



Intracranial subarachnoid hemorrhage caused by an aneurysm at the thoracic spinal region: case report and literature review

Junichi Yokosuka, Shunsuke Fukaya, Shinji Yamomoto, Keisuke Ueki & Phyo Kim

To cite this article: Junichi Yokosuka, Shunsuke Fukaya, Shinji Yamomoto, Keisuke Ueki & Phyo Kim (2020) Intracranial subarachnoid hemorrhage caused by an aneurysm at the thoracic spinal region: case report and literature review, *British Journal of Neurosurgery*, 34:6, 672-676, DOI: [10.1080/02688697.2019.1690130](https://doi.org/10.1080/02688697.2019.1690130)

To link to this article: <https://doi.org/10.1080/02688697.2019.1690130>



© 2019 The Author(s). Published by Informa UK Limited, trading as Taylor & Francis Group.



Published online: 20 Nov 2019.



Submit your article to this journal [↗](#)



Article views: 2082



View related articles [↗](#)




View Crossmark data [↗](#)



Citing articles: 2 View citing articles [↗](#)

Intracranial subarachnoid hemorrhage caused by an aneurysm at the thoracic spinal region: case report and literature review

Junichi Yokosuka , Shunsuke Fukaya, Shinji Yamamoto, Keisuke Ueki and Phyo Kim

Department of Neurosurgery, Dokkyo Medical University, Mibu, Japan

ABSTRACT

Background: The most common cause of intracranial subarachnoid hemorrhage (SAH) is an intracranial aneurysm or other vascular lesion; however, spinal lesions have also been implicated. Furthermore, vascular lesions rarely occur in the thoracolumbar region. We herein presented a case of intracranial SAH caused by an isolated aneurysm in the thoracic spinal artery.

Case presentation: A 79-year-old woman developed the sudden onset of headaches in the parietal and occipital regions followed by vomiting. Head computed tomography (CT) scans showed SAH in the basal cistern and around the parietal lobe cortex. Cerebral angiography detected no aneurysm or vascular malformation. Spinal CT on day 1 showed extensive SAH at the posterior surface of the spinal cord, which was the most prominent at the level of T9/10, and spinal angiography subsequently revealed an aneurysm fed by the T10 radicular artery. The aneurysm was resected by T8-10 laminectomy, and the patient recovered with no long-term neurological deficit.

Conclusions: A literature review revealed 17 cases of intracranial SAH from thoracolumbar vascular lesions. Most cases resulted in poor functional outcomes, which occurred in the later phase of the disease and may have been avoided with earlier diagnoses and interventions. We suggest whole spine CT as a useful tool for rapid screening of this rare lesion, and is recommended when an initial survey for intracranial lesions does not detect any likely lesions. Furthermore, ventricular reflux on head CT may lead to an accurate diagnosis in the absence of spinal symptoms.

ARTICLE HISTORY

Received 1 August 2019
Accepted 4 November 2019

KEYWORDS

Isolated aneurysm
arteriovenous malformation
SAH of unknown origin;
intraventricular haemorrhage; whole spine CT

Background

The most common cause of intracranial subarachnoid hemorrhage (SAH) is an intracranial aneurysm and the initial survey for the bleeding source focuses on intracranial vascular lesions. When it does not detect anything suspicious, further neuroimaging studies focusing on craniocervical region or cervical spine are warranted.^{1–3} However, the source of SAH can be at the thoracic or lumbar level. A previous study reported lesions at the thoracic or lumbar level in 30.5% of intracranial SAH from spinal arteriovenous shunts⁴ On the other hand, surveys for such lesions, which are rare in incidence, are technically demanding, and, thus may lead to underdiagnoses. We herein present a case of intracranial SAH caused by an isolated aneurysm in the thoracic spinal artery. The initial symptoms were headache and vomiting without spinal cord symptoms, and whole spine computed tomography (CT) was useful for identifying the region of abnormal vessels. We also conducted a literature review on intracranial hemorrhage from thoracic or lumbar vascular abnormalities, with a focus on imaging diagnoses.

Case presentation

A 79-year-old woman developed a headache in the parietal and occipital regions followed by vomiting, and was seen at a

community hospital. Head CT revealed SAH, and she was transported to our institution for further treatment. She had a previous medical history of borderline diabetes and was a hepatitis C virus carrier. On admission, she had no focal neurological signs or symptoms including those suggestive of a spinal lesion. The initial head CT scan showed SAH in the basal cistern and the parietal lobe, but not in the Sylvian fissure (Figure 1, upper). Cerebral angiography was performed the next day, but no aneurysm or vascular malformation was detected. Spinal CT was performed due to the unusual distribution of the clot, and revealed extensive SAH at the posterior surface of the spinal cord, which was most prominent at the T9/10 level. T2-weighted MRI showed a mixed intensity mass lesion at the T9-10 level (Figure 2). Spinal intraarterial CT angiography⁵ and spinal angiography was performed, that revealed an aneurysm fed by the T10 radicular artery visible only at the late venous phase (Figure 3). She developed mild weakness and numbness in the left lower extremity on day 5. To prevent re-bleeding from the aneurysm and decompress the spinal cord by hematoma removal, an open surgery was performed. The patient underwent T8-10 laminectomy on day 8. After opening the dura, a dense subarachnoid hematoma was removed meticulously until the aneurysm was visualized. After full exposure of the aneurysm including the feeding and draining vessels, those vessels from/to the aneurysm was temporary occluded. As this procedure did not cause any change in the

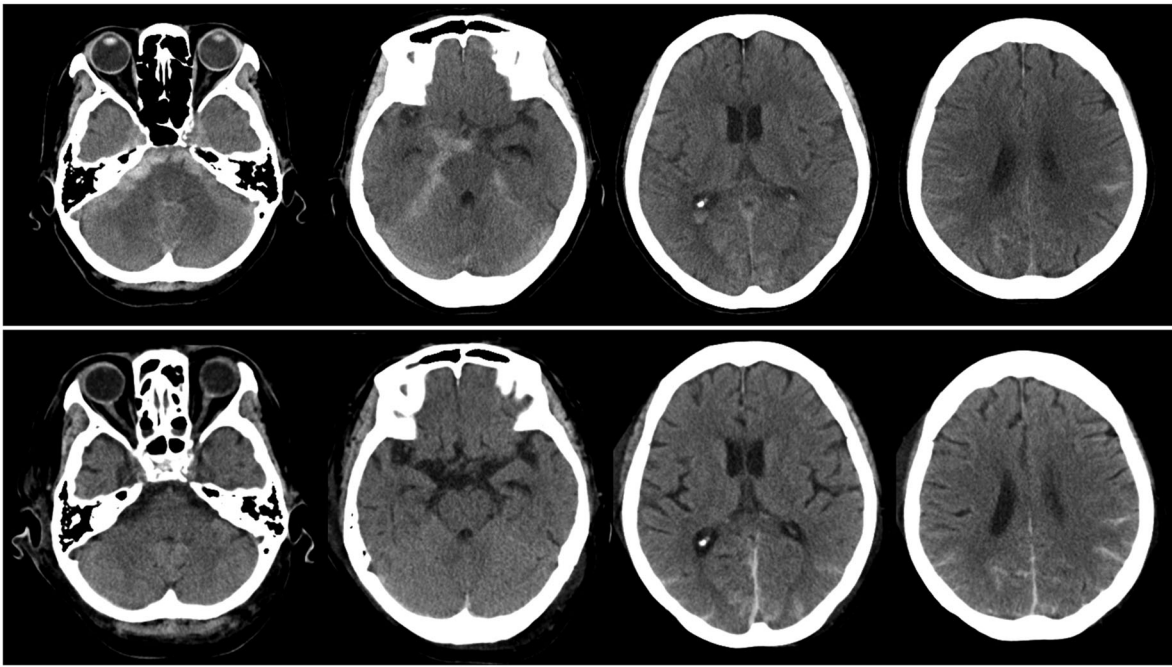


Figure 1. Upper: Initial head CT scan showing subarachnoid hemorrhage in the basal cistern and also around the parietal lobe cortex, but not in the Sylvian fissure. Lower: Head CT scan on day 5. Most of the clot was washed out, leaving a small amount at the parietal lobe sulci.

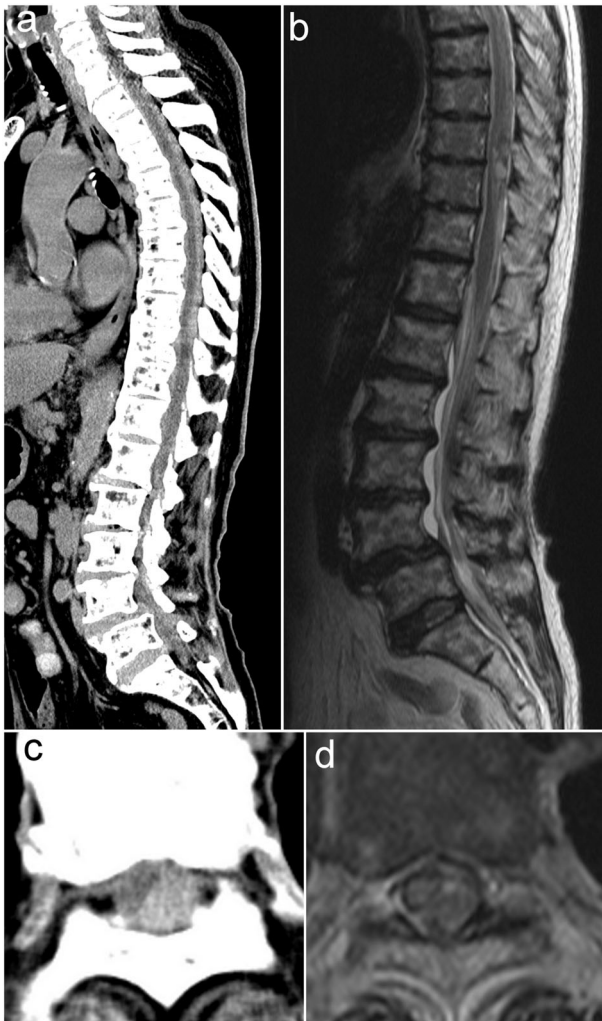


Figure 2. a & c Whole spine CT showing extensive SAH at the posterior surface of the spinal cord, most prominent at the T9/10 level. b & d T2-weighted MRI showing a mixed intensity mass lesion at the T9-10 level.

motor evoked potential, the aneurysm was resected with those vessels. Her postoperative course was uneventful. A head CT scan on day 9 showed that most of the clot was washed out, except for a small amount at the parietal lobe sulci (Figure 1, lower). Her left leg weakness gradually recovered, and, after 8 months, she was fully ambulatory with mild dysesthesia and numbness in her left leg.

Discussion

Despite its relatively low incidence, intracranial SAH from a thoracolumbar vascular lesion may require special attention because the natural course may be complicated by poor functional outcome. To the best of our knowledge, 17 of these cases have been described in the literature (Table 1), with outcomes being reported for 16 cases: 1 died, 1 developed severe developmental delay, 4 had paraplegia, and 3 had paraparesis. Many of the poor outcomes were attributed to treatment delays. In the fatal case, the thoracolumbar vascular malformation re-ruptured shortly after spinal MRI performed under general anaesthesia several days after admission. Spinal arteriovenous malformations were not diagnosed immediately in three out of the four cases of paraplegia, and a survey for a spinal lesion was only performed after the development of spinal symptoms. Three cases had multiple episodes of sudden headaches more than one year before, but remained undiagnosed until the last event. One of those three cases resulted in paraplegia after the last event and subsequent treatment of the disease. Given that intracranial SAH secondary to a cerebral aneurysm is well recognized to require immediate diagnosis and subsequent treatment to prevent re-bleeding and achieve a better prognosis, similar principles may well apply to SAH for spinal lesions. Therefore, whole spine CT may be useful and feasible when an initial survey for intracranial lesions does not detect any likely lesions. State-of-the-art CT technology allows quick examinations of the whole spine.

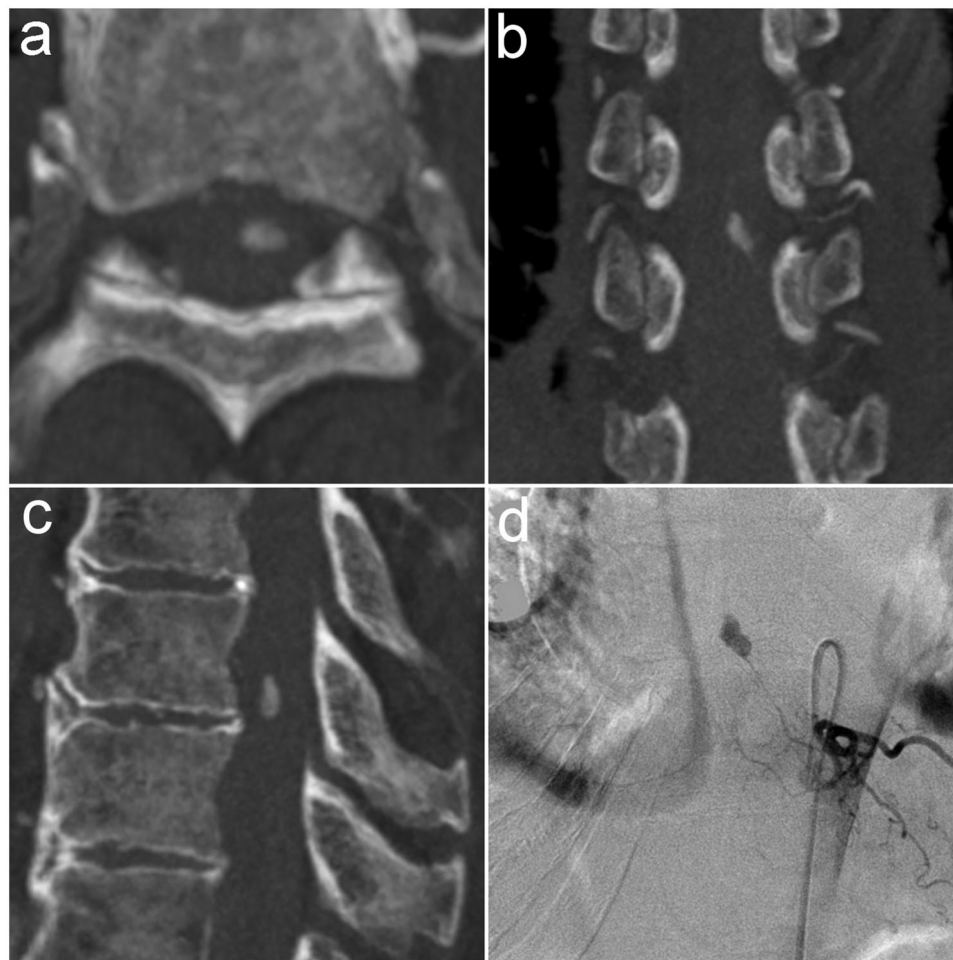


Figure 3. Spinal intraarterial CT angiography (a-c) and spinal angiography (d), showing an aneurysm fed by the T10 radicular artery.

Table 1. Cases of intracranial hemorrhage caused by a thoracic or lumbar vascular lesion.

Reference	Age (yrs, sex)	Level	Lesion type ^a	Hematoma					Hydrocephalus	Presentation	Progression of symptoms	Systemic or motor outcome	Preceding episodes
				Ventricle	Basal cistern	Sylvian fissure	Posterior cranial fossa						
Calhoun ⁶	1 mo, F	L1–3	PAVF	+	+	+	+	Yes	CPA	No	Severe developmental delay, spastic quadriplegia		
Kenning ⁷	14 mos, F	T11–12	PAVF	+	+	–	–	Yes	Fever, lethargy	Yes (paraplegia)	Paraparesis		
Kenning ⁸	2, F	T8–L2	AVM	+	–	–	–	Yes	Lethargy, vomiting, meningismus	Yes (coma)	Dead		
Clark ⁹	4, M	T9–L2	PAVF	+	+	–	–	Yes	LOC, generalized tonic seizures	No	Monoparesis		
Cogen ¹⁰	9, M	T9–12	AVM	NA	NA	NA	NA	NA	Stiff neck	No	Intact	Two SAH over 1.5 years	
Mandzia ¹¹	9, M	T12	PAVF	+	+	–	–	No	LOC	No	Intact	Two episodes of sudden onset headache, back pain, vomiting, fever, and meningismus during the past 2 years	
Herb ¹²	26, M	T5	AVM	+	–	–	–	Yes	headache, nausea, dizziness	Yes (paraplegia)	Paraplegia	Meningitis at age 12, sudden severe headache at age 17	
Barzó ¹³	28, M	T10–12	PAVF	+	–	–	–	No	Headache, LOC, transient respiratory arrest, hemiparesis	Yes (paraplegia)	Paraplegia		
van Santbrink ¹⁴	38, M	L1	PAVF or DAVF	+	+	–	+	No	Headache, neck pain, meningismus	NA	NA		

(continued)

Table 1. Continued.

Reference	Age (yrs, sex)	Level	Lesion type ^a	Hematoma					Hydrocephalus	Presentation	Progression of symptoms	Systemic or motor outcome	Preceding episodes
				Ventricle	Basal cistern	Sylvian fissure	Posterior cranial fossa						
Ohmori ¹⁵	42, M	T7–8	PAVF	–	+	–	–	no	Headache, abdominal and back pain	No	Intact		
Mohsenipour ¹⁶	59, F	T8	IA	NA	NA	NA	NA	NA	Headache, low back pain, loss of pinprick sensations at the T7–T8 level	No	Intact		
Mohsenipour ¹⁷	60, F	T11	IA	+	+	–	–	No	Headache, nausea, back and left leg pain	No	Intact		
Tan ¹⁸	61, M	T5–9	PAVF	–	+	–	–	No	Left-sided chest, arm, and leg pain, paresthesia	No	Intact		
Tanweer ¹⁹	67, F	T11	IA	NA	NA	NA	NA	NA	Paraplegia, sensory loss below T11 level	No	Paraplegia		
Yahiro ²⁰	71, F	T4–5	IA	–	–	–	+	No	Headache, vomiting, paraplegia, bilateral arm numbness, bilateral shoulder pain	No	Paraplegia		
Gonzalez ²¹	73, M	T6–7	IA	NA	NA	NA	NA	NA	Headache, back pain	NA	Intact		
Present case	79, F	T9–10	IA	+	+	–	–	No	Headache, vomiting	Yes (monoparesis)	Intact		

AVM: intramedullary arteriovenous malformation; CPA: cardiopulmonary arrest; DAVF: dural arteriovenous fistula; IA: isolated aneurysm; LOC: loss of consciousness; NA: not applicable; PAVF: perimedullary arteriovenous fistula.

^aThe type of lesion was reassessed based on images or descriptions in the literature.

One index indicating whole spinal CT may be the existence of ventricular reflux on head CT. Ten out of the 13 reported cases had ventricular reflux on initial head CT, and 3 had ventricular reflux only. Washout of the clot around the basal cistern may be quicker in SAH from a spinal lesion, leaving ventricular reflux as the only finding, even at the early stage of the event.

On the other hand, negative neurological findings suggestive of a spinal lesion are common. Half of the reported cases lacked spinal symptoms, while 44% (7/16) had back pain or bilateral sensory or motor dysfunction. Furthermore, spinal symptoms may be difficult to detect in paediatric patients or those with consciousness disturbance who require a cautious approach in order to accurately identify the spinal origin of bleeding. Even if these signs and symptoms develop during the course, cerebral vasospasm may be considered as the cause, and focusing on spinal lesions may be obscured.

Conclusion

We herein reported a case of SAH from a thoracolumbar vascular lesion. Although relatively rare, early diagnosis and treatment should improve patient outcomes. To that end, whole spine CT is a useful tool for rapid screening and is recommended when an intracranial lesion is not detected, or clot distribution patterns including ventricular reflux are detected on head CT. When suspected, spinal intraarterial CT angiography may be useful for the further detection of possible spinal vascular lesions. Physical examinations cannot rule out a spinal origin in general, and negative neurology should not be relied on to rule out.

Acknowledgment

We thank Keisuke Takai, M.D., Ph.D., Department of Neurosurgery, Tokyo Metropolitan Neurological Hospital, for his help in the reassessment of arteriovenous shunts in the literature review.

Disclosure statement

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

ORCID

Junichi Yokosuka  <http://orcid.org/0000-0002-7874-5294>

References

- Elhadi AM, Zabramski JM, Almefty KK, *et al.* Spontaneous subarachnoid hemorrhage of unknown origin: hospital course and long-term clinical and angiographic follow-up. *J Neurosurg* 2015;122:663–70.
- Maslehaty H, Petridis AK, Barth H, Mehdorn HM. Diagnostic value of magnetic resonance imaging in perimesencephalic and nonperimesencephalic subarachnoid hemorrhage of unknown origin. *J Neurosurg* 2011;114:1003–7.
- Maslehaty H, Barth H, Petridis AK, Doukas A, Maximilian Mehdorn H. Special features of subarachnoid hemorrhage of unknown origin: a review of a series of 179 cases. *Neurol Res* 2012;34:91–7.
- van Beijnum J, Straver DCG, Rinkel GJE, Klijn CJM. Spinal arteriovenous shunts presenting as intracranial subarachnoid haemorrhage. *J Neurol* 2007;254:1044–51.
- Yamamoto S, Kanaya H, Kim P. Spinal intraarterial computed tomography angiography as an effective adjunct for spinal angiography. *J Neurosurg* 2015;23:360–7.
- Calhoun AR, Bollo RJ, Garber ST, *et al.* Spinal arteriovenous fistulas in children with hereditary hemorrhagic telangiectasia. *J Neurosurg Pediatr* 2012; 9:654–9.
- Kenning TJ, Deshaies EM, Adamo MA, Waldman JB, Boulos AS. Onyx embolization of a thoracolumbar perimedullary spinal arteriovenous fistula in an infant presenting with subarachnoid and intraventricular hemorrhage. *J Neurosurg Pediatr* 2009;3:211–4.
- Marlin ES, Entwistle JJ, Arnold MA, Pierson CR, Governale LS. Thoracolumbar spinal vascular malformation as a rare cause of isolated intraventricular hemorrhage. *J Neurosurg Pediatr* 2014;14:12–5.
- Clark RS, Orr RA, Atkinson CS, Towbin RB, Pang D. Retinal hemorrhages associated with spinal cord arteriovenous malformation. *Clin Pediatr* 1995;34:281–3.

10. Cogen P, Stein BM. Spinal cord arteriovenous malformations with significant intramedullary components. *J Neurosurg* 1983;59:471–8.
11. Mandzia JL, terBrugge KG, Faughnan ME, Hyland RH. Spinal cord arteriovenous malformations in two patients with hereditary hemorrhagic telangiectasia. *Childs Nerv Syst* 1999;15:80–3.
12. Herb E, Brückmann H, Freudenberger T. Subarachnoid hemorrhage caused by spinal arteriovenous angioma. Diagnostic difficulties and therapeutic possibilities. *Nervenarzt* 1994;65:128–31.
13. Barzó P, Vörös E, Bodosi M. Intraventricular hemorrhage as a false localizing sign of a thoracolumbar arteriovenous malformation: case report. *Surg Neurol* 1999;51:430–4.
14. van Santbrink H, de Witt Hamer PC. Spinal AV malformation. *Lancet* 2003;361:1766.
15. Ohmori Y, Hamada J, Morioka M, Yoshida A. Spinal aneurysm arising from the feeding pedicle of a thoracic perimedullary arteriovenous fistula: case report. *Surg Neurol* 2005;64:468–70.
16. Mohsenipour I, Ortler M, Twerdy K, Schmutzhard E, Attlmayr G, Aichner F. Isolated aneurysm of a spinal radicular artery presenting as spinal subarachnoid haemorrhage. *J Neurol Neurosurg Psychiatry* 1994; 57:767–8.
17. Iihoshi S, Miyata K, Murakami T, Kaneko T, Koyanagi I. Dissection aneurysm of the radiculomedullary branch of the artery of Adamkiewicz with subarachnoid hemorrhage. *Neurol Med Chir* 2011; 51:649–52.
18. Tan AK, Dinesh SK, Lim WE, Ng IH. Sudden severe chest pain: thoracic dural arteriovenous fistula aneurysm rupture with intracranial subarachnoid haemorrhage. *Singapore Med J* 2010;51:e114–7.
19. Tanweer O, Woldenberg R, Zwany S, Setton A. Endovascular obliteration of a ruptured posterior spinal artery pseudoaneurysm. *J Neurosurg Spi* 2012;17:334–6.
20. Yahiro T, Hirakawa K, Iwaasa M, Tsugu H, Fukushima T, Utsunomiya H. Pseudoaneurysm of the thoracic radiculomedullary artery with subarachnoid hemorrhage: case report. *J Neurosurg* 2004;100:312–5.
21. Gonzalez LF, Zabramski JM, Tabrizi P, Wallace RC, Massand MG, Spetzler RF. Spontaneous spinal subarachnoid hemorrhage secondary to spinal aneurysms: diagnosis and treatment paradigm. *Neurosurgery* 2005;57:1127–31.