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Selective endovascular occlusion of a high-flow cervical direct vertebro-vertebral arteriovenous fistula maintaining vertebral artery patency

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Abstract

INTRODUCTION: Vertebro-vertebral arteriovenous fistulas (VVFs) are abnormal communications between the extracranial vertebral artery and one or multiple neighboring veins. We report an unusual case of a 72-year old woman with a single high-flow VVF between the left vertebral artery and the significantly dilated surrounding epidural venous plexus, located in the intervertebral foramina at the C2 level, after a craniocervical blunt injury, manifesting as progressive severe left arm paresis. METHODS: From a transarterial approach, the fistula venous site was selected with a microcatheter, and several Guglielmi detachable coils were deployed inside the venous part up to the fistula orifice until complete VVF occlusion was obtained maintaining the left vertebral arterial patency. RESULTS: Muscle strength of patient's left arm improved completely. CONCLUSION: Endovascular selective occlusion of the VVFs is the treatment of choice and every effort should be made to preserve parent vessel patency according to fistula's angioarchitecture and physician's technical experience.

Keywords: vertebro-vertebral fistula; endovascular occlusion; coiling

Abbreviations

CTA	Computed tomography angiography
DSA	Digital subtraction angiography
MRI	Magnetic resonance imaging
VA	vertebral artery
VVFs	Vertebro-vertebral arteriovenous fistulas

Introduction

Vertebro-vertebral arteriovenous fistula (VVF) is an abnormal communication between the extracranial vertebral artery (VA) (and one of its muscular or radiculam branches) and one or multiple neighbouring veins without any intervening nidus of vessels or capillaries [1]. Bruit and neck pain are common symptoms; likewise, brain and spinal cord dysfunction may also occur, attributed to blood flow steal, venous hypertension or mechanical compression of nerve roots and spinal cord [2,3]. According to the literature, VVFs are reported to be of traumatic origin in 56 % to 68 % of cases, with the remainder spontaneous [4,5]. Predisposing syndromes for spontaneous VVFs are considered to be fibromuscular dysplasia, neurofibromatosis and Ehlers-Danlos syndrome [6]. Cervical penetrating trauma or blunt injury may result in traumatic VVF that can become symptomatic several weeks later [4].

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We present an unusual case of a cervical VVF due to blunt injury manifesting as progressive severe left arm paresis, treated by selective coil embolisation and maintaining parent vessel patency.

Technique

A 72-year-old-woman presented to the outpatient clinic with a 6-month history of progressive left arm paresis after a craniocervical blunt injury. Cranial nerves were spared. Use of the Medical Research Council scale (MRC) for muscle strength [7] testing (Table 1) indicated grade 2 for the C5-C6 innervated muscles, grade 3 for the C7 and grade 4 for the C8 innervated muscles. No pathological reflexes were demonstrated. Magnetic resonance imaging (MRI) showed an epidural flow void lesion in the cervical spine region, extending from C2 to C8 level that caused mild compression of the spinal cord at the level of the nerve root exit, from the left side (Figure 1). Computed tomography angiography (CTA) and subsequently digital subtraction angiography (DSA) demonstrated a single high-flow VVF between the left VA and the significantly dilated surrounding epidural venous plexus, located in the intervertebral foramina at the C2 level (Figure 2 A,B). The fistula had clepsydroid morphology with a narrow connection between the arterial and venous side. The distal portion of the left VA was faintly and slowly opacified due to arterial flow steal phenomenon through the fistula. Temporary balloon occlusion of the left VA did not show a retrograde opacification of the left PICA through the right VA.

Under general anesthesia, a 6-F guiding catheter was advanced into the left vertebral artery; then a wire-guided microcatheter (Excelsior SL-10, Transend-X, Stryker Neurovascular) was advanced through the orifice point of the fistula into the proximal part of the venous side and 5 detachable coils (3600 GDC, Stryker Neurovascular, 8/20 - 6/15 – 4/8 – 3/6 – 2/4) were deployed inside the venous part up to the orifice (Figure 3 A,B). The high-flow cervical direct VVF was completely occluded maintaining the left vertebral arterial patency with restoration of its distal flow (Figure 4). Our usual anticoagulation strategy during coiling of brain aneurysms is to administrate 5000 IU of heparin IV after deployment of first coil, in order to achieve partial aneurysm protection and avoid thrombotic complications. Nevertheless, in this particular case the coils were deployed exclusively inside the venous part of the fistula, so we considered that the risk of thrombotic complications was low and anticoagulation therapy was not administrated. Muscle strength in the left upper extremity improved completely in a follow-up of 1 year.

Discussion

VVF is a relatively rare entity that is most frequently reported to be of traumatic origin in up to 68 % of cases [4,5]. This may be iatrogenic or secondary to other penetrating injuries such us knife wounds and gunshot injuries followed by blunt trauma [2]. Spontaneous VVFs may be related to congenital abnormalities of the arterial wall. Associated diseases with these cases include fibromuscular dysplasia (increased vascular fragility), neurofibromatosis (mesodermal dysplasia) and Ehlers-Danlos syndrome [1,8].

VVFs have been defined as truncal, with direct communication between the VA and the internal vertebral plexus or internal jugular vein, or non-truncal, with communication between muscular branches of the VA and the venous plexus [4].

VVF may be an incidental finding or be detected in patients presenting with pulsatile tinnitus, cervical bruit or vertebro-basilar insufficiency [9]. Radiculopathy due to cervical VVF is uncommon ranging from 0

%-10 % whereas a smaller percentage of patients presenting with myelopathy (4 %-8 %) have been reported in the literature [3,4,6,10] Venous congestion [11], haemorrhage, arterial steal [5] and spinal cord and/or root compression by dilated veins are possible pathogenic mechanisms.

In our case, the patient became symptomatic with slowly progressive painless left arm paresis several weeks after a blunt craniocervical injury. MRI showed an epidural flow void lesion in the cervical spine region, extending from C2 to C8 level, that caused mild compression of the spinal cord at the level of the nerve root exit from the left side without any intramedullary signal abnormality. The possible pathogenic mechanism in our case is due to a combination of direct compression of the C2 nerve root by the dilated draining vein exiting the neural foramina and distortion of the C3-C8 nerve roots in the canal and mild compression of the spinal cord by the dilated extradural venous plexus.

Surgical management of VVFs is technically difficult due to the fistula anatomical location that necessitates adequate exposure in order to achieve vessel control. Endovascular therapy is an alternative minimally invasive technique with lower risk compared to surgical intervention. In the presence of a patent artery, endovascular reconstruction of the artery using covered stents is also advocated [12,13,14,15]. This technique conveys also the theoretical advantage of preserving the parent vessel patency. This technique may offer a more economic therapeutic option. Disadvantages regarding the long term patency of covered stents used in the vertebral artery, as it is subject to compression from external structures and early cases of stent occlusion, have been described [14].

Embolisation techniques, mostly described in the literature for the treatment of VVFs, include balloon occlusion [2] or a combination of coil and balloon occlusion of the VA, [16] particularly with acute high-flow fistulae to ensure immediate treatment of the lesion. Coil deployment within the vertebral vein after navigation of a microcatheter through the orifice of the arteriovenous fistula have been described [17], resulting in successful occlusion of the fistula; however, in this particular case the formation of a venous pouch did not engaged a risk of coil migration.

In our case the clepsydroid angioarchitecture of the fistula allowed for microcatheter navigation through the narrow orifice up to the proximal venous part, which had a direct broad based communication to the dilated epidural venous plexus that engaged a high risk of coil migration. The key technical point in this case was the diameter selection of the first coil (9 mm), slightly oversized with respect to the vessel diameter, in order to attach firmly to the venous wall and prevent distal migration. The initial coil had also to be long enough in order to create a “stable coil basket” to facilitate further coiling. In this way, complete coiling of the fistula’s venous part has been achieved resulting in complete VVF occlusion and maintaining parent vessel patency. Another option for preventing coil migration could be the temporary balloon occlusion to reduce flow through the VVF. In our case, we considered that the most important risk factor for this theoretical complication was not the flow, as the VVF ostium was narrow enough, but the broad communication of the proximal venous part of the fistula to the dilated epidural venous plexus. From this point of view, we focused on creating an initial steady “coil basket”. Other options such as the double microcatheter technique could offer coil stabilization; however, this choice would be risky in our case as the VVF ostium was too narrow to pass through. Preservation of the VA has been reported in 3/8 patients (37 %) by Goyal et al [6], whereas Beaujeux et al., reported a higher preservation rate up to 93 %; however, only partial closure of 9 % of the fistulae was achieved [11]. If necessary, the VA can be

occluded if the patient tolerates the occlusion test or based on the haemodynamic information obtained by the angiography [4].

On conclusion, the VA should be preserved in case of VVF endovascular occlusion if angioarchitecture of the fistula and physician's technical experience allows for complete coiling of the proximal venous part up to the fistulous point.

Conflict of Interest

We declare that we have no conflict of interest..

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Figures

Figure 1 - (A,B). MRI (T2, A. coronal view, B. sagittal view) shows flow void at C2-C8 level that caused mild compression of the spinal cord at the level of the nerve root exit, from the left side (white arrow).

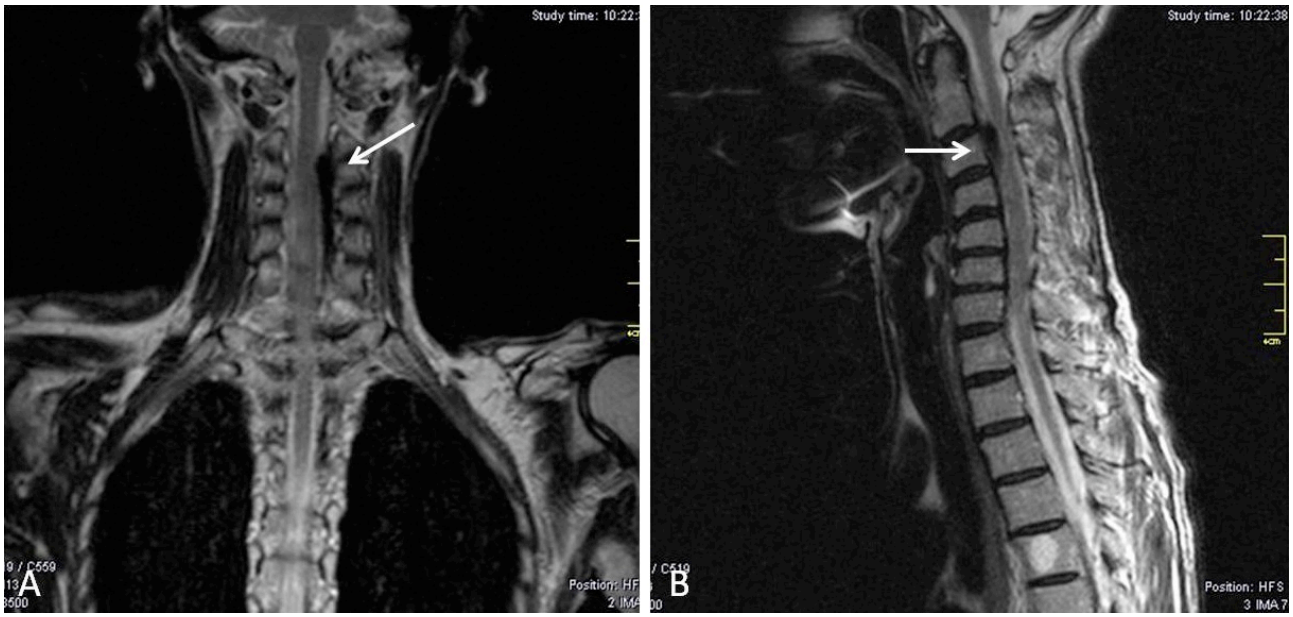


Figure 2 - (A,B). A. CTA demonstrates a single VVF between the left vertebral artery and the dilated surrounding epidural venous plexus, located in the intervertebral foramina at the C2 level (white arrow). B. DSA confirms the presence of the single high-flow VVF. The fistula has a clepsydroid morphology with a narrow connection between the arterial and venous side (white arrow). The distal portion of the left VA is faintly and slowly opacified due to arterial flow steal phenomenon.

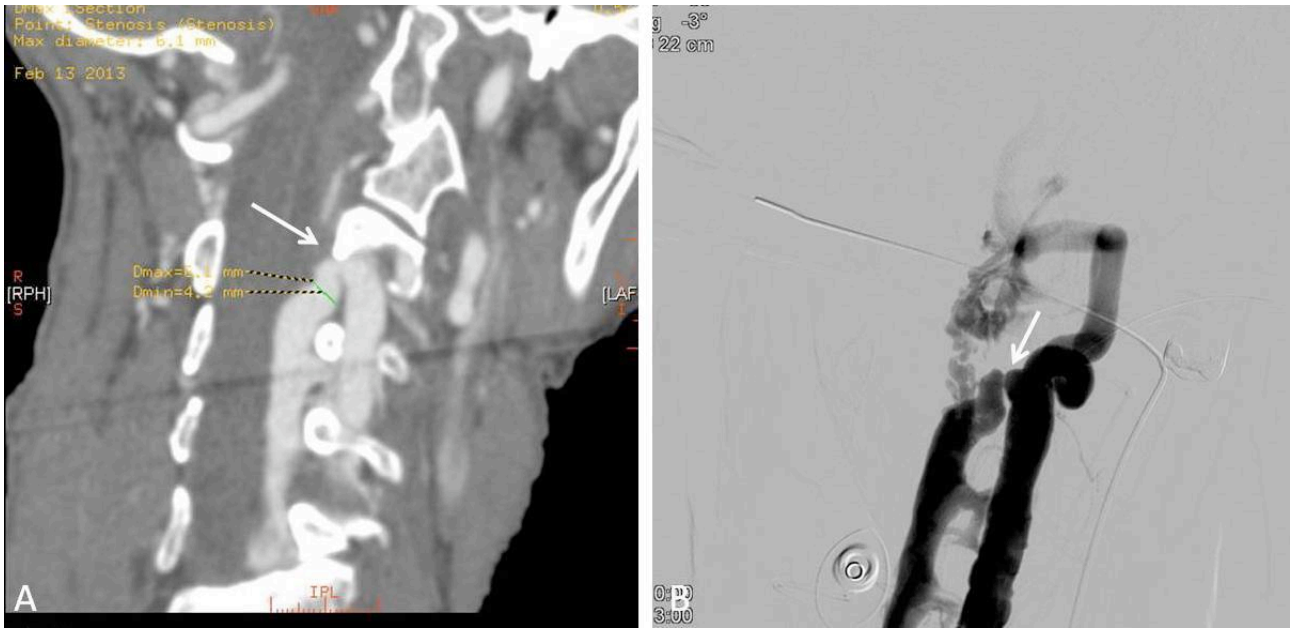


Figure 3 - (A,B). A. The tip of the microcatheter (white arrow) is navigated inside the proximal venous part of the VVF through the narrow orifice and the first coil is deployed. B. Subtracted DSA shows the coiling of the proximal venous part up to the fistulous point.

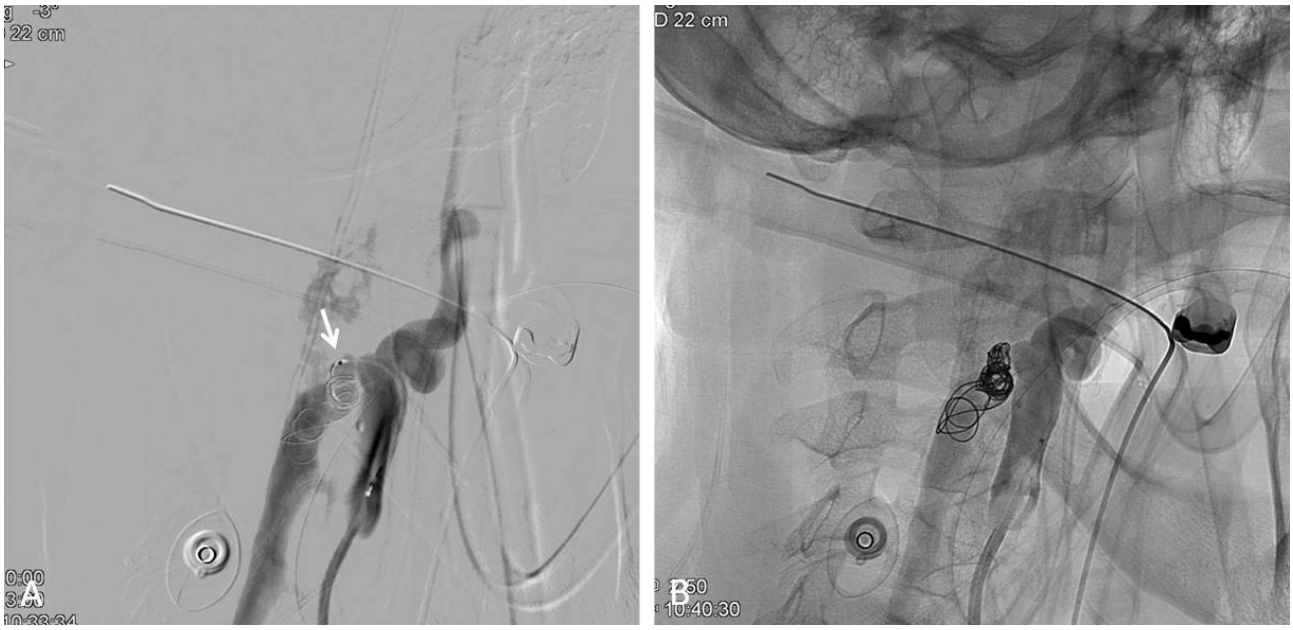
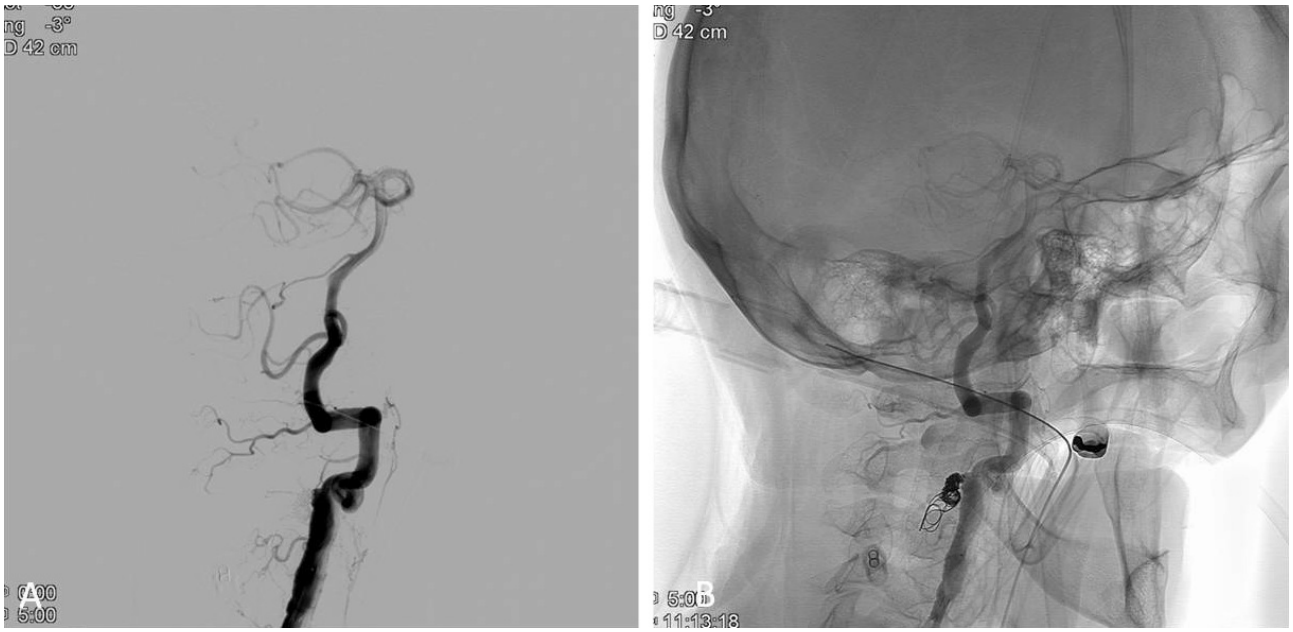


Figure 4 - (A,B). A. The high-flow VVF is completely occluded maintaining the left vertebral arterial patency with restoration of its distal flow. B. Subtracted DSA shows the coil mass and the parent vessel patency.



Tables

Table 1 - Medical Research Council (MRC) Scale for Muscle Strength

EFFORT	0-5 SCALE
Muscle contracts against full resistance	5
Strength reduced, but contraction can still move joint against resistance	4
Strength further reduced such that joint can be moved only against gravity with examiner's resistance completely removed.	3
Muscle can only move if resistance of gravity is removed.	2
Only a trace or flicker of movement is seen or felt, or fasciculations are observed,	1
No movement	0