Unusual presentation of more common disease/injury

Subarachnoid haemorrhage with bilateral intracranial vertebral artery dissecting aneurysms treated by staged endovascular stenting

David Andrew Wilkinson, Thomas J Wilson, William R Stetler Jr, Aditya S Pandey

Department of Neurosurgery, University of Michigan, Ann Arbor, Michigan, USA

Correspondence to Dr Aditya S Pandey, adityap@umich.edu

Summary

Bilateral vertebral artery dissecting aneurysms (VADAs) presenting with subarachnoid haemorrhage (SAH) are an exceedingly rare and deadly clinical dilemma. Prompt intervention in the case of unilateral VADAs is advocated to prevent rebleed; however, in the case of bilateral VADAs, the optimal therapeutic intervention is unclear. We describe the case of a patient presenting with SAH with bilateral VADAs treated by staged endovascular stenting. This led to resolution of the aneurysms with patency of both vertebral arteries.

Stent-based therapy of the symptomatic aneurysm followed by staged stenting of the asymptomatic, contralateral aneurysm appears to be a viable treatment option. This method allows the ability to preserve flow in both vertebral arteries with minimal changes in flow characteristics that may threaten the contralateral, asymptomatic VADA, and appears to be the ideal treatment in these rare cases.

BACKGROUND

Vertebral artery dissecting aneurysms (VADAs) can present with a wide variety of symptoms including neck pain, occipital pain, brainstem ischaemia, tinnitus, spinal cord ischaemia or cervical radiculopathy.¹ A small percentage of VADAs present with subarachnoid haemorrhage (SAH), as approximately 10% extend intracranially.² Intracranial VADAs with SAH present a formidable challenge. Prompt intervention is advocated to prevent further haemorrhage or rupture, since as many as 57% of those who survive a first rupture experience a rebleed within 1 week.³ Simultaneous bilateral VADAs have been reported in the literature, but the rare cases of bilateral VADAs presenting with SAH pose additional challenges from both a diagnostic and therapeutic standpoint.^{4–16} In these cases, the side of rupture may not be apparent, and when attempting to intervene, occlusion of one vertebral artery may cause increased flow in the contralateral vertebral artery with associated haemodynamic stress and possible contralateral rupture.¹³ Several therapeutic options have been proposed under these circumstances, but the ideal treatment modality would preserve flow in both vertebral arteries, secure any aneurysms present, span the length of the dissection and prevent rebleeding. We report a case in which bilateral intracranial VADAs after presentation with SAH were treated successfully with staged stent-assisted coil embolisation resulting in preservation of both vertebral arteries.

CASE PRESENTATION

A 52-year-old woman with a history of hypertension and hyperlipidaemia presented to the emergency room with acute onset right neck pain, tinnitus and severe headache. She was a lifetime non-smoker, had no family history of aneurysmal disease, intracranial pathology or vasculopathy,

and denied any recent head or neck trauma. On neurological examination, the patient was lethargic with a right cranial nerve VI palsy and nuchal rigidity.

INVESTIGATIONS

Head CT showed evidence of SAH in the interpeduncular, prepontine, suprasellar and cerebellopontine angle cisterns with more haemorrhage on the right compared to the left; hydrocephalus was also evident. Diagnostic cerebral angiography revealed a 5 mm dissecting pseudoaneurysm in the intracranial portion of the right vertebral artery and a 2 mm dissecting pseudoaneurysm accompanied by chronic dissection and fusiform dilation in the left vertebral artery (figure 1). The carotids and anterior circulation showed no abnormality.

TREATMENT

The right-side predominance of the SAH suggested that the right VADA was the origin of the bleed. In the endovascular suite, the patient was treated with stent-assisted coil embolisation using a self-expanding stent (Enterprise 4.5 mm×28 mm; Cordis Neurovascular, Inc, Miami, Florida, USA), leaving the right vertebral artery patent (figure 1). She had an uneventful hospital course with resolution of her cranial nerve VI palsy. Two months after the initial operation, the patient underwent elective stenting of her left VADA, with successful stent coverage of the entire dissecting pseudoaneurysm using a self-expanding stent (Enterprise 4.5 mm×22 mm; Cordis Neurovascular, Inc) (figure 2).

OUTCOME AND FOLLOW-UP

Surveillance diagnostic cerebral angiograms performed at 17 and 27 months following the initial treatment revealed progressive resolution of the patient's bilateral VADAs,

Figure 1 Digital subtraction cerebral angiogram at time of presentation and initial treatment of right vertebral artery dissecting aneurysm. (A and B) Anteroposterior (AP) and lateral projections with injection of left vertebral artery showing a 2 mm dissecting pseudoaneurysm (arrows). (C and D) AP and lateral projections with injection of the right vertebral artery showing a 5 mm dissecting pseudoaneurysm (arrows) as well as narrowing of its intracranial course. (E and F) Three-dimensional reconstructions showing right vertebral artery pseudoaneurysm (arrows) as well as narrowing of its intracranial course. (G and H) AP and lateral projections with injection of right vertebral artery following stent-coiling with complete occlusion of the pseudoaneurysm (arrows) and wide patency of the vertebral artery, as well as filling of the posterior inferior cerebellar artery.

with complete resolution at 27 months (figure 3). The left-sided stent was found to have approximately 50% in-stent stenosis while the right had approximately 30% in-stent stenosis. The patient has since been maintained on a regimen of aspirin and clopidogrel.

DISCUSSION

In this report, we describe the case of a patient presenting with a SAH who was found to have bilateral intracranial VADAs. We employed a treatment strategy designed to both secure the aneurysms and preserve flow in both vertebral arteries. This was accomplished by performing

staged endovascular therapy with stent-assisted coiling of the symptomatic aneurysm followed by delayed stenting of the contralateral aneurysm. To our knowledge, this is the first report of stent-based therapy utilised in bilateral intracranial VADAs in the context of presentation with SAH and the first case of bilateral VADAs where both VADAs were treated with flow-preservation. Bilateral VADAs presenting with SAH are an exceedingly rare and deadly clinical dilemma. Prompt intervention in the case of unilateral VADAs is advocated to prevent rebleed; however, in the case of bilateral VADAs, the optimal therapeutic intervention is not clear. A variety of

Figure 2 Digital subtraction cerebral angiogram at the time of treatment of left vertebral artery dissecting aneurysm, and 3 months following treatment of the right vertebral pseudoaneurysm. (A and B) Anteroposterior (AP) and lateral projections with injection of right vertebral artery showing complete occlusion of previously treated pseudoaneurysm (arrows) with no evidence of stenosis. (C and D) AP and lateral projections with injection of left vertebral artery showing a 2 mm dissecting pseudoaneurysm (arrows). (E and F) Three-dimensional reconstructions showing left vertebral artery pseudoaneurysm (arrows). (G and H) AP and lateral projections with injection of left vertebral artery following stent placement showing complete coverage of pseudoaneurysm by the stent.

Figure 3 Digital subtraction cerebral angiograms 17 and 27 months following initial presentation. (A and B) Anteroposterior (AP) and lateral projections with injection of right vertebral artery 17 months following treatment of right vertebral pseudoaneurysm showing complete occlusion of the previously treated pseudoaneurysm with approximately 50% in-stent stenosis. (C and D) AP and lateral projections with injection of left vertebral artery 14 months following treatment of left vertebral pseudoaneurysm showing obliteration of pseudoaneurysm with approximately 50% in-stent stenosis. (E and F) AP and lateral projections with injection of right vertebral artery 27 months following treatment of right vertebral pseudoaneurysm showing complete occlusion of the previously treated pseudoaneurysm with stable in-stent stenosis. (G and H) AP and lateral projections with injection of left vertebral artery 24 months following treatment of left vertebral pseudoaneurysm showing obliteration of pseudoaneurysm without any evidence of stenosis.

therapeutic approaches have previously been undertaken including both open surgical and endovascular approaches. Approaches that have been reported include coil embolisation of the aneurysms with coil occlusion of the symptomatic vertebral artery, initial conservative management without intervention, surgical trapping of the symptomatic VADA only and resection of the symptomatic aneurysm with saphenous vein interposition graft.^{9 13 16 17}

Most of the proposed interventions address the situation in a way that occludes the symptomatic vertebral artery. This leads to increased blood flow in the contralateral vertebral artery and increased stress on the wall of the artery. Increased blood flow leads to increased stress on the internal elastic lamina in patients who likely have some underlying condition causing baseline weakening in the internal elastic lamina. This increased stress ultimately damages the internal elastic lamina resulting in arterial dissection.^{18–20} Within the context of the limited number of cases reported, the highest risk of rupture for patients presenting with bilateral VADAs appears to be after intervention on one VADA.^{13 16 21–23} As in our patient, the use of stent-assisted coiling excludes the VADA without obstructing flow, thereby avoiding a dramatic increase in flow and haemodynamic stress in the contralateral VADA, which likely contributes to high mortality.

Staged, stent-assisted coiling repair of bilateral VADAs appears to be an efficacious approach. While in our patient the second VADA was repaired 2 months after the first, the optimal timing for staged bilateral repair remains uncertain. Even stent-based therapy, however, does cause some alteration in flow that may predispose enlargement of a previously existing contralateral pseudoaneurysm or even formation of a new contralateral pseudoaneurysm.²⁴ For this reason, close follow-up after the initial repair is important in order to be able to promptly recognise any

alteration in the contralateral vertebral artery and intervene prior to any catastrophic event.

In most cases, the underlying cause of spontaneous vertebral artery dissections remains unclear. There are several conditions that predispose to VADAs, but it is likely that a combination of genetic and environmental factors ultimately contribute. Conditions that predispose to VADAs include Ehlers-Danlos syndrome,²⁵ hyperhomocysteinaemia,²⁶ α 1-antitrypsin deficiency^{27 28} and fibromuscular dysplasia.^{29 30} In our case, as in most cases, none of these predisposing conditions were present and there was no family history of aneurysm, SAH or vasculopathy.

For our patient, self-expanding stents were chosen. We believe the gentle, low-pressure release minimises stress on the walls of the aneurysms and minimises the risk of intraoperative rupture. Previous reports of endovascular treatment of unruptured VADAs have suggested the use of balloon-mounted stents due to their reduced porosity in comparison with self-expanding stents, allowing greater flow diversion.²⁴ Our experience with this patient indicates that self-expanding stents provide enough flow diversion to allow thrombosis and healing of VADAs. An alternative method that has been previously described utilises a self-expanding stent as a primary stent followed by placement of a balloon-mounted, lower-porosity stent for further flow diversion.²⁴

In conclusion, we report the case of a patient with SAH with simultaneous bilateral intracranial VADAs treated by staged endovascular therapy and stent-assisted coiling of the symptomatic aneurysm followed by delayed stenting of the contralateral aneurysm. The advantage of this methodology is preservation of flow in both vertebral arteries with minimal changes in flow characteristics that may threaten the contralateral, asymptomatic VADA. This appears to be the ideal methodology for treatment of these rare cases.

Learning points

- ▶ Spontaneous intracranial vertebral artery dissecting aneurysms (VADAs) that present with subarachnoid haemorrhage warrant prompt treatment as the risk of rebleeding is extremely high.
- ▶ Treatment of unilateral intracranial VADAs can result in alteration of blood flow in the contralateral vertebral artery, resulting in increased stress and the development of a contralateral aneurysm. Patients warrant close surveillance of their contralateral vertebral artery for such aneurysms.
- ▶ When bilateral VADAs are present, treatment of the symptomatic aneurysm with a flow-preserving technique should be considered in order to minimise risk of rupture of the contralateral, asymptomatic aneurysm. The asymptomatic aneurysm can then be treated in a staged manner.

Competing interests None.

Patient consent Obtained.

REFERENCES

1. **Ansari SA**, Thompson BG, Gemmete JJ, *et al*. Endovascular treatment of distal cervical and intracranial dissections with the neuroform stent. *Neurosurgery* 2008;**62**:636–46.
2. **Schievink WI**. Spontaneous dissection of the carotid and vertebral arteries. *N Engl J Med* 2001;**344**:898–906.
3. **Mizutani T**, Aruga T, Kirino T, *et al*. Recurrent subarachnoid hemorrhage from untreated ruptured vertebralbasilar dissecting aneurysms. *Neurosurgery* 1995;**36**:905–13.
4. **Chang AJ**, Mylonakis E, Karanasias P, *et al*. Spontaneous bilateral vertebral artery dissections: case report and literature review. *Mayo Clin Proc* 1999;**74**:893–6.
5. **Cohen JE**, Gomori JM, Umansky F. Endovascular management of spontaneous bilateral symptomatic vertebral artery dissections. *AJNR Am J Neuroradiol* 2003;**24**:2052–6.
6. **Fukunaga A**, Tabuse M, Naritaka H, *et al*. Spontaneous resolution of nontraumatic bilateral intracranial vertebral artery dissections. *Neural Med Chir (Tokyo)* 2002;**42**:491–5.
7. **Funayama M**, Mimasaka S, Iwashiro K, *et al*. Spontaneous bilateral dissections of the intracranial vertebral arteries: a case report. *Nihon Hoigaku Zasshi* 1997;**51**:220–5.
8. **Garcia-Monco JC**, Fernandez Canton G, *et al*. Bilateral vertebral artery dissection in a patient with afibrinogenemia. *Stroke* 1996;**27**:2325–7.
9. **Inoue A**, Kohno K, Takechi A, *et al*. Bilateral vertebral artery dissecting aneurysm with subarachnoid hemorrhage treated with staged bilateral vertebral artery coil occlusion: a case report. *Surg Neurol* 2008;**70**:319–22.
10. **Koh JS**, Ryu CW, Lee SH, *et al*. Bilateral vertebral-artery-dissecting aneurysm causing subarachnoid hemorrhage cured by staged endovascular reconstruction after occlusion. *Cerebrovasc Dis* 2009;**27**:202–4.
11. **Leys D**, Lesoin F, Pruvo JP, *et al*. Bilateral spontaneous dissection of extracranial vertebral arteries. *J Neurol* 1987;**234**:237–40.
12. **Mass SB**, Cardonick E, Haas S, *et al*. Bilateral vertebral artery dissection causing a cerebrovascular accident in pregnancy. A case report. *J Reprod Med* 1999;**44**:887–90.
13. **Otawara Y**, Ogasawara K, Ogawa A, *et al*. Dissecting aneurysms of the bilateral vertebral arteries with subarachnoid hemorrhage: report of three cases. *Neurosurgery* 2002;**50**:1372–5.
14. **Redekop G**, TerBrugge K, Willinsky R. Subarachnoid hemorrhage from vertebralbasilar dissecting aneurysm treated with staged bilateral vertebral artery occlusion: the importance of early follow-up angiography: technical case report. *Neurosurgery* 1999;**45**:1258–63.
15. **Weidauer S**, Claus D, Gartenschlager M. Spinal sulcal artery syndrome due to spontaneous bilateral vertebral artery dissection. *J Neurol Neurosurg Psychiatry* 1999;**67**:550–1.
16. **Yasui T**, Sakamoto H, Kishi H, *et al*. Bilateral dissecting aneurysms of the vertebral arteries resulting in subarachnoid hemorrhage: case report. *Neurosurgery* 1998;**42**:162–5.
17. **Nashimoto T**, Komata T, Honma J, *et al*. Successful treatment of bilateral vertebral artery dissecting aneurysms with subarachnoid hemorrhage: report of three cases. *J Stroke Cerebrovasc Dis* 2012;**21**:422–7.
18. **Mizutani T**, Miki Y, Kojima H, *et al*. Proposed classification of nonatherosclerotic cerebral fusiform and dissecting aneurysms. *Neurosurgery* 1999;**45**:253–60.
19. **Stehbens WE**. Etiology of intracranial berry aneurysms. *J Neurosurg* 1989;**70**:823–31.
20. **Wong LC**, Langille BL. Developmental remodeling of the internal elastic lamina of rabbit arteries: effect of blood flow. *Circ Res* 1996;**78**:799–805.
21. **Caplan LR**, Baquis GD, Pessin MS, *et al*. Dissection of the intracranial vertebral artery. *Neurology* 1988;**38**:868–77.
22. **Inamasu J**, Nakamura Y, Saito R, *et al*. Endovascular treatment of ruptured vertebral artery dissection in the acute stage. *Cerebrovasc Dis* 2003;**16**:306–8.
23. **Rabinov JD**, Hellinger FR, Morris PP, *et al*. Endovascular management of vertebralbasilar dissecting aneurysms. *AJNR Am J Neuroradiol* 2003;**24**:1421–8.
24. **Komotar RJ**, Mocco J, Samuelson RM, *et al*. Rapidly successive, symptomatic, bilateral, spontaneous vertebral artery dissections: treatment with stent reconstruction. *Surg Neurol* 2009;**72**:300–5.
25. **Pepin M**, Schwarze U, Superti-Furga A, *et al*. Clinical and genetic features of Ehlers-Danlos syndrome type IV, the vascular type. *N Engl J Med* 2000;**342**:673–80.
26. **Gallai V**, Caso V, Paciaroni M, *et al*. Mild hyperhomocyst(e)inemia: a possible risk factor for cervical artery dissection. *Stroke* 2001;**32**:714–8.
27. **Guillon B**, Bousser MG. (Epidemiology and pathophysiology of spontaneous cervical artery dissection). *J Neuroradiol* 2002;**29**:241–9.
28. **Pezzini A**, Magoni M, Corda L, *et al*. Alpha-1-antitrypsin deficiency-associated cervical artery dissection: report of three cases. *Eur Neurol* 2002;**47**:201–4.
29. **Bornak A**, Milner R. Diagnosing and treating atypical arterial pathologies of aortic arch vessels: dissection and fibromuscular dysplasia. *Semin Vasc Surg* 2011;**24**:36–43.
30. **Libman RB**. Fibromuscular dysplasia with carotid artery dissection presenting as an isolated hemianopsia. *J Stroke Cerebrovasc Dis* 2008;**17**:330.

Copyright 2013 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.

Please cite this article as follows (you will need to access the article online to obtain the date of publication).

Wilkinson DA, Wilson TJ, Stetler WR Jr, Pandey AS. Subarachnoid haemorrhage with bilateral intracranial vertebral artery dissecting aneurysms treated by staged endovascular stenting. *BMJ Case Reports* 2013;10.1136/bcr-03-2012-6002, Published XXX

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