

Treatment of Dissecting Vertebral Pseudoaneurysms: A Single-Center Experience

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Abstract

Background: Dissecting vertebral artery pseudoaneurysms represent a unique clinical challenge with careful appreciation for location of the posterior inferior cerebellar artery. Limited data is available in terms of outcomes regarding the various treatment modalities.

Methods: 11 patients with dissecting pseudoaneurysms were identified from 2013-2021. Pseudoaneurysm size and morphology, clinical presentation, and treatment approach was collected. Success of treatment was recorded based on post-operative imaging as well as documented overall patient outcomes. Three primary treatment modalities emerged: coil embolization, stent assisted coiling, and flow diversion.

Results: Of the 11 patients, 5 were female and 6 were male with an age from 36 to 69.7. 7 had ruptured pseudoaneurysms at time of treatment. Size of pseudoaneurysm ranged from 3 to 6 mm. 8 were on the right and 3 were on the left vertebral artery. 8 were proximal to PICA and 3 were distal. Co-dominance of vertebral filling was seen in 5 patients, 5 with dominance through right vertebral artery, and 1 with dominance through left vertebral artery. Variability existed in treatment approaches with 4 patients undergoing coil occlusion, 5 patients undergoing flow diversion stenting, and 2 patients undergoing flow diversion stenting with jailed coiling. 1 patient had enlargement of pseudoaneurysm while inpatient and required a second flow diversion device. 1 patient had two flow diversion devices placed initially at time of treatment due to morphology of PA. 6 patients had repeat angiograms between 6 to 9 months with complete occlusion. 3 had CTA or MRA with complete occlusion for those that had flow diversion, they were transitioned from aspirin and clopidogrel to aspirin monotherapy after first repeat angiogram. 6 patients required shunt placement for hydrocephalus. 1 patient died prior to discharge due to sepsis. 2 patients died post discharge: 1 with myocardial infarction and the 2nd due to urosepsis.

Dissecting vertebral pseudoaneurysm has high morbidity and mortality if rupture occurs. Location of PICA origin influences treatment approach. Patients with poor Hunt/Hess scores upon arrival had increased risk for systemic infection and mortality.

Keywords: Vertebral pseudoaneurysm; Trauma; Microsurgery; Endovascular surgery

Introduction

Cerebral pseudoaneurysms (PA), also known as a false aneurysms, are an infrequent vascular pathology, accounting for 1% of all cerebral aneurysms.¹ Cerebral vessels lack an external lamina unlike systemic vessels making them highly vulnerable to injury [2-4]. PAs are a product of complete arterial wall injury resulting in a hemat-

oma contained by perivascular connective tissue and allowing continuous communication with the parent artery [4]. Although a rare pathology, ruptured PAs have a high rate of mortality, nearing 50% [1, 5]. The high rate of morbidity and mortality of PAs makes it critical that they are identified and treated in a timely manner.

PAs are classified by the primary form of vessel injury. Traumatic PA

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are the most common, followed by infectious, iatrogenic, connective tissue disease, vasculitis, post rupture of true cerebral aneurysm, post radiotherapy, and Moya Moya disease [6-9]. Presentation of PAs is variable although most present due to an intracranial hemorrhage prompting immediate angiography given the high mortality associated with PAs [10, 11]. Anterior circulation PAs are frequently associated with epistaxis when the intracavernous internal carotid artery (ICA) is involved and cranial neuropathies when of traumatic etiology includes microsurgical repair, endovascular intervention, and conservative observation [12]. Rapidly emerging endovascular techniques and technology have increased the options available to surgeons, although no treatment option has been studied to prove superiority.

Considering the high variability of patient presentation and treatment options, PAs continue to present a challenge for surgeons when determining management. Here we report our single-center experience of 11 patients diagnosed with dissecting vertebral artery PA and their post-operative course. In addition, we discuss the literature on existing treatment options and their challenges.

Methods

Institutional review board approval was obtained (IRB#202101872). Patients were retrospectively evaluated from University of Florida endovascular database between 2013-2021. 2013 was chosen as the initial time point of collection as this was when flow diversion embolization was introduced at our institution [11]. patients met inclusion criteria of dissecting vertebral artery pseudoaneurysm based on cerebral angiography. Demographics such as age and sex were collected. Patient's presenting symptoms and/or rupture status of pseudoaneurysm was reported. Location of pseudoaneurysm in relation to PICA, size of aneurysm, and vertebral filling pattern were documented. The type of treatment modality was noted as well as standard antiplatelet regimen utilized if applicable, and when transition to single agent occurred. If patients had post-operative angiogram, timing of angiogram and reported outcomes were presented.

Documented clinical outcomes were assessed at time of post-op follow up. Results are presented in table format. Due to limited sample size, no statistical analysis could be performed, but the three treatment patterns (coil embolization, flow diversion, and flow diversion with coiling) are presented in associated figures.

Results

Of the 11 patients, 5 were female and 6 were male. Age ranged from 36 to 69 years. 7 out of the 11 patients presented as ruptured pseudoaneurysms. Size ranged from 3 to 6 mm [8]. were on the right and 3 were on the left vertebral artery [8]. were proximal to PICA and 3 were distal. Co-dominance of vertebral filling was seen in 5 patients, 5 with dominance through right vertebral artery, and 1 with dominance through left vertebral artery. Variability existed in treatment approaches with 4 patients undergoing coil occlusion of vertebral artery and pseudoaneurysm, 5 patients undergoing flow diversion stenting, and 2 patients undergoing flow diversion stenting with jailed coiling [1]. patient had enlargement of pseudoaneurysm following placement of a flow diverting stent (pipeline) while inpatient and required placement of a second device [1]. patient had two overlapping flow diverting stents placed at time of initial treatment due to morphology of PA [6]. patients had repeat angiograms between 6 to 9 months with complete occlusion [3]. patients had follow-up CTA or MRA with complete occlusion at 6 months. For those that had flow diversion, they were transitioned from aspirin and clopidogrel to aspirin monotherapy after repeat angiogram [6]. patients required shunt placement for hydrocephalus [1]. patient died prior to discharge due to pneumonia and subsequent septic shock [1]. patient died due to urosepsis and shunt infection prior to initial follow up [1]. patient died from myocardial infarction after initial angiogram. Of surviving patients, 2 patients had residual neurologic deficits [6]. patients fully recovered neurologically. The results are presented in table 1. Figure 1 shows coil sacrifice, figure 2 shows flow diversion alone, and figure 3 shows flow diversion with jailed coils.

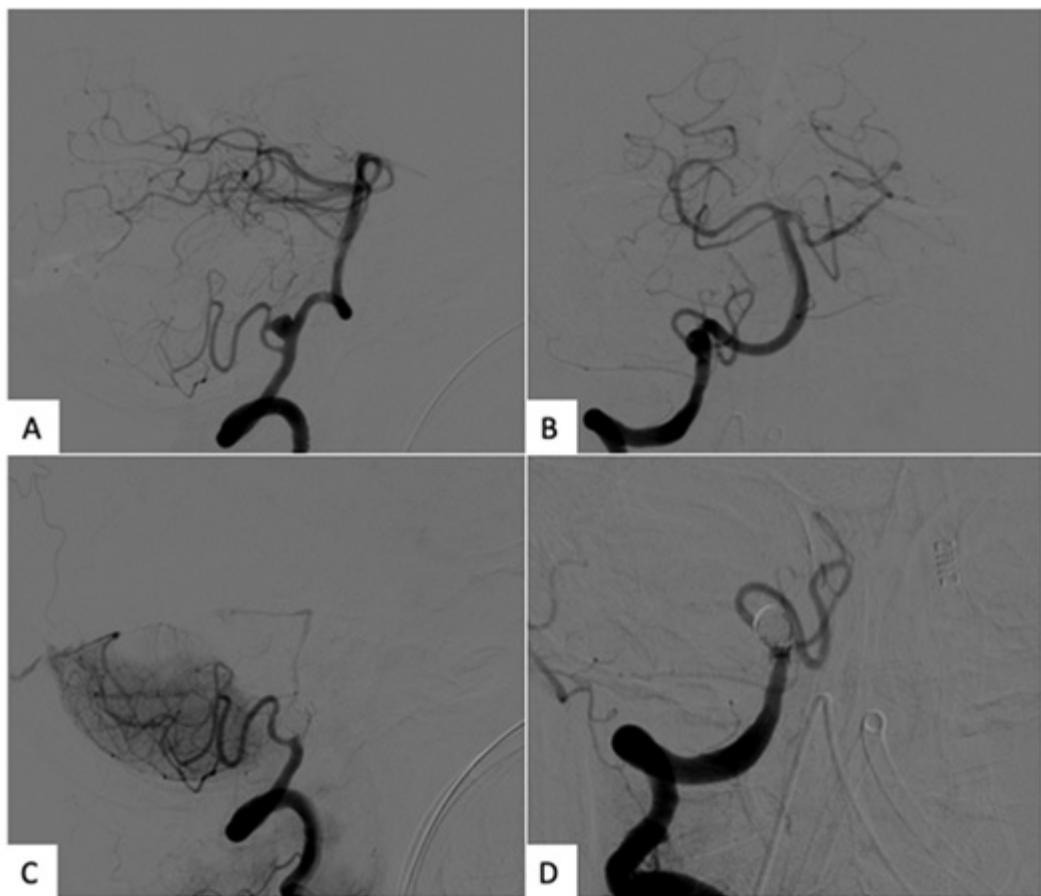


Figure 1: Coil sacrifice. A and B are pre-treatment and C and D are post.

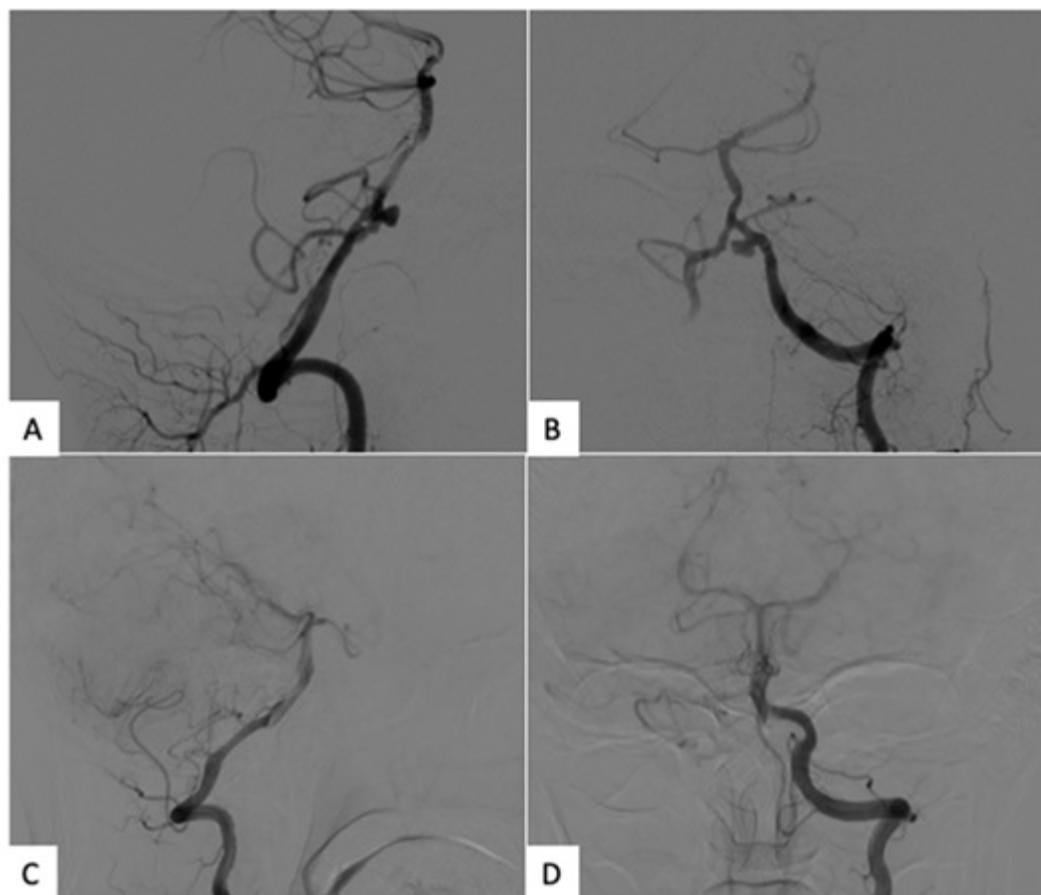


Figure 2: flow diversion stent treatment. A and B are pretreatment. C and D are 6 month follow up angiogram showing complete obliteration.

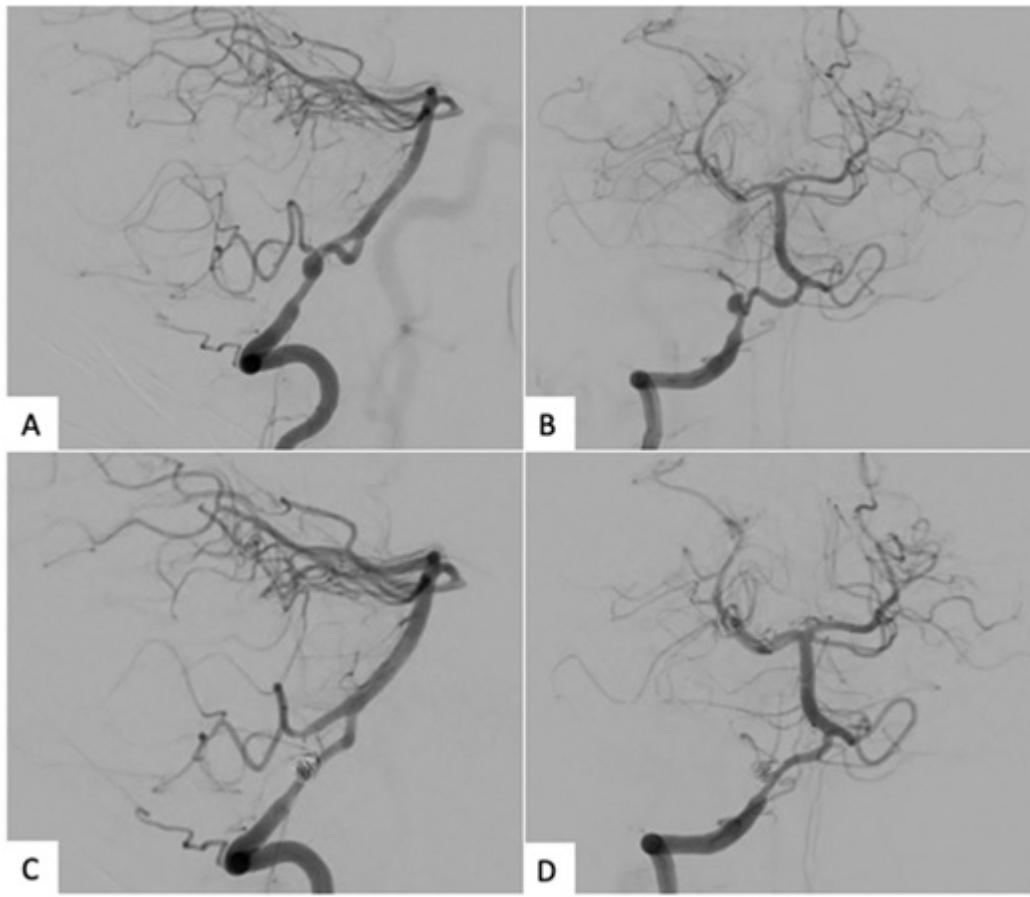


Figure 3: Stent coiling. A and B are pretreatment. C and D are 6 month follow up angiogram showing complete obliteration.

Table 1: Summary of results and patient demographics

Case	Age and Gender	Mechanism	Description	Location	Morphology	Symptoms	Size (mm)	Dominance of Vertebral Artery	Type of Treatment	Immediate Outcome	Procedure Complications	In Hospital Mortality	Discharge Destination	Follow Up Angiogram Timing	Aneurysm Occlusion	Follow-Up Outcome	Final Outcome
1	68F	Spontaneous	Ruptured Pseudoaneurysm	Right PICA proximal to pseudoaneurysm	Dissecting/Fusiform	HH3 Fisher 3/4 Pontomedullary hemorrhage Left thalamic stroke	5	Co-dominance	Coil occlusion	Successful Coil embolization of dissection	None	No	Inpatient Rehabilitation Center	No additional angiograms	At time of treatment Basilar filled from left, PICA still filling on Right (coiled distally)	Mild dysmetria on Post-op Exam	Lost to follow up
2	69M	Spontaneous	Ruptured Pseudoaneurysm	Right PICA proximal to pseudoaneurysm	Dissecting	HH3 Fisher 3/4 Diffuse SAH	3	Right	Coil occlusion	Successful Coil embolization of dissection	None	No	Inpatient Rehabilitation Center	No additional angiograms	At time of treatment Basilar filled from left, PICA still filling from Right (coiled distally)	Required shunt; Intact at Post-op Exam	MRA showed continued embolization with PICA Filling
3	59M	Spontaneous	Ruptured Pseudoaneurysm	Right PICA proximal to pseudoaneurysm	Dissecting	HH3 Fisher 3 Diffuse SAH	6	Co-dominance	Coil occlusion	Successful Coil embolization of dissection	None	No	Inpatient Rehabilitation Center	6 month	No residual aneurysm	Required Shunt: Neuro Intact	No additional scheduled follow up
4	46F	Spontaneous	Ruptured Pseudoaneurysm	Right PICA proximal to pseudoaneurysm	Dissecting	HH3 Fisher 3 Diffuse SAH Intraoperative rupture	5	Right	Coil and onyx embolization	Complete occlusion of vessel	Intraoperative rupture	No	Inpatient Rehabilitation Center	6 month	No residual aneurysm	Required Shunt; Awake, Peril, MAE x4, trach decannulation	Found altered at home: urosepsis, shunt infection, mortality
5	43F	Spontaneous	Pseudoaneurysm	Right PICA proximal to pseudoaneurysm	Dissecting	Presented with left PICA stroke/ Left vertebral occlusion in addition to pseudoaneurysm	3	Right	Pipeline stent assisted coiling	Successful coverage and occlusion	At OSH refractory vasospasm and aborted	No	Home	6 month	100% occlusion of pseudoaneurysm	Asa/Plavix with transition to Asa at angi. Neuro Intact.	Continued 100% occlusion on most recent angiogram
6	58M	Trauma	Ruptured Pseudoaneurysm	Right PICA proximal to pseudoaneurysm	Dissecting	HH4 Fisher 3 Diffuse SAH	3	Co-dominance	FRED assisted coiling	Successful coverage and occlusion	None	No	Inpatient Rehabilitation Center	6 month	100% occlusion of pseudoaneurysm	Required Shunt; Some residual hoarseness and right afferent pupillary defect	Plan for repeat angiogram in 1 year

7	55M	Iatrogenic	Pseudoaneurysm	Left PICA distal to pseudoaneurysm	Dissecting	Iatrogenic pseudoaneurysm from Anterior Cervical Discectomy Procedure	3	Co-dominance	Pipeline with balloon dilation	Successful coverage and occlusion	Vasospasm requiring IA verapamil	No	Home	No additional angiograms	At time of treatment complete occlusion of pseudoaneurysm	Transitioned to coumadin from DAPT due to DVT by time of followup; Intact on exam	CTA shows successful occlusion of pseudoaneurysm
8	40M	Fibromuscular Dysplasia	Pseudoaneurysm	Left PICA proximal to pseudoaneurysm	Dissecting	Right hemibody tingling Neck pain Right pontine infarct	4.5	Left	Pipeline	Successful coverage and occlusion	None	No	Home	6 month	100% occlusion of pseudoaneurysm	Asa/Plavix with transition to Asa at angi. Neuro Intact	Follow up PRN
9	36F	Trauma	Pseudoaneurysm	Right PICA distal to pseudoaneurysm	Disssecting	Neck trauma Pain	4	Co-dominance	Pipeline	Successful coverage and occlusion	None	No	Home	9 month	100% occlusion of pseudoaneurysm	Asa/Plavix with transition to Asa at angi. Neuro Intact.	Planned repeat CTA in 1 year
10	62F	Spontaneous	Ruptured Pseudoaneurysm	Left PICA proximal to pseudoaneurysm	Disssecting/sacular	HH4 Fisher 3 Diffuse SAH	3	Right	Two pipelines	Expansion of pseudoaneurysm	Placement of second pipeline	No	Long-term Care Center	Repeat while in house	Expanding pseudoaneurysm at which time 2nd pipeline was placed	Required Shunt; Minimally verbal with tracheostomy. Follows simple commands x4.	Patient developed respiratory decline at care center with transition to comfort care
11	55M	Spontaneous	Ruptured Pseudoaneurysm	Right PICA distal to pseudoaneurysm	Dissecting	HH4 Fisher 3 Diffuse SAH	4	Right	Two pipelines	Successful coverage and occlusion	Required ICA Surpass stent for superior hypophyseal aneurysm	Yes	Deceased	Repeat while in house	100% occlusion of pseudoaneurysm	Required Shunt; Minimally verbal with tracheostomy. Follows simple commands x4.	Patient developed pneumonia, sepsis, and subsequent mortality

Discussion

A PA is a rare cerebrovascular pathology associated with high morbidity and mortality.⁴ PAs most often result from head trauma such as closed head trauma during motor vehicle accidents or penetrating stabbing wounds, with one retrospective study finding 57% of PAs to be trauma induced.¹³ However, PAs can also arise spontaneously or due to infection, radiation and connective tissue disease that may cause vessel damage.⁷ There is no standardized protocol or guideline for the treatment of PAs. Currently, interventions range from surgical options such as resection, clipping and vascular reconstruction to endovascular techniques such as embolization, stent graft implantation and coiling [4, 14].

Surgical repair has traditionally been the treatment option of choice for patients presenting with PAs [15-18]. Direct clipping is a surgical technique where a metal clip is placed on the neck of the intracranial pseudoaneurysm to obstruct blood flow to the vulnerable vessel. Many studies have described the postoperative outcomes of the direct clipping of PAs [19-21]. The patient recovered successfully with no new deficits. Chen et al. performed direct clipping on a 25-year-old patient with a PA leading to complete occlusion of the aneurysm [20]. Ding et al. describe two patients undergoing PA clipping following intraoperative ruptures [21]. The first patient recovered nearly completely with no obvious neurological deficits, and the second patient had incomplete paralysis of left limb immediately after surgery which resolved 3 months later [21]. Clipping, however, is not typically possible for multiple reasons [22]. First, an obscure aneurysm neck and fragile wall of the PA makes satisfactory clipping incredibly difficult. Hadley et al. describes a 17-year-old patient presenting with a traumatic PA treated by clip sacrifice of the affected vertebral artery distal to the pseudoaneurysm [19]. In addi-

tion, many PAs may be in inaccessible locations such as the petrous or cavernous ICAs where clipping may be impossible.

Clip trapping is another surgical technique in which two clips are placed on either side of the PA. Studies have demonstrated successful treatment of cerebral PAs with this method [5, 6, 23]. This technique may be combined with bypass revascularization depending on the status of the collateral supply and allows for elimination of the PA if sacrifice is necessary [4]. A third technique, wrapping, is also commonly performed when clipping is not possible. Wrapping serves to support the fragile pseudo-wall and maintain the connectivity of the parent artery [24]. However, this technique is associated with a high risk of rebleeding, granuloma formation and cerebrovascular complications [25]. Although resection and end-to-end anastomosis is also possible, the literature regarding outcomes is still limited. Ultimately, surgical repair is associated with increased morbidity and mortality and is therefore reserved for lesions that fail alternative treatments or present with significant mass effect [15, 17]. Endovascular coiling is a minimally invasive procedure with the goal of blocking blood flow into the PA cavity. Many studies have demonstrated successful occlusion by employing a coiling technique [11, 26-30]. Rallo et al. achieved complete occlusion in two traumatic PAs using coiling and controlled Onyx injection [26]. Phogat et al. looked at postoperative outcomes in 14 patients diagnosed with intracranial PAs [11]. Coiling alone was performed in 8/14 patients (57.1%), and complete occlusion was achieved in all 8 patients [11]. Monteiro et al. performed a case study of a patient presenting with a postoperative iatrogenic intracranial PA treated with stent-assisted coiling [27]. Coiling led to subtotal occlusion of the PA. Al-Jehani et al. performed a similar case study of a patient presenting with a traumatic PA treated with primary coiling and achieved the same

outcome [29]. Coiling is preferred to surgical repair since the fragile PA wall has a high risk of intraoperative rupture. However, coiling still carries some risks such as microcatheter or coil perforation and potential recurrence [4, 31-34].

To avoid the well documented risks of recurrence and rebleeding in coiling of PAs, liquid and particle embolization can sometimes be used to treat PAs with a lower complication rate [35, 36]. Risks are rare and require further investigation, but include thrombosis, perioperative stroke, and delayed failure of embolization [18, 37]. PAs with narrow necks and arising from distal segments of parent artery are ideal candidates for embolization [4, 38]. Following normal balloon occlusion testing (BOT), embolization via parent vessel occlusion can quickly be carried out. Although coils can be used, particle or liquid embolic agents such as n-butylcyanoacrylate or Onyx are preferred for PAs due to their fragile vessel wall.⁴ Routine follow-up is recommended in these patients, as up to 22% of patients have been shown to experience ischemic complications even with a normal BOT [39].

Another endovascular option, stenting, is an adjunctive technique for vertebral artery PA repair [28, 40-43]. It is a technique in which a self-expanding mesh tube is placed within the vessel to open the stricture. Vezzetti et al. demonstrated that treating PA patients with stenting reduced the incidence of major adverse cardiovascular events [41]. Plou et al. describes a patient with a PA of the vertebral artery who was treated with a flow diverting stent. The patient was discharged three days after the operation and showed complete resolution of the PA 6 years later on angiography.⁴⁴ A major known limitation of flow-diverting stents is delayed aneurysm obliteration [4, 45]. Although they do have a high aneurysm occlusion rate, the process can take weeks, leaving the patient at an increased risk of rebleeding during this time [31, 46].

Senay et al. treated a patient presenting with a left internal carotid pseudoaneurysm with a pipeline embolization device [42]. The patient demonstrated near complete occlusion of the PA at 7-month follow up. Similarly, Shakir et al. describe a patient who developed a PA at the V2 segment of the vertebral artery [43]. The patient had two pipeline flow-diverting stents placed leading to complete occlusion of the PA. Another report by Cohen et al. reviewed 9 cases of vertebral artery PAs caused from blunt trauma [28]. Flow diverting stents, stent-assisted coils, overlapping covered stent implants, or balloon-expandable stents due to endoleak were placed in the patients leading to complete occlusion in all the cases. The specific use of a covered stent has multiple advantages, including minimal risk of procedure-related rupture of aneurysm, low complexity of the procedure, maintained flow through parent artery, and the generation of less mass effect compared to other procedures such as coiling

[47]. Covered stents have also been shown to decrease the incidence rate of restenosis and neointimal proliferation, but also are more difficult to deploy due to stiffness [4]. In addition, patients undergoing endovascular stenting generally require dual antiplatelet therapy (DAPT), and there is concern that this may increase the risk of intracerebral hemorrhage in patients with PAs. However, the literature has largely shown that DAPT in patients with PAs undergoing stenting is safe, with no associated episodes of rebleeding, ischemic events, periprocedural or long-term complications [47-51]. Stenting, however, cannot be used in all patients presenting with a PA. Due to the potential risk of stent-graft infection, it is contraindicated in mycotic PAs. Moreover, placement requires a stiff delivery system, limiting it to larger and less tortuous arteries [38].

Thrombin therapy is an additional non-invasive technique that causes direct clotting of the PA and is associated with a near 100% success rate [52]. Most of the literature on thrombin therapy has been for extracranial PAs and is rarely done intracranially [53-57]. The procedure consists of direct injection of thrombin into the PA, with the thrombin dose primarily determined by the size of the PA cavity. Extracranial carotid ultrasound guidance allows for visualization of both thrombin interaction with flowing blood, followed by thrombosis and loss of flow into the cavity.⁵⁸ Complications are rare and include thromboembolic events downstream of the injection site, venous thrombosis, and allergic reaction [15-17, 59, 60]. To avoid these complications, minimal amounts of thrombin are used and preprocedural allergy testing performed [61, 62]. This is only indicative for non-ruptured pseudoaneurysms.

Finally, active observation can be used as a form of conservative treatment for PAs with decreased size and flow on repeat imaging. PAs inherently carry a high rupture and subsequent mortality risk [63]. This is especially true for cerebral PAs since cerebral arteries lack an external elastic lamina, have a paucity of elastic fibers in the tunica media and thin adventitia [2]. Although spontaneous occlusion and resolution of PAs has been shown to occur in intracranial vessels, likely due to vascular remodeling or spontaneous thrombus formation, it is uncommon and unpredictable [4, 64-69]. In fact, routine follow-up should be continued well after spontaneous resolution, as reappearance has also been shown to occur in the context of thrombus dissolution [12].

6. Conclusion

Ultimately, there exists a plethora of endovascular and surgical techniques for the treatment of PAs. However, superiority has yet to be determined and therefore no guidelines exist for how to approach patients with varying presentations. Until then, treatment should continue to be tailored to each patient and the unique characteristics of their PA.

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