Spontaneous Spinal Subarachnoid Hematoma of Unknown Pathogenesis: Case Reports

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OBJECTIVE AND IMPORTANCE: The occurrence of spontaneous spinal subarachnoid hematoma of unknown pathogenesis is extremely rare. In the cases reported to date, the hematoma, located dorsal to the spinal cord (dorsal type), has caused paraplegia and has required emergency surgical intervention.

CLINICAL PRESENTATION: We examined two patients who sustained spontaneous spinal subarachnoid hematoma. Both experienced sudden back pain, but there were no symptoms of spinal cord compression. Magnetic resonance imaging revealed spinal subarachnoid hematoma located ventral to the spinal cord (ventral type).

INTERVENTION: Both patients were treated conservatively, and follow-up examinations have revealed that they have remained neurologically normal for 7 years and 6 months, respectively.

CONCLUSION: We postulate that there are two types of spontaneous spinal subarachnoid hematoma of unknown pathogenesis (ventral and dorsal types), each of which presents a distinct clinical picture and prognosis. Ventral type hematoma may be one of the causes of acute back pain, and because of its benign prognosis, surgical treatment may not be necessary. (Neurosurgery 41:691–694, 1997)

Key words: Spinal hematoma, Spontaneous hemorrhage, Subarachnoid hemorrhage, Unknown pathogenesis

Spinal subarachnoid bleeding rarely occurs. It represents less than 1% of all cases of subarachnoid hemorrhage or hematoma (20). In previous reports, there has not been a distinction between spinal subarachnoid hemorrhage and hematoma. However, in this report, we define subarachnoid hematoma as a localized congealed clot in the subarachnoid space, whereas subarachnoid hemorrhage implies a diffuse hemorrhage in the subarachnoid space. The many causes of spinal subarachnoid hemorrhage, or hematoma, include trauma (often caused by lumbar puncture), vascular lesions (arteriovenous malformations, arteriovenous fistula, aneurysm), neoplastic lesions, coagulopathy, periarteritis nodosa, systemic lupus erythematosus, hypertension, coarctation of the aorta, and Behçet's disease (1–8, 10–13, 15, 18, 20). Spontaneous spinal subarachnoid hemorrhage/hematoma of unknown pathogenesis was first reported in 1937 by Slavin (17), and it remains difficult to rule out the underlying causes of the disease without selective spinal angiography, computed tomography (CT), magnetic resonance imaging (MRI), or autopsy. In this strict sense, only seven cases of spontaneous spinal subarachnoid hematoma of unknown pathogenesis, to our knowledge, have been reported in detail to date (9, 12, 14, 15, 18, 19). Whereas the patients in all these cases developed motor and sensory deficits of the legs, we will report two additional cases with different clinical presentations.

CASE REPORTS

Patient 1

A 30-year-old woman was awakened during the night with sudden upper back pain and vomiting. The patient was admitted to the local hospital, where a spinal tap revealed bloody cerebrospinal fluid. CT of the brain disclosed nothing abnormal, and, 2 days later, the patient was transferred to us for further evaluation. At admission, she was neurologically normal, except for slight nuchal rigidity. Neither her personal medical history nor that of her family revealed any predispositions to such an occurrence. Laboratory data were within normal limits. On Day 7, MRI disclosed spinal subarachnoid hematoma at levels C7–T6. Subarachnoid hematoma was located mainly ventral to the spinal cord. Only a slight compression of the spinal cord was revealed (*Fig. 1, A* and *B*). Selective spinal angiography and four-vessel study of the cerebral vessels revealed nothing abnormal. Follow-up MRI on Day 29 disclosed no abnormalities (*Fig. 1C*). The patient was discharged 1 month after ictus with no neurological deficits. There has been no recurrence of spinal subarachnoid hematoma for 7 years. The patient was completely healthy, and repeated MRI revealed nothing abnormal at the most recent follow-up examination.

Patient 2

A 56-year-old woman experienced sudden lumbago while eating dinner and was taken to the local hospital. She underwent abdominal examinations, including CT, echography, and fiberscopic examination of the upper gastrointestinal tract. These examinations revealed nothing abnormal. The next day, a spinal tap, which was carried out

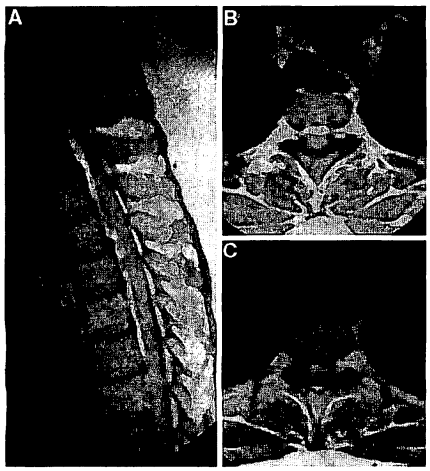

FIGURE 1. *A*, magnetic resonance images (T1-weighted spin-echo images at 0.5 T) of a 30-year-old woman. *B* and *C*, axial images of level T3. *A* and *B*, images obtained on Day 7, showing subarachnoid hematoma extending from level C7–T6 ventrally to the spinal cord. There is no severe compression of the cord. *C*, follow-up magnetic resonance images obtained on Day 29. No abnormality is revealed.

in response to the patient's complaints of a headache, revealed bloody cerebrospinal fluid. CT and MRI of the brain disclosed nothing abnormal. Spinal MRI on Day 7 revealed spontaneous spinal subarachnoid hematoma at levels T11–L2. On axial images, the hematoma was mainly located ventral and right lateral to the spinal cord (*Fig. 2, A* and *B*). The patient was transferred to us for further examination (spinal angiography) 3 weeks after ictus. At admission, the patient was completely healthy except for lumbago predominantly on the right side. She had undergone bilateral removal of an ovarian cyst approximately 30 years before our examination but was otherwise healthy. Laboratory data were within normal limits. Selective spinal angiography disclosed nothing abnormal. She was discharged 1 week later with no neurological deficits and was healthy at the follow-up examination 6 months after ictus.

DISCUSSION

Spinal hemorrhage/hematoma may be epidural, subdural, subarachnoidal, or intraparenchymal. Spinal subarachnoid hematoma rarely occurs relative

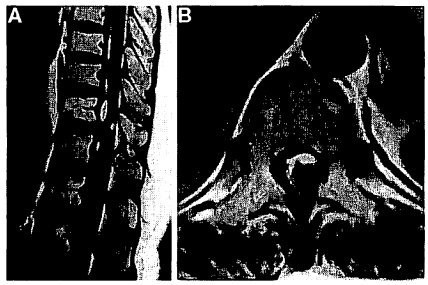

FIGURE 2. *A*, 56-year-old woman. Magnetic resonance images (T1-weighted spin-echo images at 1.0 T). The axial image is at level T11. *A* and *B*, on Day 7, subarachnoid hematoma extends from level T11–L2. Hematoma is mainly located ventrally and right laterally to the spinal cord.

to epidural and subdural hematoma (13). The spontaneous occurrence of subarachnoid hematoma of unknown pathogenesis is extremely rare; only nine cases, including the two cases described in the present report, have been reported (9, 12, 14, 15, 18, 19). Among these nine, the hematomas have been located predominantly at the thoracic or thoracolumbar levels (*Table 1*). The nine patients include five men and four women, with ages ranging from 30 to 81 years and an average age of 58 years.

Clinical manifestations include back pain, lumbago, paraparesis, sensory disturbance, urinary incontinence, loss of sphincter control, and consciousness disturbance. In the seven previously reported cases, the patients developed paraparesis or paraplegia resulting from hematoma. In this group, subarachnoid hematoma was mainly located dorsal to the spinal cord, compressing it to a degree sufficient to cause neurological deficits (dorsal type). The two cases reported here have completely different clinical presentations, including sudden back pain but no neurological deficits. In this type, the subarachnoid hematoma was located mainly ventral to the spinal cord, with only slight compression of the spinal cord (ventral type).

In the diagnosis of spontaneous spinal subarachnoid hematoma, MRI plays an important role in delineating the extension of the hematoma and in disclosing the relationship between the hematoma and the spinal cord (12, 18). CT has a more limited role because the lesion is located within the bony spinal column and, therefore, precise detection of the hematoma is less likely. Myelography has little diagnostic value in the era of MRI because it sometimes gives a false impression of the epidural mass or intraparenchymal mass (12). Computed tomographic myelography can be misleading because when the contrast material does not permeate the cleavage between the spinal cord and the hematoma, the computed tomographic myelogram's images are similar to the intramedullary mass (12). Selective spinal angiography is necessary to rule out the underlying vascular diseases, but when emergency decompressive surgery is necessary, spinal angiography should be forgone in the interest of rapid intervention (8). In this situation, spinal MRI may produce information on the vascular malformation that may be expressed as signal void.

Six of the seven previously reported cases were treated with surgical removal of the hematoma, whereas our two cases and one previously reported case were conservatively treated. Early diagnosis and rapid operative removal of the hematoma for symptomatic patients have been advocated (18). The duration between onset and surgical intervention, preoperative neurological status, and rapidity of the symptomatic presentation have all significantly influenced outcomes (9, 18). In the surgical group, good recovery has been reported in four cases, with no recovery reported in another two cases. In the conservatively treated group, good recovery has been reported in all three cases.

There have been no previously published reports on the long-term follow-up of spontaneous spinal subarachnoid hematoma of unknown pathogenesis. We have followed up Patient 1 for 7 years, with no observations of recurrence. We also followed up Patient 2 for 6 months, with no signs of recurrence. Sunada et al. (18) reported that, based on personal communications, their patient showed no recurrence and remained neurologically normal for 4 years. Thus, spontaneous spinal subarachnoid hematoma of unknown pathogenesis is essentially a nonrecurrent disease. A patient who over-

TABLE 1. Spontaneous Spinal Subarachnoid Hematoma of Unknown Pathogenesis: Review of the Literature[a]

Series (Ref. No.)	Age (yr)/Sex	Dx Tool	Hematoma extent	Hematoma location	Surgery	Symptoms	Recovery
Plotkin et al., 1966 (15)	48/M	Myelo	T6–T9?	Dorsal	Yes	Drowsy/paraparesis	Good
Plotkin et al., 1966 (15)	81/M	Myelo	T6–L3	Dorsal	Yes	Paraplegia	None
Swann et al., 1984 (19)	72/M	Myelo/angio	L4–L5	?	No	Confusion/paraparesis	Improved
Gambacorta et al., 1987 (9)	55/M	CT/myelo	T12	Dorsal	Yes	Paraparesis	Good
Hiyama et al., 1990 (12)	56/F	Myelo/angio/CT/MRI	C7–L4	Dorsal	Yes	Drowsy/paraparesis	Good
Pau et al., 1991 (14)	60/M	Myelo/angio	T3–T7	Dorsal	Yes	Confusion/paraplegia	None
Sunada et al., 1995 (18)	66/F	Angio/MRI	T2–T6	Dorsal	Yes	Paraplegia	Good
Present report, Case 1, 1996	30/F	Angio/MRI	C7–T6	Ventral	No	Pain	Good
Present report, Case 2, 1996	56/F	Angio/MRI	T11–L2	Ventral	No	Pain	Good

[a] **angio, angiography; CT, computed tomography; Dx, diagnostic; MRI, magnetic resonance imaging; myelo, myelogram.**

comes the first bleeding is unlikely to experience further bleeding.

The true mechanism of spontaneous subarachnoid hematoma reported here remains unknown. One proposed chain of causation, however, is minor trauma, which produces rapid changes of the intrathoracic and intra-abdominal pressure. This, in turn, causes increased intraluminal pressure and subsequent tearing of vessels within the subarachnoid space (16). Cerebrospinal fluid may dilute a subarachnoid hematoma. Furthermore, defibrination by pulsation of the spinal cord may reduce the likelihood of a subarachnoid hematoma forming (13). Mechanical obstacles within the spinal column such as spondylosis, disc herniation, arachnoiditis, or thickening of the yellow ligament may contribute to the formation of subarachnoid hematoma. Massive and rapid subarachnoid bleeding may also result in subarachnoid hematoma.

We think that spinal subarachnoid bleeding (either hemorrhage or hematoma) of unknown pathogenesis that causes acute back pain is more prevalent than reports suggest. Many cases do not reach medical attention because of their minor clinical symptoms. Few of them may form subarachnoid hematoma, which may or may not cause spinal cord compression. Spontaneous spinal subarachnoid hematoma of unknown pathogenesis may manifest itself in a wide variety of clinical presentations, ranging from no neurological deficits to paraplegia and altered consciousness. We postulate that there are two types of spontaneous spinal subarachnoid hematoma of unknown pathogenesis, i.e., a hematoma located ventral to the spinal cord (ventral type) that causes acute back pain and minimal neurological deficits but does not necessitate surgical treatment and can be treated conservatively and a hematoma located dorsally to the spinal cord (dorsal type) that may require surgical intervention. If the patient has come through the acute stage, recurrence of spontaneous spinal subarachnoid hematoma is less likely. Finally, an accumulation of data regarding this disease is necessary to clarify its clinical picture.

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REFERENCES

1. Arias MJ, Calero E, Gil JF, Paz J: Spinal subarachnoid hematoma in Behçet's disease. **Neurosurgery** 20:62–63, 1987.
2. Bernsen RA, Hoogenraad TU: A spinal haematoma occurring in the subarachnoid as well as in the subdural space in a patient treated with anticoagulants. **Clin Neurol Neurosurg** 94:35–37, 1992.
3. Caroscio JT, Brannan T, Budabin M, Huang YP, Yahr MD: Subarachnoid hemorrhage secondary to spinal arteriovenous malformation and aneurysm: Report of a case and review of the literature. **Arch Neurol** 37:101–103, 1980.
4. Diaz FG, Yock DH, Rockswold GL: Spinal subarachnoid hematoma after lumbar puncture producing acute thoracic myelopathy: Case report. **Neurosurgery** 3:404–406, 1978.
5. Djindjian M, Djindjian R, Houdart R, Hurth M: Subarachnoid hemorrhage due to intraspinal tumors. **Surg Neurol** 9:223–229, 1978.
6. Fody EP, Netsky MG, Mrak RE: Subarachnoid spinal hemorrhage in a case of systemic lupus erythematosus. **Arch Neurol** 37:173–174, 1980.
7. Frager D, Zimmerman RD, Wisoff HS, Leeds NE: Spinal subarachnoid hematoma. **AJNR Am J Neuroradiol** 3:77–79, 1982.
8. Gaitzsch J, Berney J: Spinal subarachnoid hematoma of spontaneous origin and complicating anticoagulation: Report of four cases and review of the literature. **Surg Neurol** 21:534–538, 1984.
9. Gambacorta D, Reale F, Falco DD: Spontaneous chronic spinal subarachnoid hematoma: Report of a case and review of the literature. **Spine** 12:716–718, 1987.
10. Hanakita J, Miyake H, Toyoda T: Acute paraplegia due to spinal subarachnoid hematoma after lumbar puncture: Case report. **Neurol Med Chir (Tokyo)** 27:1005–1007, 1987.
11. Henson RA, Croft PB: Spontaneous spinal subarachnoid haemorrhage. **Q J Med** 25:53–66, 1956.
12. Hiyama H, Shimizu T, Yato S, Bobayashi N, Ono Y, Kakinoki Y: Wide-spread spontaneous spinal subarachnoid hematoma: Case report. **Neurol Med Chir (Tokyo)** 30:842–847, 1990.
13. Kirkpatrick D, Goodman SJ: Combined subarachnoid and subdural spinal hematoma following spinal puncture. **Surg Neurol** 3:109–111, 1975.
14. Pau A, Brambilla M, Cossu M, Francaviglia N, Siccardi D, Silvestro C: Spinal subarachnoid hematoma of unknown etiology: A case report. **Neurochirurgia (Stuttg)** 34:151–153, 1991.
15. Plotkin R, Ronthal M, Froman C: Spontaneous spinal subarachnoid haemorrhage: Report of 3 cases. **J Neurosurg** 25:443–446, 1966.
16. Rader JP: Chronic subdural hematoma of the spinal cord: Report of a case. **N Engl J Med** 253:374–376, 1955.
17. Slavin HB: Spontaneous intraspinal subarachnoid hemorrhage. Report of a case. **J Nerv Ment Dis** 86:425–427, 1937.
18. Sunada I, Akano T, Kidosaki Y, Shimokawa N, Yamamoto S: Spontaneous spinal subarachnoid hematoma: Case report. **Surg Neurol** 44:133–136, 1995.
19. Swann KW, Ropper AH, New PFJ, Poletti CE: Spontaneous spinal subarachnoid hemorrhage and subdural hematoma. **J Neurosurg** 61:975–980, 1984.
20. Walton JN: Subarachnoid haemorrhage of unusual aetiology. **Neurology** 3:517–543, 1953.

COMMENTS

Komiyama et al. concisely report their experience with two ventral spontaneous spinal subarachnoid hematomas of unknown pathogenesis. Their observation

that the ventral location of spinal subarachnoid hematoma formation is perhaps more "benign" than dorsal types is of significance. The observation of such a hematoma in a patient with acute back pain and no neurological deficit should warrant a conservative nonoperative approach in most cases. The observations of Komiyama et al. should heighten the awareness of neurosurgeons regarding this entity and provide a clinical foundation toward the determination of management strategies.

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This report documents the rare occurrence of spontaneous ventral subarachnoid hematoma. Although the two reported cases had a different presentation from previously reported dorsal subarachnoid hematomas, further experience with these rare lesions will be necessary to be able to identify a distinct pathogenesis, natural history, and management strategy. Ultimately, like spontaneous epidural hematomas, the decision for surgical evacuation of a spontaneous subarachnoid hematoma will be based more on the particular clinical circumstance than the transverse location of the lesion.

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The authors demonstrate that spontaneous intraspinal extramedullary hematoma may present as acute back pain without neurological deficit and that surgical treatment may not be necessary. One of the proposed mechanisms, i.e., increased intraluminal pressure secondary to increased intrathoracic pressure, seems unlikely inasmuch as transluminal pressure would not be affected. Because two patients sustained hematoma ventral to the cord without neurological deficit does not mean that all hematomas in this location will have the same effect on the spinal cord. Neoplasms in the same location have been demonstrated to produce myelopathy.

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