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Case Report

Surgical Removal of a Ruptured Radiculomedullary Artery Aneurysm: A Case Report

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Subarachnoid hemorrhage due to a solitary spinal aneurysm is extremely rare, and diagnosis and treatment are challenging. We report a rare case of a ruptured radiculomedullary artery aneurysm in a patient with Behçet disease. A 49-year-old man presented with severe lower abdominal and leg pain. Magnetic resonance imaging was performed and an enhanced intradural-extramedullary lesion at the T12 spinal level with subarachnoid hemorrhage was identified. Diagnostic spinal angiography was performed to evaluate the vascular lesion, and a radiculomedullary artery aneurysm at the T12 level was identified. We performed surgical resection of the aneurysm and a good neurological outcome was obtained.

Keywords Spinal aneurysm, Radiculomedullary artery, Subarachnoid hemorrhage

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INTRODUCTION

Subarachnoid hemorrhage (SAH) due to a solitary spinal aneurysm is extremely rare, representing less than 1% of all cases of SAH reported in the literature. Aneurysms involving the radiculomedullary arteries are particularly rare, with only nine cases previously reported.[1-3)6)11)12)14)16)] The most common etiology of spinal SAH is spinal arteriovenous malformations (AVM); other etiologies include arterial dissection, neoplasm, moyamoya disease, systemic lupus erythematosus, and Behçet disease (BD).[12)14)] Various etiologies can contribute to the development of spinal artery aneurysm. The purpose of this case report is to describe the successful surgical treatment of a ruptured aneurysm in the radiculomedullary artery in a patient with BD.

CASE REPORT

A 49-year-old man with BD presented to our outpatient department with a 3-month history of severe lower abdomen and leg pain, with no history of trauma. The patient had a medical history of hypertension, uveitis, and oral ulcer, with use of appropriate medications for these conditions. His erythrocyte sedimentation rate (ESR) was 30 mm/h. Neurological examination revealed hyperesthesia below the level of T12 and an increased deep tendon patellar reflex, bilaterally. Magnetic resonance imaging of the lumbar spine revealed a 3 mm enhanced intradural-extramedullary mass at the T12 level with extensive SAH and cord compression due to hematoma (Fig. 1). Spinal angiogram was performed for further diagnosis of the suspected vascular lesion at T12. Selective injection into the right L2 segmental artery revealed a 5.5 × 3.2 mm

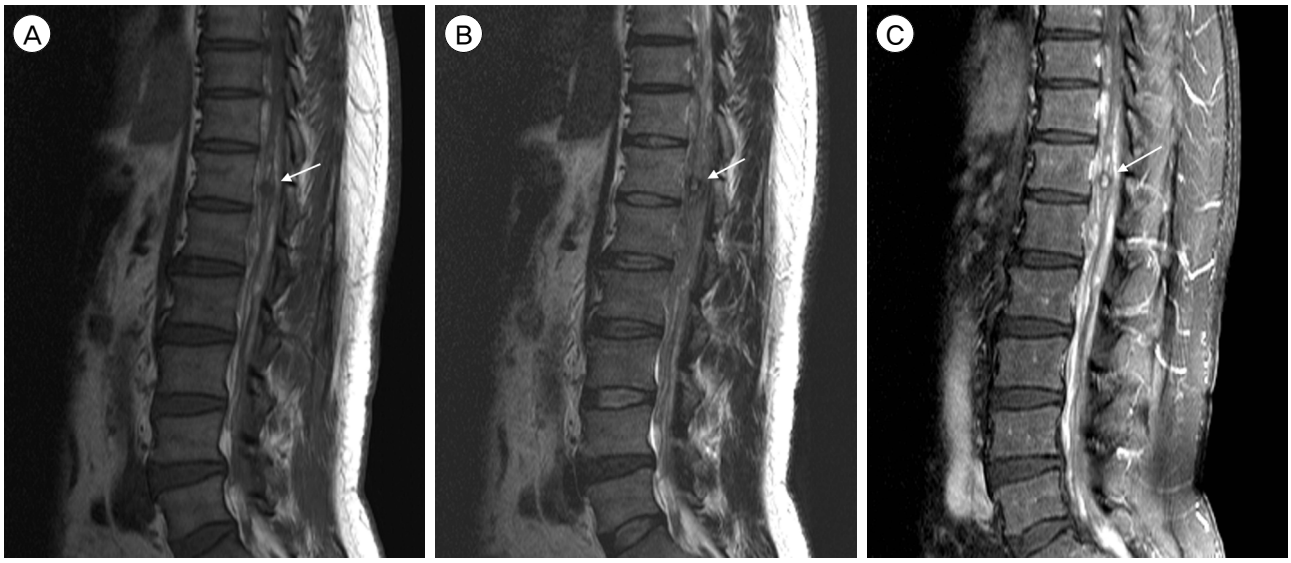

Fig. 1. A nodular lesion (arrow) with iso-intense signal on T1-weighted images (A) and hyper-intense signal on T2-weighted images (B). On enhanced T1-weighted images (C), the nodular lesion was enhanced at the T12 level.

intradural aneurysm receiving direct blood supply from the radiculomedullary artery at the T12 level (Fig. 2). The Adamkiewicz artery was visualized on left T9 segmental artery angiogram and the anterior

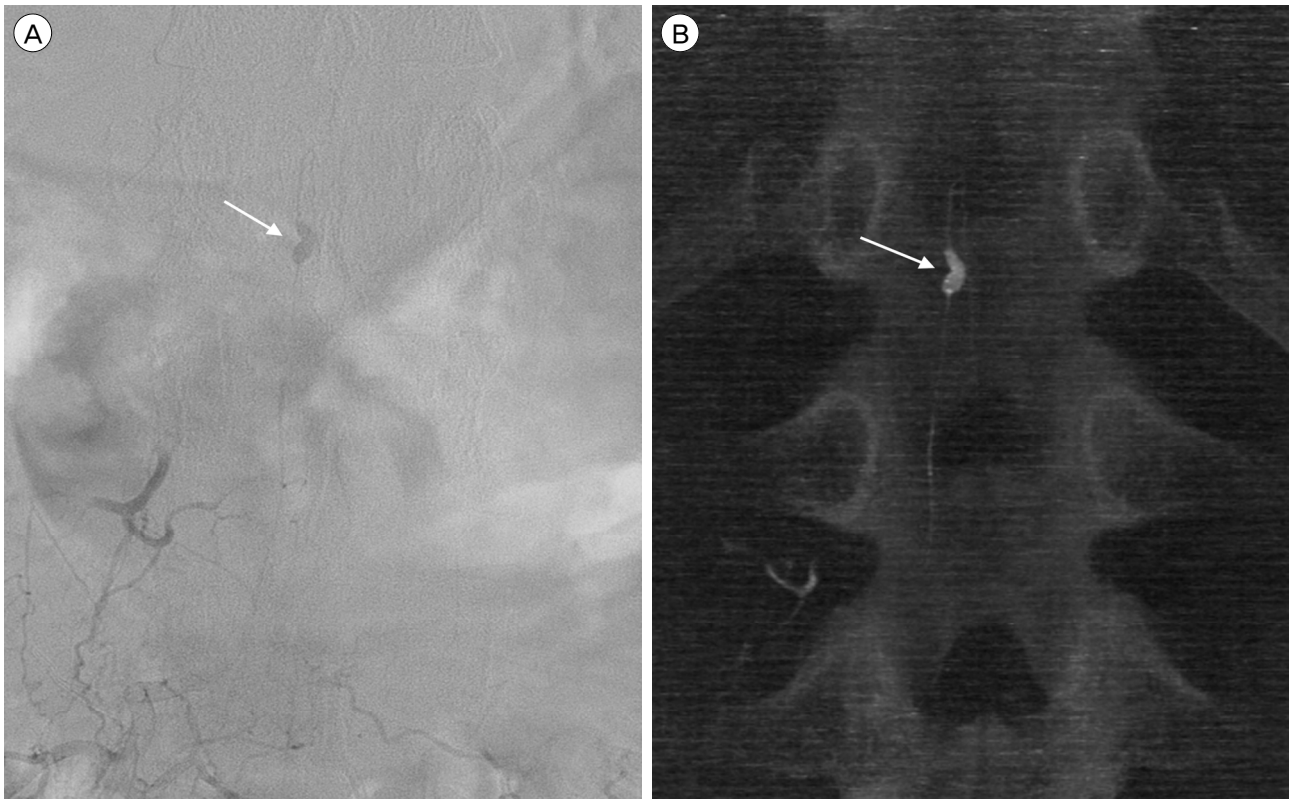

Fig. 2. (A), (B) Selective spinal angiogram showing the fusiform aneurysm (arrow) on the right L2 radiculomedullary artery at the T12 level.

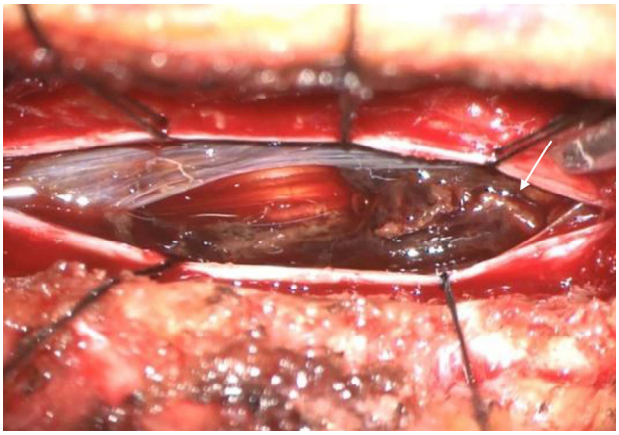

Fig. 3. Intraoperative photograph showing the aneurysm (arrow).

spinal artery (ASA) was visualized below the L2 level communicating with this artery. Based on our findings, we chose to perform surgical resection.

Laminectomy was performed from T10 to L2. After opening the dura, diffuse SAH was found and the hematoma was thick, resulting in spinal cord compression. After removal of the hematoma, the ruptured aneurysm was exposed. Active bleeding persisted as we explored the aneurysm and we identified a dissecting lesion at the distal section of the aneurysm (Fig. 3). Temporary clips were applied to the parent arteries; we then evaluated for changes in neurophysiological monitoring to identify whether complete resection would lead to ischemic complications. During the five minutes of temporary clipping, no abnormalities were detected on neurophysiological monitoring; thus, the aneurysm was completely resected.

Pathological examination of the surgical specimen revealed a degenerated vascular wall with thrombus, suggestive of an aneurysm wall without inflammatory cell infiltration (Fig. 4). No surgery-related complications were identified and the patient recovered fully after the surgery.

DISCUSSION

Aneurysms of the radiculomedullary artery are extremely rare, with nine cases identified in the literature, as summarized in Table 1.[1-3)6)11)12)14)16)] To our knowledge, this is the first case report of a ruptured aneurysm of the radiculomedullary artery, which was surgically treated in a patient with BD.

Aneurysm associated with spinal AVM is the most

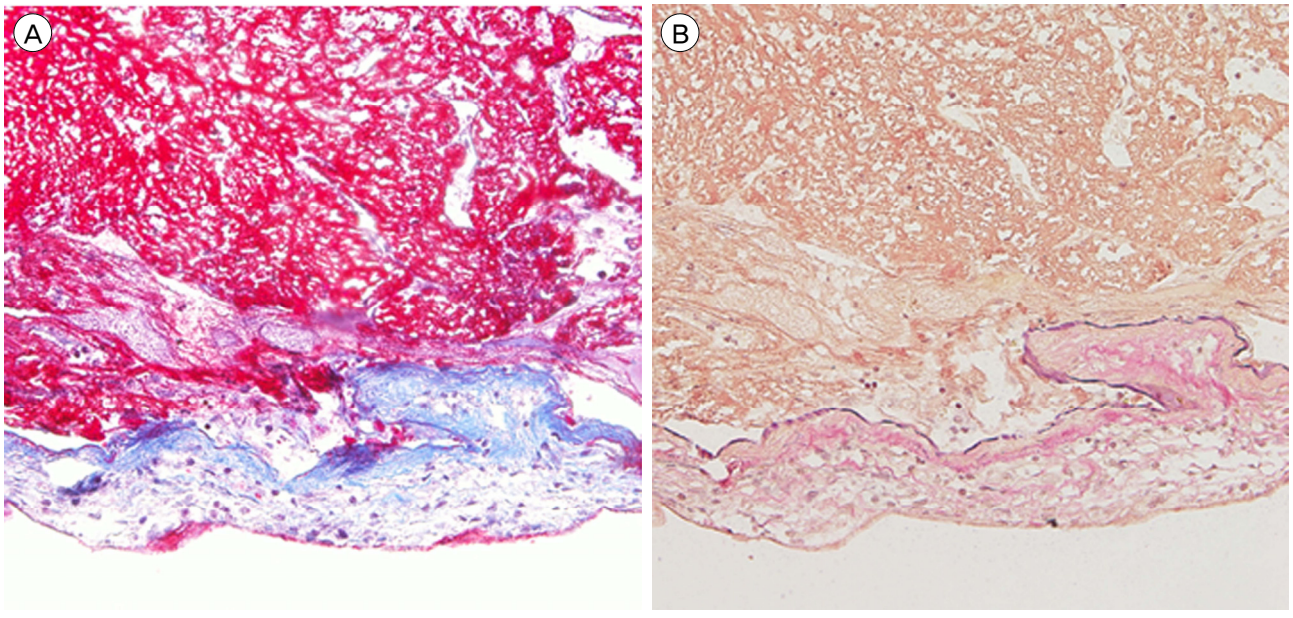

Fig. 4. Photomicrographs of the resected aneurysmal wall. (A) The section shows mild fibrosis in the degenerated vascular wall (Masson trichrome stain, ×200). (B) Thin elastic fibers in the vascular wall are demonstrated (Elastica von Giesen stain, ×200).

Table 1. Summary of all reported cases of ruptured aneurysm of the radiculomedullary artery

Author, year	Sex/age	Location	Clinical presentation	Diagnosis	Treatment	Outcome*
Garcia et al.[3], 1979	F/34	T6	Paraplegia, headache	Pregnancy	Not available	1
Bahar et al.[1], 1993	M/40	C5	Headache, vomiting	Behçet disease	Conservative	4
Vishteh et al.[16], 1997	M/30	T11	Headache, back pain	Dissection	Wrapping	5
Berlis et al.[2], 2005	M/48	T12	Abdominal pain, back pain	Infection	Conservative	5
	F/69	T12	Back pain, walking impairment	Dissection	Conservative	5
Massand et al.[11], 2005	M/30	T11	Back pain, paresthesia	Dissection	Wrapping	5
Iihoshi et al.[6], 2011	F/60	T12	Back pain, lower limb pain	Dissection	Conservative	5
Son et al.[14], 2013	F/45	T12	Headache, back pain	Dissection	Conservative	5
Nakamura et al.[12], 2015	F/59	C5	Headache, tetraparesis	Infection	Resection	4
Present	M/49	T12	Abdominal pain, leg pain	Dissection	Resection	5

F = female; M = male.
*Outcome was presented with a Glasgow Outcome Scale Score

common cause of spinal SAH.[12)13)] Other known conditions associated with spinal aneurysm include arterial dissection, neoplasm, BD, systemic lupus erythematosus, moyamoya disease, fibromuscular dysplasia, pregnancy and infection.[1-9)11-14)16)] BD is a type of systemic vasculitis that primarily affects the eyes, skin, joints, blood vessels, and nervous system.[10)15)] Vascular involvement is not uncommon with BD, with a reported prevalence ranging between 7% and 29% of all cases.[15)] Although the venous system is involved in most cases, arterial aneurysms are a leading cause of death in this clinical population. When present, arterial aneurysms typically expand rapidly and result in fatal rupture. When present in patients in BD, aneurysms most commonly develop in the aorta, and the femoral or pulmonary arteries, with involvement of branch arteries being uncommon. Spinal SAH due to ruptured solitary spinal artery aneurysm is extremely rare in BD.[1)] We found only one reported case, a patient with ruptured radiculomedullary artery aneurysm at the C5 level. In our case, based on the identification of a dissecting lesion at the distal section of the aneurysm during surgery and no evidence of inflammatory cell infiltration on pathological examination (which are specific finding of aneurysm associated with BD), we hypothesized that dissection of the aneurysm was the primary event, resulting in the formation of a pseudoaneurysm with subsequent hemorrhage.

The symptoms of ruptured spinal aneurysm seem to correlate with the level of the lesion. Ruptured aneurysm at the cervical level can cause meningeal irritation due to intracranial SAH or quadriparesis. Lesions involving the thoracolumbar levels can cause lower back pain, abdominal pain, and motor weakness or sensory changes of the lower extremities. Our patient complained of back pain and sensory changes of the lower extremities. These symptoms were suggestive of aneurysm at the thoracolumbar levels and the lesion was identified at the T12 level.

There is no standard treatment guideline for ruptured spinal aneurysms because these lesions are very rare. Therefore, the choice of proper treatment, including surgery, endovascular embolization, and conservative treatment, remains controversial. Conservative therapy has been preferred in cases of small ruptured dissected aneurysms at the ASA or Adamkiewicz artery which must be preserved to reduce neurological complications, and solitary aneurysms associated with underlying disease, such as BD, which can show spontaneous healing of the dissected aneurysms with patent flow.[1)2)6)] Endovascular treatment can be a treatment option if the surgical procedure is difficult due to the location of the lesion or the patient's general condition.[8)] However, because most spinal artery aneurysms are fusiform rather than saccular shape and endovascular

treatment has the potential risk of causing vascular injury or thrombosis that could lead a poor neurological outcome, endovascular treatment is usually not an option.[2] Surgery may be an appropriate choice for the ruptured spinal artery aneurysms, especially if the distal flow is absent or the surrounding hematoma produces a mass effect on the spinal cord. If no distal flow is confirmed on spinal angiogram, complete obliteration of the lesion can be performed to reduce the risk of re-bleeding. On the other hand, if distal flow is identified on spinal angiogram, wrapping can be an alternative treatment option that preserves the distal flow. In our case, the Adamkiewicz artery was identified on left T9 segmental artery angiogram and ASA flow was detected below the L2 level. We assumed that although the ruptured dissecting aneurysm was surgically resected, blood flow from Adamkiewicz artery to the ASA was sufficient to supply the lower L2 level. In addition, to enhance safety of the surgery, we also evaluated neurophysiological monitoring results during temporary clipping of radiculomedullary artery and during the entire procedure. There was no change in neurophysiological monitoring, and we could thus completely resect the aneurysm.

Based on our experience in this case, we propose that surgical treatment of a ruptured radiculomedullary artery aneurysm with collateral flow from the Adamkiewicz artery is feasible with application of a careful surgical technique, including temporary clipping and intraoperative neurophysiological monitoring, to achieve a good clinical outcome.

CONCLUSION

Ruptured spinal aneurysm of the radiculomedullary artery is a rare vascular lesion. With only nine cases previously reported, it is difficult to define an optimal treatment approach. However, based on our experience, we propose that early surgical resection of this aneurysm by careful surgical technique, including temporary clipping of the parent artery and intraoperative neurophysiological monitoring, when the Adamkiewicz artery is identified near the lesion, might be effective in preventing cord ischemia and improving neurological outcome.

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Informed consent from the patient is waived by the Institutional Review Board at our institution.

Disclosure

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

REFERENCES

1. Bahar S, Coban O, Gürvit IH, Akman-Demir G, Gökyiğit A. Spontaneous dissection of the extracranial vertebral artery with spinal subarachnoid heaemorrhage in a patient with Behçet's disease. Neuroradiology. 1993;35(5):352-4.
2. Berlis A, Scheufler KM, Schmahl C, Rauer S, Götz F, Schumacher M. Solitary spinal artery aneurysms as a rare source of spinal subarachnoid hemorrhage: potential etiology and treatment strategy. AJNR Am J Neuroradiol. 2005 Feb;26(2):405-10.
3. Garcia CA, Dulcey S, Dulcey J. Ruptured aneurysm of the spinal artery of Adamkiewicz during pregnancy. Neurology. 1979 Mar;29(3):394-8.
4. Gonzalez LF, Zabramski JM, Tabrizi P, Wallace RC, Massand MG, Spetzler RF. Spontaneous spinal subarachnoid hemorrhage secondary to spinal aneurysms: diagnosis and treatemtn paradigm. Neurosurgery. 2005 Dec;57(6):1127-31; discussion 1127-31.
5. Gutierrez Romero D, Batista AL, Gentric JC, Raymond J, Roy D, Weill A. Ruptured isolated spinal artery aneurysms. Report of two cases and review of the literature. Interv Neuroradiol. 2014 Dec;20(6):774-80.
6. Iihoshi S, Miyata K, Murakami T, Kaneko T, Koyanagi I. Dissection aneurysm of the radiculomedullary branch of the artery of Adamkiewicz with subarachnoid hmorrhage. Neurol Med Chir (Tokyo). 2011;51(9):649-52.
7. Kawamura S, Yoshida T, Nonoyama Y, Yamada M, Suzuki A, Yasui N. Ruptured anterior spinal artery aneurysm: a case report. Surg Neurol. 1999 Jun;51(6):608-12.
8. Kim HJ, Choi IS. Dissecting aneurysm of the posterior spinal artery: case report and review of the literature. Neurosurgery. 2012 Sep;71(3):E749-56; discussion E756.
9. Lavoie P, Raymond J, Roy D, Guilbert F, Weill A. Selective treatment of an anterior spinal artery aneurysm with endosaccular coil therapy. Case report. J Neurosurg Spine. 2007 May;6(5):460-4.

10. Li S. Analysis of 27 cases of large vascular lesions in 161 cases of Behcet's disease: clinical manifestations and treatment outcome. Clin Rheumatol. 2014 May;33(5):671-5.
11. Massand MG, Wallace RC, Gonzalez LF, Zabramski JM, Spetzler RF. Subarachnoid hemorrhage due to isolated spinal artery aneurysm in four patients. AJNR Am J Neuroradiol. 2005 Oct;26(9):2415-9.
12. Nakamura H, Kim P, Kanaya H, Kurokawa R, Murata H, Matsuda H. Spinal Subarachnoid Hemorrhage Caused by a Mycotic Aneurysm of the Radiculomedullary Artery A Case Report and Review of Literature. NMC Case Rep J. 2015 Mar;2(2):49-52.
13. Rengachary SS, Duke DA, Tsai FY, Kragel PJ. Spinal arterial aneurysm: case report. Neurosurgery. 1993;33(1):125-9; discussion 129-30.
14. Son S, Lee SG, Park CW. Solitary ruptured aneurysm of the spinal artery of adamkiewicz with subarachnoid hemorrhage. J Korean Neurosurg Soc. 2013 Jul;54(1):50-3.
15. Tsuda K, Ohkura K, Shintani T, Saito T, Shiiya N. Endovascular treatment of a ruptured innominate artery aneurysm in Behcet disease. Ann Vasc Surg. 2016 May;33:230.e1-4.
16. Vishteh AG, Brown AP, Spetzler RF. Aneurysm of the intradural artery of Adamkiewicz treated with muslin wrapping: technical case report. Neurosurgery. 1997 Jan;40(1):207-9.