Case report

Hemorrhagic lumbar facet cysts accompanying a spinal subdural hematoma at the same level

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Context: We present a rare and interesting case of hemorrhagic lumbar facet cysts accompanying a spinal subdural hematoma at the same level suggesting a possible mechanism by which spinal subdural hematomas can arise.

Findings: A 71-year-old man presented with persistent sciatic pain and intermittent claudication. Magnetic resonance imaging demonstrated a multilocular mass lesion that showed high signal intensity in both T1- and T2-weighted images, and was located both inside and outside of the spinal canal. Computed tomographic myelography showed a cap-shaped block of the dural tube at L5 and computed tomography with L5–S facet arthrography demonstrated cystic masses. The patient was diagnosed with lumbar radiculopathy caused by hemorrhagic facet cysts, and then progressed to surgical treatment. Surgery revealed that the cysts contained blood clots, and intraoperative findings that the inside of the dural tube appeared blackish and that the dural tube was tensely ballooned after removal of the cysts led us to explorative durotomy. The durotomy demonstrated concentrated old blood pooling both in the dorsal and ventral subdural space, and these spaces were subsequently drained. After surgery, his sciatic pain and intermittent claudication resolved. There was no evidence of cyst mass recurrence at 2 years of follow-up.

Conclusion: We propose a newly described mechanism for the formation of spinal subdural hematomas. We recommend surgeons be alert to epidural lesions causing repeated acute compression of the dural tube, which can cause spinal subdural hematoma, and consider the possible coexistence of these lesions in diagnosis and strategic surgical decisions.

Keywords: Epidural hematoma, Hemorrhagic facet cyst, Multilocular cysts, Spinal subdural hematoma

Introduction

Lumbar facet cysts are lesions, which when generated in the intraspinal canal can compresses the cauda equina and cause various neural symptoms such as lumbago, sciatic pain, muscle weakness, and intermittent claudication. Some lumbar facet cysts necessitate surgical treatment.^{1–3} Moreover, some patients are reported in whom intracystic bleeding causes the rapid growth of cysts, producing an occupation of the spinal canal space, severely compressing nerves.^{4–7} However, spinal subdural hematomas (SSDH) are very rare pathological entities. They occur mostly in the thoracic to thoracolumbar region, and cause rapid progressing nerve palsy in the acute phase,^{8,9} or similarly produce spinal

canal stenosis leading to symptoms of nerve irritation in the chronic phase.^{10–12} SSDHs are mysterious entities because the spinal subdural space contains very few blood vessels, and it remains unclear how and where SSDHs arise. In this report, we present a rare and interesting case of hemorrhagic lumbar facet cysts accompanying a spinal subdural hematoma at the same level. We also review the literature to determine the association between these two pathological conditions.

Report of the case

Informed consent for publication of this report was obtained from the patient.

History

A 71-year-old man presented with a 5-month history of sciatic pain with lumbago, beginning with pain in his

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right buttock and developing into continuous numbness in his right sole and finally intermittent claudication after just a 10-m walk. He also complained of pollakiuria. He first visited our hospital 5 months after the onset of symptoms. He had no history of anticoagulant therapy or trauma associated with his symptoms.

Examination

Severe weakness, grade 1 in a manual muscle test, was noted in dorsiflexion of his right great toe, and mild weakness, grade 4, was noted in plantarflexion of his right great toe and right knee flexion. Mild hypoalgesia was found in his right L5 and S1 dermatomes. Proprioception of his right great toe was reduced. Deep tendon reflexes were normal except for that in his right ankle which had disappeared. Japanese Orthopaedic Association (JOA) score was 9/29.^{13,14} A blood test demonstrated that his blood coagulation, including the international ratio of prothrombin time, activated prothrombin time, bleeding time, and platelet numbers were all within normal limits.

Roentgenography showed no evidence of lumbar spine instability or spondylolisthesis. Magnetic resonance imaging (MRI) demonstrated a multilocular mass lesion, extending 5 cm craniocaudally, located both inside and outside of the spinal canal from the level of L4–S1, which showed high signal intensity in both T1- and T2-weighted images (Fig. 1A–E). A computed tomographic myelogram showed a cap-shaped block of the dural tube at L5 (Fig. 2A) and computed tomography (CT) with right L5–S1 facet arthrography demonstrated cystic masses both inside and outside of the spinal canal (Fig. 2B–D). Osteoarthritic change of the right L5–S1 facet joint was found in CT (Fig. 2C).

Diagnosis and surgery

Based on these clinical observations, the patient was diagnosed with lumbar radiculopathy caused by hemorrhagic facet cysts originating from the right L5–S1 facet joint, and then progressed to surgical treatment. We performed a L5 laminectomy, right L5–S1 facetectomy, and cystectomy. Because of potential spinal instability after the facetectomy, we also performed L5–S1 transforaminal lumbar interbody fusion with posterior instrumentation. The surgery revealed cysts (Fig. 3A, arrow) that contained blood clots with the consistency of a dark-red paste (Fig. 3A, arrow heads). After carefully removing the scar tissue covering that was tightly adhered to the entire dorsal surface of the dural tube from L4 through S1 (Fig. 3B, arrow), we found that the dural tube was tensely ballooned and that the inside of the tube appeared blackish through the dura

mater (Fig. 3C). An explorative durotomy was conducted, and concentrated old blood flooded out from the dorsal subdural space (Fig. 3D). The arachnoid membrane that retained pooled old blood below the dura appeared milky-white; scar tissue was widespread in the dorsal subdural space (Fig. 3E, arrow). The cerebrospinal fluid was crystal clear; however, close observation deep inside the subdural space by parting the spinal nerve roots showed that old blood had also pooled in the ventral subdural space (Fig. 3F); these spaces were subsequently drained. There was no connection between extradural cysts and SSDH.

Clinical and radiological outcome

The patient was fitted with a corset after surgery and began rehabilitation 2 days later. His radiculopathy resolved immediately and muscle weakness gradually resolved such that he was able to walk for a long period without claudication. His JOA score increased to 27/29.^{13,14} MRI performed 2 years postoperatively showed adequate decompression of the cauda equina, and no evidence of cyst recurrence.

Histopathological outcome

Histopathological examination after surgery proved that the cysts were so-called “organized hematomas” with a thick cyst wall made of fibrous tissue, and there were histiocytes containing hemosiderin in the wall of the cysts. Although synovial lining cells were not found in the cyst wall, cysts contained some synovial tissue forming granulation.

Discussion

Facet cysts are known to be slowly growing lesions; however, they can become acute nerve compressing lesions in the spinal canal if they suddenly expand because of bleeding into them. There are two hypotheses regarding the origins of intracystic bleeding. The first is that proliferated synovium following osteoarthritis of facet joints generates vulnerable neo angiogenic vessels having a tendency to break and bleed^{15,16}; the second is that vessels in the cyst wall could be the source of the bleeding.^{6,15} Most authors have considered that hemorrhagic cysts form from secondary bleeding into preexisting cysts.^{4,7,15} The histopathological findings in our case show that the cysts were organized hematomas containing granulating synovial tissues. We can interpret this as a possible mechanism by which bleeding into preexisting cysts caused organization of synovial cysts, and if bleeding repeats, cysts may grow by repeating the rupture and granulation processes. That the cyst formation was multilocular in the present case also suggests

Figure 1 (A, B) Magnetic resonance imaging. Sagittal (A) T1-weighted image, (B) T2-weighted image, showing a high signal intensity mass lesion, extending 5 cm craniocaudally, located in the intraspinal canal from L4 through S1 level (arrowheads) (C–E) Axial (C) T1-weighted image, (D) T2-weighted image, and (E) T2*-weighted image, showing a multilocular mass lesion located both inside and outside (arrowheads) the spinal canal at L5–S1.

a repeated bleeding and healing processes. In all cases, it is true that facet cysts can rapidly transform themselves into expanding extradural masses with bleeding. Indeed, there have been reports of hemorrhagic facet cysts causing acute cauda equina syndrome,⁷ with consequent acute paralysis.⁶

SSDH can be produced by acute compression of the dural tube. Although SSDH have been reported to be influenced by factors including trauma, vascular malformation, and anticoagulant therapy, their essential etiology including the source of bleeding remains a mystery.^{8–12} We considered acute compression of the dural tube by some related influences. There have been some reports concerning postsurgical SSDH. Gehri *et al.*¹⁷ reported that SSDH occurred with dural tearing in microdiscectomy surgery. By contrast, Chang *et al.*¹⁸ Reinsel *et al.*,¹⁹ and Sakai *et al.*,²⁰ did not find dural tearing in a patient who suffered SSDH after discectomy nor in three patients who underwent lumbar decompression and fusion surgery. We speculated that the cause of SSDH was surgical trauma

where the dural tube might be dislodged in a procedure decompressing the dural tube and while also stripping adherent tissues from the dura. There are interesting reports of SSDH associated with acute compression of the dural tube where SSDH occurred after large-volume epidural blood patches performed to treat post-dural puncture headache.^{21,22} This treatment may be similar to surgical trauma because epidural injection of blood that is relatively more viscous than anesthetic agents might temporarily cause acute compression of the dural tube.

The SSDH in our patient may have been produced by epidural hematomas originating from a degenerated facet joint. In the present case, there were two rare lesions adjacent to one another: one was a hemorrhagic facet cyst, the other a SSDH. The two rare hemorrhagic lesions coexisting inside and outside of the dura mater at the same level, suggested a causal relationship between them. We infer the pathogenic mechanism in this case from the etiology and characteristics of each lesion: repeated bleeding in the facet cysts enlarged the cysts

Figure 2 (A) Reconstructed sagittal computed tomographic myelogram (A) showing a dural tube capping shape block at L5–S1 (B, C) Reconstructed sagittal (B), axial (C), and reconstructed coronal CT image with facet arthrography of the right L5–S1 showing a multilocular mass lesion located both inside and outside (arrowheads) the spinal canal at L5–S1. Osteoarthritic change of the right L5–S facet joint is noted (C).

by repeating the rupture and granulation processes; this produced rapid compression of the dural tube, and sequentially generated a SSDH beneath it. Each hemorrhagic lesion probably remained as a chronic hematoma because of repeated bleeding or because the hematoma absorption responses did not function properly, or both. The probability of SSDH formation may increase with repetition of epidural or intracystic bleeding because this increases the frequency of trauma to the dural tube, and also because posthemorrhagic inflammation induces more vigorous angiogenesis, which can provide a new source of bleeding. Additionally, assuming that mere acute compression of the dural tube could be a cause of SSDH, acutely prolapsed disc herniation should also produce it; however, to our knowledge there have been no reports demonstrating any relationship between them. This supports our hypothesis that repeated bleeding and compression increases the risk of generating a SSDH, and raises another hypothesis that whether compression of the dural tube is

ventral or dorsal might be important, because compression by a herniated disc would commonly occur from the ventral side. The latter hypothesis is in good agreement with other conditions that indicate both surgical trauma and an epidural blood patch cause compression dorsally.

The two hemorrhagic lesions in the present case were thought to be chronic hematomas. These hematomas exhibited high signal intensity both in T1- and T2-weighted MRI. This finding indicates that they are in a subacute phase. However, intraoperative findings that the cysts contained blood clots that appeared like dark-red colored paste and SSDH consisting of concentrated old blood indicated that these two hematomas at least included blood in a chronic phase. This viewpoint can also explain the patient's history in which persistent sciatic pain had continued for over 6 months since onset until surgical treatment. Some studies of intracranial chronic subdural hematoma found an association with various factors including vascular endothelial growth

Figure 3 Intraoperative photographs. (A) The right side of each photograph is in the cranial direction. Facet cyst (arrow) was filled with blood clots with the appearance of dark-red colored paste (arrowheads). (B) Removing the scar tissue covering and tightly adherent to the entire dorsal surface of the dural tube from L4 to S1 (arrow). (C) The dural tube was tensely ballooned and the inside of the tube appeared blackish through the dura mater. (D) Concentrated old blood flooded out from the dorsal subdural space after explorative durotomy. (E) The arachnoid membrane was a milky-white color, and scar tissues were widespread in the dorsal subdural space (arrow). (F) Close observation of the deep subdural space through parted spinal nerve roots (arrows) revealed that old blood also pooled in the ventral subdural space.

factor, basic fibroblast growth factor, chemokines, and cytokines.^{23–25} By clarifying the mechanisms of subdural hematoma formation, we may be able to develop innovative treatments that can prevent chronic hematoma formation and promote tissue healing responses to absorb hematomas within the acute phase.

Conclusion

We present a rare and interesting case of hemorrhagic lumbar facet cysts accompanying a spinal subdural hematoma at the same level. From the experience of this combined lesion, we propose a newly described mechanism by which SSDHs can arise. We recommend surgeons be alert to epidural lesions causing repeated acute compression of the dural tube, which can cause SSDH, and consider the possible coexistence of these lesions in diagnosis and strategic surgical decisions. Moreover, surgeons should be alert to SSDHs as a post-surgical complication that can be generated by surgical trauma.

Disclaimer statements

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Conflicts of interest The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this manuscript.

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