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Case Report

Spinal Subarachnoid Hemorrhage Caused by a Mycotic Aneurysm of the Radiculomedullary Artery: A Case Report and Review of Literature

Homare Nakamura,[1] Phyo Kim,[1] Hideaki Kanaya,[1] Ryu Kurokawa,[1] Hidetoshi Murata,[1] and Hadzki Matsuda[2]

We report a case of spinal subarachnoid hemorrhage (SAH) caused by rupture of a mycotic anerurysm. A 59-year-old woman was admitted to our hospital with a sudden onset of headache and tetraparesis. Computed tomography (CT) scan of the brain revealed SAH, and magnetic resonance imaging (MRI) of the cervical spine showed an acute intradural hematoma. On angiogram, a saccular aneurysm was found on the C5 radiculomedullary artery, which arose from the left ascending cervical artery. Subsequently, her consciousness status deteriorated due to rebleeding, and she was brought to surgery. An aneurysm was found at the cephalad aspect of the left C5 root. On histological examination, it showed typical characteristics of mycotic aneurysms. Spinal mycotic aneurysm is a very rare entity with scant description in the literature. It can be extremely brittle and therefore warrants expeditious surgical treatment. When encountering spinal origin of subarachnoid hemorrhage, it should be included in the differential diagnosis

Keywords: spinal cord, hemorrhage, mycotic aneurysms, micro-abscess, arteriovenous malformation

Introduction

Spinal subarachnoid hemorrhage (SAH) is a rare condition and often takes place in the presence of spinal arteriovenous malformations (AVMs).[1–13] Spinal SAH has been reported in 6% of cases harboring AVM, and 75–90% of the AVM causing SAH are accompanied by aneurysms.[2,4,14] Although solitary aneurysms are less common to occur in the spinal vasculature than those accompanying AVMs, several conditions predispose to the generation of aneurysms. The underlying pathology includes dissection, coarctation of the aorta, neoplasm, systemic lupus erythematosus, Behçet's disease, Moyamoya disease, pseudoxanthoma elasticum, and fibromuscular hyperplesia.[1,3,6,10,15–21] We report a rare case of spinal SAH associated with mycotic aneurysm.

Case Report

In February 2006, a 59-year-old female suffered a sudden onset of headache and tetraparesis. When transported to our hospital, she was alert, afebrile, and vital signs were stable. Neurological examination showed upper and lower extremity weakness which was more dense on the right side. She was complaining of hypesthesia on the lower extremities. Deep tendon reflexes were diminished in the upper limbs and brisk in the lower limbs. Past medical history included leg vein varices for which she had been taking antiplatelets. There was no history of trauma. Routine laboratory analysis including coagulation profile revealed no abnormality. Bleeding time was normal. There was no hemorrhagic tendency. Computed tomographic (CT) scan of the head showed SAH predominantly in the posterior fossa. Magnetic resonance imaging (MRI) of the cervical spine revealed an intradural hematoma on the dorsal aspect of the cord (Fig. 1). Angiography was performed. Aortography was unremarkable. Cerebral studies including the posterior circulation was negative for aneurysms. Selective injection into the thyrocervical trunk revealed a small aneurysm at the C5 level arising from the left ascending cervical artery branching off from the trunk (Fig. 2). The neurological findings improved slightly during the imaging studies. On the next day, her level of consciousness deteriorated suddenly. Glasgow Coma Scale (GCS) was 6 (eye opening 1, verbal response 1, best motor responses 4). She was intubated and started on mechanical ventilation. CT scan of the head revealed an increase of the SAH compared to the study taken on the previous day. MRI T_2-weighted image of the cervical spine revealed a hyperintense signal in the spinal cord (Fig. 3). Decision was made to extirpate the small aneurysm and to remove the hematoma emergently.

I. Operation

C2 to C6 myoarachitectonic spinolaminoplasty was performed in a prone position.[22] The dural tube was tensely expanded and discolored with hematoma. A thin membrane consistent with an arachnoid membrane was immediately found after opening the dura. The clot was thicker on the left side, and the spinal cord was compressed. Following meticulous removal of the clot, the aneurysm was identified at the cephalad aspect of the left C5 root (Fig. 4). SAH was most dense around the aneurysm, substantiating that the aneurysm was the source of the bleeding. The neck of the aneurysm was obliterated and coagulated. The dome was resected and sent for histological examination.

After the hematoma was removed, the dura was closed in a watertight fashion. Pulsation of the dural tube was recovered by the end of the operation. The laminae and the spinous processes of C2 to C6 were reconstructed using hydroxyapatite implants.[22] Her neurological status did not improve after

Departments of Neurologic Surgery[1] and Pathology,[2] Dokkyo University Hospital, Mibu, Tochighi

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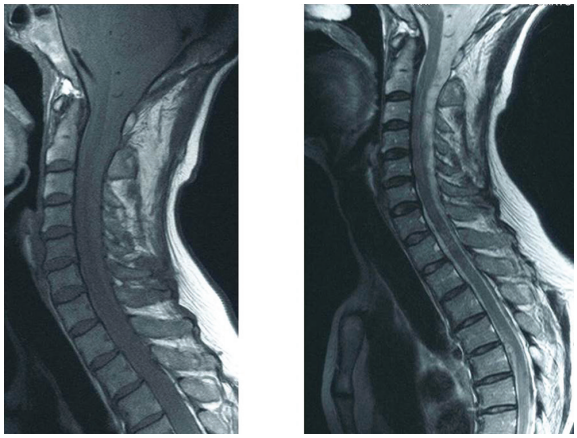

Fig. 1 The spinal hematoma extending from the posterior fossa to C7, T_1-weighted images (T_1WI) of magnetic resonance (left) demonstrated an iso-intense signal mass located predominantly posterior to the cord. On T_2-weighted images (right), the lesion was a hyper-intense signal compared to the spinal cord. The subarachnoid space is obliterated.

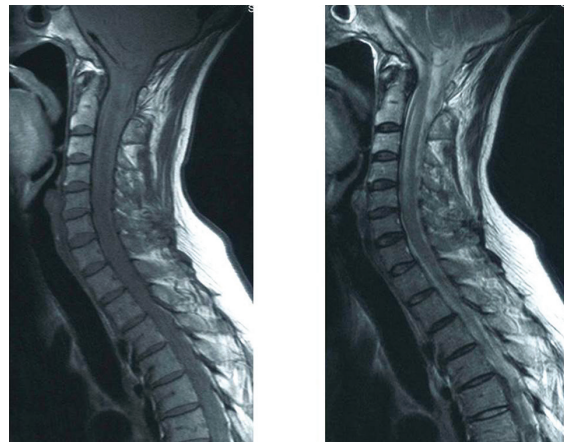

Fig. 3 Magnetic resonance imaging after rebleeding. On T_2-weighted image, the hematoma is visualized as a mixed hypo- and hyper-intense signal. Hyper-intense signal is detected inside the spinal cord from C2 to C5, presumably representing an ischemic change.

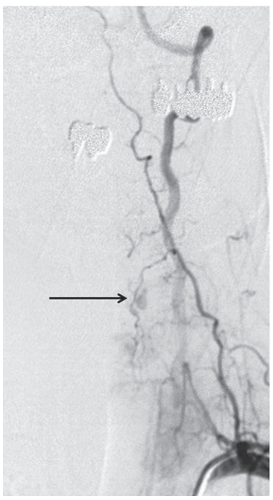

Fig. 2 Selective spinal angiogram showing the saccular aneurysm (arrow) on the left C5 radiculomedullary artery, a branch of the ascending cervical artery. The staining remained into the late venous phase.

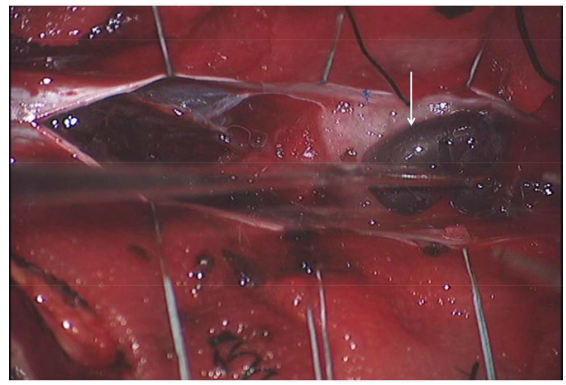

Fig. 4 Intraoperative photograph showing the aneurysm (arrow) retrieved from the cephalad aspect of the left C5 root.

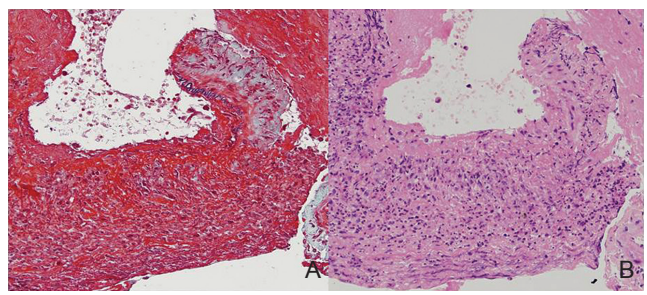

Fig. 5 Photomicrographs of the resected aneurysmal wall. A: The section showing disruption of the internal elastic layer. Elastica–Masson stains, ×200. B: Infiltration of the inflammatory cells is evident. Hematoxylin-Eosin stains, ×200.

surgery, and the patient died 3 days later. Autopsy was not obtained.

II. Histological findings

Pathological examination of the resected aneurysm showed partially defective dome as a result of the rupture. Elastica–Masson staining demonstrated that the internal elastic lamina was disrupted. On hematoxylin and eosin (HE) stains, inflammatory cell infiltration, mainly by neutrophils, was prominent in the extensively destroyed aneurysmal wall, with notable micro abscesses (Fig. 5). Gram stain of the specimen was negative. The findings were compatible with a diagnosis of mycotic aneurysm.

Discussion

SAH of a spinal origin is a rare clinical condition.[1–3,6,9,10,12,14] The most common cause of spinal SAH is an AVM.[1–13] Incidence of accompanying aneurysms in the presence of spinal AVMs is reported to be 2.2–7.7%, and possibility of aneurysmal rupture has to be taken into consideration.[7,23] Other conditions known to be associated with solitary spinal aneurysm include dissection of the aorta, coarctation of the aorta, neoplastic lesion, systemic lupus erythematosus, Behçet's disease, Moyamoya disease, pseudoxanthoma

Table 1 Summary of all reported cases of subarachnoid hemorrhage caused by solitary spinal aneurysm (not accompanying arteriovenous malformations)

Case no.	Author	Age	Sex	Etiology	Level	Operation	Pathology	Deterioration	Outcome
1	Walz et al.[21]	58	M	Moyamoya	C4	endovascular	-	-	no change
2	Gonzalez et al.[3]	30	M	NA	T11	+	-	NA	excellent
3		73	M	NA	T6–7	+	-	-	excellent
4		54	M	dissection	T12	+	dissection	-	excellent
5		69	M	NA	NA	+	-	NA	excellent
6	Massand et al.[20]	30	M	dissection	T11	+	-	-	excellent
7		69	M	dissection	L1	+	dissection	-	poor
8		54	M	dissection	T12	+	dissection	-	excellent
9		73	M	NA	T7	+	-	-	NA
10	Berlis et al.[1]	62	F	dissection	T5	+	-	-	excellent
11		48	M	autoimmune disease	T12	-	-	-	no change
12		69	F	dissection	L1	-	-	+	excellent
13	Yahiro et al.[12]	71	F	pseudoaneurysm	T4–5	+	pseudoaneurysm	-	no change
14	Kawamura et al.[5]	42	M	NA	C1	+	-	-	excellent
15	Rengachary et al.[9]	50	F	autoimmune disease	T12	+	autoimmune disease	-	no change
16	Bahar et al.[15]	40	M	Behçet's disease	C5–6	-	-	-	excellent
17	Goto et al.[4]	53	M	true saccular	C2	+	true saccular aneurysm	-	excellent
18	Hino et al.[18]	45	F	coarctation	C5–6	-	-	-	no change
19	Saunders et al.[10]	48	F	FMD	T1	+	+	-	excellent
20	Smith et al.[11]	29	M	NA	T12, L1	+	-	-	no change
21	Kito et al.[19]	37	F	PXE	T9–10	-	-	-	excellent
22	Moore et al.[8]	30	F	NA	C1	+	-	-	no change
23	Vincent [13]	30	F	NA	C2	+	-	-	no change
24	Kormos et al. [6]	31	F	hemangioblastoma	C1	+	false aneurysm	-	excellent
25	Fody et al.[17]	50	F	SLE	midthoracic	-	autopsy	+	death
26	Garcia et al.[24]	34	F	infection	T6	-	infectious, autopsy	+	death
27	Banna et al.[16]	40	M	coarctation	C6–7	-	autopsy	-	death
28	Our case	59	F	infection	C5	+	infectious	+	death

FMD: fibromuscular hyperplesia, SLE: systemic lupus erythematosus, PXE: pseudoxanthoma elasticum, NA: date not available.

elasticum, and fibromuscular hyperplesia.[1,3,6,10,15–21] Reported cases of SAH caused by solitary spinal aneurysms and the underlying conditions are summarized in Table 1. There were 28 cases, 15 men and 13 women, with age ranging from 30 years to 73 years (average 49.2 years). Location of the aneurysms was cervical in 10, thoracic in 15, and lumbar in 11 patients. Surgical interventions including endovascular procedures were performed in 20 cases, and histological examination was performed in 12 cases including autopsy cases. The outcome was excellent in 12, poor or death in 5, and fair with stabilized status in 8 cases. Neurological deterioration during the course of treatment was described in 4 cases. Notably, two of these four cases were of mycotic aneurysms with rebleeding.[24] Risks of rebleeding may be high with the spinal mycotic aneurysms, as has been known with the intracranial mycotic aneurysms.[25] On the other hand, accurate diagnosis of mycotic aneurysms may be difficult when it occurs in the spinal region. Intracranial mycotic aneurysms most commonly affect peripheral arteries, and the demography is different from the common saccular aneurysms. Spinal aneurysms are themselves rare, and location seems to be of little help in identifying the mycotic nature. Therefore, it may be judicious to conduct surgical exploration when seeing a solitary spinal aneurysm causing SAH.

Conflicts of Interest Disclosure

The authors report no conflict of interest concerning the materials and methods used in this study or the findings specified in this article. All authors who are members of The Japan Neurosurgical Society (JNS) have registered online Self-reported COI Disclosure Statement Forms through the website for JNS members.

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Corresponding author:
Homare Nakamura, MD, Department of Neurosurgery, St. Marianna University School of Medicine, Yokohama City Seibu Hospital, 1197-1 Yasashi-cho, Asahi-ku, Yokohama, Kanagawa 241-0811, Japan.
✉ homare@marianna-u.ac.jp