

Fenestration deformity of the basilar artery trunk with an aneurysm

A case report

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Abstract

Rationale: Fenestration of the basilar artery is most common in the proximal portion near the vertebrobasilar artery junction. Conversely, fenestration of the middle and distal portions of the basilar artery is not common, and fenestration of the basilar artery with an aneurysm is even less common.

Patient concerns: This study reports the case of a 37-year-old woman with basilar artery fenestration malformation and an aneurysm at the mid-distal junction; her symptoms included sudden headaches with nausea and vomiting.

Diagnoses: Head digital subtraction angiography showed fenestration at the junction of the middle and upper portions of the basilar artery associated with an aneurysm, and spontaneous pseudoaneurysm formation could not be excluded.

Interventions: The patient underwent stent-assisted fenestration and channel occlusion.

Outcomes: Five months later, no abnormalities were found by head magnetic resonance imaging. The stents were well positioned, and no occluded branches or aneurysms were present.

Lessons: For mid-distal basilar artery fenestration malformation with an aneurysm, occlusion of the lesion channel is relatively safe when there are no perforating vessels in the fenestration channel and the lesion channel is a nondominant channel. Overall, more attention should be paid to the possibility of pseudoaneurysm formation in the diagnosis and treatment of this type of aneurysm.

Abbreviations: BA = basilar artery, CT = computed tomography, DSA = digital subtraction angiography, L-PCA = left posterior cerebral artery, L-VA = left vertebral artery, MRI = magnetic resonance imaging, R-PCA = right posterior cerebral artery, R-VA = right vertebral artery.

Keywords: aneurysm, basilar artery fenestration, embolization, pseudoaneurysm

1. Introduction

Fenestration is a common variation of intracranial arteries. The fenestration pathway begins with a common origin and splits into 2 endothelial cell-lined ducts that then reconnect distally.^[1,2] Basilar artery fenestration is the 2nd most common intracranial artery fenestration after anterior communicating fenestration.^[1] Fenestration deformity of the basilar artery is most common near the vertebrobasilar artery junction. In contrast, fenestration deformity of the middle and distal basilar arteries is not common, and fenestration deformity with an aneurysm is even less

common.^[3,4] This study reports the case of a patient with basilar artery fenestration deformity and an aneurysm at the mid-distal junction. While pseudoaneurysm formation could not be excluded,^[5,6] it is very rare in the clinic. Currently, little is known about this condition, and relatively few data based on experience with interventional therapies are available. In this case report, we describe our experience regarding the diagnosis and treatment of this condition with a review of the relevant literature.

2. Case report

The patient, a 37-year-old woman, was admitted to our hospital on July 7, 2018, after 5 days of sudden headaches with nausea and vomiting. On July 3, 2018, with no obvious cause for symptoms, her family sent her to the local hospital for treatment. On July 3, 2018, head computed tomography (CT) showed subarachnoid hemorrhage (Fig. 1). On July 4, 2018, CT angiography revealed no obvious abnormalities. After she underwent conservative treatment at a local hospital for 5 days, she achieved relief from the headaches. To obtain a diagnosis and further treatment, she visited our hospital. Physical examination revealed a positive meningeal irritation sign and a positive Kernig sign, and the 2 transverse fingers displayed positive cervical rigidity; no other obvious abnormalities were found. There was no history of hypertension, heart disease, diabetes, or other conditions. She denied a history of trauma or surgery. Head digital subtraction angiography (DSA) performed at our hospital

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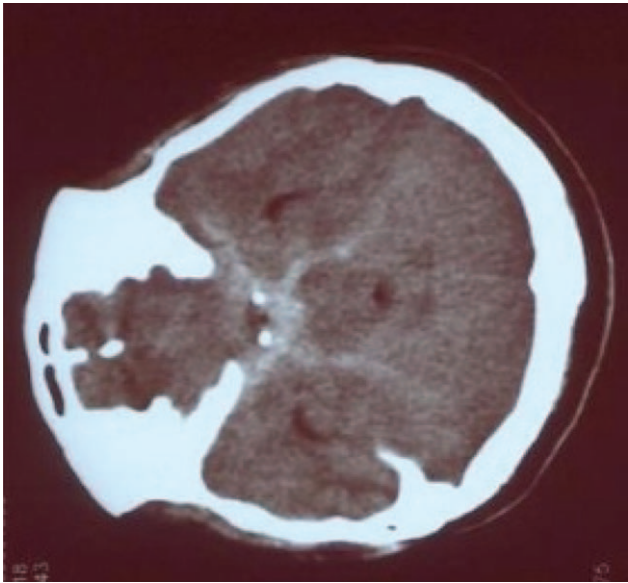


Figure 1. Head computed tomography indicated subarachnoid hemorrhage.

on July 8, 2018, showed fenestration at the junction of the middle and upper portions of the basilar artery with an aneurysm; spontaneous pseudoaneurysms could not be excluded, the bilateral posterior communication was obviously open, and the posterior cerebral artery was well developed (Fig. 2A, B).

2.1. Interventional therapy

The patient underwent stent-assisted fenestration and channel occlusion under general anesthesia. An Enterprise stent 4.5×22 mm in size was placed in the normal fenestration malformation

channel with the 2 ends of the stent completely covering the openings of the lesion channel. Target 360 NANO ($1.5 \text{ mm} \times 3 \text{ cm}$) and helical ($1.5 \text{ mm} \times 2 \text{ cm}$, $1 \text{ mm} \times 2 \text{ cm}$) spring coils were placed to fill in the fenestration malformation channel. Intraoperative routine heparin saline irrigation and continuous anticoagulation therapy were performed. After stent placement, tirofiban was administered intravenously at 10 mL/kg/h to prevent angiogenesis in the stents. Following the procedure, the stents were found to be well positioned, with no lesion channel or aneurysm visible (Fig. 3A, B). After embolization, the patient was lucid and exhibited good limb movement. At 24 hours after the operation, the oral administration of 100 mg of aspirin and 75 mg of Plavix once per day was prescribed to continue the anticoagulation treatment and prevent thrombosis in the stents. Five months later, no abnormalities were found by head magnetic resonance imaging (MRI). The stents were well positioned, and no occluded branches or aneurysms were present (Fig. 4A, B).

2.2. Ethics statement

Informed written consent was obtained from the patient for publication of this case report and accompanying images.

3. Discussion

Intracranial vascular fenestration is a common anatomical variation, with an incidence of 0.3% to 0.9%.^[2] Fenestration refers to paired longitudinal nerve-artery fusion failure or bridging artery degeneration during embryonic development.^[1-4,7-12] Basilar artery fenestration is the 2nd most common intracranial artery fenestration after anterior communicating fenestration.^[1,7] Fenestration can occur at any part of the basilar artery, but it usually occurs at the proximal end near the vertebrobasilar artery junction. Although fenestration at the middle and distal portions has been reported, it is very rare.^[1,3,4,7,8,11] The incidence of basilar artery fenestration is



Figure 2. (A and B) Fenestration deformity of the basilar artery trunk with an aneurysm.

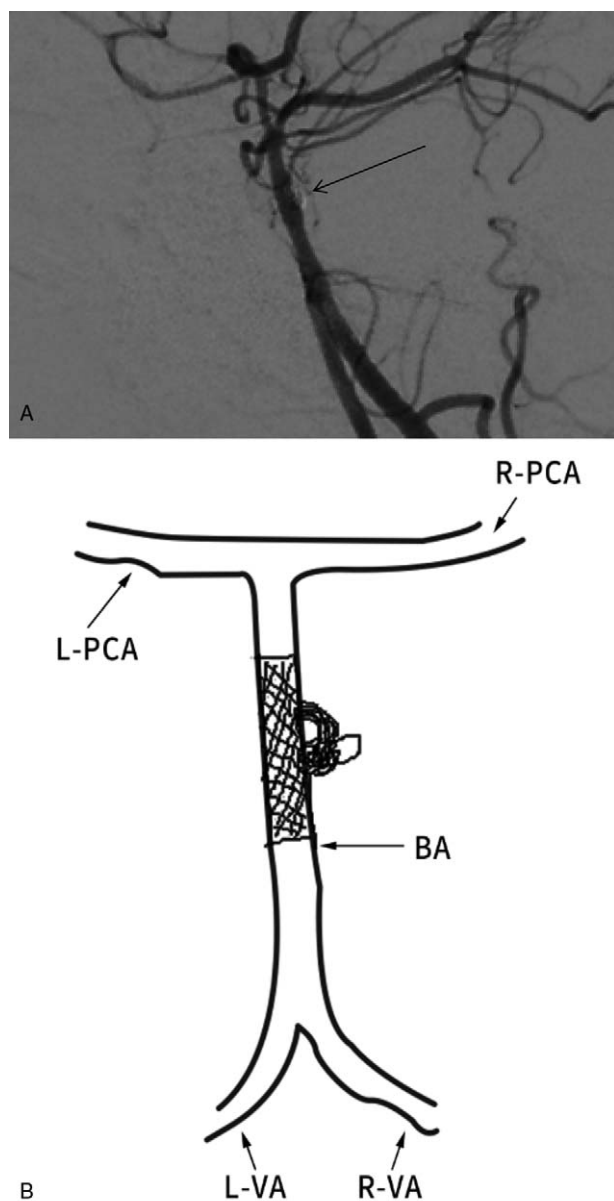


Figure 3. (A) Stent-assisted lesion channel occlusion and (B) pattern diagram. BA=basilar artery, L-PCA=left posterior cerebral artery, L-VA=left vertebral artery, R-PCA=right posterior cerebral artery, R-VA=right vertebral artery.

1% to 2.07% by magnetic resonance angiography, 0.28% to 6% by autopsy, and 0.02% to 0.6% by angiography.^[2,4,8,9,12]

Fenestration malformation may be associated with aneurysms, venous malformations, arteriovenous fistulas, cavernous hemangiomas, and smoking-related vascular changes; they may also be associated with cerebral ischemic events.^[2,4,8,9,13] Histologic changes have been found in fenestration-related deformities, including partial loss of the medial membrane at both ends of the fenestration, discontinuity of elastin near the fenestration, thickening of the subendothelial layer at the distal end with thinning of the proximal end, resulting in weakening of the arterial wall,^[1,4,10–12] and changes related to high-speed turbulence and high-shear-stress hemodynamics at

the fenestration fork. These factors contribute to aneurysm formation and growth.^[1,4,7,8,10,12] Here, we report the very rare case of a patient with fenestration malformation and an aneurysm at the middle/distal junction of the basilar artery. The only case found in the PubMed database was previously reported by Tasker and Byrne.^[10]

This study reports the case of a patient with fenestration malformation and an aneurysm at the mid-distal junction of the basilar artery. The patient was diagnosed with distal fenestration malformation of the basilar artery by head DSA. The proximal end of the fenestration malformation lesion channel showed a nonuniform, filling, and lobulated aneurysm, in accordance with the characteristics of pseudoaneurysms.^[5] The formation of pseudoaneurysms after aneurysm rupture has been reported in the literature.^[5] Pseudoaneurysms do not have a normal vascular wall structure and are prone to rebleeding, endangering the patient's life.^[5,6] However, there are no similar reports in PubMed. Endovascular therapy is currently the best treatment for basilar artery fenestration with an aneurysm. However, the possibility of pseudoaneurysm formation needs to be excluded. Clinically, we consider sacrificing lesion channels therapeutic. Reports in the literature indicate that when angiography does not reveal the origin of the perforator of the fenestration passage and the anatomical relationship is complex, the fenestration passage can be sacrificed.^[11] Moreover, some studies have explored fenestration malformation at the vertebrobasilar artery junction with aneurysm occlusion and fenestration malformation channels. It should be noted that neurologic dysfunction can occur after surgery.^[9,11,14] Nonetheless, there have been no reports of cases in which the fenestration pathway in the middle and distal portions of the basilar artery was sacrificed in the presence of an aneurysm. In general, stent-assisted fenestration and channel occlusion are performed based on comprehensive consideration of the patient. In the case presented here, an Enterprise stent 4.5 × 22 mm in size was placed in the normal fenestration malformation channel; the 2 ends of the stent exceeded both ends of the fenestration malformation and completely covered the opening of both ends of the lesion channel.

Spring coils were also placed in the fenestration lesion channel to prevent drift, which can lead to ischemic events. Because anticoagulants are not administered to patients with a ruptured aneurysm before surgery, in this case, an intravenous infusion of tirofiban was performed during the operation, and oral anti-coagulants were administered after the operation to prevent thrombosis.^[15] After the surgery, the patient was lucid and had good limb movement, and there was no neurologic dysfunction. At 5 months after the operation, no abnormalities were found by head MRI, and head DSA showed no fenestration channels or aneurysms associated with the lesion.

Fenestration in the middle and distal portions of the basilar artery is not common, and fenestration with an aneurysm is rare; hence, experience with interventional treatments for these conditions is relatively insufficient. In the diagnosis and treatment of basilar artery fenestration malformation with an aneurysm, physicians should be vigilant with regard to the possibility of pseudoaneurysm formation. When there are no perforating vessels in the fenestration channel and the lesion channel is nondominant, occlusion of the lesion channel is relatively safe.

Author contributions

Data curation: Yingli Feng.

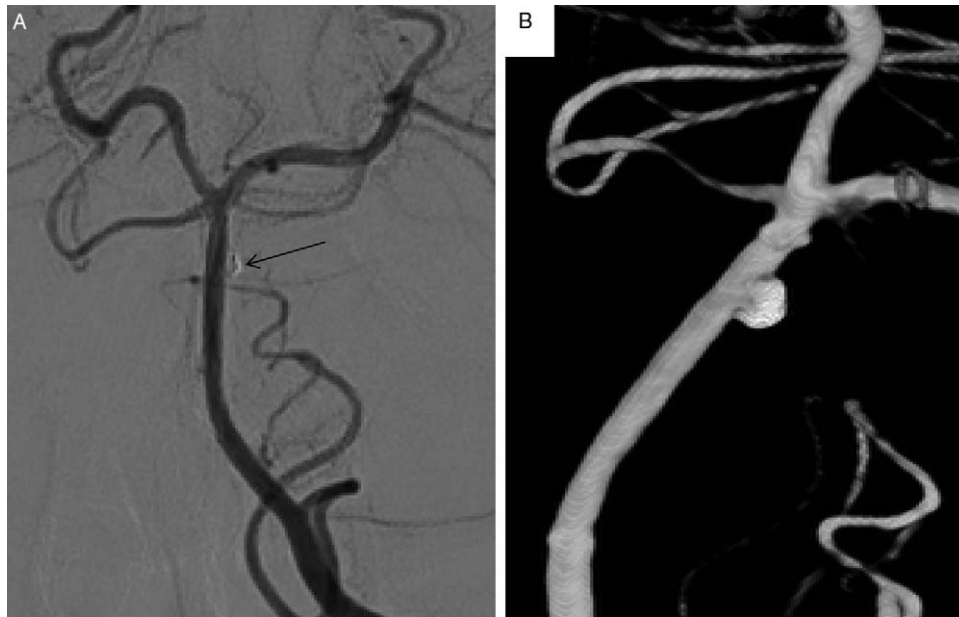


Figure 4. (A and B) Angiography showed that the stent was well positioned and there were no occluded branches or aneurysms.

Methodology: Honglei Wang.

Project administration: Honglei Wang.

Writing – original draft: Donghuan Zhang.

Writing – review & editing: Ning Xu.

References

- [1] Trivelato FP, Abud DG, Nakiri GS, et al. Basilar artery fenestration aneurysms: endovascular treatment strategies based on 3D morphology. *Clin Neuroradiol* 2014;26:73–9.
- [2] Patel MA, Caplan JM, Yang W, et al. Arterial fenestrations and their association with vertebrobasilar cerebral aneurysms. *J Clin Neurosci* 2014;21:2184–8.
- [3] Sogawa K, Kikuchi Y, O'Uchi T, et al. Fenestrations of the basilar artery demonstrated on magnetic resonance angiograms: an analysis of 212 cases. *Interv Neuroradiol* 2013;19:461–5.
- [4] Gao LY, Guo X, Zhou JJ, et al. Basilar artery fenestration detected with CT angiography. *Eur Radiol* 2013;23:2861–7.
- [5] Nomura M, Mori K, Tamase A, et al. Pseudoaneurysm formation due to rupture of intracranial aneurysms: case series and literature review. *Neuroradiol J* 2017;30:129–37.
- [6] Turan N, Butler S, Mason A. Nontraumatic, posterior circulation pseudoaneurysm of the basilar artery summit with complete spontaneous resolution: case report and literature review. *Surg Neurol Int* 2017;8:50.
- [7] Bentura JE, Figueiredo EG, de Monaco BA, et al. Vertebrobasilar artery junction aneurysm associated with fenestration. *Arq Neuropsiquiatr* 2010;68:312–4.
- [8] Cooke DL, Stout CE, Kim WT, et al. Cerebral arterial fenestrations. *Interv Neuroradiol* 2014;20:261–74.
- [9] Tanaka S, Tokimura H, Makiuchi T, et al. Clinical presentation and treatment of aneurysms associated with basilar artery fenestration. *J Clin Neurosci* 2012;19:394–401.
- [10] Tasker AD, Byrne JV. Basilar artery fenestration in association with aneurysms of the posterior cerebral circulation. *Neuroradiology* 1997;39:185–9.
- [11] Islak C, Kocer N, Kantarci F, et al. Endovascular management of basilar artery aneurysms associated with fenestrations. *AJNR Am J Neuroradiol* 2002;23:958–64.
- [12] Foroni LH, Figueiredo EG, Teixeira MJ, et al. Saccular aneurysms at middle basilar trunk fenestration. *Arq Neuropsiquiatr* 2010;68:309–11.
- [13] Wu X, Lin A, Zhu J, et al. Basilar artery fenestration: an unusual possible cause of ischaemic stroke? *BMJ Case Reports* 2018;2018: bcr-2017-222910.
- [14] Kai Y, Hamada J, Morioka M, et al. Endovascular treatment of ruptured aneurysms associated with fenestrated basilar artery. Two case reports. *Neurol Med Chir (Tokyo)* 2006;46:244–7.
- [15] Kim S, Choi JH, Kang M, et al. Safety and efficacy of intravenous tirofiban as antiplatelet premedication for stent-assisted coiling in acutely ruptured intracranial aneurysms. *AJNR Am J Neuroradiol* 2015;37:508–14.