Dural Dissection Cyst: a More Accurate Term for Extradural Meningeal Cyst

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Keywords

Meningocele; Spinal cyst; Dural dissection; Extradural.

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Received 8 January 2014; revision 29 January 2014; accepted 11 February 2014

SUMMARY

Aims: Spinal extradural meningeal cyst (EMC) aetiology remains unclear. Based on our in-depth analysis of EMC clinical characters, we propose the alternative term 'dural dissection cyst' (DDC), which is more consistent with its aetiology and pathological manifestations.

Methods: We examined the clinical, imaging and operative findings of four patients with spinal EMC (type I A) and analysed the aetiology and treatment of the cyst. **Results:** Spinal DDC was observed between T10 and L3 in our series. Patients presented with low back pain, lower extremity numbness and weakness, and segmental muscle atrophy. Small clefts were found on the inner wall of all cysts. Microscopic suture of the cleft successfully improved patient's symptoms and neurological deficits. **Conclusions:** Spinal EMC (type I A) is characterized by dural dissection, so the term DDC can best reflect its aetiology. Because it is a dissection cyst, the most reasonable treatment is to suture the fistula.

doi: 10.1111/cns.12246

Introduction

Spinal extradural meningeal cyst (EMC) is a very rare condition [1]. Nabors et al. [2] divided spinal meningeal cysts into three categories: EMC without nerve root fibres (type I), EMC with nerve root fibres (type II) and intradural meningeal cyst (type III). EMCs can be intra- or extraspinal and mostly occur in the mid-lower thoracic spine [3] and compress the spinal cord anteriorly [4,5]. The cysts usually cause pain, weakness of the lower back and amyotrophy in lower extremities [6]. To date, EMC aetiology remains unclear, although most investigators consider it to be congenital. The arachnoid membrane herniates from the congenital defect of the dura and forms an epidural arachnoid cyst. Suspected reasons for cyst expansion include the secretory function of the cells in the inner layer of the cyst, the osmotic pressure gradient between the cyst and the subarachnoid space, the pulsation of the b (CSF) and the flapper effect of the orificium fistulae [5–9]. Here, we describe four patients with EMCs (type I A according to the classification of Nabors) treated by a single surgeon and an analysis of the clinical, radiographic, operative and pathological findings. Based on our in-depth analysis of EMC (type I A) clinical characters, we propose the alternative term 'dural dissection cyst' (DDC), which is more consistent with its aetiology and pathological manifestations.

Materials and methods

We retrospectively analysed the clinical symptoms, imaging and operative findings, and follow-up results of four patients.

Results

Case 1

A 52-year-old woman presented with low back pain and left lower extremity weakness that had persisted for about 2 years. Magnetic resonance imaging (MRI) of the lumbar spine identified an EMC. Laminectomy, cystectomy and cystoperitoneal shunt were subsequently performed but failed to relieve her symptoms. After the patient was referred to our institution, subsequent MRI revealed an EMC extending from T11 to L2 compressing the conus and cauda equina (Figure 1A). Lumbar lateral X-ray revealed a cystoperitoneal shunt tube at the L1 level (Figure 1B). The patient's left lower extremity muscle strength was grade 4 with bilateral hyperreflexia and positive Babinski sign.

During surgery, we eliminated the shunt and detected the thin semitranslucent inner wall of the cyst, through which we could observe the spinal cord and nerve root. A 10.0-mm dural cleft inferior to the left L1 root sleeve that drained CSF was then identi-

Figure 1 The imagings and intraoperative photograph of case 1. **(A)** Sagittal MRI after the first operation showing a spinal extradural meningeal cyst still existed extending from T11 to L2 compressing the conus and cauda equina. **(B)** Lumbar lateral X-ray revealing a cystoperitoneal shunt tube (hollow arrow) at the L1 level. **(C)** Intraoperative photograph demonstrated a 10.0-mm dural cleft (black arrow) inferior to the left L1 root sleeve was identified and used to drain the CSF, and the nerve root was adherent to the cleft. **(D)** Postoperative MRI demonstrating that the cyst completely disappeared, and spinal cord compression resolved.

fied, and the nerve root was adherent to the cleft (Figure 1C). We used 8-0 nylon to suture the cleft. Three months after surgery, the patient's symptoms had improved significantly. At the 11-month follow-up visit, MRI showed that the cyst had completely disappeared, and spinal cord compression had remitted (Figure 1D).

Case 2

A 27-year-old military officer who engaged in high-intensity training presented with low back pain and bilateral lower extremity weakness that he had been experiencing for the previous year. MRI revealed a left-oriented EMC extending from T11 to L1 that dilated the left foramen, squeezed the pedicle and compressed the conus and cauda equina (Figure 2A). He had undergone laminectomy and cystectomy 3 months before admission to our hospital, but improvement was minimal. MRI demonstrated that the EMC persisted (Figure 2B), and computed tomography (CT) myelography showed that the contrast filled the cyst diffusively (Figure 2C). He had decreased superficial sensation below the left T12 level and bilateral hyperreflexia of the lower extremities.

During surgery, we removed the residual T12 lamina and detected the thin inner wall of the cyst. A 5.0-mm dural cleft inferior to the left T12 root sleeve that leaked CSF was identified, and the nerve root was adherent to the cleft (Figure 2D). The cleft was fully repaired during microscopic surgery. The patient had an excellent recovery, and the 8-month follow-up MRI showed complete cyst resolution (Figure 2E).

Case 3

A 57-year-old woman complained of low back pain accompanied with bilateral lower extremity weakness for 2 years. MRI revealed a left-sided EMC extending from T11 to T12 and compressing the dural sac (Figure 3A). It extruded out of the left T11 foramen (Figure 3B). She had normal sensory function and muscle strength but exhibited bilateral lower extremity hyperreflexia.

Because the cyst wall was strongly adherent to the lamina, when we performed a left laminectomy of T11-12, the cyst wall was dissected with it and was sent for pathological examination. The inner wall of the cyst was thin. We found that the left T11 nerve root in the cyst had a very thin sleeve. A 4.0-mm cleft was

Figure 2 The imagings and intraoperative photograph of case 2. **(A)** Sagittal MRI before the first operation showing a spinal extradural meningeal cyst extending from T11 to L2. **(B)** Sagittal MRI after the first operation showing the EMC still existed compressing the conus and cauda equina. **(C)** In CT myelography, the contrast filled the cyst diffusively. **(D)** Intraoperative photograph demonstrated 5.0-mm cleft (black arrow) under the left T11 nerve root on the inner cyst wall. **(E)** Postoperative MRI showing complete resolution of the cyst.

identified under the left T11 nerve root on the inner wall of the cyst, and CSF flowed out from the cleft with nerve root protruding from the cleft (Figure 3C). The cleft was repaired with microscopic sutures. The patient had a satisfactory recovery, and 2-month follow-up MRI displayed complete resolution of the cyst and demonstrated that spinal cord compression had been relieved (Figure 3D).

Case 4

A 31-year-old woman with a 1-year history of lumbar strain presented with low back pain and bilateral lower extremity weakness. MRI revealed an EMC at T12-L1 causing vertebrae deformity (Figure 4A). CT myelography confirmed an EMC that was entrapping the nerve root (Figure 4B). On examination, she had decreased superficial sensation below the left T12 level, grade 4 muscle strength, tendon hyperreflexia of both lower extremities, and positive Babinski sign.

The left semilamina of T11-L1 was removed during surgery, and the wall of the cyst was noted to be thin. Partial cyst wall dissection was carried out, and a 10.0-mm cleft was found below the left T12 nerve root sleeve on the inner wall of the

cyst. CSF was determined to flow out from the cleft, with the nerve root protruding from it (Figure 4C). The cleft was repaired with 8-0 nylon suture, and the patient had a successful recovery. The 3-month follow-up MRI showed that the cyst had disappeared and the spinal cord shape had returned to normal (Figure 4D).

Discussion

Aetiology

Spinal EMCs are rare and are also referred to as 'spinal extradural arachnoid cysts', 'pouches', or 'diverticula', so the nature of the cyst lesion is not clear, and these definitions do not accurately reflect the underlying mechanism and provide no appropriate treatment methods [9–12]. The hypothesis proposed for cyst formation is that congenital or traumatic dural defects lead to arachnoid membrane herniation, which gradually dilates to form an extradural arachnoid cyst [13–15]. Suspected reasons for cyst expansion include osmotic pressure gradient between the cyst and the subarachnoid space, CSF pulsation and high hydrostatic pressure [16]. Some authors proposed that the flapper effect of the ori-

Figure 3 The imaging and intraoperative photograph of case 3. (A) Sagittal MRI demonstrated an EMC extending from T11 to T12. (B) Axial MRI showing that the cyst extruded out of the left T11 foramen. (C) Intraoperative photograph demonstrated a 4.0-mm cleft (black arrow) under the left T11 nerve root on the inner cyst wall. (D) Postoperative MRI confirmed complete elimination of the cyst.

Figure 4 The imaging and intraoperative photograph of case 4. (A) Sagittal MRI showed an EMC at T12-L1. (B) Axial CT myelography demonstrated an EMC entrapping the nerve root sleeve. (C) A 10.0-mm cleft below the left T12 nerve root sleeve was found during operation (black arrow). (D) The 3-month follow-up MRI confirmed that the cyst had disappeared.

ficiu fistulae as another possible reason; indeed, Rohrer observed such an effect during surgery [6].

In many case reports, the pathological findings of the resected cyst wall described both arachnoid membrane epithelial cells and compact fibrous connective tissue [17,18]. We also get the same pathological finding on the cyst wall. These pathological findings suggest that the outer wall of the cyst is not limited to the arachnoid membrane but also contains dural components; hence, the cyst lesions located inside the dura, so the reasonable explanation of this findings is that the nature of the cyst is a kind DDC. Saito described the dura had two layers, the inner and the outer layer, which can be easily separated, in his spinal meningioma surgeries, and there was a potential plane between the two layers [19]. So it is possible that dissection cysts form between the two layers of the dura.

The intraoperative findings confirmed our speculation of the character of the cyst. In our four patients, we found that the outer layer of the cyst wall merged together with the inner layer at the rostral and the caudal ends of the cyst. There is no inner layer of the cyst wall which can be separated and removed from the dura, the dura which become much thinner than normal composed the inner layer of the cyst wall. The inner layer of the cyst wall migrated to the inner layer of the nerve root sleeve on the surface of nerve root. By analysing cyst morphology, we confirmed that the cysts were DDCs; the inner layer of the cyst wall was made up of the inner layer of the dura, and the outer layer of cyst was the outer layer of the dura. Single longitudinal clefts of different lengths were found on the inner cyst walls in all four cases, and all clefts were located under the nerve root sleeve. The cleft connected the

DDCs with the subarachnoid space, and CSF drained into the cyst through the cleft.

A possible mechanism for DDC formation is that some congenital defect or trauma could break a small cleft on the inner layer of the dura, but leave the outer dural layer intact. There is a potential plane between the inner and outer dural layers, when the inner layer is broken but the outer layer is intact, CSF can fill into the potential space, and the hydrostatic pressure and pulsation of the CSF can enlarge the space to form a DDC (Figure 5). The formation mechanism is similar to that of artery wall dissection, but the impact force of CSF is much smaller than that of artery blood flow. For this reason, DDC formation is slower than artery wall dissection, and symptoms develop slowly. Cleft formation on the inner dural layer may be related to lumbar spine trauma. One patient in our series had a clear lumbar sprain history, and another patient was a military officer, who had lot of lumbar activities due to his training. Histopathologic analysis demonstrated that the cyst outer wall was composed of dense fibrous tissue (Figure 6), similar in appearance to the dura. This result supports the hypothesis that the cyst is intradural but not outside the dura. Based on our findings, we suggest that the term DDC more accurately reflects the nature and formation mechanism of EMC (type I A).

Treatment

Due to the unclear nature of these cystic lesions, treatment is controversial. Some articles have reported that total or partial resection of the cyst wall or cyst shunt is an effective treatment [20]. In the series described here, laminectomy, cystectomy and cystoperitoneal shunt were performed on case 1 but without any benefit. In case 2, the patient underwent cystectomy, but the surgeons who performed the first surgery did not detect and close the cleft, and the cyst did not changed after operation. Both patients underwent further surgery, during which we identified and repaired the

Figure 5 The schema graph of the formation of dural dissection cyst (DDC). (A) The arachnoid membrane and inner layer of the dura is torn, but the outer layer is intact, (SC, spinal cord; NR, nerve root; ID, inner layer of dura; OD, outer layer of dura; AM, arachnoid membrane). (B) The pulsatile and hydrodynamic force of CSF infiltrate and expand the potential space between the inner and outer dural layers. (C) Eventually, this influx of CSF expands the cyst to form a DDC.

Figure 6 Histopathology showing that the outer cyst wall is composed of dense fibrous tissue.

clefts. It thus appears that microscopic suturing of the clefts is the most crucial step in treating DDC. That is, treatment of DDC is similar to that employed to correct artery wall dissection; the goal is close the fistula.

As DDCs tend to involve multiple spine segments and can induce vertebral deformation in susceptible thoracolumbar segments, we do not recommend multiple-level laminectomy because spinal stability should be considered as a priority. We have reviewed some related literature that report that there is usually only one cleft; if its location can be identified, a limited laminectomy and microscopic suturing are possible. Ball *et al.* [21] proposed dynamic CT myelography as a useful modality for locating the cleft of ventral spinal intradural dissecting meningoceles, and they did so successfully in three of four described cases. However, detailed techniques, such as the consecutive times and interval of CT scans, were not described and should be studied in future research. Similarly, Neo *et al.* [9] used cine MRI to detect a DDC cleft in one patient. These reports illuminate the possibility of minimally invasive treatment for DDC, which is especially significant in cases where multiple levels are involved.

In our observation, all four DDC cases occurred in the thoracolumbar junction, among which, two cases involved five vertebral segments, one case involved four, and one case involved three. Because all the DDCs were mainly in the left side of the spinal canal and showed more obvious left nerve root foramen expansion, we suspect that the cleft was on the left side of the DDC. During surgery, we detected clefts on the left-middle parts of the DDCs. One was identified under the left T12 root sleeve, and the other three were under the root sleeves of L1. For the 3rd and 4th cases, a semilamina resection technique was applied during surgery. The spinal process and lamina on the contralateral side were intact, so spinal stability was not significantly affected. This surgical strategy could address the limited accuracy of imaging performed to identify the cleft and reduce spinal destabilization. As to why the cleft tends to occur in the thoracolumbar junction, we hypothesize that the larger range of motion in this area may tear the inner dural layer under the nerve root sleeve or that this area is vulnerable to congenital dural hypoplasia.

Conclusion

Based on intraoperative observations and imaging analyses, we propose that the mechanism of EMC (type I A) formation involves inner dural wall breakage caused by trauma or congenital defect, and it is enlarged by the pulsatile and hydrodynamic CSF forces. We propose that EMC (type I A) should be termed DDCs, which is more descriptive of their pathological mechanism and subsequent surgical strategy. The identification and microscopic suturing of the cleft are the most crucial steps in treating DDCs.

Acknowledgments

We thank Li-Yan Wang for information collection of patients. We also thank Yan Liu for revising the article.

Conflict of Interest

The authors declare no conflict of interest.

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