Flow Diverter for the Treatment of Pseudoaneurysms of the Extracraneal Vertebral Artery: Report of Two Cases and Review of the Literature

Pedro Plou[1], Federico Landriel[1], Sofía Beltrame[1], Santiago Hem[1], Oscar Peralta[2], Ricardo García-Mónaco[2], Claudio Yampolsky[1]

Key words
- Spine flow diverter
- Spine pseudoaneurysm
- Vertebral artery pseudoaneurysm

Abbreviations and Acronyms
AVF: Arteriovenous fistula
CT: Computed tomography
PA: Pseudoaneurysm
VA: Vertebral artery

From the [1]Neurosurgery Department and [2]Diagnostic Imaging Department, Hospital Italiano de Buenos Aires, Buenos Aires, Argentina

To whom correspondence should be addressed: Federico Landriel, M.D.
[E-mail: fedelandriel@gmail.com]

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BACKGROUND: **Pseudoaneurysms (PAs) of the extracranial vertebral artery (VA) are rare lesions, representing less than 1% of all aneurysms. Although these lesions may resolve spontaneously, they present a high rupture rate, so early preventive treatment is advised.**

CASE DESCRIPTION: ***Case 1*: A 48-year-old woman presented with pain and cervical rigidity. An angiotomography showed a PA of the left VA at the level of the C2 transverse foramen, with mural thrombosis and bone remodeling of the left lateral mass. The PA was treated with the endovascular placement of a flow diverter stent. The patient was discharged 3 days after the procedure without complications. The last vascular imaging follow-up was performed 6 years after the procedure showing a patent left VA, with complete resolution of the aneurysm. *Case 2*: A 57-year-old woman was admitted referring cervicalgia after a polytraumatism. An angiotomography revealed a fracture of the C1 posterior arch, lateral mass, and left transverse foramen, on top of a left VA thrombosis due to a vascular dissection. Eleven months after the trauma, a left VA V3 segment arteriovenous fistula developed. It was treated with hydrocoils, with no complications. One month after the embolization, a left VA V3 segment PA was observed and treated with a flow diverter stent. An angiographic follow-up 2 years after the procedure showed a patent left VA, with complete resolution of the PA.**

CONCLUSION: **The use of flow diverters seems to be a safe and effective therapeutic option for the treatment of PAs of the extracranial VAs.**

INTRODUCTION

Pseudoaneurysms (PAs) of the vertebral artery (VA) are rare lesions, representing less than 1% of all arterial ecstasies; they are usually located in the intracranial segment of the artery.[1,2] PAs of the extracranial segment are generally the result of a traumatic or iatrogenic injury of the vessel or associated with connective tissue disorders weakening the arterial wall.[1-5] These lesions may be diagnosed as an imaging finding or as a result of symptoms originated from the compression of the adjacent structures, cerebral ischemic events, or their rupture associated with bleeding complications.[5,6] Although the natural history of these lesions is unknown, some authors report aneurysm growth, worsening symptoms, and even death; therefore, their adequate diagnosis and early preventive treatment may be considered.[1]

Given the low incidence of this type of PAs, there are few reports in the literature regarding the best approach for treating them. Several procedures have been described, both surgical and endovascular, ranging from occlusion of the abnormal artery to the reconstruction of the vascular wall with exclusion of the aneurysm. With the advancement of endovascular techniques, the therapeutic trend has increasingly leaned toward the use of endovascular devices, such as stents, with or without coils, or flow diverters.[7]

The purpose of this report is to present the treatment and long-term follow-up of 2 patients with PA of the left extracranial VA treated with flow diverters, and to review the literature.

CASE DESCRIPTION

Case 1

A 48-year-old woman, with no previous medical disorders and surgical or trauma history, presented at our institution with a 1-year history of pain and cervical rigidity, predominantly on the left side, with a normal neurological physical examination. She had been previously evaluated at another hospital, where a noncontrast magnetic resonance of the cervical spine revealed a C2 left lateral mass lesion. A percutaneous biopsy under computed tomography (CT) guidance was performed at the same institution, which complicated with severe bleeding. Based on that history, an angio-CT was performed at our hospital, confirming an 11 × 19 × 20 mm PA of the left VA (Figure 1A and B) with mural thrombosis and bone remodeling of the left lateral mass at the level of the C2 transverse foramen, associated with the arterial dissection. Endovascular therapy was indicated to treat the PA.

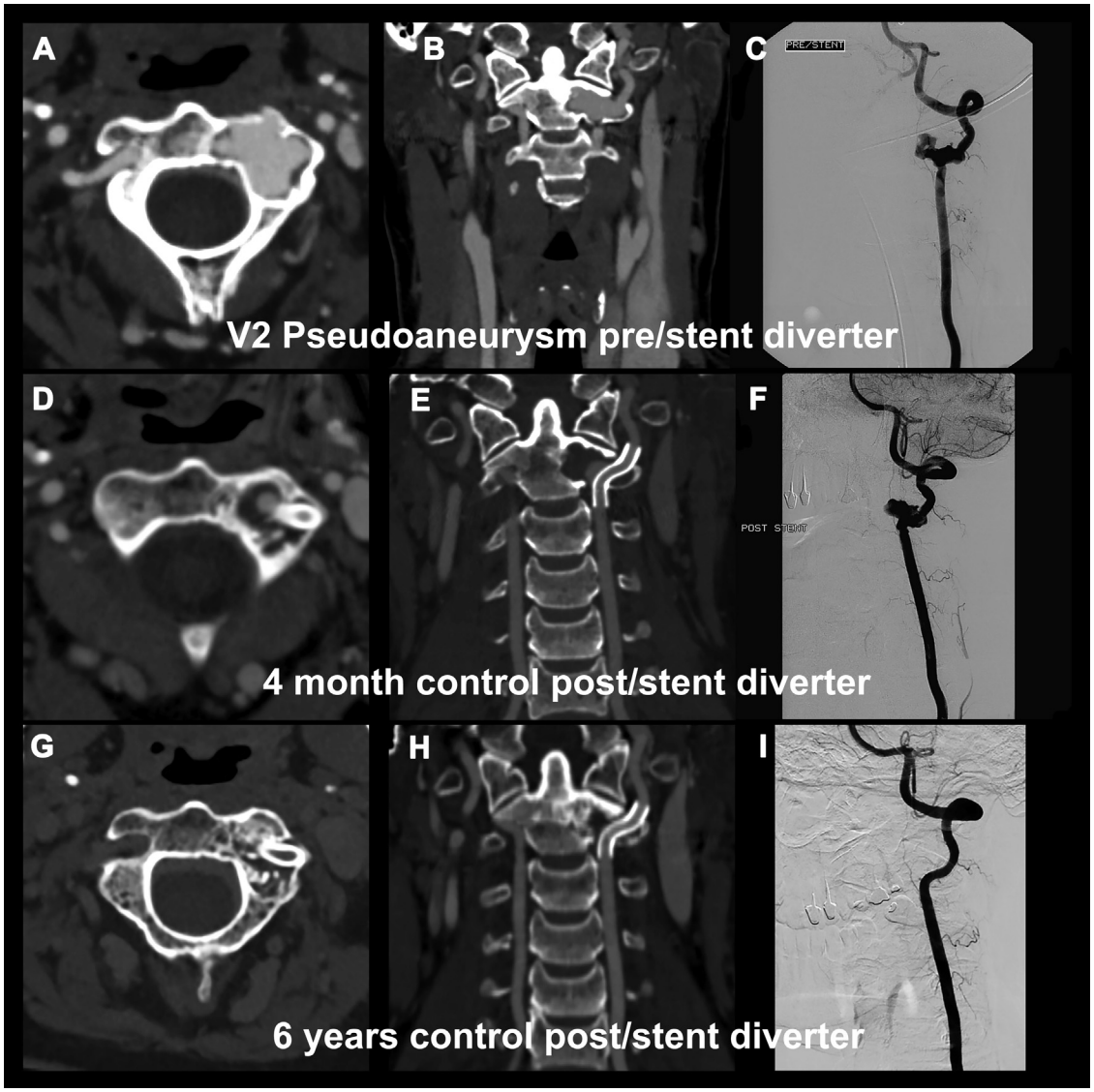

Figure 1. (**A, B**) Diagnostic angiotomography: axial and coronal section of the cervical spine, respectively. In the C2 left transverse foramen, there is evidence of a PA affecting the left VA with irregular contours and minimal mural thrombosis. The PA measures 11 mm × 19 mm × 20 mm in the longitudinal, anteroposterior, and transverse diameters, accordingly. It produces a reshaping of the bone walls of the foramen, with a small continuity solution, connecting the PA with the vertebral channel. The PA has a 4-mm-diameter neck and has a sharp luminal irregularity, with an image of pseudodissection in the vascular wall. (**C**) Digital angiography of the left VA before placing the diverter; lateral view, showing in the cervical trajectory, between V2/V3 segments, a PA dilation, measuring 30 mm in its larger diameter. The edges are rough and irregular. Distally to this lesion, the VA presents adequate size, with some signs of arterial dysplasia. (**D, E**) Cervical spine angiotomography 4 months after the stent placement; axial and coronal sections, respectively. Flow is preserved in the left VA, with a patent stent. There continues to be a minimal flow within the PA, with a significant size reduction (9 mm × 7 mm). (**F**) Follow-up digital angiography showing a patent VA with normal flow. The PA had a successful outcome after treatment, with a 95% size reduction. A small portion remains, with minimal flow within the lesion. (**G, H**) Follow-up digital angiotomography 6 years after the stent placement in the left VA V2 segment, at the level of the C2 lateral mass. The stent was patent, the vascular size was preserved, and there was no evidence of flow inside the PA. Increased re-ossification of the C2 lateral mass is observed when compared with previous images. (**I**) Digital angiography of the left VA showed preserved size with no lesions. Specifically, no traces of a PA are observed at the C2 level after treatment with the flow diverter stent. PA, pseudoaneurysm; VA, vertebral artery.

Under general anesthesia and percutaneous femoral approach, a 6-French guiding catheter (Envoy Cordis, DePuy Synthes, Raynham, MA, USA) was coaxially inserted in the left VA. A microcatheter (Marksman, Medtronic, Minneapolis, MN, USA) was coaxially introduced inside the guiding catheter, through which a flow diverter stent (Pipeline; PED, 4 × 30 mm,

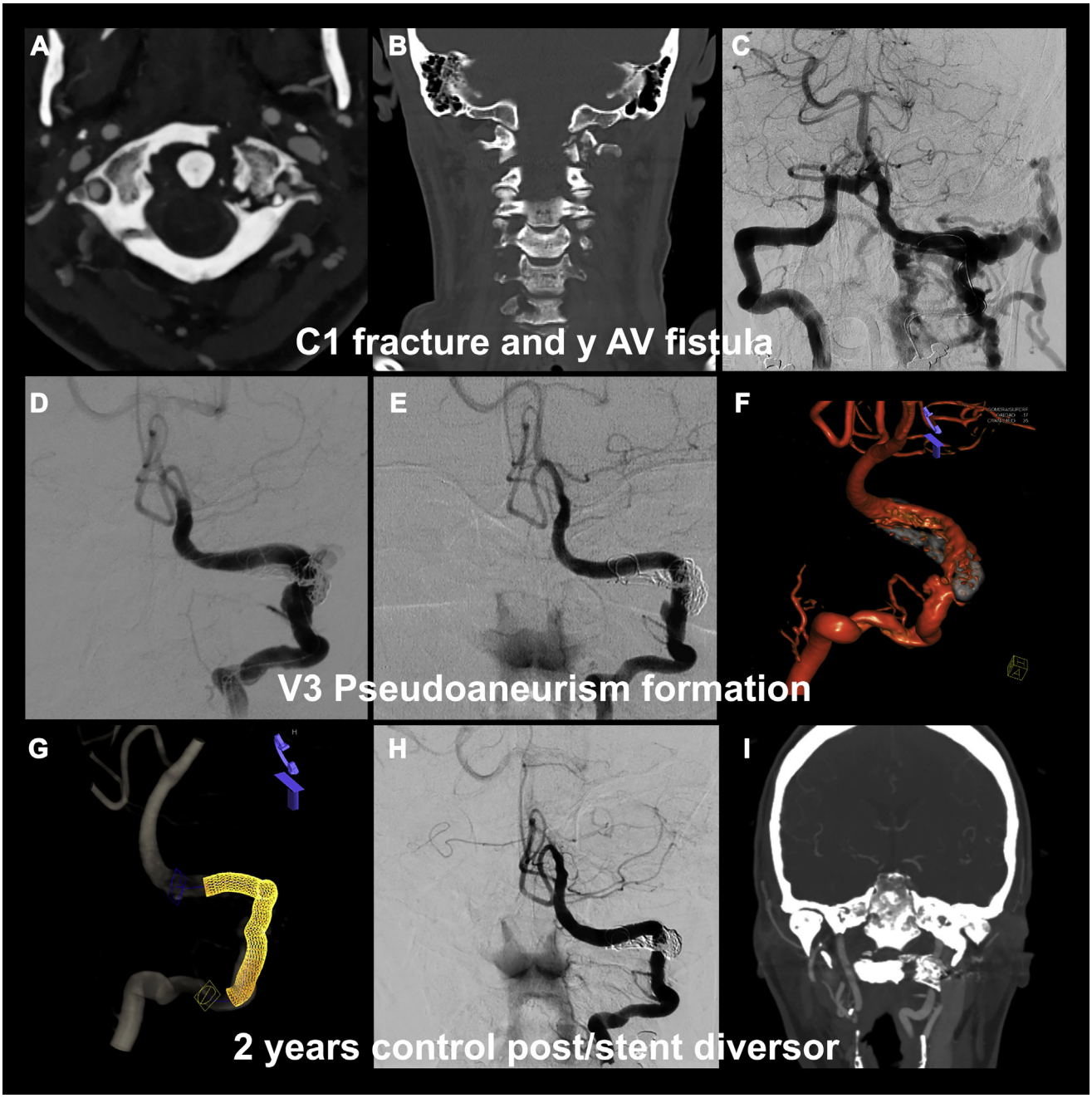

Figure 2. (**A, B**) Computerized tomography of the cervical spine; axial and coronal sections, respectively. Multiple traces of fractures are observed, extending through the C1 anterior and posterior arches, with the presence of a small bone fragment that may be associated with the avulsion of the insertion of the transverse ligament. (**C**) Digital angiography of the VA showing an AVF diverting much of the vessel flow; it was not possible to assess the posterior intracranial circuit. (**D, E**) Digital angiography showing a completely occluded fistula and a small arterial dilation consistent with a V3 segment PA. (**F**) A 3D reconstruction enables the visualization of a 3.75 mm lesion; the coils occluding the previously treated fistula can be seen in gray color. (**G**) Digital angiography planning the stent placement. (**H**) Digital angiography follow-up after 2 years; anterior view, showing a patent left VA, with no evidence of stenotic or aneurysmatic lesions, with complete occlusion of the AVF, and patent stent with no evidence of intrastent stenosis. (**I**) Angiotomography after 2 years, showing embolization material and the stent placed in the topography of the left VA V3 segment, with adequate postcontrast vascular filling, and no evidence of a pseudoaneurysmatic lesion. AVF, arteriovenous fistula; PA, pseudoaneurysm; VA, vertebral artery.

Table 1. Etiology, Localization and Treatment of Reported Vertebral Artery PA

	Localization				
	V1	V2	V3	Not Specified	Total
Etiology					
Traumatic	10	20	13	5	48
Iatrogenic					
Spine surgery	1	16	6	2	25
IJV catheterization	7	1	0	0	8
SCV catheterization	2	0	0	0	2
Radiotherapy	0	0	1	0	1
Chiropractic manipulation	0	3	0	0	3
Collagen disease	0	1	1	0	2
Not specified	1	9	2	1	13
Total	21	50	23	8	102
Treatment					
Endovascular (78)					
VA embolization	4	13	9	4	30
Stent	6	18	3	2	29
Coils	1	4	6	2	13
Stent + coils	2	4	0	0	6
Surgery (18)					
VA ligation	4	6	1	0	11
VA direct repair	3	1	0	0	4
Bypass	0	1	1	0	2
C1–C2 fixation	0	0	3	0	1
VA manual compression	0	0	1	0	1
Conservative	1	2	1	0	4
Not specified	0	1	0	0	1
Total	21	50	23	8	102

IJV, internal jugular vein; PA, pseudoaneurysm; SCV, subclavia vein; VA, vertebral artery.

Medtronic) was advanced into the injured vessel. The stent was positioned across the lesion and was gradually released. Angiographic control after stent placement showed vessel patency and intra-aneurysmal blood ectasia (Figure 1C). Clinical follow-up was uneventful and the patient was discharged 3 days after the procedure, with antiplatelet medication and neck physical therapy.

Follow-up angio-CT tomography at 4 and 12 months showed a significant flow reduction of the PA (Figure 1D–F); at 6 years, the patient is asymptomatic and both angio-CT and direct angiography confirmed disappearance of the PA and bone defect of C2 (Figure 1G-I).

Case 2

A 57-year-old woman, without medical background, was admitted to the emergency unit after a polytraumatism referring intense cervicalgia without neurological deficit. A CT showed an stable fracture of the anterior and posterior arch, left lateral mass, and left transverse foramen of C1 (Figure 2A and B). An angio-CT was performed demonstrating thrombosis of the left VA due to vascular dissection. The vertebral fracture was treated conservatively (immobilization with a Philadelphia collar for 2 months), and the patient was discharged 3 weeks after with acetylsalicylic acid 100 mg/daily.

One year after trauma, a fremitus and murmur in the left submastoid region were detected. An angiography was performed demonstrating an AVF of the left VA at the previous cervical traumatized level (Figure 2C); no distal flow was detected in the left intracranial VA after ipsilateral injection or retrograde filling with the injection of the right VA. An embolization was performed using hydrocoils, with complete occlusion of the AVF and VA anterograde flow recovery. One month after the embolization, a control angiography showed a 3.84 mm PA of the left V3 segment (Figure 2D–F). Endovascular treatment was indicated and a flow diverter stent (Pipeline; FRED, 4.5 mm × 25 mm, Medtronic) was placed across the opening of the PA (Figure 2G).

Follow-up angio-CT tomography at 4 months shows that the flow diverter was patent, with complete occlusion of the PA (Figure 2H). Two years after the device placement, an angio-CT demonstrates complete resolution of the PA and persistence of the left VA patency (Figure 2I).

LITERATURE REVIEW

To identify all cases of PA of extracranial VA described in the available medical literature, we searched in PubMed (U.S. National Library of Medicine) and Cochrane Database of Systematic Reviews (Cochrane Collaboration resources) under the following keywords: "vertebral aneurysm," "pseudoaneurysm," "traumatic aneurysm," and "extracranial vertebral artery."

Since the first reported case by Early et al in 1966 until now, we identified 102 cases in 80 articles.[1-26] Patients' mean age was 41.3 years (range, 4–86 years), we found a female predomination 60.4% (n = 61); in 1 case, the authors did not specify age or gender.[4] Etiology, location in the artery segment, and treatment of these lesions are summarized in Table 1.

DISCUSSION

Pseudoaneurysms are caused by a disruption of the arterial wall, which leaves the

Table 2. Reported Cases of Pseudoaneurysms of the Extracranial Vertebral Arteries Treated with Flow Diverters

Author, Year	Age/Sex	Clinical Presentation	Initial Assessment	Arterial Segment/Size	Treatment	Imaging Follow-Up
Ambekar et al, 2014[3]	47/M	Pulsatile mass at the surgical site, on the second postoperative day for C1—C2 posterior fixation	CAT	V3/left	EV: flow divider stent	Complete occlusion at 6 months
Kerolus et al. 2018[26]	27/M	Cervicalgia and dysphagia 3 years after penetrating neck trauma	CAT	V2/left	EV: flow divider stent.	Occlusion of aneurysm at 3 months
Shakir et al, 2016[7]	60/M	Quadriparesis at 36 hours postoperative for discectomy and C3—C7 anterior arthrodesis	CAT	V2/left	EV: flow divider stent	Complete occlusion at day 4 (no follow-up specified)
Dolati et al. 2015[30]	71/M	Intraoperative arterial bleeding for C1—C2 posterior fixation	DA	V3/left	EV: flow divider stent	Complete occlusion at 3 months
Cohen et al, 2016[4]	52/M	Nonpenetrating neck trauma	CAT	V2	EV: flow divider stent	Complete occlusion at 6 and 12 months
	17/M	Nonpenetrating neck trauma	CAT	V2	EV: flow divider stent	Complete occlusion at 6 and 12 months
	19/M	Nonpenetrating neck trauma	CAT	V2	EV: flow divider stent	Complete occlusion at 6 and 12 months
	23/M	Nonpenetrating neck trauma	CAT	V2	EV: flow divider stent	Complete occlusion at 6 and 12 months
	32/F	Nonpenetrating neck trauma	CAT	V2	EV: flow divider stent and coils.	Complete occlusion at 6 and 12 months
	29/M	Nonpenetrating neck trauma	CAT	V2—V3	EV: AVF embolization with coils and occlusion of PA using a flow divider stent	Complete occlusion at 6 and 12 months

AVF, arteriovenous fistula; CAT, computed angiotomography; DA, digital angiography; EV, endovascular; PA, pseudoaneurysm.

blood contained by a thin layer of adventitia or underlying connective tissue, making them lesions that lack a true vascular wall and neck.[3] From the morphological point of view, they can be classified into saccular, resulting from a focal lesion to the vessel wall, or fusiform PA that occur after dissection of the artery causing in thinning of the adventitia and dilation of the vessel.[3,8]

In the cervical trauma, either open or closed, V2 is the most frequently affected segment; this may be caused by either the close anatomical relationship of the VA with the transverse foramen, with the potential artery injury by bone fragments, or the instability of the cervical spine.[9,10] Regarding the surgical procedures, the most vulnerable segment is V3, reporting an incidence of lesion of this segment between 0% and 8% in the occipitocervical fixation surgery,[11,27] followed by the lesion in the V2 segment, documented in 0.5% of the anterior approaches between C2 and C6.[28] The connective tissue disorders may also predispose to the development of PAs, as they may weaken any or all of the arterial wall layers.[12-14] On occasions, such as in our case 1, the cause is idiopathic, being slow-growing progressive lesions with no evident background of trauma or surgery.[3,4,8] Although these lesions may evolve toward spontaneous resolution,[15,16] they also could grow, make the symptoms worse, bleed, and even cause patient death; this is the reason why early treatment is recommended.[1,7]

In the past, these lesions were treated via the exclusion of the entirety of the affected arterial segment, whether through endovascular or surgical procedures[3]; however, this approach may lead to severe neurological complications, such as cerebellar infarction, Wallenberg syndrome, quadriparesis, hemiparesis, or even death.[29] Such complications mainly depend on the inadequate perfusion of the contralateral VA, which may not compensate the sudden lack of flow of its counterpart. The right and left VAs are hypoplastic in 8.8% and 5.7% of the general population, respectively.[17] However, as adequate as the contralateral vascularization may seem, the incidence of infarction may reach up to 16%.[18]

With the emergence of endovascular devices, the current recommendation is the reconstruction and reinforcement of the vessel wall with the exclusion of the PAs, maintaining vascular permeability via the use of stents or flow diverters. Selective embolization of the PAs is not recommended because of the fragile nature of its wall[19]; on the other hand, as the vascular wall is not reconstructed, this may increase the risk of bleeding or lesion growth.[20] The primary complication for these

devices is ischemia.[21] Stents were initially used to sustain the coils, thus preventing their movement.[30] Although balloon-expanding stents possess greater radial strength, their rigidity makes them less maneuverable in twisted vessels, compared with self-expanding stents.

Flow diverters are a new generation of stent-like devices: they reconstruct the wall, maintaining the patency of the efferent vessels, whilst excluding and favoring blood stasis and thrombosis inside the dilated area.[3,22] However, these devices require dual antiplatelet therapy, as they are typically highly thrombogenic, which limits their use in patients traumatized with hemorrhagic lesions, the main PA etiology. Another characteristic limiting their use for this type of PAs is the limited capacity to maintain the vascular diameter due to their diminished radial strength, meaning that they would not be a good therapeutic option in PA with associated stenotic lesions.[4] The primary complication documented with flow diverters is arterial stenosis and device migration.[23] Promising results have been reported with their use, with an obliteration rate of 82.9% for intracranial aneurysms and 87.5% for intracranial PAs.[23-25] Given the low frequency of PAs of extracranial vessels, like the VA, there is less experience with the use of flow dividers for their treatment. Ambekar et al[3] reported in 2014 a case of successful treatment with complete occlusion of a V3 segment pseudoaneurysm at 10 months. Similar results were published for the treatment of PA of the V2 and V3 segments.[7,26,30] Cohen et al[4] published in 2016 a series of 9 patients with pseudoaneurysms due to nonpenetrating cervical spine traumas, 4 of them treated with flow diverters with complete occlusion at 6 months; patients treated with flow diverters are shown in **Table 2**. In both cases reported in this paper, we attained complete occlusion of the lesions, with a 24-month follow-up in case 2 and a 72-month follow-up for case 1. Even though the available literature shows good therapeutic results with a low morbid-mortality rate using these devices, the number of cases reported is low, with a short follow-up, precluding us to make a therapeutic recommendation.

CONCLUSION

The use of flow diverters may be considered as a therapeutic option for the treatment of PA of the extracranial VA.

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