control by means of bipolar coagulation and increasing the risks of further neurologic damage. (**D**) After positioning pial stitches and coagulating the superficial veins, the cord is incised longitudinally in the midline and split down through the median raphe, to gain access to the ventral surface of the intradural space. The cyst cavity is opened at the T3 level. (**E**) The cord splitting is carefully extended rostrally and caudally to improve the access. The ventral wall of the cyst is fenestrated, the cyst wall (CW, *black arrow*) is visible, and the ventral dura (VD, *white arrow*) can be identified. Cerebrospinal fluid (CSF) freely flows across the split. (**F**) A shunt tube is introduced through the split in the cord and fed cranially toward the superior extent of the cyst cavity. The opposed end of the shunt is inserted at the right sixth intercostal space through a small fenestration in the pleura (L, lung; P, pleura; R, Rib), to grant permanent cyst diversion. (**G**) The spinal cord is now no longer under tension and its lateral surface can be seen, with CSF free flowing around it. (**H**) A watertight dural closure is performed and the proximal end of the shunt is sutured to the dura (*black arrow*) to secure it against the risk of displacement.

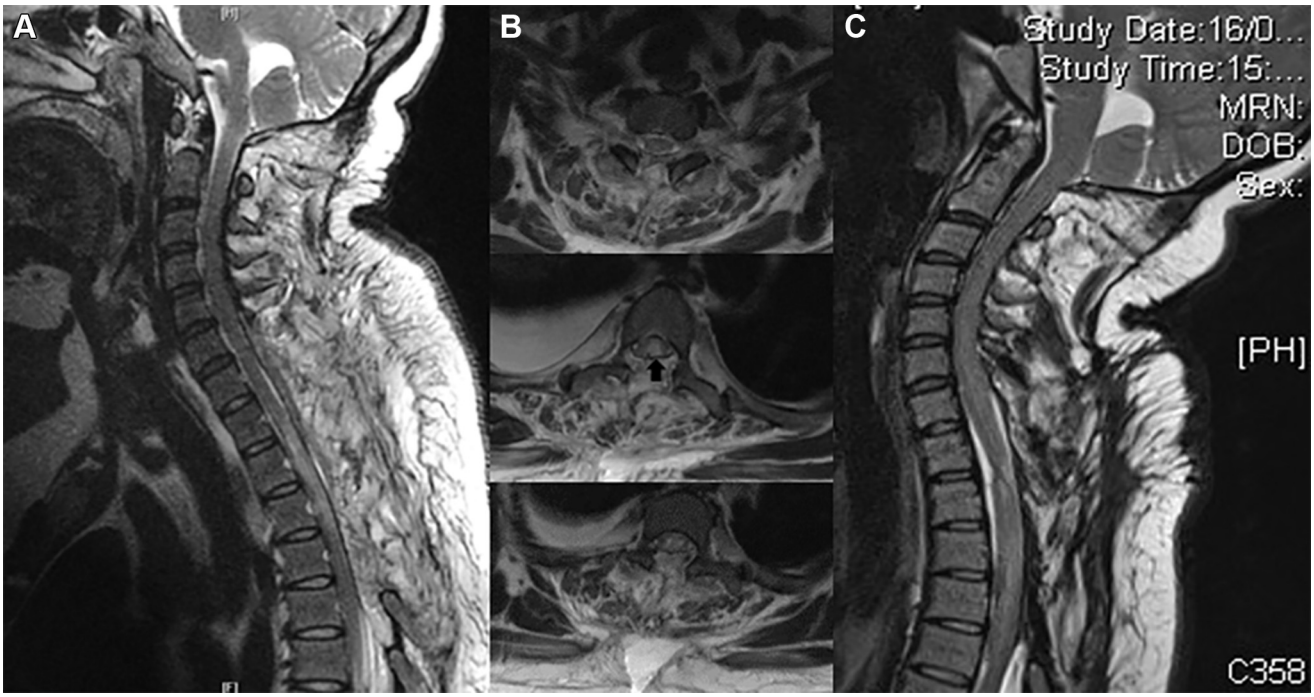

Figure 4. (**A** and **B**) Early postoperative magnetic resonance imaging (MRI). On the left, sagittal T2 images, showing cord reexpansion with no residual cyst left. The spinal cord has significantly reexpanded. On the right, axial images. At the top level, cord expansion appears almost complete (*black arrow*). At the bottom end, the split is still partially visible. (**C**) 2 years postoperative MRI. On the left, sagittal T2 image confirms fully stable cord expansion with no evidence of cyst relapse.

right. Bladder and bowel disturbance were unchanged. An early postoperative MRI showed full cord reexpansion with collapse of the cyst cavity (Figure 4). Three weeks postoperatively the patient was able to walk with a Zimmer frame, and after 1 month was unsupported. Urinary continence recovered incompletely, bowel function was regular, and anal tone was present. There was a slight improvement in the sensory level, which was now T7. A follow-up MRI at 2 years showed no recurrence of the cyst, and the patient was neurologically intact and independent (see Figure 4).

DISCUSSION

Spinal cysts are rare lesions.[1,3,6,8,10] They can be found either extradurally or intradurally, most frequently in the thoracic spine.[17] Their clinical presentation is varied, with the lesion's location being strategical for symptoms and treatment.[13,17-20] Arachnoid cysts represent >80% of these lesions, followed by neuroenteric cysts and posttraumatic/postinflammatory adhesive collections.[1,3,5,8,11,14,18]

Several different classifications have been attempted on the basis of histopathology, pathogenesis (congenital vs. acquired), and etiology (idiopathic, posttraumatic, inflammatory, parasitic, etc.). In 1988 Nabors suggested a comprehensive classification based on surgical, radiologic, and histologic criteria. He identified 3 main cyst types: 1) extradural without spinal nerve root fibers, 2) extradural with spinal nerve root fibers, and 3) intradural.[5]

The largest series reported in the literature are by Fortuna[14] (18 patients, 9 arachnoid, 9 neuroepithelial), Alvisi[13] (17 cases, all arachnoid cysts) and Bassiouni[5] (21 cases, 16 arachnoid cysts, 4 neuroepithelial cysts, and 1 cervical nerve root cyst). All these authors agree that, when feasible, the aim of surgery should be complete cyst removal to relieve pressure on the spinal cord or nerve roots and to reduce the risk of lesion reexpansion and recurrence.[4,18,20,21] Large multilevel cysts, especially when ventrally located, represent a complex surgical challenge, with a risk of further neurologic deterioration.[7,11,12,15] Alternative strategies such as cyst marsupialization; shunting to peritoneum, pleural space, or atrium; and multiple fenestrations have been reported with good results.[11,12]

Dorsal cord cysts can be successfully approached and removed by laminectomy, hemilaminectomy, or laminotomy.[2,13,17] Ventral cervical arachnoid cysts are extremely rare. Only 14 cases have been reported.[12] In his 1974 paper Palmer described 2 out of 6 patients affected by ventral cervical cysts.[22] In the first case, a 19-year-old female, simple fenestration of the cyst through a C1-4 laminectomy led to permanent improvement of quadriparesis. In the second case, a 3-year-old boy, a cervical laminectomy followed by needle aspiration through the median raphe allowed a transient neurologic improvement. Unfortunately, the final diagnosis was reached postmortem, 2 operations later. A large arachnoid cyst compressing the ventral surface of the upper cervical spinal cord was found. Laminectomy and division of the denticulate ligament with subsequent gentle cord retraction and

rotation have become the standard approach for ventral cysts, but all the authors agree that in these patients attempts at complete excision are associated with an increased risk of spinal cord injury. This is particularly true in cases where the spinal cord has been molded into a C-shape and is displaced posteriorly so that it occupies not only the dorsal but also the entire lateral surface of the intradural space. Cyst fenestration through laminectomy can be chosen in case of lesions extending over multiple vertebral levels.[12] Obviously, in these patients fenestration might be limited to the point of maximal cyst extension or, in case of multiple cysts, used in 2–3 different places. Unfortunately this poses higher risks of damage beyond control. Also, the main disadvantage is that a laminectomy spanning several levels may negatively affect spinal stability, especially if the area of surgical interest is placed at the column's junctional levels. Partial cyst resection can be used as an alternative, but its effectiveness has been questioned; results can be quite unsatisfactory and lead to fast relapse.[11,17] Some authors have reported good results by the use of marsupialization (i.e., shunting to the pleural or peritoneal space, occasionally even to the atrium).[11,12] The main problem in these cases is that the surgeon needs at the same time to enter the cyst to put a catheter inside it and get enough space to avoid the risk of shunt malpositioning. Nonetheless, the concept of fenestration is based on the creation of a communication as large as possible between the cyst and the intradural space, interrupting the arachnoidal veil that adheres to the cord, to maximize the chance of reestablishing a more physiologic CSF flow, to which the catheter is an adjunct. Unfortunately, a large opening may facilitate to and fro movements of the catheter itself, putting it at risk of displacement or malfunction. To avoid this, a catheter can be sutured to the dura at the time of closure. In 2008 Muhammedrezai[12] reported the first case of corpectomy, cyst removal, and complex spine reconstruction as treatment for a C7 ventral cervical cyst, with a good outcome. Since this report, this approach has been confirmed as a safe and effective surgical option. Its main disadvantage is the technical complexity associated with gaining access, the requirement for reconstruction of the anterior spinal column with cages (grafts) and implants, and the risk of a postoperative CSF fistula.[23]

An anterior transthoracic approach can be used for ventral thoracic intradural cysts.[24] The surgery has greater approach-related morbidity in both the short and long term, related to thoracotomy, vertebral body resection, and complex reconstruction. The dura is difficult to repair, and a postoperative CSF leak into the pleural cavity (subject to negative pressure) can prove problematic. Access to and reconstruction of the upper levels of the thoracic spine is particularly challenging due to the unfavorable anatomy. These ventral approaches have been used because even when laminectomy is extended laterally via a transfacet, transpedicular, or costotransversectomy approach, reaching the anterior surface of the thecal sac remains challenging and, in rare cases, impossible.

The approach we describe in this paper has its foundations on all the previous considerations. In both cases we have described, the method was adopted only after an attempt at gaining access via a posterolateral approach, which was unsuccessful. These 2 patients had severe and progressive preoperative neurologic compromise, and in both patients the anterior-posterior diameter of the spinal cord was significantly reduced. The considerable experience in treating intramedullary pathology in our center, together with the sophisticated and reliable intraoperative neurophysiologic monitoring with SEPs and MEPs, allowed this approach to be adopted with minimum risk to the patients. Using a D-wave electrode placed subdurally, we were also able to get continuous feedback on conduction in the motor pathways.

In our cases we used a midline split of the raphe in a thinned cord, which allowed the ventral cysts to be approached along a straight trajectory with minimal retraction and manipulation of cord tissue. The pial stitches applied as in conventional midline cord openings for tumors or intramedullary cysts exerted only a minimal retraction on the cord itself, much less so than if the stitches had been used to rotate and retract the spinal cord with lesser risk of exacerbating preexisting neurologic deficits.

In both cases there was no evidence of even a transient worsening in the neurophysiologic spinal cord monitoring. In fact, an improvement in both SEPs and MEPs followed the decompression of the spinal cord. The immediate postoperative imaging demonstrated cord reexpansion, which was associated with a rapid, significant, and permanent neurologic improvement. Twenty-four months following surgery, both patients maintained their neurologic recovery and repeat imaging showed no evidence of cyst regrowth.

One important risk associated with this approach is an inadvertent injury to the anterior spinal artery with consequent anterior cord infarction. In the first case the cyst was adherent to the ventral cord and the artery was not evident. In the second case careful dissection of the cord and ventral pia allowed the cyst cavity to be opened under direct vision, and we suggest that provided the surgeon is mindful of this anatomic relationship, the risk of inadvertent damage is minimal.

Injury to the anterior corticospinal tracts was avoided because these are not midline structures. As written earlier, we also monitored motor tract function throughout with MEPs and by using an intradural D-electrode, which provides continuous monitoring of the corticospinal tracts. The anterior pia was divided under direct vision (high magnification), and we were able to see and avoid the anterior spinal artery. Neither patient had a significant sensory deficit as a consequence of division of the decussating spinothalamic fibers. We attribute this to the short vertical length of the myelotomy and location of the lesions and because minor thoracic sensory deficits are unlikely to be noticed by the patients.

CONCLUSIONS

In conclusion, we have presented these cases because we believe that the use of a dorsal midline approach splitting the spinal cord longitudinally down the median raphe approach provides surgeons with another option in patients affected by ventrally located intradural cysts of the thoracic or cervicothoracic junction that cannot be accessed dorsally or dorsolaterally. In our hands, this technique has proven both safe and effective.

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