

## CASE REPORT

# Surgical Management of Recurrent Spontaneous Spinal Epidural Hematoma With 3 Episodes

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**Study design.** A case report.

**Objective.** We report a case of spontaneous spinal epidural hematoma with 3 bleeding episodes and discuss the surgical management of recurrent spontaneous spinal epidural hematoma.

**Summary of Background Data.** Spontaneous spinal epidural hematoma is a rare condition that causes spinal cord compression and neurological deficits. However, the cause of bleeding remains unclear and recurrent bleeding is very seldom reported.

**Methods.** A 39-year-old female patient was referred to Kyoto University hospital with sudden back and right upper extremity pain. She was also treated conservatively at the local hospital 19 and 4 months previously because of same episodes. Magnetic resonance images demonstrated right spinal epidural hematoma at the C6–T1 level. In the first 2 episodes, magnetic resonance images revealed spinal epidural hematomas at exactly the same level.

**Results.** In the third episode, the patient's neurological condition was not worse than it had been in the first 2 episodes, and we initially managed her conservatively. To identify the cause of the hematoma, surgery was performed 15 days after the third onset. Microscopic examination revealed the development of a venous plexus around the old hematoma in the dorsal epidural space. The patient was discharged without any further neurological deficits, and recurrent bleeding has not occurred for 6 months after surgery.

**Conclusion.** This is the first report of operative and histological observation of recurrent spontaneous spinal epidural hematoma caused by a posterior venous plexus. In a case of recurrent spontaneous spinal epidural hematoma, surgery might be necessary to prevent further hematomas.

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**Level of Evidence:** N/A

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Spontaneous spinal epidural hematoma (SSEH) is an uncommon condition that causes spinal cord compression and neurological deficits. The cause of SSEH remains unclear<sup>1</sup> and recurrent bleeding rarely occurs.<sup>2–5</sup>

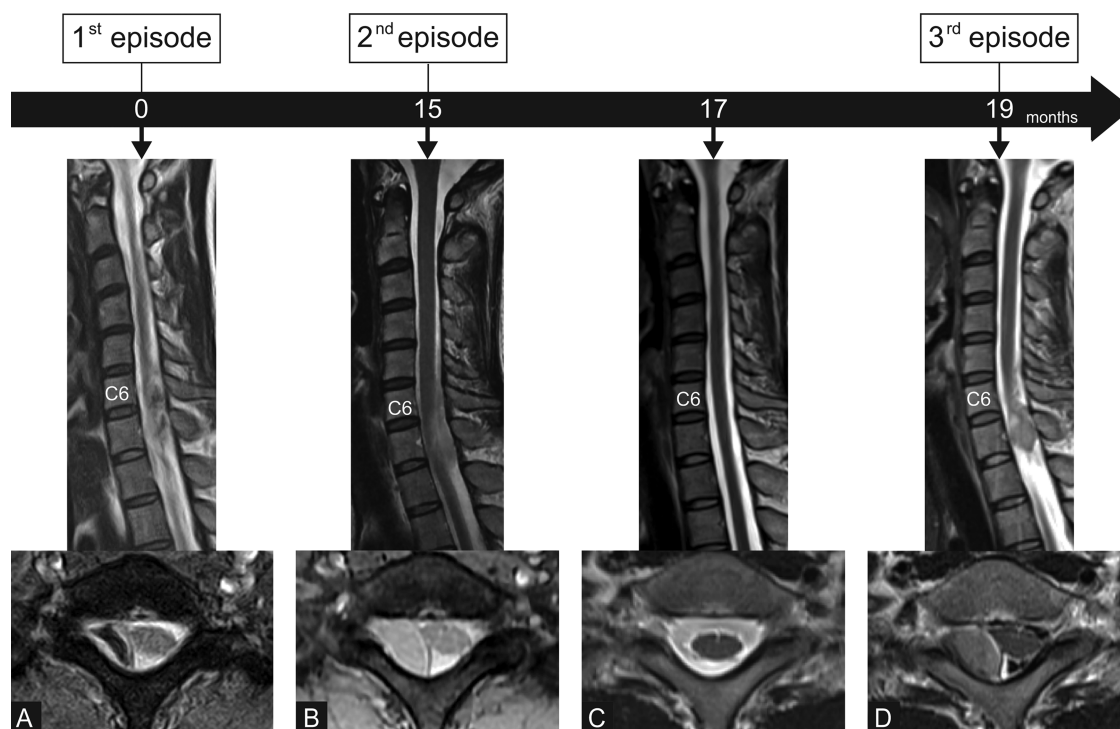
We report a case of recurrent SSEH in a patient who had 3 bleeding episodes, and we discuss the surgical management.

## CASE REPORT

A 39-year-old female presented with sudden back and right upper extremity pain and she was delivered at her local hospital. Magnetic resonance (MR) images revealed a spinal epidural hematoma, and she was referred to our hospital the following day. Because of the same episodes, she had also been treated conservatively at the local hospital 19 and 4 months previously. During the interval between the second and third episodes, she was admitted to our hospital for inspection, but the cause of hematoma was not identified. An interview revealed that she did not have any chronic illness, take any medication, or have any history of injury. The blood examination, including coagulation, was normal. An onset condition common to 3 episodes was not observed.

She exhibited no neurological deficit during the interval between 3 episodes. On admission, she complained of severe back and right upper extremity pain (right C5–T1 area). She displayed no hyper-reflexia or motor weakness or bladder or rectal disturbances.

MR images demonstrated a right epidural hematoma at the C6–T1 level (Figure 1). At the first 2 episodes, MR images revealed epidural hematomas at exactly the same level (Figure 1A, B). However, during the interval between the second and third episodes, no lesions were observed in MR images, even with enhancement (Figure 1C). At the third episode, epidural hematomas were also revealed at exactly the same level (Figure 1D). To identify the cause of recurrent bleeding, repeated digital subtraction angiography during the interval between episodes and at the third episode was performed but did not demonstrate vascular malformations or abnormality.



**Figure 1.** Magnetic resonance (MR) images at each episode. In all 3 episodes of bleeding, the hematoma occupied the right dorsolateral epidural space at exactly the same level, C6–T1 (**A, B, D**). At the interval between the second and third episodes, MR images revealed no lesions (**C**). Upper: Sagittal plane of MR T2-weighted images. Lower: Axial plane of MR T2-weighted images at the C7 level.

Plain radiograph and computed tomographic scan of the cervical spine revealed no remarkable pathological changes.

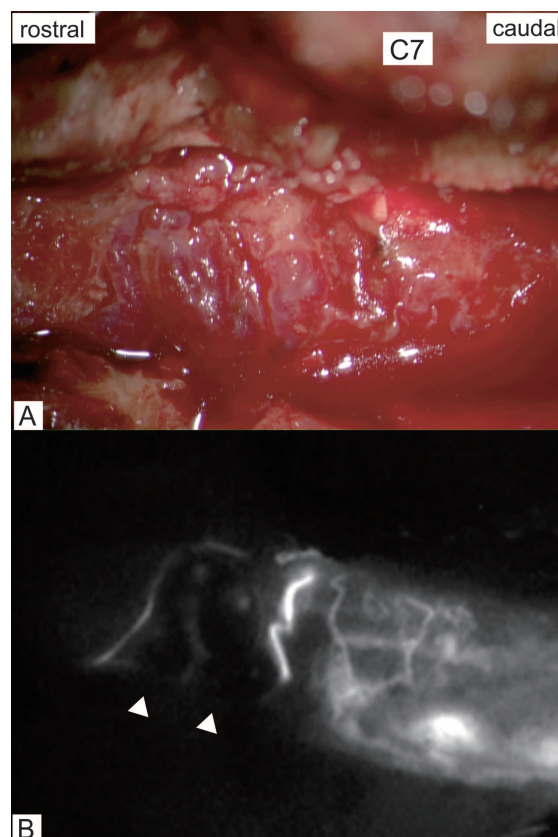
During the first 2 episodes, her symptoms recovered within 2 weeks, and in the third episode, her neurological condition was not worse than that during previous episodes, and we managed her conservatively. To identify the cause of the hematoma, we obtained informed consent and performed surgery 15 days after the third onset.

The patient underwent right hemilaminectomy at the C7–T1 level and right partial hemilaminectomy at the T2 level. Using a microscope, we observed the development of a venous plexus around the old hematoma in the dorsal epidural space (Figure 2A), whereas spines, including yellow ligaments, were normal. An arteriovenous fistula was not confirmed even with the use of a Doppler microprobe or intraoperative indocyanine green video angiography (Figure 2B). The vessels were removed, and histological examination revealed an enlarged vein and hemosiderin around the vein (Figure 3).

She was discharged 16 days after surgery without any further neurological deficits. A recurrent hematoma has not occurred for 6 months after surgery.

## DISCUSSION

SSEH typically presents as acute onset pain in the back, occasionally followed by compressive myelopathy.<sup>3</sup> The cervicothoracic region is the most common location of these hematomas,<sup>2,6,7</sup> most of which arise dorsal to the spine because the dorsal epidural space is occupied by epidural fat. In the majority of previous studies, the source of epidural hematoma was



**Figure 2.** A venous plexus was observed to develop in the dorsal epidural space (**A**). Intraoperative indocyanine green video angiography (**B**) did not demonstrate early filling of the venous plexus (white arrowheads).



**Figure 3.** Histological examination revealed an enlarged vein, and hemosiderin was observed around the vein (arrow) (hematoxylin and eosin, 100 $\times$ ).

postulated to be the fragile valveless vertebral venous plexus, located within the epidural space, which is unprotected from sudden fluctuations of intrathoracic and intra-abdominal pressure;<sup>2,4,7</sup> however, certain investigators have suggested that the hematoma originates in epidural arteries.<sup>1,8</sup> In our case, the results of repeated spinal digital subtraction angiography and coagulation data were normal. The patient did not take any anticoagulant therapy or have any history of injury and, therefore, SSEH was diagnosed. In our case and previously reported cases on recurrent SSEH,<sup>2-5</sup> for the first time, the posterior venous plexus was considered a cause of recurrent hematomas, based on the operative view. The duration of recurrent bleeding was 15 and 4 months, respectively. In previously reported cases, recurrent bleeding occurred within 1 year,<sup>2-5</sup> and thus we hypothesize that recurrent bleeding could occur within a short span.

Surgery can be performed to remove compressive myelopathy. No cutoff time has been established, but various studies have shown that decompressive surgery is most effective when performed within a narrow time window, such as within 8 hours of onset of weakness.<sup>1,3,9</sup> Conservative therapy can be considered in patients who exhibit mild neurological deficits or whose symptoms have improved rapidly.<sup>9,10</sup> Our case was managed conservatively in the first 2 episodes because the symptoms were mild. The symptoms were also mild in the third episode, but we performed surgery to identify the source of hematoma. In a case reported previously,<sup>5</sup> complete recovery occurred after the first 2 episodes, but at the third episode, the patient exhibited severe neurological deficits. Thus, if bleeding were to occur the next time in our case, complete recovery might not be expected. Consequently, the probable source of hematoma was removed without complications. In previous reports,<sup>11,12</sup> surgery of spinal epidural hematoma can

be performed safely (neurological complications 0%). Recurrent SSEH with more than 2 episodes might apply to surgical procedure, but further case accumulation is warranted for fixing when the surgical treatment of recurrent SSEH should be performed.

In summary, recurrent SSEH reported here originated from a posterior venous plexus. In the case of recurrent SSEH, surgery might be required for preventing further hematomas.

## ➤ Key Points

- ❑ Recurrent spontaneous spinal epidural hematoma is extremely rare, and we report, for the first time, that operative and histological observation of recurrent spontaneous spinal epidural hematoma caused by a posterior venous plexus.
- ❑ Recurrent bleeding could occur within a relatively short span.
- ❑ In a case of recurrent spontaneous spinal epidural hematoma, surgery might be required for preventing further hematomas.

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