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Case Report

Spinal Extradural Arachnoid Cyst: Significance of Intrathecal Infusion after Fistula Closure

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The spinal extradural arachnoid cyst is a rare entity. Obtaining the correct diagnosis and detecting the fistula location are critical for providing effective treatment. A 41-year-old man had numbness in the soles of his feet for 2 years with accompanying gait disturbance, and a defecation disorder. Computed tomography myelography performed at another hospital revealed an epidural arachnoid cyst from Th11 to L2. He received a sub-arachnoid-cyst shunt at the rostral part of the cyst. However, his symptoms worsened and he was admitted to our hospital. Neuroradiological investigations revealed the correct location of the fistula at the level of Th12. We performed partial removal of the cyst wall with fistula closure via right hemilaminectomy of Th11 and 12. The complete closure of the fistula was confirmed by intrathecal infusion of artificial cerebrospinal fluid through the shunt tube. The shunt tube was removed with the sutures. The patient's symptoms improved, although numbness remained in his bilateral heels. There has been no recurrence in 15 months since the surgery. Fistula closure may work as a balanced therapeutic strategy for spinal extradural arachnoid cyst, and intrathecal cerebrospinal fluid infusion is useful for the confirmation of complete fistula closure.

Key words: fistula closure, intrathecal infusion, microscopic surgery, preoperative evaluation, recurrence

S pinal extradural arachnoid cyst (SEAC) is a rare entity that causes various symptoms [1] such as motor deficits, impaired sensation, and bladder and bowel dysfunction, although some patients with SEAC have no symptoms and require no treatment. There are several articles on how SEAC should be treated [2-6]. The therapeutic strategy consists of cyst shunting, cyst wall fenestration, cyst wall removal and fistula closure [2]. Here, we report the case of a SEAC patient whose perioperative images were informative for the diagnosis and treatment of SEAC. Fistula closure might be a balanced treatment that can achieve a complete cure with

relatively little invasiveness. Furthermore, the intrathecal infusion of artificial cerebrospinal fluid (CSF) after fistula closure has been shown to confirm that there is no unclosed fistula.

Case Presentation

A 41-year-old man had numbness in the soles of his feet for 2 years with accompanying gait disturbance, and a defecation disorder. He had no history of trauma, lumbar puncture, surgery, or spinal infectious disease. At another hospital, magnetic resonance (MR) images (Fig. 1A) and computed tomography (CT) myelography

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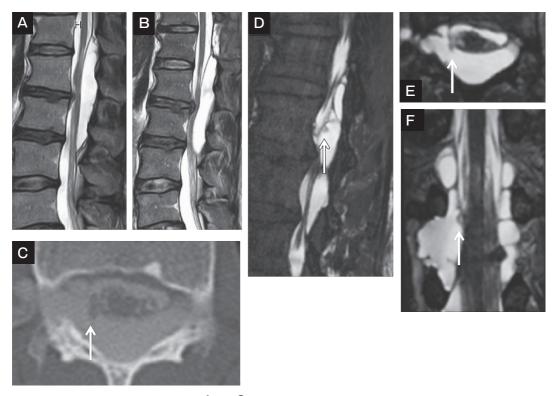


Fig. 1 Preoperative neuroradiological evaluation. A and C, Magnetic resonance (MR) images before and after the insertion of the subarachnoid cyst shunt showing the spinal extradural arachnoid cyst compressing the spinal cord; B, CT myelography showing the cauda equina protruding into the cyst (white arrow); D-F, Sagittal, axial and coronal CISS images revealing the fistula location (white arrow). T2-weighted MR images showing arachnoid cyst from Th11-L2.

(Fig. 1B) were used to diagnose thoracolumbar SEAC from the level of Th11 to L2. He received a subarachnoid-cyst shunt at the rostral part of the cyst. However, his symptoms worsened with no neuroradiological improvement. Three months after surgery, he was admitted to our hospital. MR images on admission revealed an unchanged cystic legion with cord compression (Fig. 1C). It was predicted that the fistula from the intradural subarachnoid space to the SEAC was located in the dorsolateral part of the dural sac at the level of Th12 (Figs. 1D-F). The herniated cauda equina was identified using CT myelography and constructive interference in steady state (CISS) MR images (Figs. 1B, E). Partial removal of the cyst wall with fistula closure was planned via right hemilaminectomy of Th11 and 12. High intracystic pressure was revealed by the leakage of CSF at the cyst wall incision (Fig. 2A). A 5-mm-length fistula was found, as expected (Fig. 2B). The cyst wall around the fistula was removed, and the dural defect was closed using a vascular closure staple clip (LeMaitre Vascular GK, Tokyo, Japan). The complete closure of the fistula was confirmed by the intrathecal infusion of artificial CSF (ARTCEREB, Otsuka Pharmaceutical Factory, Tokushima, Japan) through the shunt tube (Fig. 2C). The closed fistula was covered using Neoveil (Gunze, Kyoto, Japan) and fibrin glue. The shunt tube was removed with the sutures to prevent CSF leakage. The patient's symptoms improved after surgery, with MRI showing that the SEAC had disappeared (Figs. 3A, C) through a small fenestration of the lamina (Fig. 3B), although numbness remained in his bilateral heels. Thirteen months after the removal of the SEAC, the symptoms have not recurred, and the patient shows unchanged neuroradiological amelioration (Fig. 3D).

Discussion

A previous report of several SEAC cases hypothesized that there was likely a one-way valve mechanism underlying the condition [7]. SEAC dural defect is usually found in idiopathic cases, such as in our patient,

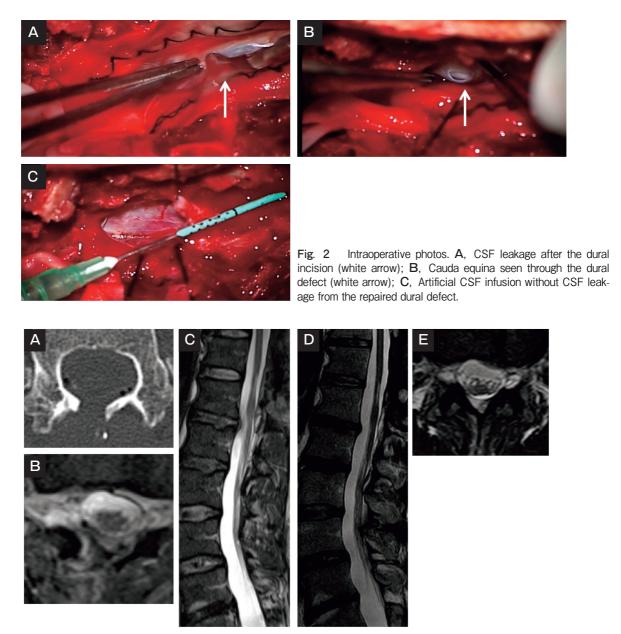


Fig. 3 Postoperative neuroradiological images. A, C, Postoperative MR images taken at 1 week (A and C) after surgery showing disappearance of the extradural arachnoid cyst; B, CT image taken 1 day after surgery showing the small fenestration in the lamina; D, Postoperative MR images taken 8 months after surgery showing unchanged decompressed cauda equina.

although secondary cases exist following trauma, infection, or lumbar puncture [8]. Several surgical options have been proposed, including cyst wall fenestration, cyst wall removal, and a shunt between the cyst and the pleural, peritoneal, and subarachnoid spaces [9,10]. However, the most reasonable and balanced surgical treatment might be fistula closure, as it is less invasive than these other options and shows high curability. In patients undergoing cyst shunting or cyst wall fenestration, SEAC might recur through occlusion of the shunt or fenestration through arachnoid adhesion. Additionally, to achieve efficient therapeutic effects, the outflow through the shunt or fenestration should be greater than the inflow through the original fistula. The surgical results of 12 patients with SEAC treated at a single institute showed that fistula closure was not supe-

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rior in terms of postoperative neurological recovery [2]. However, fistula closure is less invasive with less kyphotic deformity than cyst removal. To prevent kyphotic deformity, cyst removal with laminoplasty was performed in a recent case series, although fistula closure was suggested as the main surgical goal in that report [6]. There are several reports of patients with multiple SEACs [11] in whom the closure of each fistula was required. In our patient, intrathecal infusion of artificial CSF through the previously placed shunt tube showed no CSF leakage after fistula closure, indicating that our patient had a single SEAC. We performed fistula closure with partial cyst wall removal via right hemilaminectomy, and kept the facet intact. To achieve this using a small incision, it is critical to identify the fistula location. Similarly, a minimal skipped hemilaminectomy was used to achieve cyst wall removal and fistula closure [12]. To detect the fistula location, CT myelography [7], cine MRI [13], and MR myelography [14] were reported to be useful. In our patient, the correct location of the fistula was confirmed by previous CT myelography performed at another hospital and by CISS images taken from 3 directions at our hospital. The preoperative information corresponded to the intraoperative findings.

Conclusions

In conclusion, fistula closure might function as a balanced therapeutic strategy for SEAC with relatively little invasiveness and high curability. Intrathecal infusion of artificial CSF can help to confirm complete fistula closure.

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