



Original Article

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INTRODUCTION

Spinal epidural meningeal cysts (SEMCs) are rare pathological entities that account for about 1% of epidural lesions.¹ Most of the literature comprises case reports. The etiology, pathogenesis, and treatment of the disease remain controversial. Although cystectomy, shunting, and marsupialization are common surgical procedures that have been successfully reported,²⁻⁵

most scholars believe that fistula closure is key in preventing recurrence.⁶⁻⁹

The difficulty in treating this disease is the exploration of the fistula. However, at present, there are limited studies on the location distribution of fistulas with a small sample size.¹⁰⁻¹² Based on our previous studies,^{7,11} we retrospectively analyzed the data of 30 patients with SEMCs at a single center to discuss the features of fistula location distribution and the surgical strategy for

Location Distribution of Fistulas and Surgical Strategies for Spinal Extradural Meningeal Cysts: A Retrospective Analysis of 30 Cases at a Single Center

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Objective: This study aimed to illustrate the features of fistula location distribution, surgical strategies, and outcomes in spinal epidural meningeal cysts (SEMCs).

Methods: The authors searched and reviewed the medical records for cases of SEMCs. Imaging features, operative reports, and media were reviewed to accurately describe the surgical techniques employed. We recorded the level and laterality of the fistula according to the operative report and the media. Consistency analysis was performed on the dominant laterality of the cyst on preoperative axial magnetic resonance imaging and laterality of the fistula in the operative media or report. When cyst and fistula lateralities were the same, they were considered consistent. Finally, the Japanese Orthopedic Association (JOA) score was used to obtain patient-reported results at each follow-up.

Results: Thirty patients with SEMCs were identified. Fistula repair was performed in all patients. Two patients experienced cyst recurrence after surgery and were repaired during the second surgery. Based on imaging findings, SEMCs mostly occurred in the thoracolumbar junction. Most of the fistulas (87.88%) were at the T12 or L1 levels. In patients with multiple adjacent SEMCs, the fistula may be at one end of the cyst rather than in the middle level of the cyst. A fistula laterality of 72.72% was consistent with cyst laterality. The JOA improvement rate was $61.84\% \pm 26.63\%$.

Conclusion: Most fistulas were always located at the T12–L1 level as well as the middle level of the cyst, which is always consistent with cyst laterality. In patients with multiple adjacent SEMCs, the fistula may be at one end of the cyst. Cleft closure is key to healing SEMCs.

Keywords: Spinal extradural meningeal cyst, Surgical strategy, Spinal dural dissection cyst, Spinal extradural arachnoid cyst, Hemilaminectomy, Fistula location

this disease, which, to our knowledge, is the largest sample size of SEMCs ever studied at a single center.

MATERIALS AND METHODS

1. Patient Population

The nomenclature and pathogenesis of the disease are controversial. Early on, we considered Nabors' type I cyst (SEMC without nerve root fibers) as a type of spinal dural dissection cyst (SDDC).⁷ At a later stage, we suggested that SEMC also forms as a result of arachnoid herniation through the dural defect in the epidural space, namely, spinal extradural arachnoid cysts (SEACs). Thus, in this study, the term SEMCs is divided into SDDCs and SEACs. For fear of omission, we searched the medical records of all patients diagnosed with SEMCs, SEACs, spinal dural cyst, or SDDCs in the 10-year period from January 2011 to January 2021 at Xuanwu Hospital. All patients diagnosed with SEMCs, SEACs, SDDCs, or spinal dural cysts were included, including several cases we previously reported.^{7,11} We screened the surgical cases that the authors participated in and excluded other cystic disorders of the spine, such as intradural cysts, intramedullary cysts, and cystic tumors.

There were 12 men and 18 women with an average age of 39 years (range, 7–80 years) enrolled in the study. Twenty-five patients underwent the initial surgery at our center. Five patients underwent revision surgery at our department after magnetic resonance imaging (MRI) indicated cyst recurrence outside the hospital. The details were as follows: A patient achieved relief of symptoms after a cystoperitoneal shunt was performed at the local hospital. However, he had undergone peritoneal end dredging of the shunt tube 8 times due to repeated shunt obstruction. Three patients underwent cystectomy at a local hospital. One patient underwent cystectomy and peritoneal cyst shunt at a local hospital, but the symptoms did not improve significantly after surgery (Table 1).

2. Preoperative Imaging Examination and Diagnosis

MRI plays an important role in the diagnosis and differentiation of lesions. The diagnosis of SEMC with low intensity on T1-weighted imaging and high intensity on T2-weighted imaging is consistent with the cerebrospinal fluid (CSF) signal and no enhancement in the lesion. Hyperintense-signal epidural fat on the both cranial and caudal sides of the hypointense-signal SEMC on sagittal T1-weighted imaging indicates the epidural location.² On T2-weighted imaging, the cyst walls can sometimes be seen at its superior and inferior poles. The curvature

of the normal dura above the cyst continues with the curvature of the ventral wall of the cyst, where the subarachnoid space gradually narrows, which further indicates that the cyst is extradural. Preoperative T2-weighted MRI showed that the flow void of the CSF could be used to locate the fistula level, and the size of the left or right laterality of the cyst on the axial MRI was significant for the fistula location. However, subdural cysts are mostly arachnoid or enterogenous cysts of spinal origin, which generally do not invade the dura. Thus, the dural wall is generally not deformed on MRI.

Preoperative computed tomography (CT) and x-rays were used to examine the instability and deformity of the spine, pedicle thinning, and previous implants.

The purpose of myelography, CT myelography (CTM), and real-time digital subtraction myelography is to locate the fistula, to identify the cyst lesion, and to identify whether the cyst communicates with the subarachnoid space, and to determine whether adjacent cysts communicate with each other. The SEMC has a fistula on the ventral wall, and the cyst is often visualized by delayed CTM, whereas a subdural lesion, such as an enterogenous cyst, has no communication with the subarachnoid space. Since myelography does not allow contrast to enter the cyst, the myelography findings show a filling defect in the spinal canal.

3. Operation

Posterior hemilaminectomy and fistula repair were performed in most patients during the first surgery (revision surgery was not included). As most of the cysts were in the thoracolumbar segment, the T12/L1 segmental approach was performed for most patients based on our experience. For others, the approach is usually performed at the middle level of the cysts. Single-segmental semilaminectomy was performed to expose the cyst according to the dominant laterality of the cyst on axial MRI. We released the CSF by cutting the dorsal cyst wall and searching for the cleft of the ventral layer (Fig. 1A). The nerve would be returned to the subarachnoid space if it were incarcerated at the fistula. Finally, the fistula was continuously sutured with 8-0 Prolene suture (Johnson & Johnson, New Brunswick, NJ, USA) or combined with muscle mud and fibrin sealant (Fig. 1B). If no fistula was found, hemilaminectomy was performed at an adjacent level or with extended bilateral hemilaminectomy or total laminectomy. After suturing, the patient's airway pressure was increased to confirm the watertight suture. A small portion of the dorsal wall of the cyst was carefully removed for pathological examination.

Table 1. Data of 30 patients with spinal epidural meningeal cysts

Case No.	Sex/age (yr)	Clinical presentation			Imaging data			Operation data			Follow-up duration (mo)	Preoperative JOA up	Final follow-up JOA	Complication			
		Operation history	Symptom duration (mo)	Symptoms (at surgery)	Symptoms (final)	Flow void	Cyst span	Contrast examination outcome	Cyst laterality	Cleft level					Nerve root incision	Resected lamina	
1	F/26	None	120	Low back and right lower limb pain	Low back pain	None	T12-L1	-	Right	Right	L1	None	Right T12-L1 hemilaminectomy	19	26	27	None
2	M/39	None	240	Bladder dysfunction	Bladder dysfunction	None	T12-L2	-	Equilibrium	Left	L1	None	Right T12-L1, Left T12 hemilaminectomy	24	23	26	None
3	M/20	None	12	Low back pain, numbness of bilateral lower limbs, bowel and bladder dysfunction	Low back pain	None	T7-L2	Cyst recurrence	Equilibrium	Right	T11-T12	None	Left T11-L1, Right T11-T12 hemilaminectomy	9	23	23	Early cyst recurrence, nervous system infection
4	F/43	None	12	Low back and bilateral lower limbs pain	Low back pain	None	T11-L1	-	Left	Left	T12	None	Left T11-L1 hemilaminectomy	16	26	28	None
5	M/7	None	0.2	Weakness of lower extremities, dysuria	Low back pain	L1	T11-L4	-	Left	Left	L1	Yes	Left T12-L1 hemilaminectomy	16	18	28	None
6	M/19	Spinal cord detethering, cystoperitoneal shunt	84	Bowel and bladder dysfunction, hypoaesthesia of lateral lower extremities	None	T12	T9-S1	-	Right	Right	T12	None	Right T12-L1 hemilaminectomy	43	23	29	None
7	M/54	Cystectomy	216	Weakness and numbness of lower limbs, gait disturbance, bowel and bladder dysfunction	Loss to follow-up	None	T12-L2	-	Right	Right	L1	None	Revision surgery	Loss to follow-up	19	-	Loss to follow-up
8	F/11	None	60	Low back and lower limbs pain, gait disturbance, bowel and bladder dysfunction	None	None	T6-10, T11-L2, L2-4	No communication between cysts	Left	Left	T9, T12, L2	None	Left T6-9, T12-L1, L2-3 hemilaminectomy	42	24	29	Cyst recurrence

(continued)

Table 1. Data of 30 patients with spinal epidural meningeal cysts (continued)

Case No.	Sex/age (yr)	Clinical presentation			Imaging data			Operation data			Follow-up duration (mo)	Preoperative JOA	Final follow-up JOA	Complication			
		Operation history	Symptom duration (mo)	Symptoms (at surgery)	Symptoms (final)	Flow void	Cyst span	Contrast examination	Cyst laterality	Cleft level					Cleft laterality	Nerve root incision	Resected lamina
9	F/33	None	36	Low back pain, weakness of both limbs	Weakness of both limbs	T12	T11-L2	-	Left	Left	L1	Yes	Left L1-L2 hemilaminectomy	25	26	28	None
10	M/16	None	6	Low back pain	Low back pain	None	T10-L1, L1-L3	-	Equilibrium	Right	T11, L1	None	Right T11-L3 hemilaminectomy	48	25	28	None
11	M/28	Cystectomy	12	Weakness and atrophy of lower extremities	Weakness and atrophy of lower extremities	None	T11-L3	No fistula was found	Left	Left	T12	None	Revision surgery	8	23	25	None
12	F/57	None	36	Low back and right lower limb pain, left lower limb numbness	Left lower limb pain, left lower limb numbness	T11	T10-T12	-	Left	Left	T11	None	Left T11-T12 hemilaminectomy	3	25	27	None
13	F/32	None	18	Low back pain, bowel and bladder dysfunction	Low back pain	None	T11-L1	No fistula was found	Left	Left	T12	Yes	Left T11-T12 hemilaminectomy	3	25	27	None
14	F/55	Cystectomy, cystoperitoneal shunt	24	Low back pain, left lower extremity weakness	Loss to follow-up	None	T11-L2	-	Left	Left	L1	None	Revision surgery	Loss to follow-up	23	-	Loss to follow-up
15	F/80	None	120	Low back and bilateral lower limbs pain	Low back pain	None	T12-L2	-	Right	Right	L1	None	Right T12-L1 hemilaminectomy	16	20	26	None
16	M/55	None	60	Left lower limb weakness	Left lower limb weakness	None	T8-L2	No fistula was found	Left	Left	T12	None	T11-T12 laminectomy, fixation	18	24	25	Progression of kyphosis
17	M/61	None	1	Low back and right lower limb pain	None	None	L1-2	-	Right	Right	L1	None	Right L1 hemilaminectomy	14	25	27	None

(continued)

Table 1. Data of 30 patients with spinal epidural meningeal cysts (continued)

Case No.	Sex/age (yr)	Clinical presentation			Imaging data			Operation data			Follow-up duration (mo)	Preoperative JOA	Final follow-up JOA	Complication		
		Operation history	Symptom duration (mo)	Symptoms (at surgery)	Syptoms (final)	Flow void	Cyst span	Contrast examination	Cyst laterality	Cleft level					Cleft laterality	Nerve root incision
18	F/34	None	96	Low back pain	None	T12-L3	-	Right	Right	T12	None	Right T12-L1 hemilaminectomy	8	27	29	None
19	F/30	None	36	Low back pain and right lower limb numbness	Low back pain	T12-L3	-	Right	Right	L1	Yes	Right L1 hemilaminectomy	5	25	27	None
20	F/22	None	0.5	Low back and bilateral lower limbs pain	Low back pain	T12-L2	-	Left	Right	L1	None	Left T12-L2, Right L1 hemilaminectomy	2	26	28	None
21	F/35	None	36	Low back and bilateral lower limbs pain	Low back pain	T11-L3	No fistula was found	Right	Right	L1	None	Right L1-L2 hemilaminectomy	2	25	28	None
22	M/46	None	1	Low back pain	Low back pain	T12-L2	No fistula was found	Left	Left	L1	Yes	Left T12-L2 hemilaminectomy	20	24	26	None
23	M/52	None	50	Low back and left lower limb pain	Loss to follow-up	T11-L1	-	Left	Left	T12	Yes	Left T11-T12 hemilaminectomy	Loss to follow-up	27	-	Loss to follow-up
24	F/39	None	120	Back and left lower extremity pain	Loss to follow-up	T11-L3	-	Left	Left	L1	Yes	Left L1 hemilaminectomy	Loss to follow-up	23	-	Loss to follow-up
25	F/48	None	5	Low back pain	Loss to follow-up	T11-L2	-	Left	Left	T12	Yes	Left T12-L1 hemilaminectomy	Loss to follow-up	26	-	No Loss to follow-up
26	M/43	None	5	Back pain	Loss to follow-up	T12	-	Left	Left	T12	None	Left T12 hemilaminectomy	Loss to follow-up	25	-	Loss to follow-up
27	F/64	Cystectomy	156	Low back pain and bilateral lower limb weakness, Right lower limb atrophy	Loss to follow-up	T11-L1	-	Equilibrium	Left	T12	None	Revision surgery	Loss to follow-up	24	-	Loss to follow-up

(continued)

Table 1. Data of 30 patients with spinal epidural meningeal cysts (continued)

Case No.	Sex/age (yr)	Clinical presentation			Imaging data			Operation data				Final follow-up JOA	Complication				
		Operation history	Symptom duration (mo)	Symptoms (at surgery)	Symptoms (final)	Flow void	Cyst span	Contrast examination outcome	Cyst laterality	Cleft laterality	Cleft level			Nerve root incarceration	Resected lamina	Follow-up duration (mo)	Preoperative JOA
28	F/47	None	6	Weakness of lower extremities, Low back pain	Low back pain	T12	T10-L2	-	Left	Right	T12	Yes	T12 bilateral hemilaminectomy	23	23	27	None
29	F/29	None	5	Right lower limb numbness	None	None	T12-L3	-	Right	Right	L1	Yes	T12-L1 hemilaminectomy	19	27	29	None
30	F/62	None	72	Low back and bilateral lower limb pain	Low back pain	None	T11-L2	-	Right	Right	T12	None	Right T12 hemilaminectomy	21	22	27	None

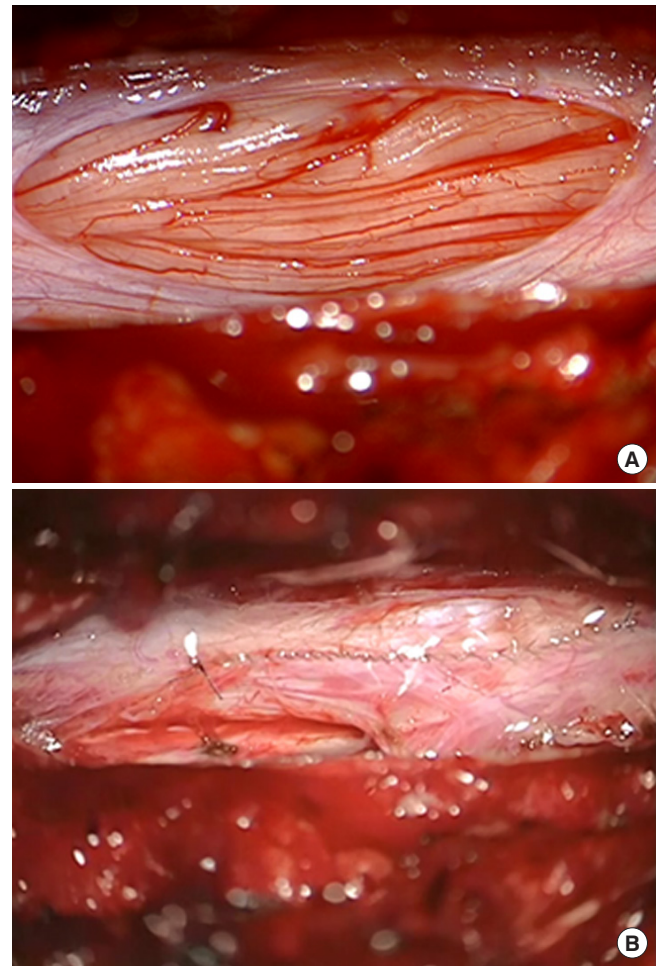


Fig. 1. Intraoperative views. (A) The fistula of patient No. 6 on the ventral wall of the cyst at T12 level was 20 mm in length and 10 mm in width. (B) The fistula of patient No. 6 was sutured with 8-0 Prolene suture.

4. Study Parameters

The clinical presentations, radiographic data, surgical reports, and pathological reports of the patients were recorded. The radiographic data included cyst quantity, cyst span, flow void level on sagittal MRI, cyst dominant laterality on axial MRI, and contrast examination outcome. According to the cyst dominant laterality on axial MRI, the SEMCs were divided into left, right, and equilibrium. The surgical data included the location of the resected lamina (revision surgery was not included), the level and laterality of the fistula, and the nerve root incarceration. The Japanese Orthopedic Association (JOA) score was used to assess neurological improvement, and the JOA improvement rate was calculated as follows: JOA improvement rate = (JOA at last follow-up – preoperative JOA) / (29 – preoperative JOA) × 100%.

5. Statistics

Statistical analyses were performed using IBM SPSS Statistics ver. 22.0 (IBM Co., Armonk, NY, USA). The parameters with normal distribution were expressed as the mean ± standard deviation. The median (interquartile range) was used to express the non-normally distributed data. Consistency analysis was performed on the dominant laterality of the cyst on preoperative axial MRI and laterality of the fistula in the operative media or report. When cyst and fistula lateralities were the same, they were considered consistent.

Table 2. Preoperative clinical symptoms of the 30 patients

Clinical symptoms	No. (%)
Motor disorders	
Lower extremity weakness	8 (26.7)
Lower extremity atrophy	2 (6.7)
Gait disturbance	2 (6.7)
Sensory disorders	
Low back pain	20 (66.7)
Backache	2 (6.7)
Lower extremity pain	11 (36.7)
Lower extremity numbness	5 (16.7)
Urination and defecation disturbance	
Bladder disturbance	7 (23.3)
Bowel disturbance	5 (16.7)

RESULTS

1. Clinical Symptoms

Clinical symptoms of the patients are presented in Table 2. The 3 most common symptoms were low back pain (66.7%), lower extremity pain (36.7%), and lower extremity weakness (26.7%).

2. Imaging Outcome

1) MRI

In the axial view, the cyst is usually located posterolaterally, compressing the spinal cord. Only 4 cysts were located directly behind the spinal cord and were classified as equilibrated cysts. Unilateral dominance on the axial view was seen in the others (Table 3). In 3 cases, the cyst completely surrounded the lateral side of the spinal cord, eroding the posterior margin of the vertebral body.

In the sagittal view, the incidence of 2 or 3 cysts was the same at 3.33% (1 of 30), and the number of cysts was also confirmed

Table 3. The cyst and cleft laterality

Cleft laterality	Cyst laterality			Total
	Right	Equilibrium	Left	
Right	10	2	2	14
Left	0	2	14	16
Total	10	4	16	30

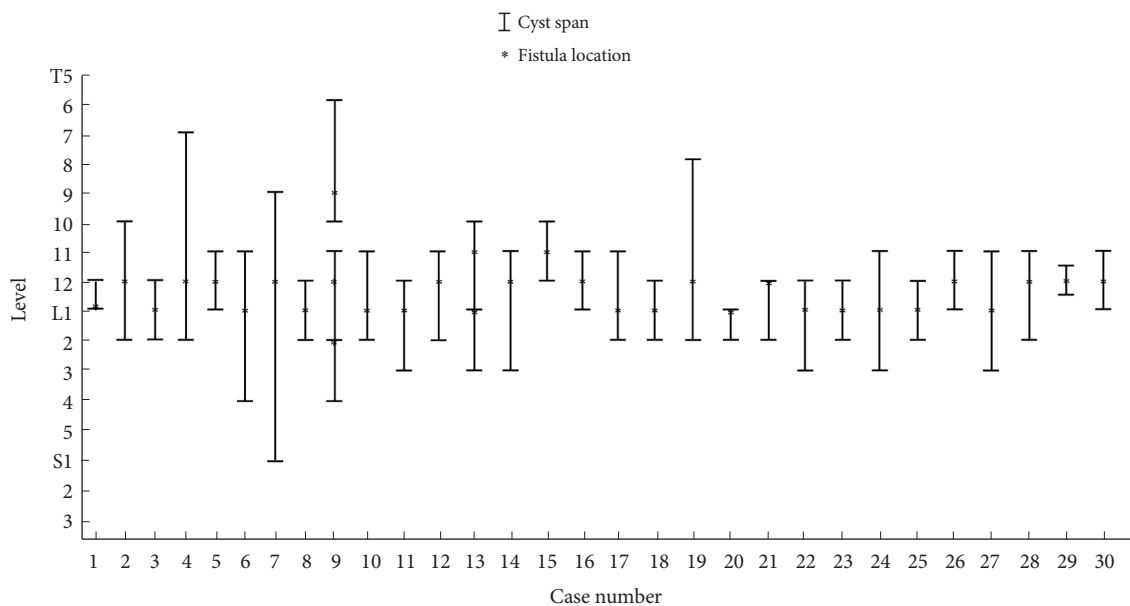


Fig. 2. Cyst span and fistula location.

intraoperatively. Patients with single cyst was common, accounting for 93.33% (28 of 30) of all patients. Most of the cysts occurred in the thoracolumbar region (Fig. 2), 3.03% (1 of 33) of the cysts were located in the thoracic canal, 6.61% (2 of 33) in the lumbar canal, and 3.03% (1 of 33) involved the sacral canal. The median (interquartile range) cyst span was 4 (3–5).

In 18.18% (6 of 33) of the cysts, CSF flow voids were observed on preoperative MRI (Table 1).

2) Radiological examination

Two patients had kyphosis preoperatively.

3) Contrast examination

Seven patients underwent contrast examinations. The examinations did not reveal any clefts. One case of recurrent cyst was confirmed by myelography. No communication between the 2 adjacent cysts was identified in 1 case.

3. Surgical Outcomes

Fistula repair was performed in all patients. The dorsal wall of the cyst was excised in 1 patient. Two patients had cyst recurrence after surgery and were sutured under the microscope during the second operation. Recurrence in 1 patient was due to slack suturing, and cyst recurrence was confirmed by myelography 20 days after surgery inside our department. The other was a patient with multiple cysts and fistulas, one of which was missed during the primary operation. One patient developed postoperative nervous system infection.

Most of the fistula levels were T12 or L1, accounting for 87.88% (29 of 33) of cases, followed by T11 (6.06%, 2 of 33) (Fig. 2). Nerve root incarceration occurred in 27.28% (9 of 33) of the fistulas. A total of 72.72% (24 of 33) of the fistulas were consistent with cyst laterality (Table 3).

Two patients had kyphosis preoperatively and were treated with internal fixation to prevent the progression of kyphosis. Hemilaminectomy was performed in 25 patients, which were extended to contralateral hemilaminectomy in 4 patients. The median (quartile range) of the semilamina (resected a half) was 2 (2–3).

4. Pathological Findings

We performed pathological examination on 11 patients, which showed fibrous tissue. Among them, arachnoid epithelial cells were found in 2 cases, and dense connective tissue was clearly indicated in 5 cases. Hyaline degeneration was identified in 3 cases, focal calcification was found in 2 cases, and ossification in 2 cases. One patient underwent immunohistochemistry: GFAP (-), CD31 (blood vessel +), CD34 (blood vessel +), D2-40 (blood vessel +), vimentin (+), cytokeratin (-), and epithelial membrane antigen (+).

5. Follow-up

Seven patients were lost to follow-up. The remaining patients were followed up for 16 months (8–23 months), and their symptoms improved significantly during the follow-up period, with a mean JOA improvement rate of $61.84\% \pm 26.63\%$.

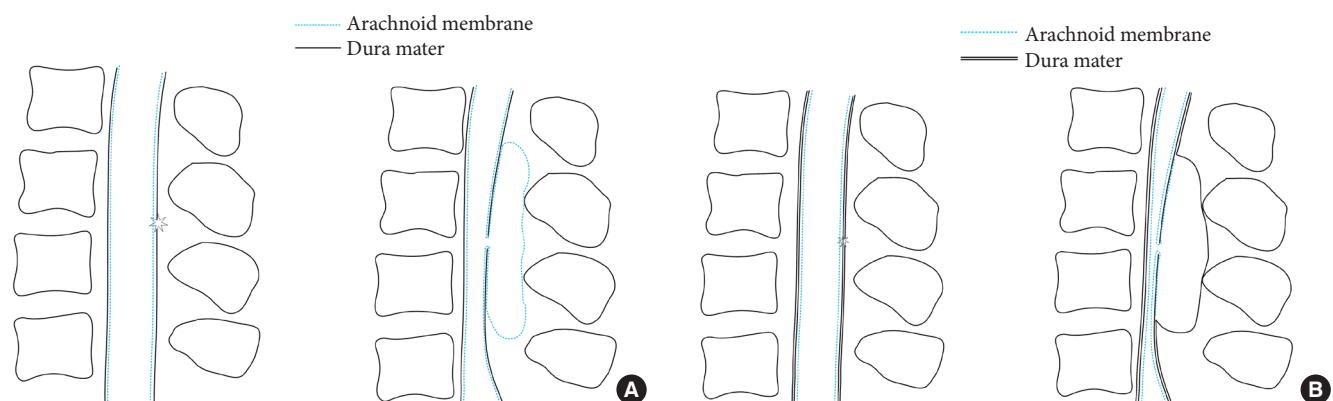


Fig. 3. Illustration of 2 types of SEMC: SEAC and SDDC. (A) An SEAC is caused by herniation of the arachnoid space either due to congenital reasons or trauma-induced defect involving both dural layers. (B) SDDC is formed due to a defect in the inner layer of the dura mater, and the potential gap between the inner and outer layers of the dura mater is affected by the fluctuation of CSF, resulting in the DDC between the inner and outer layers of the dura mater. SEMC, spinal extradural meningeal cyst; SEAC, spinal extradural arachnoid cyst; SDDC, spinal dural dissection cyst; CSF, cerebrospinal fluid; DDC, dural dissection cyst.

DISCUSSION

1. Etiology and Pathogenesis

At present, the etiology and pathogenesis of SEMCs remain controversial. Ogura et al.^{13,14} reported familial *FOXC2* gene mutation and fibronectin type III-related gene mutation-induced epidural arachnoid cysts. The widely accepted theory is that an extradural cyst is caused by herniation of the arachnoid through a congenital or trauma-induced dural defect (Fig. 3A).¹⁷

In 2014, the author reported the diagnosis and treatment of 4 cases of Nabors' type I cyst and believed that naming the cyst as dural dissection cyst (DDC) could better define the nature of

the cyst and guide the treatment. The formation of the cyst is due to a defect in the inner layer of the dura mater, and the potential gap between the inner and outer layers of the dura mater is affected by the fluctuation of CSF, resulting in the DDC between the inner and outer layers of the dura mater (Fig. 3B).⁷

In 2017, Klekamp¹⁵ proposed that both types of lesions, dural diverticulum, and DDC, have defects in the dura, leading to the impact of CSF on the epidural or dural interlayers, forming an SEAC or SDDC.

In our opinion, SDDCs and SEACs were both present in our study on SEMCs. In patient No. 5, we found that the dorsal wall of the cyst was thin, similar to the arachnoid membrane (Fig.

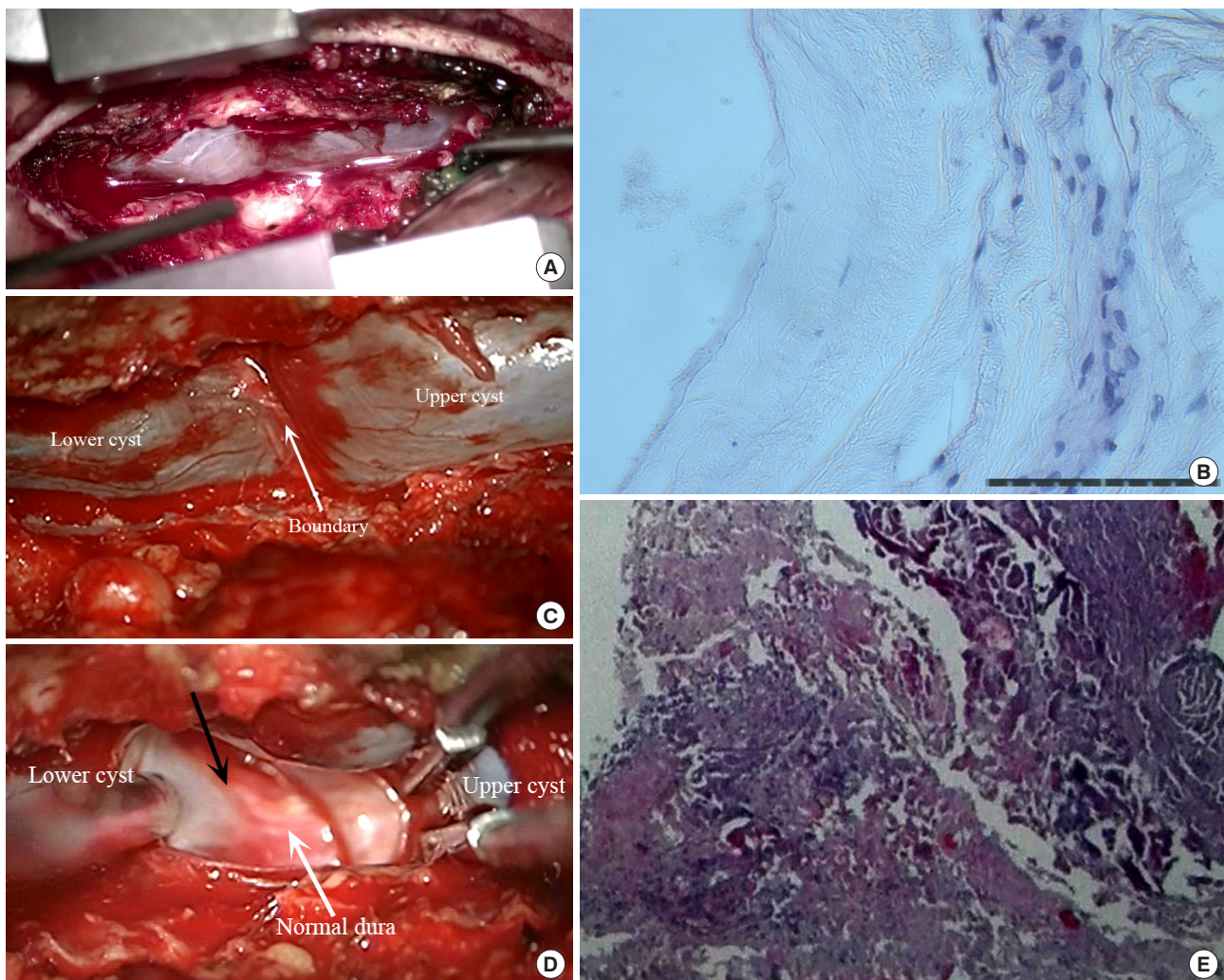


Fig. 4. Patient No. 5 and No. 10: histopathology and intraoperative photos. (A) The dorsal cyst wall of patient No. 5 was found to be thin, resembling an arachnoid layer during surgery. (B) Photomicrograph demonstrating a thin layer of arachnoid cells, collagen fibers, and singular fibroblasts. (C) Intraoperative photograph of patient No. 10 shows the boundary between the cysts. (D) After separating the 2 cysts, the dorsal wall of the lower cyst was found to be combined with the normal dura (white arrow) and could not be dissected from it (black arrow). (E) Histopathology showing that the dorsal wall of the cyst was composed of dense fibrous tissue, accompanied by hyaline degeneration, similar in appearance to the dura (H&E).

4A), and the pathological outcome of the cyst's dorsal wall showed a thin epithelial lining (Fig. 4B). As a result, we considered it an SEAC. Differently, in a study by Liu et al.² they did not perform fenestration for SEAC. Instead, they selected *en bloc* cystectomy by ligating and cutting the fistula outside the cyst as there is a space between the cyst wall and dural sac that allowed dissociation of the cyst. In the case of patient No. 10 with double cysts, we found that the dorsal wall of the cyst was thicker than the normal arachnoid and thinner than the dura (Fig. 4C), and the dorsal wall of the lower cyst was combined with the dura and could not be dissected (Fig. 4D). The dorsal wall of the cyst showed dense connective tissue (Fig. 4E), and we considered this to be an SDDC. Similarly, in a study by Qi et al.¹⁶ the cyst walls were attached tightly to the dura, which prohibited total cystectomy.

Imaging examinations do not distinguish between the 2 types of lesions, and pathological examination is probably the best. The exact number of SEMCs or SDDCs could not be determined in our study due to the incomplete implementation of pathological examination. The best method for identifying a DDC is to collect a specimen at the junction of the cyst and normal dural mater for pathological examination. Microscopically, a complete layer of the dural mater, divided into 2 lobes, is shown to be a DDC. However, this would have increased the dural damage, so this was not performed.

2. Surgical Strategy

Regardless of SDDC or SEAC, there was one cleft in the ventral wall of the cyst.^{7,15} The effect of removing the walls of the

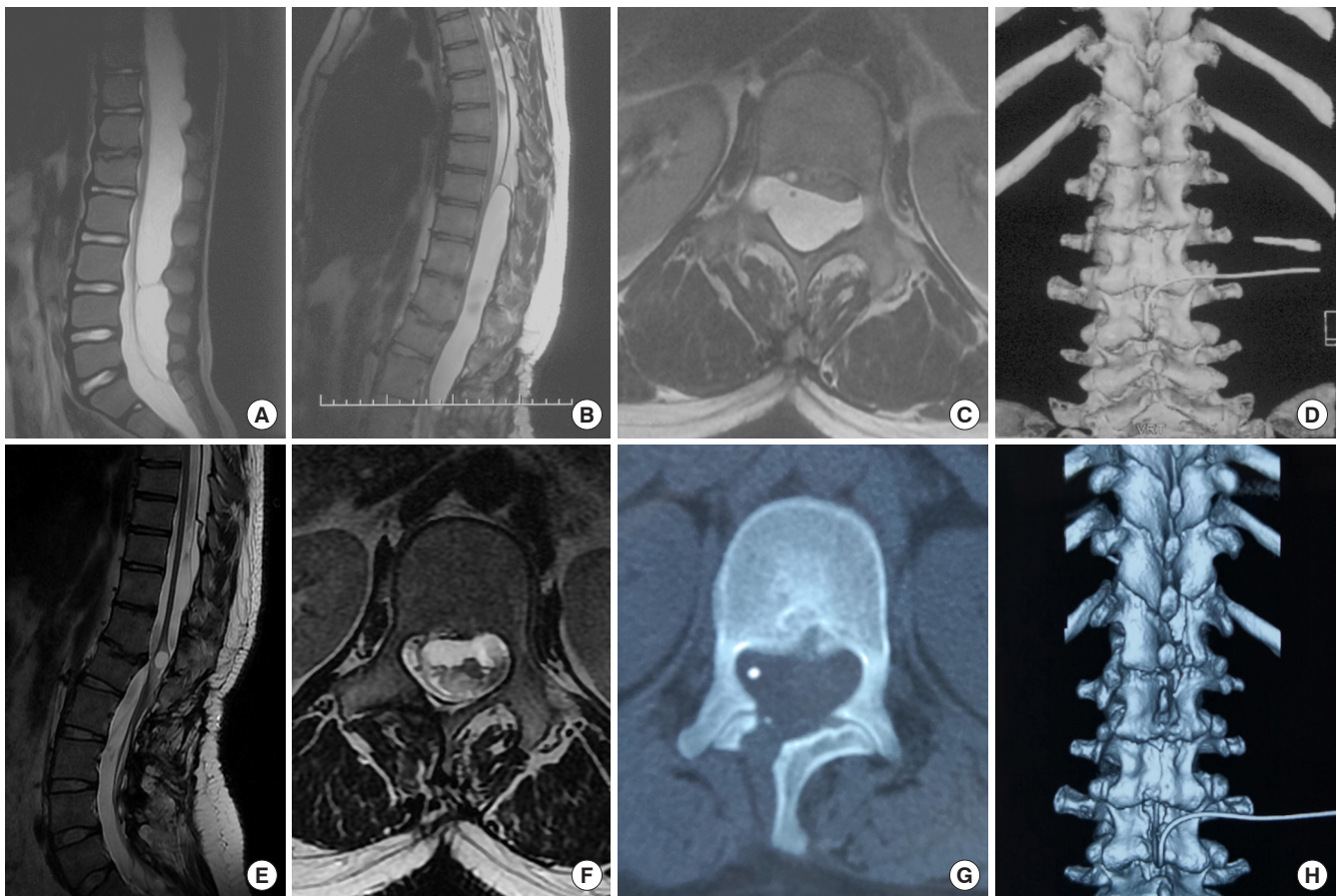


Fig. 5. The images of the patient No. 6. (A) Sagittal magnetic resonance imaging (MRI) before both spinal cord detethering and cystoperitoneal shunt showed that the cyst terminated at S1. (B–D) Postoperative views after cystoperitoneal shunt: (B) sagittal MRI revealed that the cyst extended from T9, (C) axial MRI showed that the right-laterality dominant cyst compressed the spinal cord severely and that another cyst were in the spinal cord, (D) computed tomography (CT) 3-dimensional reconstruction demonstrated the end of the shunt was located at the L3 level. (E–H) Postoperative views after suture of cleft: (E, F) MRI showed shrinkage of the cyst, spinal cord decompression and enlarged intramedullary cyst, (G, H) CT showed hemilaminectomy at the T12–L1 level.

cyst and placing a shunt tube in the cyst is not appropriate due to the presence of a cleft. Finding and suturing the fistula is the most effective treatment strategy.^{6,9}

A key difficulty in the surgery is the accurate detection of fistula. We present our strategy for different conditions as well as the cleft site feature as follows.

1) *Surgical strategy for SEMCs in the thoracolumbar spine*

For patients with multisegmental SEMCs in the thoracolumbar spine, locating the fistula is key for reducing invasion. If the level and laterality of the fistula can be determined, limited hemilaminectomy can be used to explore the fistula, and microsurgery can be performed. After statistical analysis, a cyst usually involves 4 segments. If the thoracolumbar lamina is extensively removed to explore the fistula on the ventral wall of the cyst, the stability of the thoracolumbar spine of the patient will be affected, resulting in kyphotic deformity.^{10,17} Therefore, it is vital to locate the fistula segment. CTM and cine-MRI have been used to locate the fistula of the ventral wall of the cyst in previous studies, but it has not been completely successful.^{10,18} Based on our experience, large SEMCs can lead to subarachnoid obstruction and poor flow of CSF, and the timing of performing CT scan after contrast agent injection is uncertain, making it difficult to determine where the contrast agent enters the cysts. In both our study and study of Gu et al.,^{1,2} the effect of CTM was not satisfactory, and no fistula was found. Ying et al.⁸ reported a successful digital subtraction myelography case of finding the fistula by injecting the contrast agent into the subarachnoid space and simultaneously releasing the fluid in the cyst to reduce the cyst pressure by 2-needle-puncture. However, we failed in 1 patient. Lee et al.⁶ determined the cleft location according to the flow void of CSF, but the flow void does not always exist.

Our experience is that the fistula is mostly located at the T12–L1 level and dominant cyst laterality.¹¹ The reason for this remains uncertain. A potential reason is that the T12 nerve root moves in the direction of the 12th rib, which has a small range of motion due to the restriction from the 12th rib. However, L1 nerve roots are not restricted by ribs, so they have a greater range of motion and are more caudally inclined. Therefore, the change in dural stress between the T12 and L1 nerve roots may lead to dural injury in this area. The CSF impinges through the fistula and causes the spinal cord to shift forward and laterally. Thus, the cyst is eccentric in the spinal canal (Table 3). By the consistency analysis of the fistula and the dominant laterality of the cyst, we found a high consistency of up to 72.72% (24 of 33).

Therefore, our strategy was to perform hemilaminectomy on the dominant side of the cyst at the T12–L1 segment to have the highest chance of detecting the cyst. If the fistula is not detected, the side and segment where the cyst fistula is located can be further determined by carefully observing the direction of CSF flow in the cyst to minimize the range of laminectomy.

In the early stage, we performed total laminectomy or laminoplasty and found that the fistula was mostly lateral and in close proximity to the nerve root, which prompted us to use hemilaminectomy to locate the fistula later. We empirically performed 2-level hemilaminectomy at the T12–L1 level, which is usually successful in finding the fistula and suturing. Hemilaminectomy is performed as minimally as possible to prevent thoracolumbar kyphosis.

An illustrative case for SEMC in the thoracolumbar spine: A 17-year-old male presented with bowel and bladder dysfunction for 6 years. He had undergone spinal cord detethering and a cystoperitoneal shunt at a local hospital. The cyst-peritoneal shunt could partially relieve the patient's symptom; however, he had received 8 peritoneal end dredging of the shunt tube due to repeated shunt obstruction. Physical examination after admission revealed hypoesthesia and decreased muscle strength in both feet, with a positive Babinski sign. MRI revealed an SEMC at the T9–S1 level and an intramedullary cyst at the T12–L1 level, and the morphology of the cyst and the degree of compression of the spinal cord did not change on preoperative and postoperative MRI (Fig. 5A–C). Thus, shunts can only reduce the pressure in the cyst and partially relieve the neurological symptoms (Fig. 5D), but have no therapeutic effect on the cyst. We performed left T12–L1 hemilaminectomy and found a large fistula (Fig. 1A, B). At the last follow-up, MRI showed that spinal cord compression was relieved (Fig. 5E, F). The patient did not complain of any discomfort and achieved a 100% improvement in JOA.

2) *Surgical strategy for SEMCs in the thoracic spine*

Thoracic SEMCs are rare. For isolated SEMC in the thoracic spine, we performed hemilaminectomy at the middle level of the cyst to explore the fistula. This is because fluctuations in CSF pressure can lead to bidirectional dilation of the epidural meninges, leading to cephalic and caudal accumulation of CSF.

3) *Surgical strategy for multiple SEMCs*

In addition, cases of multiple cysts were very rare, accounting for only 6.67% (2 of 30) of the cases in this series. It is also rarely reported in the literature. In the study of Lee et al.⁶ multiple

cysts were defined as more than 3 cysts, accounting for 11.7% (6 of 51) of cases. A fistula was found in each of the multiple cysts. Unstitching or slack stitching may lead to recurrence. For multiple adjacent SEMCs found on MRI, CSF may only accumulate in the opposite direction to the adjacent cysts due to the obstruction of adjacent cysts. In these patients, the fistula may be at one end of the cyst rather than at the middle level. If 2 adjacent SEMCs are found on preoperative MRI, cystography can detect whether there is communication between them, which was successful in our 1 case No.3. Intraoperative hemilaminectomy can also be performed at the boundary level to determine the number of cysts and the presence of communication. The fistula was then probed, especially at the boundary of the adjacent cyst septa, as well as around T12/L1.

An illustrative case for multiple SEMCs: A 16-year-old boy presented with low back pain for 6 months. The symptoms were aggravated during exercise and coughing. The patient had normal sensory function and muscle strength. MRI revealed 2 SEMCs extending from T10 to L3 and compressing the conus and cauda equina. A linear boundary can be found at the L1 lev-

el on sagittal MRI (Fig. 6A). Unfortunately, we did not find a dural defect on the preoperative MRI. The cysts were equilibrated on axial MRI, but extruded out of the right T11–L1 foramen (Fig. 6B). Thus, the right semilamina of L1 was removed during surgery. The purpose was to expose the linear boundary observed on preoperative MRI. We pushed the adjacent cysts aside to reveal the normal dural sac in the middle, and the dorsal wall of the lower cyst was combined with the normal dura and could not be dissected. Then, the wall of the lower cyst was opened to release the CSF under the microscope. Along the flowing CSF, we found a defect on the ventral wall of the lower cyst below the linear boundary of L1 (Fig. 6C). The defect was repaired using an 8-0 nylon suture. Then, we opened the wall of the upper cyst and looked for another defect. Finally, a dural defect inferior to the right T11 root sleeve with leaked CSF was identified. The defect was fully repaired using an 8-0 nylon suture (Fig. 6D). After the operation, the patient's symptoms improved significantly. The 3-month follow-up MRI showed that the cyst had disappeared, and the spinal cord had remodeled to a normal shape (Fig. 6E, F). The patient's symptoms disappeared completely.

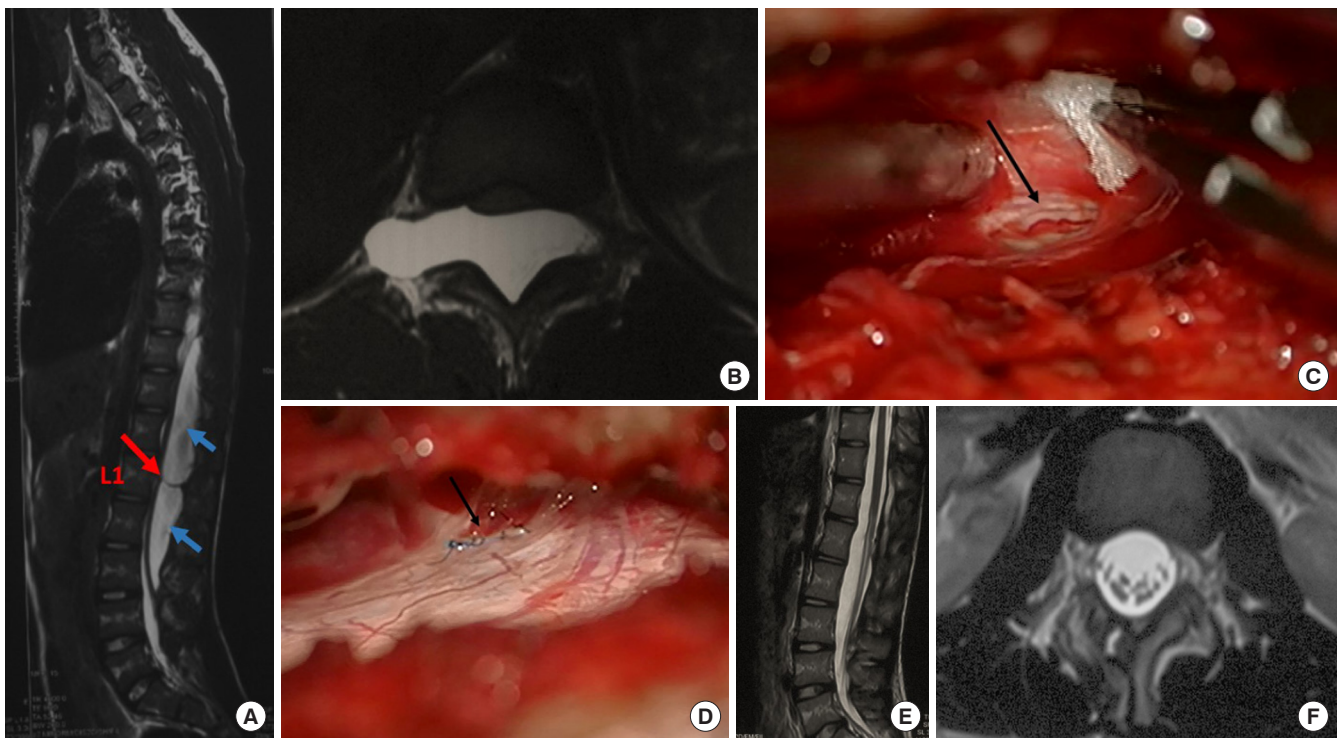


Fig. 6. Patient No. 10: images and intraoperative photograph. (A) Magnetic resonance imaging (MRI) revealed 2 cysts extending from T10 to L3 and compressing the conus and cauda equina. Red arrow shows a linear septum is found at the L1 level. Two blue arrows show 2 cysts. (B) The equalizing cyst extrudes out of the right foramen. (C) The defect of the lower cyst (black arrow). (D) The defect of the upper cyst is repaired with 8-0 nylon suture (black arrow). (E, F) The 3-month follow-up MRI showed that the cyst had disappeared and the spinal cord shape had returned to normal.

4) Surgical strategy for recurrent SEMCs

Early postoperative MRI showed an air signal and cyst outline behind the dural sac due to the removal of the space-occupying effect of the cyst and the incomplete reduction of the dural sac. One patient experienced nosocomial recurrence, which was diagnosed using myelography. For patients with early recurrence, MRI shows a CSF flow void and air signal with cyst outline, which is of low diagnostic value for early recurrence. We performed myelography. The contrast medium showing the outline of the cyst proved the recurrence of the cyst and suggested a flaw in the microscopic suture.

Laminectomy is recommended for revision surgery because of scar adhesions, unclear anatomical structure, and difficulty in determining the dominant laterality of the cyst on axial MRI. For patients with spinal instability, internal fixation should be performed.

5) Prevention for postoperative residual cyst

A few patients with a preoperative large cyst still had a residual cyst at follow-up on MRI. On sagittal MRI, the cyst showed a slim stripe of equal-CSF signal behind the filled dural sac, and on axial MRI, a slim-arc equal-CSF signal behind the filled dural sac. However, the residual cyst is small enough not to compress the spinal cord. Residual cysts may be caused by large preoperative cysts and incomplete CSF release during surgery, rather than postoperative cyst secretion. Therefore, after the fistula is sutured, the CSF in the cyst can fluctuate by changing the patient's body position and increasing the patient's airway pressure to fully aspirate the CSF in the cyst.

Liu et al.² advocated the complete removal of the cyst at the same time of suturing the fistula to avoid the recurrence of the cyst. We believe that this is unnecessary, because postoperative pathology showed that the cyst wall was a nonsecreted functional fibrous tissue, and the cyst would not continue to expand after suturing the fistula. Removal of multiple levels of SEMC will inevitably affect the stability of the spine and lower the clinical recovery effect.⁶ In addition, Aoyama et al.³ and Hamburger et al.⁴ reported that cyst shunt achieved a good effect, but the outcome was not good in our case, so we believe that this method cannot be used to treat the cause. We performed selective laminectomy/hemilaminectomy and microrepair of the fistula with satisfactory clinical outcomes. Symptomatic remission was achieved in most patients with a JOA median improvement rate of $61.84\% \pm 26.63\%$.

Unfortunately, we did not study the changes in thoracolumbar curvature due to the short follow-up time, which is very the

limitation of this study. However, hemilaminectomy has been shown to preserve the contralateral posterior structure and to have a low incidence of kyphosis.¹⁹

CONCLUSION

Most fistulas were always located at the T12–L1 level as well as the middle level of the cyst, which is always consistent with cyst laterality. In patients with multiple adjacent SEMCs, the fistula may be at one end of the cyst. Cleft closure is key to healing SEMCs.

NOTES

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