Transforaminal endoscopic excision of bi-segmental non-communicating spinal extradural arachnoid cysts: A case report and literature review

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Abstract

BACKGROUND
Spinal extradural arachnoid cysts (SEACs) are a rare cause of spinal cord compression. Typically, these cysts communicate with the intradural subarachnoid space through a small defect in the dural sac. For symptomatic SEACs, the standard treatment is to remove the cyst in total with a (hemi)laminectomy or laminoplasty. We present a rare case of bi-segmental non-communicating SEACs and describe our experience of using an endoscopic minimal access technique to remove bi-segmental non-communicating SEACs.

CASE SUMMARY
A 79-year-old female presented with pain related to bi-segmental SEACs at the T11-L1 segments. She underwent sequential transforaminal percutaneous endoscopic thoracic cystectomy of the SEACs. Following her first procedure, spinal magnetic resonance imaging demonstrated complete excision of the cyst at the T12-L1 segment. However, the cyst at the T11-T12 segment was still present. Thus, a second procedure was performed to remove this lesion. The patient’s right-sided lumbar and abdominal pain improved significantly postoperatively. Her Japanese Orthopaedic Association score increased from 11 to 25, her visual analogue scale score was reduced from 8 to 1. The physical and mental component summary of the 36-item short-form health survey (SF-36) were 15.5 and 34.375 preoperatively, and had increased to 79.75 and 77.275 at the last follow-up visit, respectively.

CONCLUSION
Bi-segmental non-communicating SEACs are extremely rare. Endoscopic surgery is a safe, effective, and reliable method for treating these cysts. In the event of bi-segmental SEACs, it is important to identify whether both cysts are communicating before surgery, and if not, to remove both cysts separately during the
A 79-year-old female patient presented with severe back and right-sided abdominal pain of one year duration. The patient did not complain of myelopathic symptoms such as heaviness or stiffness. She had no history of trauma.

**History of present illness**

The patient presented with severe lumbar and right-sided abdominal pain of one year duration, which had become increasingly severe over the preceding week. The patient

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**Core Tip:** Spinal extradural arachnoid cysts (SEACs) are a rare cause of spinal cord compression. Typically, these cysts communicate with the intradural subarachnoid space through a small defect in the dural sac. To date, few articles have reported SEACs that are not in direct communication with the subarachnoid space. For symptomatic SEACs, the standard treatment is to remove the cyst in total with a (hemi)laminctomy or laminoplasty. We present a rare case of bi-segmental non-communicating SEACs and describe our experience of using an endoscopic minimal access technique to treat them.

**INTRODUCTION**

Spinal extradural arachnoid cysts (SEACs) are a rare cause of spinal cord compression [1,2]. A mass effect is created by a collection of cerebrospinal fluid (CSF) that originates through a small defect in the dural sac [3,4]. Variation in nomenclature for this pathology including “meningoceles”, “arachnoid cysts”, and “CSF diverticula pseudo-meningocele” has led to significant confusion in their classification [5]. It has been estimated by Klekamp et al [6] and Tokmak et al [7] that these cysts account for as few as 1% of all primary spinal mass lesions. These lesions can extend as a single cyst across several spinal segments, or they may occur in the form of multiple cysts with each accompanied by a corresponding dural defect [4,6-9]. In 1988, Nabors et al [10] proposed a classification of arachnoid cysts comprising three categories: SEACs without spinal nerve root fiber involvement (Type I); Type I are further divided into two subtypes, extradural arachnoid cysts (Type IA) and sacral meningoceles (Type Ib); SEACs with spinal nerve root fiber involvement (Type II); and spinal intradural arachnoid cysts (Type III). Despite multiple efforts to better define these groups, there remains a lack of consensus about how they should be classified [11,12]. SEACs in the spinal canal are usually located on the lateral or posterior side of the dural sac [6,8]. A majority of SEAC cases reported in the literature involve one segment, with very few reporting disease across multiple segments [4,6-9,13-18]. In this case report, we present a rare case of bi-segmental SEACs, and introduce an important practice consideration in foraminal endoscopic surgery.

**CASE PRESENTATION**

**Chief complaints**

A 79-year-old female patient presented with severe back and right-sided abdominal pain of one year duration.

**History of present illness**

The patient presented with severe lumbar and right-sided abdominal pain of one year duration, which had become increasingly severe over the preceding week. The patient did not complain of myelopathic symptoms such as heaviness or stiffness. She had no history of trauma.
History of past illness
The patient’s past medical history included hypertension, type 2 diabetes mellitus, and coronary heart disease. She had previously presented to two other hospitals with similar symptoms, but they advised her that she was too high risk for open surgery.

Personal and family history
No relevant personal and family history.

Physical examination
Physical examination revealed lower back tenderness and percussion pain, which radiated to the intercostal region. Superficial sensation across the T12 dermatome on the right-side of the abdomen was decreased and strength in the lower extremities muscle groups was grade four. The right knee-tendon reflex and Achilles-tendon reflex could not be elicited bilaterally, but both the planter reflex and ankle clonus were negative. The patient had no abnormality in muscle tone. There is no urinary or faecal abnormalities. At baseline, the Japanese Orthopaedic Association (JOA) and visual analogue scale (VAS) scores were 11 and 8 points, respectively. The physical and mental component summary of the 36-item short-form health survey (SF-36) were 15.5 and 34.375, respectively.

Imaging examinations
Magnetic resonance imaging (MRI) of the whole spine revealed cystic lesions located adjacent to the nerve roots at the T11-L1 level. The lesion showed a low-intensity signal on T1WI and a high-intensity signal on T2WI. The cystic lesion had caused spinal nerve root compression and foraminal enlargement, without compression of the conus medullaris (Figure 1A-C). MRI with gadolinium (Gd) contrast demonstrated no enhancement of the cysts. No apparent communications between the cyst and the subarachnoid space were detected on MRI. Computed tomography (CT) scan of the thorax, abdomen and pelvis revealed bony erosion, foraminal enlargement, and enlargement of the spinal canal at the level of the cystic lesions (Figure 1D).

FINAL DIAGNOSIS
Clinical diagnosis was SEACs.

TREATMENT
Surgical method
The patient underwent surgical excision of the lesion using a minimally invasive endoscopic technique. We were unable to determine whether the cysts were connected on preoperative MRI. It was our expectation that after removing the cyst at T12-L1, the adjacent cyst at T11-T12 would collapse. If a dural tear were to be found during the operation, we planned to close this with a dural patch and gelfoam without thoracic drainage.

Surgical procedure
The operation performed was similar to a percutaneous endoscopic lumbar discectomy. Under endoscopic vision, the T12-L1 cyst was observed directly by manipulating the nucleus pulposus using specialised forceps. The nerve root was carefully protected, and the extradural cyst was excised using nucleus pulposus forceps piece by piece. A radiofrequency probe was used to ensure hemostasis in the spinal canal. Under direct vision, we observed that the surface of the dural sac was intact, with no visible defects. The autonomic beat of the nerve root was observed, suggesting intact function. After confirming full decompression of the nerve root and hemostasis, the wound was sutured without any drainage.
Figure 1 Magnetic resonance imaging and computed tomography scan. A-C: Preoperative magnetic resonance imaging (MRI) showed cystic lesions (white arrows) at T11-L1 level, T1WI demonstrated a low-intensity signal, and T2WI demonstrated a high-intensity signal. The cyst caused spinal cord compression, moving to the left. Coronal MRI showed that the cystic lesions were in the nerve root (white arrows) axilla at the level of T11-L1; D: Preoperative computed tomography scan revealed bone erosion (white arrows), foraminal enlargement, and enlargement of the spinal canal; E: MRI after the first surgery showed the cyst at T11-T12 (white arrows); F: 2-year follow-up MRI showed no recurrence.

OUTCOME AND FOLLOW-UP

Second operation

Postoperative MRI demonstrated that the cyst at T12-L1 had been excised in total, but a second cyst at T11-T12 still remained in situ (Figure 1E). A biopsy from the wall of the cyst demonstrated fibrous tissue without evidence of arachnoid features (Figure 2A). The cyst was filled with a blood clot, without any epithelial or stromal components (Figure 2B). No disc materials, nerve tissues, or tumor cells were found in any of the histopathological specimens.

The patient’s symptoms recurred on the third postoperative day, and she responded to nerve root block therapy for just one day. Therefore, a decision was made to return for a second transfornaminal percutaneous endoscopic cystectomy. The same procedure was carried out using the endoscopic system; however, this time at the level of the T11-T12 disk space instead of T12-L1.

Results and a follow-up visit

Postoperatively, the patient’s right-sided back and abdominal pain had largely resolved. The VAS and JOA scores were improved to 1 and 25 points, respectively. We encouraged the patient to do lower extremity muscle strengthening exercises to prevent muscle atrophy, and allowed the patient to walk from the third day onwards. The physical and mental component summary of SF-36 had increased to 79.75 and 77.275 by the time of the first postoperative follow-up visit. The patient remained asymptomatic during the following two years, and no recurrence was found on MRI (Figure 1F).
DISCUSSION

PubMed and Web of Science were used to search for articles published before April 2020. The keywords and MeSH terms for retrieval were: “spinal extradural meningeal cysts”, “SEMC”, “spinal extradural arachnoid cysts”, “SEAC”, “spinal extradural cysts”, “arachnoid cysts”, “spinal canal”, “extradural arachnoid cyst”. The language of the search study was restricted to English.

Various cystic lesions can be found in the spinal canal, including intramedullary, intradural, epidural, perineural, synovial, and intervertebral disc cysts. As the reliability of MRI has developed, SEACs can now be easily distinguished from other spinal cysts. Characteristically their composition may include fibrous connective tissue and inner single-cell arachnoid lining (although this lining is sometimes not present upon histopathological examination). Most SEACs reported in the literature affect just one segment. In this case report we diagnosed and treated a rare presentation of simultaneous bi-segmental SEACs.

Mechanisms of pathogenesis

The pathogenesis of SEACs remains unclear. Authors have described links to congenital abnormalities or acquired, degenerative changes secondary to trauma; however, the majority seem to be idiopathic. Ogura et al. reported that the transcription pathway mediated by HOXD4 and FOXC2 may play an important role in the developing dura mater, and therefore could have a role in the pathogenesis of SEACs. Trauma and local mechanical stress, infection, or degenerative changes may all cause acquired dural defects. These defects may allow the arachnoid and its closed subarachnoid space to protrude through the dura, where SEACs are formed.

In this case, the patient had no history of trauma and was an older adult. As such, we suspected that the cyst was idiopathic.

Mechanism of cyst enlargement

The mechanism of cyst enlargement also remains unclear. Some mainstream hypotheses include a one-way valve system, hyperosmolar fluid concentration within the cyst, or secretion of fluid from the cyst lining. Rohrer et al. reported that a one-way valve can be caused by the meninges folding at the ostium of the cyst. However, in a series by Morizane et al., in 7 of 12 patients the nerve root fiber may have acted as an alternative valve mechanism. This 'one-way valve' may prevent or hinder the CSF from flowing back into the intradural space. Gradual expansion of the cyst may cause erosion of surrounding bony structures through repetitive micro-stress. Many authors have argued against the hyperosmolar fluid concentration theory because the cyst is likely to have the same fluid concentration as CSF. However, Gortvai et al. found xanthochromic fluid in SEACs which may increase the osmolarity, in support of this theory. The theory of fluid secretion is considered the least likely because cystic walls largely consist of simple connective tissue and often lack an inner arachnoid lining.
Diagnosis

The clinical symptoms of SEACs are related to the size and location of the cyst. In addition to possible pain involving the dermatome of the corresponding segment, cysts may also cause symptoms of spinal cord, nerve root and/or cauda equina compression. As the cyst grows, typically the associated symptoms are also exacerbated[31]. Diagnosing SEACs from symptoms alone is difficult and imaging examination is essential. X-ray imaging typically demonstrates a mass effect of the cystic lesion including spinal bone erosion, foraminal enlargement, and spinal canal enlargement[1]. MRI is the most useful modality for imaging SEACs because this technique can determine the location, extent and relationship of the cysts to the spinal dura mater. Thin slice and contrast-enhanced MRI are also helpful in excluding other diagnoses, such as cystic tumors, synovial cysts and inflammatory cysts[1,32]; typically SEACs show no enhancement after Gd administration[19]. In this case, whilst physical examination demonstrated tenderness in the lower back, we believe that this was unlikely to be due to the cyst itself. There was no compression of the conus medullaris on imaging. The strength in the lower extremity muscle groups was grade four. This was likely to be due to the patient’s age (79 years), with no features suggestive of an upper motor neuron lesion or lower motor neuron lesion. The knee-tendon reflex and Achilles-tendon reflex were abnormal, which again may be due to expected variation between patients rather than directly related to the SEAC.

Treatment

At present, (hemi)laminectomy or laminoplasty with closure of the dural defect is considered the standard method of treating SEACs[2,33]. However, several reports suggest that closure of the dural defect without resection of the cyst may be as effective as a complete cyst resection, whilst maintaining a minimally invasive approach[8]. Lee et al.[34] proposed a "twist technique" as another treatment method, but Shanbhag et al.[31] responded that it may be dangerous to twist the cyst wall without a thorough examination of the inside of the cyst and this has not yet been widely performed.

Spinal endoscopic surgery is now widely used for the treatment of many lumbar degenerative diseases such as foraminal stenosis and lumbar disc herniation[35,36]. In this case report, because we were unable to assess whether the two cysts were continuous or discontinuous preoperatively, the patient had to undergo a re-do transfemoral percutaneous endoscopic procedure to remove the second cyst. If possible, we recommend that future clinicians presented with bi-segmental disease determine whether the cysts are connected before proceeding to surgery. Compared with traditional open surgery, endoscopic spinal surgery has several advantages, including preserving the paraspinal muscle structure, less blood loss, and faster postoperative recovery[37-39]. The endoscope can reach the lesion through a small puncture wound to effectively remove a cyst. This approach preserves the integrity and stability of the spine as the vertebral plate is not removed. Finally, direct vision under endoscopy allows the surgeon to ensure the cyst has been removed in total. Dural tear has been a disadvantage of endoscopic spinal surgery[40], but new methods proposed by Kim et al.[40] reduce this risk significantly. In this case report the patient recovered rapidly with a clear improvement in symptoms postoperatively; we attribute this positive outcome to the use of foraminal endoscopic surgery.

Strangely, no cyst or dural sac communication was found during the operation, and postoperative MRI showed no CSF leakage indicating a dural defect. A review of the literature revealed few other cases of non-communicating SEACs[9,41]. Liu et al.[9] hypothesised that non-communicating cysts may originate from SEACs, but enlargement of the cyst eventually disrupts the communication with the subarachnoid space due to Laplace’s law[42]. Proliferation of arachnoid cells may eventually lead to closure of the dural defects leaving a non-communicating cyst[9]. This is more likely in thoracic segment disease as the CSF pressure is close to zero in the upright position, which is beneficial for early closure[9]. Kim et al.[41] reported a case of huge non-communicating SEACs with myelopathy. These authors believed that the communication was likely to have closed as the cysts continued to expand. Compared with communicating SEACs, surgeons treating non-communicating SEACs do not need to deal with any communication between the cyst and the dura, such as dural defects, arachnoid pedicles or fistulas[41]. We hypothesise that non-communicating SEACs likely develop directly from communicating SEACs. When SEACs develop and begin eroding bony structures surrounding them, the pressure increases inside the cysts. At this time, the pressure in the arachnoid space becomes greater than that in the cyst, so the CSF cannot open the ‘one-way valve’ and enter the cyst to fill further. As time
progresses, the channel gradually closes, and the defect disappears.

Limitations
This article has several limitations. Firstly, this was a single case report at risk of bias. Multicentre studies of this treatment method are required to obtain more valid results. Secondly, endoscopic cystectomy surgery may not be suitable for multi-segment SEACs (e.g., involving more than three segments). Thirdly, constructive interference in steady-state MRI and CT myelography with delayed scanning can be used to explore a communication between cysts. However, these examinations were not performed in our case. Finally, in the first operation, only one segment was treated, and the cyst in the second segment was not excised. As a result, the patient was still symptomatic after the index operation and had to return for a second procedure.

CONCLUSION
In conclusion, bi-segmental non-communicating SEACs are extremely rare. Endoscopic surgery is a safe, effective, and reliable method for treating SEACs. However, in the event of bi-segmental SEACs, it is important to identify whether both cysts are communicating before surgery, and if not, to remove both cysts separately during the index surgery to avoid re-operation.

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