Late-developing posttraumatic dural arteriovenous fistula of the vertebral artery: illustrative case

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BACKGROUND A dural arteriovenous fistula (dAVF) involving the vertebral artery (VA) is a rare vascular pathology that can result from damage to the VA, most frequently following cervical spine trauma. In most traumatic cases, the dAVF develops and manifests shortly after trauma.

OBSERVATIONS A patient was admitted after a fall from the stairs causing neck pain. Computed tomography of the cervical spine revealed a Hangman’s fracture, and angiography showed a left VA dissection. The patient was treated with a cervical brace and clopidogrel. Three weeks after trauma, the patient was admitted because of bilateral leg ataxia, dizziness, and neck pain. Repeat imaging revealed increased displacement of the cervical fracture and a dAVF from the left VA with retrograde filling of the dAVF from the right VA. Embolization of the dAVF using coils proximally and distally to the dAVF was performed prior to placing a halo brace. At 6 months, all symptoms had disappeared and union of the cervical spine fracture had occurred.

LESSONS This case report emphasizes the need for follow-up angiography after traumatic VA injury resulting from cervical spine fracture and underlines important treatment considerations for successful obliteration of a dAVF of the VA.

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KEYWORDS dural arteriovenous fistula; posttraumatic; embolization; vertebral artery

Illustrative Case

A 69-year-old male patient with an extensive cardiovascular history presented to the emergency department after a fall from the stairs. On admission, the patient reported neck pain without neurological symptoms.

Investigation

Computed tomography (CT) of the cervical spine showed a Hangman’s fracture type 2 extending into the left vertebral foramen and a fracture through the spinous process of C3 (Fig. 1). A CT angiography (CTA) revealed a contrast-filling defect of the left VA at the level of C2–C3, suggesting a dissection of the VA, and occlusion of the right internal carotid artery (ICA) (Fig. 2). Pharmacological therapy with clopidogrel was advised (75 mg, 1 × /day). In the absence of neurological symptoms and signs of ligament injury, a cervical brace was recommended with follow-up imaging 2 weeks later. The patient remained stationary for continuous heart monitoring of the suspected heart arrhythmias.
Three days later, the patient developed binocular diplopia, which was related to a vertebrobasilar insufficiency, most likely as a result of the dissection of the VA. Treatment with clopidogrel was continued. Symptoms spontaneously resolved a few days later. After 7 days, the patient was discharged from the hospital.

At the outpatient follow-up visit 2 weeks after trauma, follow-up radiographs of the cervical spine showed increased anterior displacement of the C1–C2 complex in relation to C3 and reduced lordosis (Fig. 3). The absence of neck pain and neurological symptoms warranted observation. Six days later, readmission occurred due to neck pain, bilateral leg ataxia, and dizziness. Repeated radiography and CT of the cervical spine showed further progression of the anterior displacement of C2 in relation to C3 (Fig. 3), and CTA revealed venous dilation, large varices, and possible aneurysmal dilation of the left VA at the level of C2–C3 (Fig. 2).

**Treatment**

Subsequent digital subtraction angiography (DSA) demonstrated abnormal early venous filling and dilated congestive veins in combination with a venous aneurysm at the level of the dissection associated with a dAVF and an absent antegrade filling of the basilar artery from the left VA (Fig. 4). The dAVF was occluded by coil embolization with the use of multiple detachable neuro coils (Stryker Target XL, standard and soft) ranging from 3 to 6 mm and placed proximal and distal to the dAVF.

**Outcomes**

Follow-up DSA demonstrated absent early venous filling and no residual flow in the coil-embolized part of the left VA, suggesting complete obliteration of the fistula. Collateral flow to the posterior circulation via the right VA was sufficient, and retrograde filling of the posterior inferior cerebellar artery (PICA) on the left side was
observed (Fig. 4). Thereafter, the unstable cervical C2 fracture was stabilized externally with a halo brace for 3 months. At 6 months, all symptoms had resolved and radiography demonstrated improved cervical lordosis (Fig. 3).

Patient Informed Consent
The necessary patient informed consent was obtained in this study.

Discussion
Observations
We described the case of a patient developing a dAVF of the VA following cervical spine trauma. Here, it is likely that the patient’s cardiovascular comorbidities, instability of the cervical fracture with increasing displacement, and proximity of the fracture to the transverse foramen preceded development of the fistula. More specifically, the initial trauma was likely to have caused the dissection, which, in turn, may have led to thrombus formation (as suggested by the short episode of symptoms denoting infarction in the posterior circulation). This may have resulted in altered venous hemodynamics, promoting pathological recanalization in the form of a fistula. To improve our understanding of this rare entity, we reviewed and summarized the literature on traumatic dAVFs of the VA (Table 1). Fistulas involving the VA can be asymptomatic due to paired vessel perfusion compensation (Table 1; cases by Andersen et al. and Sarath Chander et al.) or present with symptoms of vertebrobasilar insufficiency, including tinnitus (case by Karakoyun et al.), vertigo, visual changes, ataxia, or lowered level of consciousness. Enlarged or aneurysmatic fistulas with vascular steal manifest as radiculopathy, neck and occipital pain, or myelopathy (Table 1; cases by Avellanosa et al. and Karakoyun et al.). In all but one case, the dAVFs were diagnosed within a few hours to 3 days after admission. In one case, a dAVF developed 8 months after cervical spine trauma with the sudden onset of quadriplegia caused by an epidural hematoma at C1–C5 originating from a dAVF at the level of C1. In that case, however, one cannot exclude the presence of a dAVF at the time of trauma, as no angiography was performed initially. Our case

FIG. 4. Digital subtraction angiography (DSA) of the left VA illustrating the dAVF. Lateral DSA (A) showing an aneurysmal dilation (red arrow) at the level of the left VA with early filling of ectatic veins (blue arrows). Coronal DSA (B) after contrast injection to the right VA showing filling of the left VA and the left posterior inferior cerebellar artery (PICA; yellow arrow), thereby confirming adequate collateral perfusion via the right VA. Coronal DSA (C) after contrast injection to the right VA showing flow from the right VA to the left VA with retrograde flow towards the dAVF (red arrow). Coronal view (D) after contrast injection to the right VA and following distal embolization of the dAVF, showing an absence of filling of the left VA, while flow to the left PICA (yellow arrow), is preserved.
<table>
<thead>
<tr>
<th>Authors &amp; Year</th>
<th>Age (yrs)</th>
<th>Sex</th>
<th>Fracture Type</th>
<th>AVF Location, Side</th>
<th>Timing of Diagnosis of AVF</th>
<th>Presenting Symptoms</th>
<th>AVF Treatment</th>
<th>Cervical Fracture Treatment</th>
<th>Treatment Order</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Avellanosa et al., 1977</td>
<td>54</td>
<td>F</td>
<td>Hangman's fracture, C1 arch fracture</td>
<td>C1, rt VA</td>
<td>1 day later</td>
<td>Increased muscle tone upper extremities</td>
<td>Open vertebral &amp; intracranial clipping after 4 wks</td>
<td>Cervical collar</td>
<td>1st fracture healing, then AVF</td>
<td>Partial neurological symptom remission: remaining ataxia</td>
</tr>
<tr>
<td>Hayes et al., 1980</td>
<td>76</td>
<td>M</td>
<td>C2 anterolisthesis w/ regard to C3, spinous process fracture C4-C5</td>
<td>C2-C3, lt VA</td>
<td>3 days after injury</td>
<td>Neck swelling, cervical bruit, left-sided paresis</td>
<td>Surgical ligation of VA at subclavian level</td>
<td>C2-C3 wiring</td>
<td>1st fracture, then AVF</td>
<td>Respiratory decompensation &amp; death due to pneumonia</td>
</tr>
<tr>
<td>Okuchi et al., 1994</td>
<td>45</td>
<td>M</td>
<td>C1-C2 fracture w/ C2 subluxation</td>
<td>C1-C2, rt VA</td>
<td>Shortly after admission</td>
<td>Uncontrollable nasal bleeding, ataxic respiration, low blood pressure</td>
<td>Failed direct balloon occlusion of fistula; trapping of both proximal &amp; distal ends of involved VA from C5 to C1</td>
<td>Halo</td>
<td>1st AVF, then fracture</td>
<td>Unknown</td>
</tr>
<tr>
<td>Vankan et al., 2004</td>
<td>23</td>
<td>M</td>
<td>Odontoid fracture type I &amp; fracture through both atlantal post arches</td>
<td>C1, lt VA</td>
<td>8 mos after trauma</td>
<td>Sudden upper &amp; lower limb weakness &amp; paresis</td>
<td>Coil embolization</td>
<td>Halo brace followed by cervical collar</td>
<td>1st halo, later dAVF</td>
<td>Neurologically stable</td>
</tr>
<tr>
<td>Heuer et al., 2008</td>
<td>23</td>
<td>M</td>
<td>Bilat C2 pedicle &amp; lat mass fracture toward transverse foramen</td>
<td>C2, rt VA</td>
<td>6 hrs after admission</td>
<td>Decreased strength in upper &amp; lower extremity</td>
<td>Endovascular coiling proximally &amp; distally to transaction</td>
<td>Halo</td>
<td>1st AVF, then fracture</td>
<td>Full neurological symptom remission</td>
</tr>
<tr>
<td>Nishihiro et al., 2016</td>
<td>66</td>
<td>M</td>
<td>C3 fracture toward transverse foramen</td>
<td>C2, rt VA</td>
<td>On admission after trauma</td>
<td>Cervical bruit</td>
<td>Coil embolization</td>
<td>Conservative management</td>
<td>NA</td>
<td>Full neurological symptom remission</td>
</tr>
<tr>
<td>Shiban &amp; Meyer, 2018</td>
<td>87</td>
<td>F</td>
<td>C5-C6 dislocation fracture</td>
<td>Thyrocerical trunk, rt VA</td>
<td>Immediate</td>
<td>Paraparesis lower extremity</td>
<td>Not fully treated due to fistula size &amp; hemorrhage</td>
<td>Pst stabilization C3-C4-T10-T2</td>
<td>1st dissection (partially successful), then op</td>
<td>Death</td>
</tr>
<tr>
<td>Young et al., 2019</td>
<td>55</td>
<td>M</td>
<td>C1-C4 pst cortex fractures</td>
<td>V2-V3, lt VA</td>
<td>Directly at admission</td>
<td>GCS 3</td>
<td>NBCA embolization of recipient vein &amp; endovascular coiling ligation of VA</td>
<td>Occipito-cervical stabilization</td>
<td>1st AVF, then fracture</td>
<td>Partial neurological symptom remission GCS 10</td>
</tr>
</tbody>
</table>

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### TABLE 1. Case reports on traumatic vertebral artery arteriovenous fistulas

<table>
<thead>
<tr>
<th>Authors &amp; Year</th>
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<th>Treatment Order</th>
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</tr>
</thead>
<tbody>
<tr>
<td>Zanin et al., 2020¹³</td>
<td>37</td>
<td>F</td>
<td>C2-C3 dislocation w/ retrolisthesis C2-C3</td>
<td>C3, lt VA</td>
<td>At admission</td>
<td>Neck pain, paraparesis, impaired sensation lower body half</td>
<td>Endovascular embolization proximally &amp; distally to rupture site</td>
<td>Pst cervical decompression of C2-C7</td>
<td>1st AVF, then fracture</td>
<td>Full neurological symptom remission</td>
</tr>
<tr>
<td>Andersen et al., 2021¹⁴</td>
<td>70</td>
<td>M</td>
<td>C1-C2 (lt facet, transverse foramen)-C3 fractures</td>
<td>C2, lt VA</td>
<td>Diagnosis 3 days after</td>
<td>None</td>
<td>Endovascular coiling</td>
<td>Cervical collar</td>
<td>NA</td>
<td>No complications or other symptoms</td>
</tr>
<tr>
<td>Sarath Chander et al., 2021¹⁵</td>
<td>70</td>
<td>M</td>
<td>C2 pars fracture, C2-C3 disc disruption</td>
<td>C2, lt VA</td>
<td>Diagnosis on admission</td>
<td>None</td>
<td>Endovascular coils (Amplatzer device, NBCA)</td>
<td>C2-C3 ACDF</td>
<td>1st AVF, then fracture</td>
<td>Full neurological symptom remission</td>
</tr>
<tr>
<td>Karakoyun et al., 2022¹⁶</td>
<td>65</td>
<td>M</td>
<td>Hangman's fracture C2, C3 corpus, C1 anterior arch</td>
<td>C1-C3, lt V2-V3 segment</td>
<td>Immediate</td>
<td>Directly, weakness lt upper limb, tinnitus lt ear</td>
<td>Endovascular stent coiling</td>
<td>Pst stabilization C1 lat mass, C2 transpedicular, C3-C4 lat mass screws</td>
<td>1st fracture, then AVF</td>
<td>Improved neurological deficit, no complications</td>
</tr>
</tbody>
</table>

ACDF = anterior cervical discectomy and fusion; AVF = arteriovenous fistula; dAVF = dural arteriovenous fistula; GCS = Glasgow Coma Scale; NA = not applicable; NBCA = N-butyl cyanoacrylate; pst = posterior; VA = vertebral artery.

All but one case described AVFs diagnosed immediately at admission up until 3 days after trauma. One case described the occurrence of a symptomatic AVF 8 months after trauma. Nine of the 12 reported cases described endovascular treatment of the AVF with partial to full remission of symptoms. Open surgery was associated with partial recovery and death in two cases.
features the rare, late development of a dAVF. The exact timing of the development of the dAVF, albeit delayed, is nonetheless unknown. The dAVF was not seen on the initial CTA at admission, and no CTA was performed at the 2-week follow-up when the patient remained asymptomatic. It is, however, clear that the patient developed symptoms of the dAVF at a delayed stage (i.e., 3 weeks after trauma). The initial VA injury was grade II (dissection with luminal narrowing) according to the Fassett classification, which progressed into a symptomatic grade V lesion (arteriovenous fistula [AVF]).17 Biffi et al.18 reported that the progression of VA injury occurred in 51% of their series of 97 patients with VA injury. Therefore, repeat CTA 7–10 days after initial diagnosis of a grade I or II VA injury is recommended.18 In our case, no cervical auscultation was performed, which could have raised the suspicion of an AVF before the development of clear symptoms and prompted earlier imaging and treatment.17

The preferred treatment of high-grade VA injuries including dAVFs is endovascular and may include the embolization of fistulas or aneurysms with or without balloon support or (flow-diverting) stent placement (Table 1).19–21 Ideally, the dAVF gets occluded with preservation of the parent vessel. In this case, the extensive dissection, proximal aneurysm formation, and high flow (as suggested by the absence of flow from the left VA to the posterior circulation) over the AVF necessitated occlusion of the left VA. Additional imaging of the right VA was therefore performed, demonstrating adequate flow to the posterior circulation and contralateral PICA (Fig. 4), thereby confirming that it was safe to perform segmental occlusion of the VA while preserving the PICA.

Lessons

This case reinforces that VA injury should be suspected in patients with a traumatic upper cervical spine fracture, especially in cases of a subluxation and involvement of the transverse foramen. VA injury can progress from a low-grade injury to a high-grade injury, thus necessitating follow-up angiography. Subsequent development of a (d)AVF can manifest shortly after cervical spine fracture, but our case demonstrates that late manifestation can also occur. In either case, complete obliteration of the dAVF is warranted, and endovascular embolization has been shown to be an effective treatment with high success rates. Finally, occlusion of the VA may be necessary for complete obliteration of the dAVF. Therefore, careful assessment of flow from the contralateral VA is required to confirm adequate perfusion of the posterior circulation.

References


Disclosures

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

Author Contributions

Conception and design: Schenck, Fodor, Haeren. Acquisition of data: Fodor, Wagemans. Analysis and interpretation of data: Fodor. Drafting of the article: Schenck, Fodor, Wagemans. Critically revising the article: all authors. Reviewed submitted version of the manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Schenck. Administrative/technical/material support: Wagemans. Study supervision: Haeren.

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