CASE REPORT

Ruptured aneurysm of the artery of Adamkiewicz: is conservative management the standard of treatment in the current era?

Pedro Aguilar-Salinas,¹ Jussie Lima,² Leonardo B C Brasiliense,³ Ricardo A Hanel,¹ Eric Sauvageau¹

► Additional material is published online only. To view please visit the journal online (<http://dx.doi.org/10.1136/neurintsurg-2017-013194>).

¹Department of Lyrerly Neurosurgery, Baptist Neurological Institute, Jacksonville, Florida, USA

²Department of Neurology, University of Connecticut, Farmington, Connecticut, USA

³Department of Surgery, Division of Neurosurgery, University of Arizona, Tucson, Arizona, USA

Correspondence to

Dr Eric Sauvageau,
Eric.Sauvageau@bmjjax.com

Accepted 3 July 2017

SUMMARY

Spinal aneurysms are rare lesions, and their natural history is not well known. In particular, aneurysms arising from the artery of Adamkiewicz (AA) are an extremely rare phenomenon, and given the lack of solid evidence its management remains unclear. We present the case of a 54-year-old woman with spinal subarachnoid hemorrhage. A spinal angiogram demonstrated a fusiform aneurysm arising from the AA. Based on the location, the absence of evident collateral circulation to the anterior spinal artery, and the available literature supporting spontaneous thrombosis, we opted for conservative management. Fifteen days after the diagnosis, a spinal angiogram demonstrated complete occlusion of the aneurysm with no visualization of the AA. At 10-month follow-up, the patient was able to walk without assistance. Management should be done on a case-by-case basis with the goal of preserving spinal cord perfusion. Similar to previous articles, our report reinforces conservative management as a valid strategy.

BACKGROUND

Subarachnoid hemorrhage (SAH) of the spinal cord accounts for approximately 1% of all SAH cases. Non-traumatic bleeding is most often caused by vascular malformations, followed by intradural neoplasms and rarely ruptured aneurysms.^{1–3} Spinal aneurysms (SAs) are usually found after patients become symptomatic and are often associated with vascular malformation. Ideal management remains challenging due to complex anatomy, especially if

an aneurysm involves the artery of Adamkiewicz (AA). Finding an isolated aneurysm in the AA is a rare event and the treatment remains unclear.^{1 4–8} We present the case of an isolated aneurysm of the AA, rationale for treatment, and an overview of the literature.

CASE PRESENTATION

A 54-year-old woman with history of hypertension presented to the hospital with severe back pain accompanied by headache, nausea, and vomiting. Initial head CT was negative, but a lumbar puncture revealed an SAH. Subsequently, the patient developed lower left extremity (LLE) weakness (motor strength of 3/5). A spinal MRI revealed an SAH at the T8 level, which was compressing and displacing the spinal cord to the right ([figure 1A,B](#)). Cerebral and spinal angiograms were performed. The spinal angiogram demonstrated a fusiform aneurysm measuring 4 mm at its widest point and 8 mm in length arising from the AA. The AA was supplied by the left T10 segmental artery and its diameter at origin measured 0.5 mm ([figure 2A,B](#)). There was no evidence of collateral vessels to the anterior spinal artery (ASA).

INVESTIGATIONS

Differential diagnosis and treatment

Based on previous case reports, aneurysm location, and absence of collateral circulation to the ASA, we discussed with the patient and recommended surveillance with no intervention unless there was evidence of lesion growth. We determined that although re-hemorrhage would unlikely result in death, it could cause paraplegia, and we estimated the risk of spinal cord infarct would be similar for surgical treatment and watchful surveillance. Conservative management consisted of close neuromonitoring, maintaining a systolic blood pressure between 110 and 140 mm Hg, and use of corticosteroids to decrease spinal cord inflammation and edema. Aneurysm was monitored with spinal angiograms and Magnetic Resonance Angiographies (MRAs, [figure 3](#)). On day 3 postdiagnosis, a spinal angiogram demonstrated a partially thrombosed aneurysm with no change in size nor morphology. On day 15 postdiagnosis, the patient's LLE motor function acutely worsened. A spinal angiogram demonstrated complete occlusion of the aneurysm with no visualization of the AA

Figure 1 Thoracic T2-weighted MRI demonstrating intradural extramedullary hypointense signal (black and white arrows), which is displacing the spinal cord at the T8 level on axial (A) and sagittal (B) views.

CrossMark

To cite: Aguilar-Salinas P, Lima J, Brasiliense LBC, et al. *BMJ Case Rep* Published Online First: [please include Day Month Year]. doi:10.1136/bcr-2017-013194

Figure 2 Unsubtracted (A) and subtracted (B) spinal angiogram at the level of left T10 intercostal artery demonstrating a fusiform aneurysm arising from the artery of Adamkiewicz (black arrow) prior to joining the anterior spinal artery (double white arrows).

(figure 4A,B). The patient was later discharged to rehabilitation with 3/5 motor strength in her LLE.

Outcome and follow-up

At 10-month follow-up, the patient could walk without assistance. Her LLE motor strength improved to 4/5 (Glasgow Outcome Scale of 5). Spinal MRI demonstrated no evidence of residual aneurysm but subtle changes of residual cord edema (figure 5).

DISCUSSION

The AA supplies the vascularization of the anterior two-thirds of the thoracolumbar segment. It arises from one of the 8th–12th intercostal arteries and joins the ASA. The AA is mostly found on the left side (up to 75%) and can be found in approximately 75% of the cases from T9 to T12. Although the pathogenesis remains unknown, the data available suggest two factors are involved in non-traumatic SAs: (1) increased blood flow in the spinal circulation, causing shear vessel stress, and (2) inflammatory disorders compromising the vessel wall.¹⁷⁹

Understanding the spine vasculature is paramount in deciding the appropriate management. A spinal angiogram is the first step to evaluate the aneurysm's characteristics, including whether it is associated with vascular malformations. Any intervention carries the risk of AA injury, leading to spinal cord ischemia. Preservation of the parent vessel and the ASA is essential; therefore, surgical risk should be weighed against conservative management. Unlike intracranial aneurysms, most SAs are fusiform, located at non-bifurcation sites, and originating from small parent arteries, making them difficult to treat without a discernible neck to clip or aneurysm sac to coil. Other strategies should be considered, including parent vessel sacrifice, lesion resection, or wrapping. However, all isolated aneurysms of the AA have been described as fusiform in morphology (table 1). From a technical point of view, either microsurgical or endovascular intervention could result in sacrifice of the parent vessel (0.5 mm) and/or thrombosis of the ASA, possibly leading to paraplegia. Although muslin wrapping has been successfully performed on one patient,⁴ we believe the risks associated with surgery itself outweigh the 'wait-and-see' strategy. In our patient, we observed decreased filling of the aneurysm on day 3 postdiagnosis, and the aneurysm was thrombosed by day 15 when the patient's LLE motor function declined.

Figure 3 Spinal Magnetic Resonance Angiography (MRA) obtained 2 days after the diagnosis showing a fusiform aneurysm arising from the artery of Adamkiewicz (single white arrow). The anterior spinal artery (double white arrows) may be seen coursing along the spinal cord.

Based on similar cases, we can merely speculate the management of aneurysms located in the AA (table 1). Including our case, there are eight isolated aneurysms reported in the literature. The mean age of presentation was 49.8 years (range, 30–69). The first case was discovered postmortem, six cases were conservatively managed (including ours), and one was surgically treated. Most cases were female (6/8). There was no evidence of hormone replacement therapy, and with the exception of one case with

Figure 4 Unsubtracted (A) and subtracted (B) spinal angiogram 15 days after diagnosis demonstrating obliteration of the aneurysm and no visualization of the anterior spinal artery.

systemic mycosis,¹ aneurysms occurred as a consequence of vessel dissection. Although observation has been favored in comparison to surgery to a ratio of 6:1, the current body of information does not provide sufficient evidence to establish the optimal management since spontaneous resolution may eventually occur.

LIMITATIONS

This manuscript is limited to a single case report and results may not be generalizable to other patients. Our literature review was restricted to English-language manuscripts, and a comparison between historical cases does not provide evidence to suggest an ideal treatment. Unlike other manuscripts, we found only one surgical case.⁴ Care should be taken when reviewing the literature since the same surgical case has been reported as part of larger series in other articles.^{3 10}

Figure 5 Spinal MRI at 10-month follow-up with no evidence of residual aneurysm and demonstrating subtle changes of residual cord edema with minimal T2 hypointensity that can potentially represent hemosiderin within the thoracic cord.

Table 1 Summary of cases with isolated aneurysms of the artery of Adamkiewicz

Study (year)	Age	Gender	Clinical presentation	Associated comorbidities	Location	Morphology	Aneurysm size (mm)	Treatment	Aneurysm outcome	ASA preserved	Clinical outcome
Garcia <i>et al</i> ^{1*}	34	F	Ruptured during pregnancy, paraplegia	Gestational hypertension	T6	Fusiform	5×3	N/A	N/A	N/A	N/A
Vishteh <i>et al</i> ⁴	30	M	Headache, severe back pain, and paresthesias in lower extremities	Not relevant	T11	Fusiform	N/A	Muslin wrapping	No imaging follow-up	Yes	Full recovery
Berlis <i>et al</i> ¹	48	F	Abdominal pain and paraplegia	Systemic mycosis (<i>Candida</i>) Reduced CD8 T-cell function	T12	Fusiform	5×7	Conservative	Complete occlusion	No	Residual paresthesia
	69	F	Back pain and walking impairment	Not relevant	T12	Fusiform	2.5×8	Conservative	Complete occlusion	Yes	Full recovery
Iihoshi <i>et al</i> ⁶	60	F	Severe headache, back pain, and left lower extremity weakness	Hypertension and hyperlipidemia	T12	Fusiform	2×8	Conservative	Complete occlusion	Yes	Full recovery
Son <i>et al</i> ⁶	45	F	Headache, back pain, and motor weakness in lower extremities	Not relevant	T12	Fusiform	3×11	Conservative	Not reported/Residual intradural collection	Not reported	Full recovery
Dobershtein <i>et al</i> ⁶	59	M	Severe back pain and walking impairment	T-cell lymphoma and Parkinson's disease	T11	Fusiform	N/A	Conservative	Not reported	Not reported	Full recovery
Present case	54	F	Severe headache and back pain	Hypertension	T10	Fusiform	4×8	Conservative	Complete occlusion	No angiographically visualized	Full recovery

* Aneurysm discovered postmortem.
ASA, anterior spinal artery; F, female; M, male; N/A, not applicable.

CONCLUSIONS

Aneurysms arising from the AA are extremely rare, and given the paucity of case reports their standard of treatment remains unclear. Decision on management should be done on a case-by-case basis with the goal of preserving spinal cord perfusion. Our report reinforces conservative management as a reasonable strategy, although options should be considered and discussed with patients.

Contributors PA-S, JL, and LBCB were responsible for study concept and design. JL contributed to acquisition of the data. All the authors were responsible for analysis and interpretation of the data. PA-S, JL, and LBCB contributed to drafting of the manuscript. RH and ES contributed to critical revision of the manuscript for important intellectual content. All the authors were responsible for administrative, technical and material support. RH and ES contributed to study supervision.

Competing interests RH is a consultant for Covidien, Stryker, Codman, and MicroVention. The other authors have no personal, financial, or institutional interest in any of the drugs, materials, or devices described in this article.

Patient consent Obtained.

Ethics approval The Institutional Review Board of Baptist Medical Center.

Provenance and peer review Not commissioned; externally peer reviewed.

© BMJ Publishing Group Ltd (unless otherwise stated in the text of the article) 2017. All rights reserved. No commercial use is permitted unless otherwise expressly granted.

REFERENCES

- 1 Berlis A, Scheufler KM, Schmahl C, *et al.* Solitary spinal artery aneurysms as a rare source of spinal subarachnoid hemorrhage: potential etiology and treatment strategy. *AJNR Am J Neuroradiol* 2005;26:405–10.
- 2 Madhugiri VS, Ambekar S, Roopesh Kumar VR, *et al.* Spinal aneurysms: clinico-radiological features and management paradigms. *J Neurosurg Spine* 2013;19:34–48.
- 3 Gonzalez LF, Zabramski JM, Tabrizi P, *et al.* Spontaneous spinal subarachnoid hemorrhage secondary to spinal aneurysms: diagnosis and treatment paradigm. *Neurosurgery* 2005;57:1127–31. discussion 27–31.
- 4 Vishteh AG, Brown AP, Spetzler RF. Aneurysm of the intradural artery of adamkiewicz treated with muslin wrapping: technical case report. *Neurosurgery* 1997;40:207–9.
- 5 Son S, Lee SG, Park CW. Solitary ruptured aneurysm of the spinal artery of adamkiewicz with subarachnoid hemorrhage. *J Korean Neurosurg Soc* 2013;54:50–3.
- 6 Iihoshi S, Miyata K, Murakami T, *et al.* Dissection aneurysm of the radiculomedullary branch of the artery of adamkiewicz with subarachnoid hemorrhage. *Neurol Med Chir* 2011;51:649–52.
- 7 Garcia CA, Dulcey J, Dulcey J. Ruptured aneurysm of the spinal artery of Adamkiewicz during pregnancy. *Neurology* 1979;29:394–8.
- 8 Doberstein CA, Bouley A, Silver B, *et al.* Ruptured aneurysms of the intradural artery of adamkiewicz: angiographic features and treatment options. *Clin Neurol Neurosurg* 2016;146:152–5.
- 9 Klingler JH, Gläsker S, Shah MJ, *et al.* Rupture of a spinal artery aneurysm attributable to exacerbated Sjögren syndrome: case report. *Neurosurgery* 2009;64:E1010–. 1; discussion E11. doi.
- 10 Massand MG, Wallace RC, Gonzalez LF, *et al.* Subarachnoid hemorrhage due to isolated spinal artery aneurysm in four patients. *AJNR Am J Neuroradiol* 2005;26:2415–9.

Copyright 2017 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow