

Basilar Artery Fenestration Aneurysms: Endovascular Treatment Strategies Based on 3D Morphology

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Abstract

Purpose Basilar artery is the second most common site of fenestration, after the anterior communicating region. It is believed this variation predisposes a patient to posterior aneurysm formation and increases the complexity of the surgical anatomy. Endovascular management has become the first option to treat these aneurysms. We retrospectively evaluated eight patients, who underwent endovascular treatment for fenestrated basilar artery related aneurysms (fBA-AN). Additionally, based on our findings and on literature review, we developed a treatment strategy based on a proposed classification of fBA-AN.

Methods Between June 2010 and September 2012, eight patients harboring nine basilar artery fenestration aneurysms were consecutively treated. Based on aneurysm morphology (neck size) and its relationship to the fenestration (sparing or not one channel) characterized by 3D-DSA, we proposed a simple classification and treatment strategies. Additionally, a literature review was performed.

Results All patients received endovascular treatment. Most aneurysms involved the vertebrobasilar junction and both channels of the fenestration. A total of 5 aneurysms had wide neck while 4 had narrow neck. Overall, 5 (55.5%) an-

eurysms were treated with stent assisted coiling, 3 (33.3%) aneurysms with selective coiling, and 1 (11.1%) aneurysm with balloon assisted coiling. We had only 1 (11.1%) complication, named aneurysm rupture.

Conclusion Basilar artery fenestration aneurysms are rare and complex lesions. Endovascular treatment appears to be safe and efficient. The detailed understanding of the aneurysm morphology and its relation to the fenestration is strongly recommended to treatment planning. Further studies are necessary to validate the utility of the proposed classification and treatment strategy.

Keywords Aneurysm · Basilar artery · Embolization · Fenestration

Introduction

Fenestration of cerebral arteries is a rare but well-known vascular variation that begins with a common origin, splits into two distinct endothelium-lined channels, which then rejoin distally [1]. Basilar artery is the second most common site of fenestration, after the anterior communicating region [1]. The frequency of basilar artery fenestration is reported to range from 0.6 to 2.33% depending on the diagnostic modality employed [1, 2]. It is believed this variation predisposes a patient to posterior aneurysm formation [1]. Aneurysms associated with a fenestrated basilar artery are very uncommon; their incidence is reported to be 0.33% of all intracranial aneurysms [3].

Surgery of basilar artery aneurysms is difficult and challenging, owing to the complex surrounding anatomy, proximity of lower cranial nerves, difficulty in obtaining adequate surgical exposure, and the presence of blood vessel perforators running from the artery to the brainstem.

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Furthermore, fenestration increases the complexity of the surgical anatomy. Therefore, endovascular management has become the first option to treat these aneurysms [3–5]. Regarding this scenario, three-dimensional rotational angiography has been shown to be a mandatory tool to plan the endovascular treatment.

We retrospectively evaluated eight patients, who underwent endovascular treatment for fenestrated basilar artery related aneurysms (fBA-AN). Additionally, based on our findings and on literature review, we developed a treatment strategy based on a proposed classification of fBA-AN [3–24].

Methods

Study Population

Between June 2010 and September 2012, nine patients harboring ten basilar artery fenestration aneurysms were consecutively admitted in two Brazilian centers, Instituto Neurovascular, Belo Horizonte, Minas Gerais, and Hospital das Clínicas of University of São Paulo, Ribeirão Preto, São Paulo.

All patients were evaluated with computed tomography scan (CT) and catheter angiography, with special emphasis on three-dimensional rotational reconstruction (3D-digital subtraction angiography (DSA)). Both ruptured and unruptured aneurysms were included. Patients with fenestrated basilar artery but distant aneurysms, not directly related to the fenestration, were excluded.

One patient had two aneurysms related to the same basilar fenestration. One patient with a very small aneurysm was not included and conservative management was proposed. Therefore, 8 patients with 9 aneurysms were treated by endovascular approach.

Basilar Artery Fenestration Aneurysms Classification

Based on aneurysm morphology and its relationship to the fenestration characterized by 3D-DSA, we proposed a simple classification. Two parameters were considered essential to determine the type of aneurysm: (1) narrow or wide neck, and (2) base of implantation in the proximal bifurcation of fenestration or asymmetrically in one channel of fenestration, sparing the other.

A wide neck aneurysm was defined as having a neck size of ≥ 4 mm or a dome to neck ratio of < 2 . So, patients were allocated into four types of fBA-AN (Fig. 1).

The fenestration could be located at the proximal, median or distal basilar artery.

Choice of Modality of Treatment

All patients were submitted to endovascular treatment. Management strategy was defined according to the type of fBA-AN mentioned above. Type 1A and type 1B aneurysms were treated using simple coiling. On the other hand, type 2A and 2B aneurysms were treated with stent or balloon assisted coiling. In these cases, stents were preferentially avoided in ruptured aneurysms, unless the balloon remodeling technique was not possible.

Preservation of both loops of the fenestration was aimed in all cases, even if the aneurysm was restricted to only one channel.

Simple Coiling Procedure

All procedures were performed under general anesthesia and heparinization.

Selective coiling procedure was done in a standard fashion. Embolization was carried out with GDC platinum coils (Stryker Neurovascular, Fremont, CA). Coated coils were not used.

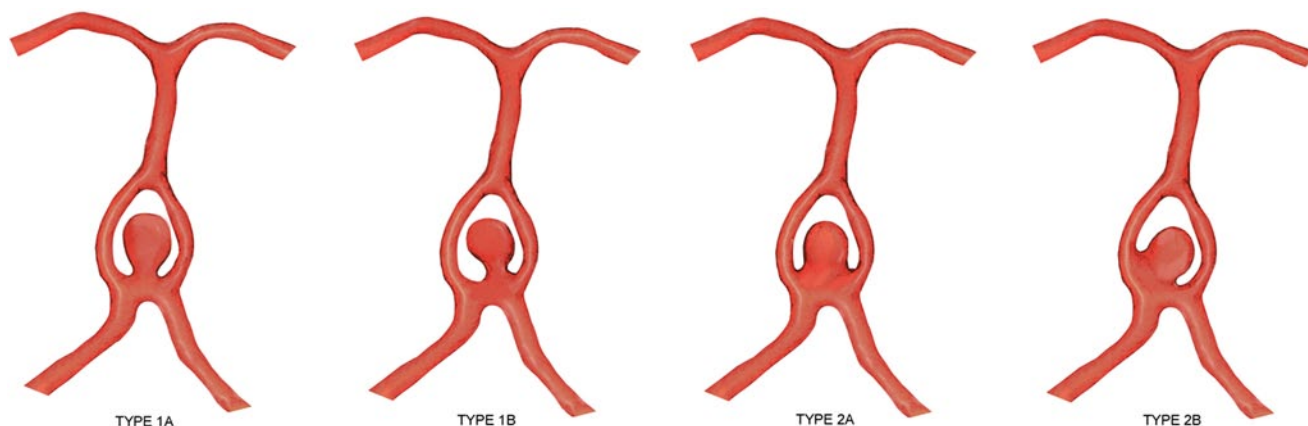


Fig. 1 Types of fenestrated basilar artery aneurysms. 1A: Narrow neck, symmetrically at the bifurcation. 2A: Narrow neck, spars one loop. 1B: Wide neck, involves both loops. 2B: Wide neck, spars one loop

Balloon Assisted Procedure

Balloon remodeling technique (BRT) was performed using a compliant balloon, Hyperform (ev3, Irvine, CA). The balloon guide wire was placed in the loop of fenestration where the aneurysm was located; or in case of bifurcation aneurysm, in the loop closer to the aneurysm neck. No occlusion tests were performed.

Stent Assisted Procedure

Stent assisted coiling was performed under dual antiplatelet therapy. Aspirin and clopidogrel were started 5 days prior to the procedure in case of unruptured aneurysms, and administered just in the beginning of the embolization while dealing with ruptured aneurysms.

Open-cell stent, Neuroform (Stryker Neurovascular, Fremont, CA), was used in all cases. Just one stent was used in each case. Stent was employed under the following strategy regarding three situations: (1) one-channel restricted aneurysm (type 2B)—stent deployment through the involved channel; (2) distal or median fBA-AN located at bifurcation (type 2A)—stent deployment from the proximal basilar artery through the dominant channel; and (3) proximal fBA-AN located at bifurcation (type 2A)—stent deployment from the dominant vertebral artery, crossing the aneurysm neck, to the contralateral channel.

Data Collection

All data were collected retrospectively. Preoperative data included age, sex, initial presentation, aneurysm topography, neck length, dome/neck ratio, and modified Rankin score (mRs). Operative data included modality of treatment, occlusion grade (Montreal scale), and complications. Follow-up evaluation included rebleeding rate, recanalization, and need for retreatment, and outcome assessed by the mRs. A good outcome was defined as a mRs score of 0–2.

We also searched MEDLINE database (1996 to Week 18, May 2013) using the following terms: [(fenestrated OR

fenestration) AND (basilar artery) AND (intracranial OR cerebral aneurysm)]. The reference lists of all papers dealing with fBA-AN were subsequently searched. Papers involving exclusively surgically treated aneurysms were excluded. Aneurysms treated conservatively or not directly related to the fenestration were also excluded. All articles were reviewed and relevant data, as mentioned above, were extracted.

The search yielded 22 articles [3–24]. Of these, there were 13 case reports, 5 nonspecific case series and 4 specific case series.

Results

Patient Demographics

A total of 9 patients harboring 10 fBA-AN were admitted to our institutions from June 2010 and September 2012. All patients, except one with a very small unruptured aneurysm, were endovascularly treated and entered into the study.

There were 7 (87.5%) women and 1 man (12.5%). The mean age was 56.5 ± 9.81 years (median 54.5 years; range 42–75 years). Clinical presentation included subarachnoid hemorrhage in 4 (50%) patients, chronic headache in 1 (12.5%) and was an incidental finding in 3 (37.5%).

Basilar artery fenestration aneurysms were located at the vertebrobasilar junction, the so-called proximal basilar aneurysms (PBA), in 5 (62.5%) patients and at the distal basilar artery (DBA) in 3 (37.5%) patients. One patient had two aneurysms (PBA) pediculated in the bifurcation of the same fenestration, but with independent necks. Therefore, they were analyzed separately.

Overall, 5 aneurysms had wide neck (mean dome/neck ratio of 1.38) while 4 had narrow neck (mean dome/neck of 2.7). According to the previously proposed morphological classification, we observed 2 (22.2%) type 1A aneurysms, 1 type 1B aneurysm (11.1%), 5 (55.5%) type 2A aneurysms and 1 type 2B aneurysm (Table 1).

Table 1 Summary of patients harboring fBA-AN submitted to endovascular treatment

Patient no	Age, sex	Presentation	Site	Type	EVT	Occlusion rate ^b	Complication
1	52, F	SAH	DBA	2A	BRT	B	None
2	64, F	H/A	PBA	2B	Coiling + stent	A	None
3	42, M	SAH	PBA	1A	Coiling	B	None
4	51, F	Incidental	DBA	2A	Coiling + stent	A	None
5	54, F	SAH	DBA	2A	Coiling + stent	B	Perforation
6 ^a	55, F	Incidental	PBA	2A	Coiling + stent	A	None
			PBA	2A	Coiling + stent	A	None
7	75, F	SAH	PBA	1A	Coiling	A	None
8	59, F	Incidental	PBA	1B	Coiling	A	None

SAH subarachnoid hemorrhage, H/A headache, DBA distal basilar artery, PBA proximal basilar artery, BRT balloon remodeling technique

^aTwo aneurysms

^bMontreal scale

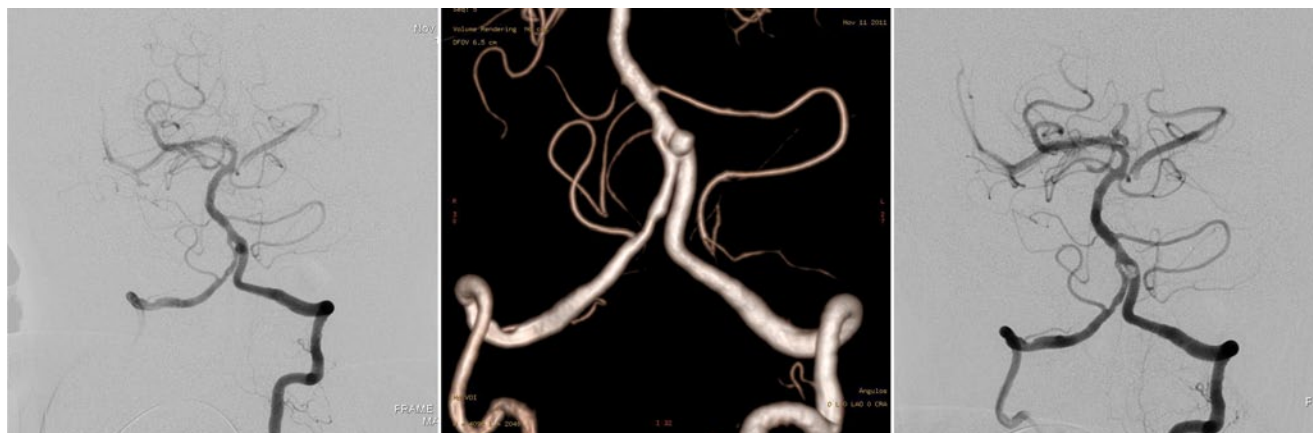


Fig. 2 *Left:* Aneurysm located at the proximal basilar artery fenestration. *Middle:* 3D angiography shows a narrow neck bifurcation aneurysm (type 1A). *Right:* Control angiography demonstrates total aneurysm occlusion and preservation of both channels



Fig. 3 *Left:* Aneurysm located at the distal basilar fenestration. *Middle:* 3D angiography shows a broad neck bifurcation aneurysm (type 2A). *Right:* Control angiography reveals total aneurysm occlusion and patency of both limbs

Endovascular Procedure

Patients with hemorrhagic presentation were treated during the acute phase, between 1 and 9 days after bleeding. On the other hand, unruptured aneurysms were treated electively.

All patients completed endovascular treatment according to the modality previously chosen, named simple coiling, balloon remodeling technique, or stent assisted coiling. A total of 5 (55.5%) aneurysms were treated with stent assisted coiling: 4 type 2A and 1 type 2B; 3 (33.3%) aneurysms with selective coiling: 2 type 1A and 1 type 1B; and 1 (11.1%) aneurysm with balloon assisted coiling: type 2A (Figs. 2 and 3).

Although one patient had a ruptured aneurysm, we decided to perform stent assisted coiling instead of balloon remodeling, once the neck was too large.

Immediate Anatomic Results

Evaluation of postoperative anatomic results indicated total occlusion in 6 (66.6%) aneurysms and neck remnants in 3 (33.3%) aneurysms. Among those uncompleted occluded, it was observed 1 type 1A and 2 type 2A (one submitted to stent assisted coiling and other to balloon remodeling).

Preservation of both channels of the fenestration was possible in all cases.

Complications

We had only one (11.1%) complication related to treatment. In a patient with HH-grade III subarachnoid hemorrhage, harboring a distal ruptured fBA-AN, stent was successfully deployed but while positioning the third coil we experienced aneurysm rupture. Bleeding was controlled deploying more coils inside the aneurysm sac. Despite initial neurological stability, patient developed severe vasospasm and died.

Table 2 Review of patients harboring fBA-AN submitted to endovascular treatment

Reference	Patients/Aneurysms	Hemorrhagic presentation ^b	Site	Type	EVT	Complication
Graves [10]	3/3	1	PBA	2A	Coiling	Thrombus (1)
Pierot [8]	1/1	1	PBA	1A	Coiling	None
Nichols [9]	1/1	1	PBA	1A	Coiling	None
Tasker [14]	5/5	4	4 PBA, 1 MBA	1A, 2A, NM (3)	NM	Failure (1)
Nagashima [6]	1/1	1	MBA	1A	Coiling	None
Islak [7]	10/11	7	9 PBA, 2 DBA	1A (6), 2A (4), 2B (1)	5 Coiling 6 BRT	Sacrifice one loop (1)
Saatci [4]	1/2	1	PBA	1A	Coiling	None
Yoon [5]	4/5	3	PBA	1A (3), 2A (1), 1B (1)	Coiling	Rt VA thrombosis (1)
Kai [11]	2/2	2	PBA	1A, 1B	Coiling	None
Fujimoto [22]	2/2	1	PBA	1A, 2B	Coiling	None
Peluso [12]	7/7	7	PBA	1A, 1B, NM (5)	Coiling	None
Krings [24]	3/6 ^a	2	PBA	2A	Coiling	Late ischemia (1)
Im [20]	1/2	0	DBA	1A	Coiling	None
Albanese [13]	1/1	1	PBA	2A	BRT + SAC + Onyx	None
Juszkat [15]	1/1	1	DBA	1A	Coiling	None
Foroni [16]	1/2	0	MBA	1A	Coiling	None
Gruber [18]	1/1	0	PBA	2A	Waffle cone stent	None
Chung [17]	3/3	2	PBA	2A (1), NM (2)	Coiling, 2 SAC	None
Tanaka [3]	4/4	3	PBA	2A (2), NM (2)	Coiling	Ischemia (2)
Alurkar [19]	1/1	1	PBA	1A	Coiling	None
Kam [21]	1/1	0	PBA	2A	Double stent	None
Stark [23]	1/1	1	PBA	1A	Y stent	Ischemia

DBA distal basilar artery, PBA proximal basilar artery, MBA median basilar artery, BRT balloon remodeling technique, NM not mentioned, SAC stent assisted coiling, EVT endovascular treatment, VA vertebral artery

^aThree patients harboring three kissing aneurysms—one aneurysm treated in each case

^bRelated to the fenestrated artery aneurysm

Clinical Outcome and Follow-up

Three (37.5%) patients with ruptured aneurysms died during the acute phase of hemorrhage. One of them experienced a perioperative ruptured as mentioned above. Other patient presented ventriculitis related to external ventricular drainage and despite adequate treatment, he succumbed. The last one had a refractory vasospasm and died.

Follow-up information for survivors was available at a mean of 16.2 months (median 9 months; range 6–48 months). No patient was lost to follow-up. Five (62.5%) patients had a good outcome. Among them, 4 are totally asymptomatic and 2 with slight disability (mRs=2).

No rebleeding was observed. One subtotally occluded aneurysm was found to have an increasing neck remnant and retreatment was performed without complications.

Literature Data Analysis

Literature search yielded 55 patients with 63 fBA-ANs. Including the present series, we achieved 63 patients with 72 aneurysms. Of these, 40 (72.7%) patients presented with

hemorrhage, followed by 7 (12.7%) cases with headache. Other manifestations included mass effect, distant hemorrhage, and subarachnoid hemorrhage associated with another aneurysm. It was an incidental finding in three patients (Table 2).

The mean age of the patients was 48.4±13.38 years (median 50 years; range 18–75 years). There were 29 (52.7%) woman and 26 (47.3%) men. The aneurysms were located at proximal basilar artery in 48 (87.3%) patients, at distal basilar artery in 4 and median basilar artery in 3.

Among the detailed cases, the aneurysms could be classified as Type 1A in 25 cases, type 1B in 3 cases, type 2A in 21 cases, and type 2B in 2 cases. Regarding the treatment modality, 43 aneurysms were treated with simple coiling, 6 with balloon assisted coiling, and 3 with standard stent assisted coiling. Other strategies included Y stent, waffle-cone technique, double stent technique, and sacrifice of one channel (Table 2).

According to the aneurysm type, they were preferentially treated with the following techniques (detailed cases): (1) type 1A, simple coiling (20/24); (2) type 1B, simple coiling (3/3); (3) type 2A, simple coiling (11/17) and some

technique involving stent (4/17); and (4) type 2B, equally simple coiling and balloon assisted coiling.

Analysis of immediate anatomical results in types 1A, 1B, 2A, and 2B revealed total occlusion of 96, 100, 53, and 100%, respectively. Owing to the neurological status on admission and non-availability of any long-term follow-up, every analysis was very limited.

Discussion

Basilar artery fenestration is an uncommon anatomic variant caused by a failure of fusion of the paired longitudinal neural arteries during the 5th week of fetal life [3]. Although widely used in literature, the term fenestration may not be the most appropriate. It should be applied when a single artery presents with two luminal channels. The condition described in this paper, a lack of fusion of embryologically paired vessels, is more appropriately called segmentally unfused basilar artery [24].

The observation of basilar artery fenestration depends a lot on which diagnostic tool is used. Its incidence is reported to be 2.33% in computed tomography angiography (CTA), 1.0% in magnetic resonance angiography (MRA) and 0.6–1.7% based on digital subtraction angiography [1–3]. Since it has a frequency of 5.26% in autopsy [1], it is expected that more reliable studies, like three-dimensional rotational angiography could more frequently diagnose that variation.

It is known that histologic alterations are present at the fenestrations, the media is absent locally, with discontinuity of elastin at the proximal end of the fenestration and the sub-endothelium is thickened distally and thinned proximally. These morphological changes, together with flow and hemodynamic alterations predispose the formation of aneurysms in the fenestrations [2–4]. Therefore, the presence of an aneurysm occurs in as many as 7% of cases of fenestration [5]. In posterior circulation, most of them involve the vertebrobasilar junction and present with hemorrhage.

The treatment of basilar artery fenestration aneurysms can be challenging. This is in part because of the complex and unusual anatomy of the fenestration and in part due to the intention to preserve both channels of the fenestration. Even if perforator vessels originating from one loop cannot be recognized in angiography, there is no guarantee that its occlusion can be done safely.

Endovascular therapy is no longer reserved exclusively for narrow neck aneurysms. Instead, an increasingly proportion of complex aneurysms have being treated. Basilar artery fenestration aneurysms offer a great opportunity for neuro-interventionalists to put in practice their whole armamentarium, including simple coiling, balloon remodelling technique and several techniques of stent assisted coiling [3–24].

The majority of fBA-AN in literature was treated with simple coiling (78.2%), even those with wide neck. This could be explained because many patients were treated before the development of balloon and stent assisted coiling [3–23]. Regarding balloon remodeling technique, it was employed in only one series, in six aneurysms. Immediate anatomical results were excellent, although sacrifice of a fenestration loop occurred in one case. Two recanalizations were observed on follow-up [7]. Since 2009, initially with Albanese [13], stent-assisted embolization have been frequently used. Many modalities have been described, including Y stent, waffle-cone technique, and double stent technique. In all cases, except one, total occlusion was reached [13,17, 18, 21, 23]. Despite of that, we believe that the use of only one stent per case, with the standard technique is highly efficient and related to lower complications rates, compared to the modified techniques. Even not covering the entire aneurysm neck, the stent transforms a broad neck aneurysm into a narrow neck one.

Endovascular treatment of fBA-AN appeared to be very safe. Literature analysis revealed an overall complication rate of 14.5%. It occurred in two cases of inadvertent occlusion of one loop of the fenestration, but it was well tolerated in both cases. Additionally, 4 cases of ischemic complications, 1 of technical failure, and 1 of thromboembolic event without ischemia were observed. All cases of ischemia were associated to thromboembolic complications of endovascular treatment or vasospasm. No ischemia was demonstrably related to basilar perforators occlusion. Nevertheless, owing to their embryological development, both channels of the unfused basilar artery carry brainstem perforating arteries to their respective sides [24].

Conversely, in our series we had only one (11%) complication. A patient experienced aneurysm rupture during coiling. Despite a high rate of all-causes mortality (37.5%, all ruptured aneurysms), just in the mentioned case, the procedure could be directly responsible for the inadvertent effect. All survivors were independent on follow-up (mRS < 3).

Despite overall good anatomical results, independently of the chosen technique, any attempt was made to systematize the decision making process, maybe because of the limited number of aneurysms in each series.

We proposed a simple and reproducible classification for fBA-AN based exclusively on aneurysm morphology and its relation to the fenestration (established by 3D-DSA). It provides an easy way to characterize these aneurysms. Furthermore, it allows therapeutic planning beyond the simplistic concept of using simple coiling in narrow neck aneurysms and balloon or stent assisted coiling in wide neck aneurysms. While dealing with complex aneurysms and unusual anatomy, to standardize where the balloon and stent should be positioned is very important, as mentioned in *Patients and Methods* section.

Some authors describe a specific type of aneurysm, so-called “kissing” or “mirror” aneurysms, that is, in fact, two aneurysms located at the same fenestration, but with individual necks and pointing for opposites sides [4, 5, 7, 16, 20, 24]. We decided to classify each aneurysm singly.

For a rare condition, a significant number of patients were accrued. We could identify one of the four types of fBA-AN in all cases. Although endovascular treatment was conducted based on this classification, no superiority of this methodology could be identified. So, the real value of this classification and treatment strategy must be validated with prospective and comparative studies, mainly involving all embolization modalities. However, the low incidence of fBA-AN limits the ability to identify outcome differences.

Conclusion

Basilar artery fenestration aneurysms are rare and complex lesions. Endovascular treatment appears to be safe and efficient. The detailed understanding of the aneurysm morphology and its relation to the fenestration is strongly recommended to treatment planning. Further studies are necessary to validate the utility of the proposed classification and treatment strategy.

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Conflict of Interest On behalf of all authors, the corresponding author states that there is no conflict of interest.

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