Endovascular Treatment of Vertebro-Vertebral Arteriovenous Fistula

A Report of Three Cases and Literature Review

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SUMMARY – *This report describes endovascular approaches for occlusion of vertebro-vertebral arteriovenous fistula (VV-AVF) in a series of three cases and a review of the literature. Complete neuroimaging assessment, including CT, MR and DSA was performed in three patients (two female, one male) with VV-AVF. Based on DSA findings, the VV-AVF were occluded by endovascular positioning of detachable balloons (case 1), coils (case 2), or a combination of both (case 3) with parent artery patency in two out of three cases. In this small series, endovascular techniques for occlusion of VV-AVF were safe and effective methods of treatment. To date, there are no guidelines on the best treatment for VV-AVF. Detachable balloons, endovascular coiling, combined embolization procedures could all be considered well-tolerated treatments.*

Introduction

A direct communication between the extracranial vertebral artery or its muscular/radicular branches and epidural venous intervertebral plexus, without intervening capillaries, is termed vertebro-vertebral arteriovenous fistula (VV-AVF) [1,2].

We describe the endovascular approaches used for occlusion of VV-AVF in three patients and the selection criteria, and compare our experience with literature reports of VV-AVF treatments.

The clinical and neuroradiological records of all patients referred to our Institute for the diagnostic and therapeutic work-up of a craniospinal vascular malformation in the decade 2000-2010 were retrospectively reviewed. All patients underwent a complete neuroimaging assessment, including computed tomography angiography (CTA) or magnetic resonance angiography (MRA) of the supraortic vessels and digital subtraction angiography (DSA) to evaluate the best treatment options for fistula occlusion. The clinical and radiological characteristics of our series are summarized in Table 1.

Case 1

A 61-year-old man, with previous multiple fractures at C2-C6 level and epidural hematoma at C1-C4 after a recent cervical trauma, presented rigor nucalis and inferior limbs weakness. MRA showed a right ectasic vertebral artery (VA) at the anterior epidural level (Figure 1A). DSA showed a direct VV-AVF between the right VA at C2 level and periradicular venous plexus (Figure 1B).

An endovascular approach was chosen. Systemic heparinization was given seven days before the procedure. Before sacrificing the vessel an occlusion test was performed. Due to a good collateral circulation, a right VA occlusion was planned. An 8 Fr introducer sheath (Cordis) was placed in the right common femoral artery route and the right VA was catheterized

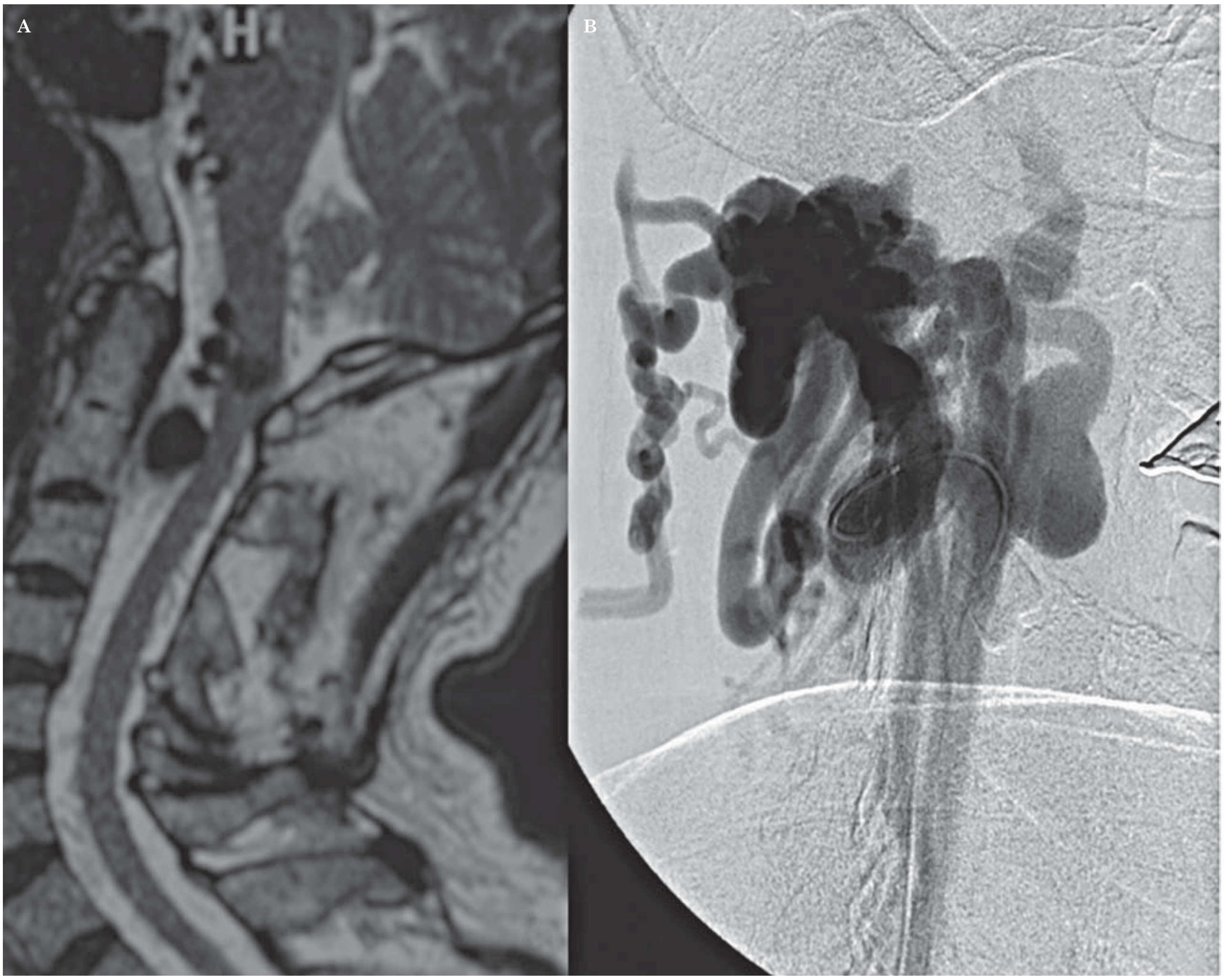

Figure 1 A) Sagittal T2-w MRI shows a right ectasic vertebral artery at the anterior epidural level; B) Preoperative right vertebral artery DSA shows a direct VV-AVF between the right VA at C2 level and the periradicular venous plexus.

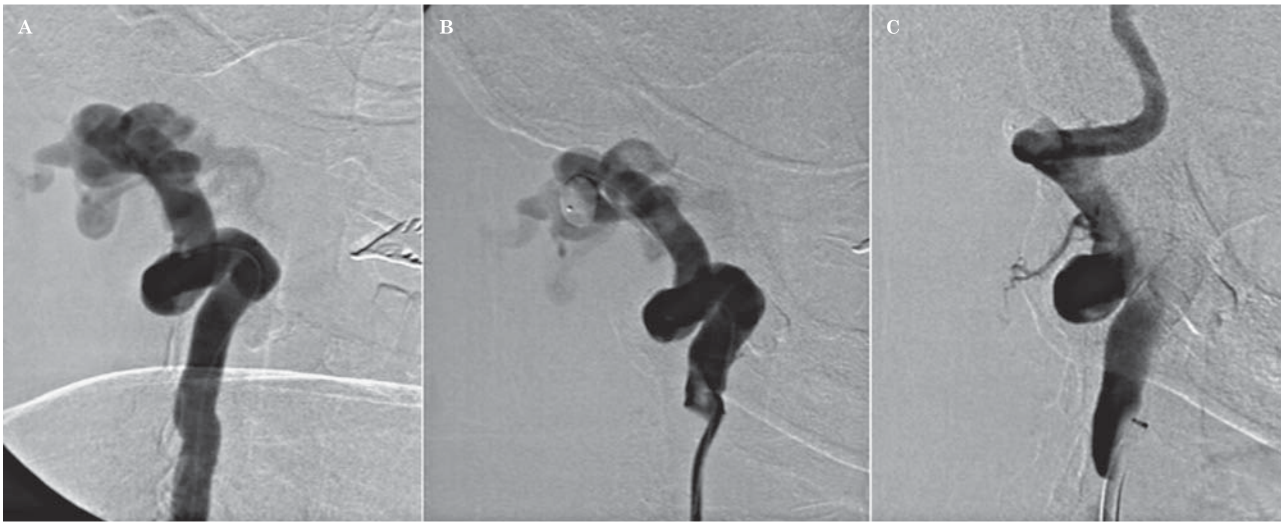

Figure 2 Operative DSA (A-C): the balloon is detached at the fistula point with preservation of parent vertebral artery patency.

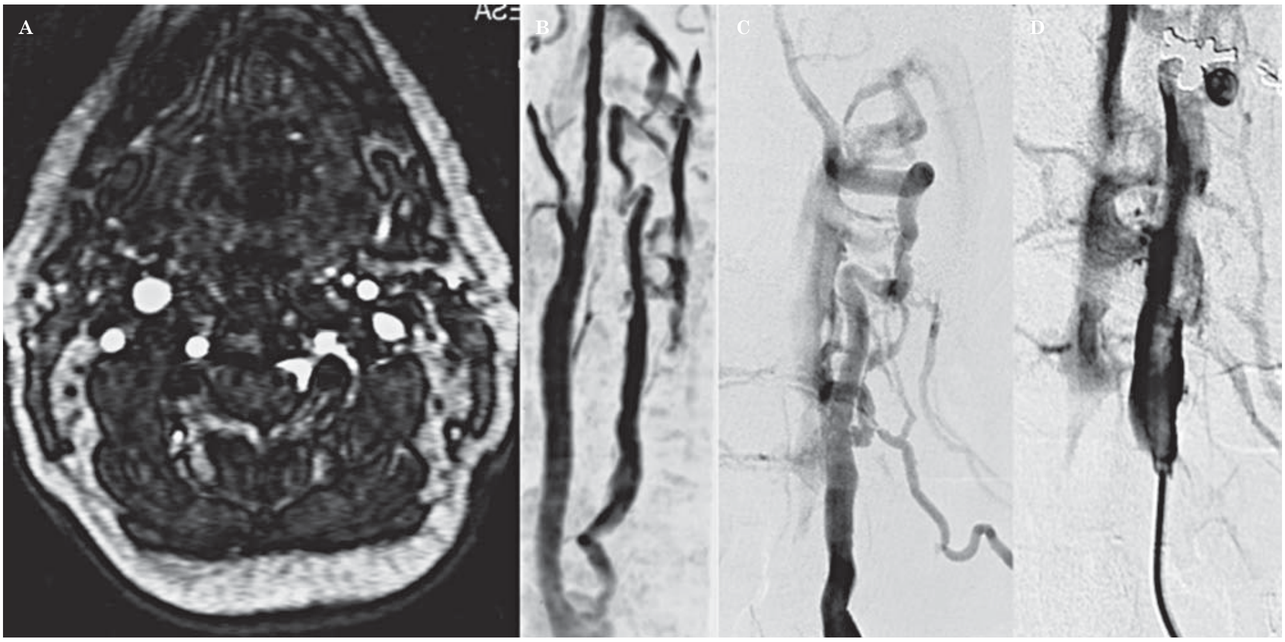

Figure 3 Axial MRA TOF 3D (A) and MIP reconstruction (B) showing an extradural fistula of the vertebral artery involving the perivertebral venous plexus at C3-C4 level. Vertebral artery DSA (C,D) shows a dilated extracranial segment of the right vertebral artery between the muscular branches of the vertebral artery and a dilated periradicular venous plexus.

with a 8 Fr. The site of the fistula was less evident, the large size of the single arterial communication and the high flow prompted an endovascular balloon occlusion of the VV-AVF. A BAL1XRAY (Balt, Extrusion, Montmorency, France) mounted on a MABDTE (Balt Extrusion, Montmorency, France) microcatheter was maneuvred into the right VA and positioned in the fistulous tract under fluoroscopic control. The balloon was inflated with 50% of 300 mg I/ml iohexol and 50% of saline, and after satisfactory occlusion of the AVF, was detached at this site. Postembolization angiograms showed immediate occlusion of the fistula with no early filling of the epidural veins (Figure 2A-C).

Case 2

A 56-year old woman presented with cervical pain radiating down to the right arm. She also reported a pulsatile bruit on the right side of the neck which, in auscultation, was spreading over the chest. Physical and neurological examinations were otherwise unremarkable and medical history was negative for surgical or endovascular procedures. A history of chiropractic manipulation to alleviate the pain was referred. MRA revealed an extradural fistula of the vertebral artery at the C3-C4 level (Figure 3A,B). DSA showed a dilated extracranial segment of the right VA and a high flow communication between the muscular branches of the VA and the periradicular venous plexus (Figure 3C,D).The intervention was performed under general anesthesia and the systemic heparization protocol was the same as adopted in the first patient. A 6 Fr introducer sheath (Cordis) was placed in the right common femoral artery route and the right vertebral artery was catheterized with a 6 Fr. A microcatheter (Cordis, Prowler plus 0.14) was placed proximal to the arterial edge of the fistula and coiling was attempted with two electrodetachable coils (Micrus, 4×15 and 6×25, respectively). Immediate occlusion of the fistula was reported. Angiographic post-procedural evaluation confirmed complete occlusion of the fistula with preservation of right vertebral artery patency. The patient experienced no complications and had a total regression of symptoms (Figure 4A-C).

Case 3

A 34-year-old woman with neurofibromatosis type 1 (NF1) presented with a four-month history of bilateral tinnitus, objective vertigo and right arm paresthesia. No history of trauma was referred. A supraaortic vessels color Dop-

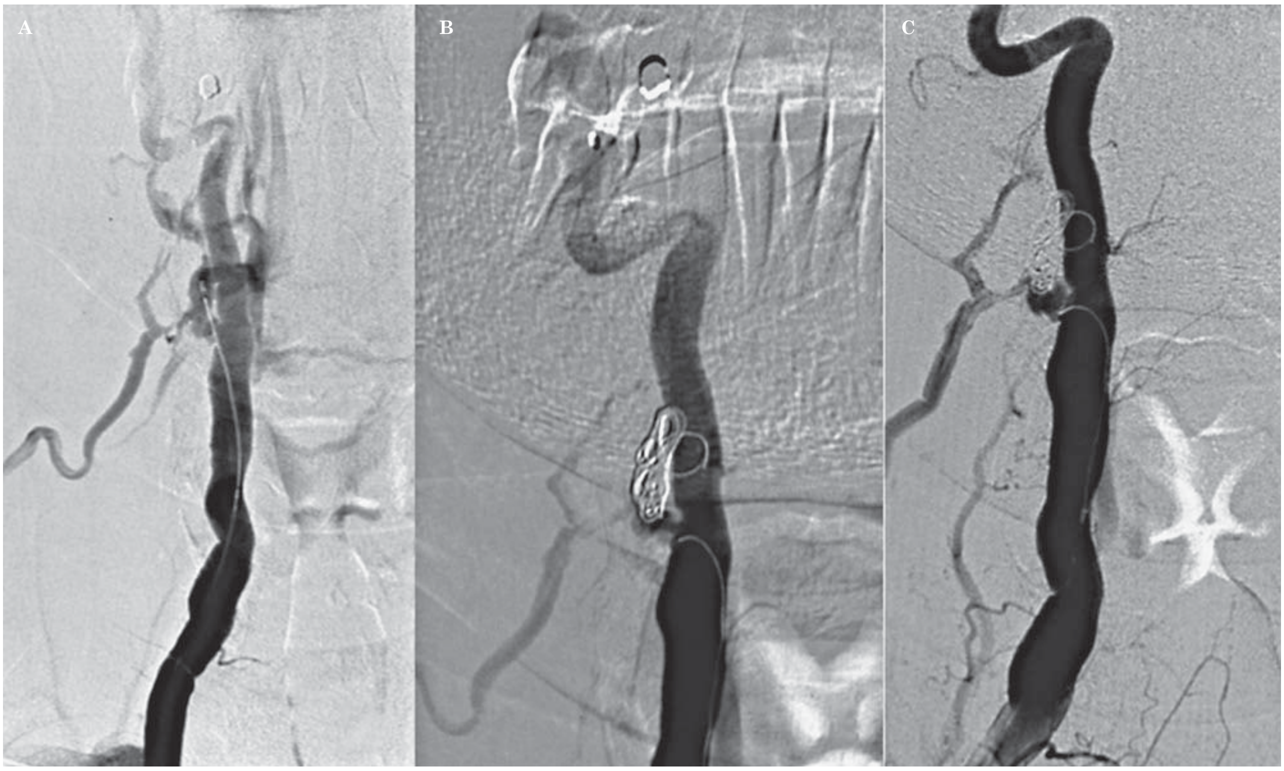

Figure 4 Vertebral artery operative DSA (A-C) shows complete occlusion of the fistula with coils and preservation of right vertebral artery patency.

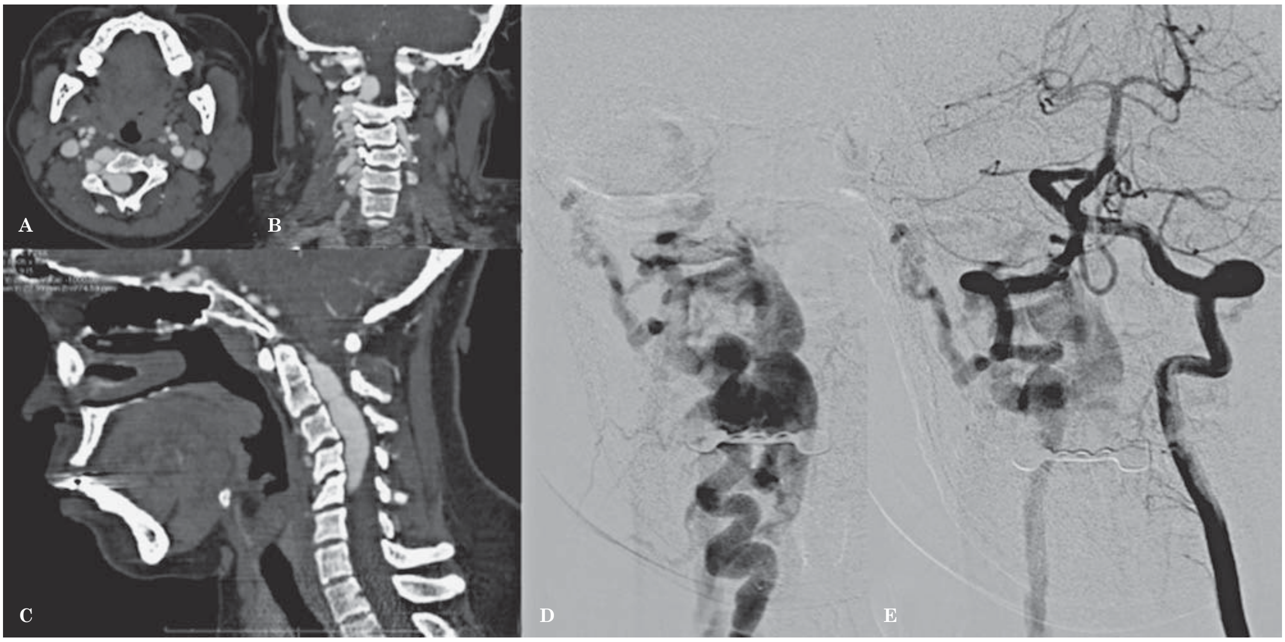

Figure 5 Contrast-enhanced CT angiography, MPR reconstruction (A) axial (B) coronal (C) sagittal, shows an extradural fistula of the right VA with multiple arteriovenous high-flow shunts at C2-C5 level. Pre-operative DSA right (D) and left (E) VA injection: large VVAF in NF1 with flow steal.

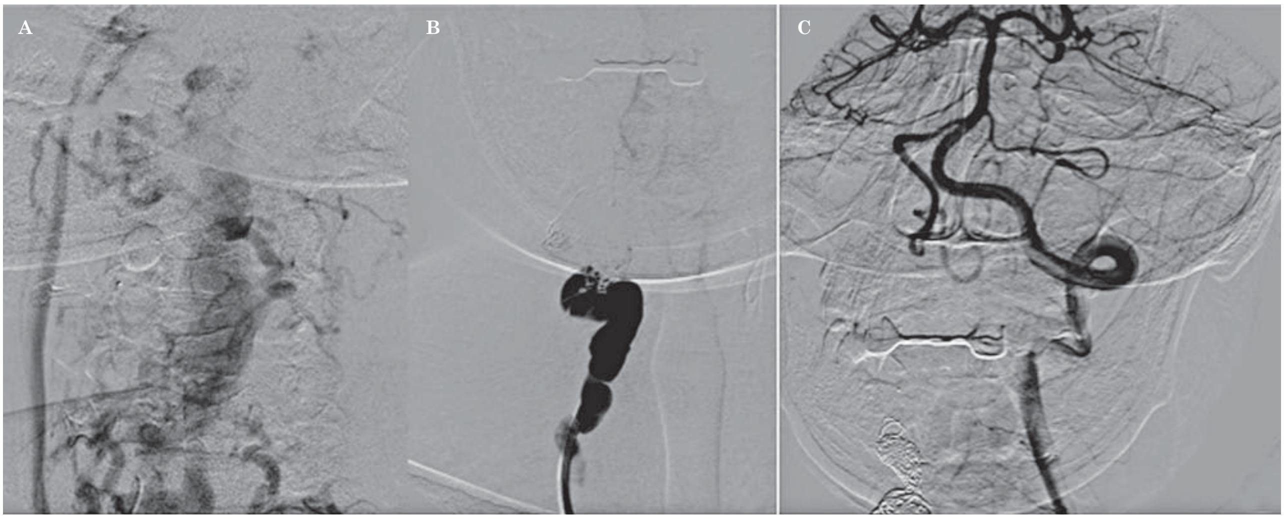

Figure 6 Operative DSA: partial occlusion with detachable balloon (A). Complete VA and fistula occlusion with further coiling (B). C) Post-embolization DSA by controlateral VA injection shows patency of the right PICA.

pler examination was negative. An arterial puff was discovered on the jugular vein. CTA showed an extradural fistula of the right VA with multiple arteriovenous high flow shunts at C2-C5 level (Figure 5A-C). DSA confirmed the diagnosis (Figure 5D,E).

An endovascular approach was performed. An 8 Fr introducer sheath (Cordis) was placed in the right common femoral artery route and the right vertebral artery was catheterized with a 8 Fr A BAL1XRAY detachable latex balloon (Balt, Extrusion, Montmorency, France) mounted on a MABDTE (Balt Extrusion, Montmorency, France) microcatheter maneuvered into the right VA and positioned at the fistulous tract under fluoroscopic control. The VA was sacrificed due to a good collateral circulation (Figure 6A). DSA control after two months showed partial reperfusion of the VV-AVF so endovascular VA occlusion was undertaken with coils (Figure 6B). Control angiograms from the controlateral VA showed complete occlusion of the fistula and opacification of the right posterior inferior cerebellar artery (PICA) from the left VA (Figure 6C). The peri and postprocedural periods were uneventful and the patient experienced no complications and had a total regression of symptoms.

Discussion

VV-AVF is uncommon and consists of an abnormal communication between the extracranial vertebral artery or its branches and the deep venous plexus [1]. The most common cause of VV-AVF is penetrating neck injury, blunt cervical trauma, iatrogenic forms of trauma including direct percutaneous puncture of a carotid or vertebral artery for diagnostic angiography, or vertebral artery injury during insertion of central venous catheters [2,3]. Dysplastic conditions of the vascular wall such as NF1, fibromuscular dysplasia or Ehlers-Danlos should be considered predisposing factors [4-11]. Whatever the pathogenesis, VV-AVF may be asymptomatic or become symptomatic due to compression on the spinal cord or roots, bleeding, or vascular steal.

A surgical approach to VV-AVF is no longer warranted in most circumstances. The classic surgical procedure is based on direct surgical ligature of punctiform fistulas and the use of bypass venous graft from the subclavian to the distal vertebral artery for large, complex fistulas with several arterial feeders. Steal flow symptoms, massive hemorrhages and injuries to surrounding structures have been associated with surgical treatment [2].

As symptoms are usually progressive, prompt treatment is needed: the endovascular occlusion of the VV-AVF is considered the first therapeutic option, with a variety of devices developed to achieve fistula occlusion while possibly preserving the parent VA patency [2,9]. As with surgery, periprocedural and postprocedural ischemic or thromboembolic complications and vessel injuries can also occur [2,12]. Detachable balloons [3,13-14], stent grafts [15-20], and detachable

Table 1 Clinical and radiological characteristics of patients.

	Age/Sex	*Location*	*Etiology*	*Symtoms*
1	61/M	Right VA C2-C3	Previous multiple fractures at C2-C6 level.	Rigor nucalis and inferior limbs weakness
2	56/F	Right VA C3-C4	Spontaneous; chiropractic manipulation	Cervical pain radiating down to the right arm
3	34/F	Right VA C2-C3	Secondary (NF1)	Bilateral tinnitus, objective vertigo, right arm paresthesia

Table 2 Review of the literature.

Author	*Patients*	*Device*	*Flow/size*	*Level*	*Cause*	*Parent VA*
Modi et al. [4]	1	Balloon	High/Large	C3	Spontaneous	Patent
Reddy et al. [5]	1	Coils (failed); Surgery	–	C2-C3	Traumatic	Sacrificed due to coiling
Shirikawa et al. [7]	1	Coils	High/Large	C6	Traumatic	Sacrificed
Sadato et al. [8]	1	Stent	High/Large	C4-C5	Spontaneous	Patent
Gonzalez et al. [9]	1	Stent	High/Small	C7	Iatrogenic	Patent
Benndorf et al. [15]	1	Coils (failed); Coils (transvenous)	High/Giant	Low	NF1	Sacrificed
Kahara et al. [16]	1	Coils	High/Large	C3-C4	NF1	Sacrificed
Siddharta et al. [17]	2	Coils (1); Balloon (1)	High/Large (both)	C5 (both)	NF1	Sacrificed (both)
Hauck et al. [18]	1	Coils (failed); Coils and surgery.	–/Large	C3-C4	NF1	Sacrificed
Gao et al. [19]	2	Balloon, coils and glue;	High/Large (Both)	C2-C3 C5	NF1	Sacrificed
Paolini et al. [20]	1	Balloon (failed); Surgery	High/Large	C2-C3	NF1	Sacrificed
Patro et al. [21]	1	Surgery (failed); Coils	High/Large	C5-C6	NF1	Sacrificed
Briganti et al. [22] and Present Report	3	Balloon (1) Coils (1) Balloon and coils(1)	High/Large High/Small High/Large	C1-C4 C3-C4 C2-C5	Traumatic Chiropractic NF1	Sacrificed Patent Sacrificed
Taylor et al. [24]	2	Coils (1); Balloon (1)	High/Large (Both)	C1-C6 C5-C6	Traumatic Iatrogenic	Sacrificed (both)
De Keukeleire et al. [25]	1	Coils	High/Large	C2	Spontaneous	Sacrificed
Wang et al. [26]	1	Coils and glue	High/Large	C3	Spontaneous	Sacrificed
Mortimer et al. [27]	1	Coils	–	Low	Traumatic	Sacrificed
Kai et al. [28]	2	Coils (venous)	Low/ Small	C6 C1	Iatrogenic	Patent (Both)
Herrera et al. [29]	4	Balloon	Low/Small	Various	Traumatic (all)	Sacrificed (All)
Heuer et al. [30]	1	Coils	High/NA	C2	Traumatic	Sacrificed
	29					

coils [21-27] are the embolic materials that have been used to treat VV-AVF. Particulate embolic agents and cyanoacrylate glue are avoided in the treatment of high-flow fistulas, as they are likely to be swept away by rapid flow without occlusion of the fistula. Endovascular treatment with a detachable balloon to occlude the fistula is preferred over microsurgical ligation, coil embolization, or stent-grafting. Detachable balloons are most suitable for occlusion of fistulas because they can be repeatedly inflated and deflated prior to their detachment so that precise placement and optimal occlusion of the fistula can be achieved [3,13-14]. However the balloon could deflate with a recurrence of the fistula. When the fistula is not well seen or masked by ectasic vessels, detachable coils, used in our case 2, show advantages such as electrically induced thrombosis with no recanalization and retrievability when the position is not satisfactory. These advantages help to reduce the risk of embolic complications, especially in high-flow fistulas, preserving the patency of the adjacent artery [21,28]. Experience with endovascular stent-graft repair is limited [15-20]. These devices are feasible and offer a therapeutic alternative in the treatment of VV-AVF, particularly for preserving vessel patency. However, these devices are currently very rigid and not suitable if vessels are tortuous [15-20]. This minimally invasive treatment may become of the choice management of this kind of lesions, but at the moment should be considered experimental [18,29-33].

In particular situations, endovascular treatment can be very difficult and a complex approach is required. Gao et al. [9] reported two cases of VV-AVF in two patients with NF1, successfully treated with a combined complex approach. In one of these patients, both right and left VAs were catheterized, respectively: a detachable balloon was placed in the VV-AVF varix to provide flow control, which allowed to three coils to be deployed together for obliteration; an appropriate mixture of glue was then used to avoid coil migration.

Reviewing the recent literature (Table 2), endovascular treatment with detachable balloons, alone or with other devices, was used in 12/29 patients (41%): all VAs, except one [3], were sacrificed with no clinical outcome. In all cases this approach was therapeutic, except for one which required surgery [10]. Endovascular coiling alone or with other devices was performed in 16/29 patients (55%): all VAs were sacrificed except for three [21,27] with no neurological symptoms. Endovascular stenting was performed in 2/29 (6%) patients: both VAs were patent after the procedure [15,16]. Detachable balloons, endovascular coiling, or combined embolization treatment should be chosen by evaluating the fistula and parent vessel anatomy, hemodynamics and flow (high or low) [29,30,34-37]. According to our experience if the AVF flow is low and the point of fistula is evident at DSA, as in our case 2, progressive coil occlusion is recommended. For high flow VV-AVF, when the VA sacrifice is possible and well-tolerated and the point of fistula is less evident, as in our cases 1 or 3, balloon occlusion or combined treatment (coil and glue; coil and balloon) are advised.

Conclusion

In this small series, endovascular techniques for occlusion of VV-AVF were safe and effective methods of treatment. In expert hands, even the sacrifice of the parent VA can be accomplished without complications. To date, there are no guidelines on the best treatment of VV-AVF, and detachable balloons, endovascular coiling, and combined embolization procedures can all be considered well-tolerated treatments. No rules for flow diverter devices are currently available.

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