



Spontaneous Obliteration of a Dissecting Aneurysm of Recurrent Artery of Heubner Monitored by Serial Magnetic Resonance Vessel Wall Imaging

Asuka Nakazaki^{1,2} Masaki Ito^{1,2} Masanori Isobe² Takeshi Takahashi³ Taichi Nomura⁴
Fumiaki Fujihara⁵ Toyohiko Isu² Taku Sugiyama¹ Toshiya Osanai¹ Miki Fujimura¹

¹ Department of Neurosurgery, Hokkaido University Hospital, Sapporo, Japan

² Department of Neurosurgery, Kushiro Rosai Hospital, Kushiro, Japan

³ Department of Radiology, Kushiro Rosai Hospital, Kushiro, Japan

⁴ Department of Neurology, Asahikawa Red Cross Hospital, Asahikawa, Japan

⁵ Department of Neurosurgery, Sasebo Chuo Hospital, Sasebo, Japan

Address for correspondence Masaki Ito, MD, PhD, Department of Neurosurgery, Hokkaido University Hospital, North 14, West 5, Sapporo 060-8648, Japan (e-mail: masakiitou-nsu@umin.ac.jp).

AJNS 2022;17:331–336.

Abstract

Aneurysms of the recurrent artery of Heubner (RAH) are known to be one of the uncommon cerebral aneurysms, predominantly presenting with bleeding symptoms. Previously, nine cases of the RAH aneurysms have been reported, all of which were treated surgically or endovascularly and most cases developed postoperative cerebral infarct in the ipsilateral caudate nucleus. Herein, we report a man presenting with transient ischemic attack due to diffuse cerebral vasospasm from a minor non-disabling subarachnoid hemorrhage (SAH) from an RAH aneurysm. He visited our hospital 7 days after the first experience of a thunderclap headache complaining with transient unilateral motor weakness and thin SAH in the right sylvian fissure. Diagnostic catheter angiography revealed a dissecting fusiform aneurysm (8 mm in size) originating from the left RAH contralateral to the thin SAH. Contrast-enhanced magnetic resonance vessel wall imaging (MR-VWI) helped to identify the ruptured nature of the RAH aneurysm. Owing to his delayed ischemic condition after minor SAH, he was conservatively treated with serial MR-VWI monitoring. The aneurysm was spontaneously obliterated with an asymptomatic lacunar infarct in the ipsilateral caudate nucleus in a month. Together, this case was considered as the dissecting aneurysm of RAH with a favorable outcome after the conservative management. Although long-term follow-up is mandatory because the disappearance of the vessel wall enhancement does not necessarily secure the permanent cure of the lesion, serial MR-VWI is helpful to diagnose the ruptured nature and monitor its chronological change in combination with conventional radiological imaging techniques.

Keywords

- ▶ MRI
- ▶ recurrent artery of Heubner
- ▶ vascular disorder
- ▶ vessel wall imaging

DOI <https://doi.org/10.1055/s-0042-1750712>.
ISSN 2248-9614.

© 2022. Asian Congress of Neurological Surgeons. All rights reserved.

This is an open access article published by Thieme under the terms of the Creative Commons Attribution-NonDerivative-NonCommercial-License, permitting copying and reproduction so long as the original work is given appropriate credit. Contents may not be used for commercial purposes, or adapted, remixed, transformed or built upon. (<https://creativecommons.org/licenses/by-nc-nd/4.0/>)

Thieme Medical and Scientific Publishers Pvt. Ltd., A-12, 2nd Floor, Sector 2, Noida-201301 UP, India

Key Messages

Spontaneous obliteration of a dissecting aneurysm of recurrent artery of Heubner could be monitored by serial magnetic resonance vessel wall imaging as a clear contrast enhancement of the affected vessel.

Introduction

Aneurysms of the recurrent artery of Heubner (RAH) are known as uncommon cerebral aneurysms located nearby the horizontal portion of the anterior cerebral artery at the base of the brain. To the best of our knowledge, only nine cases of the RAH aneurysms have been reported, all of which were treated surgically or endovascularly. In most cases, hemorrhagic presentation was the clinical symptom and clinically silent, postoperative cerebral infarcts were observed in the ipsilateral caudate nucleus, except for two cases with unfavorable outcome.¹⁻⁷ Previous reports mostly claimed that RAH aneurysms can be treated surgically because RAH

occlusion might only cause clinically silent cerebral infarct, which is debatable based on the two above-mentioned unexpected cases. Herein, we report a man with a dissecting fusiform aneurysm of RAH, monitored by repeated magnetic resonance (MR) imaging of the vessel wall, which was helpful in identifying the ruptured nature of this aneurysm as a clear contrast enhancement of the affected vessel.

Case History

A 58-year-old man presented to the emergency room of our hospital after transient weakness in his left extremities. He had a thunderclap headache a week ago without any neurological deficit. He had essential thrombocytosis and was on once-daily low-dose aspirin, while his past brain health check-up screening showed no abnormalities in his brain and cerebral arteries. A brain computed tomography (CT) scan showed no subarachnoid hemorrhage. Transient ischemic attack (TIA) due to reversible cerebral vasoconstriction syndrome (RCVS) was suspected at this time. One week later,

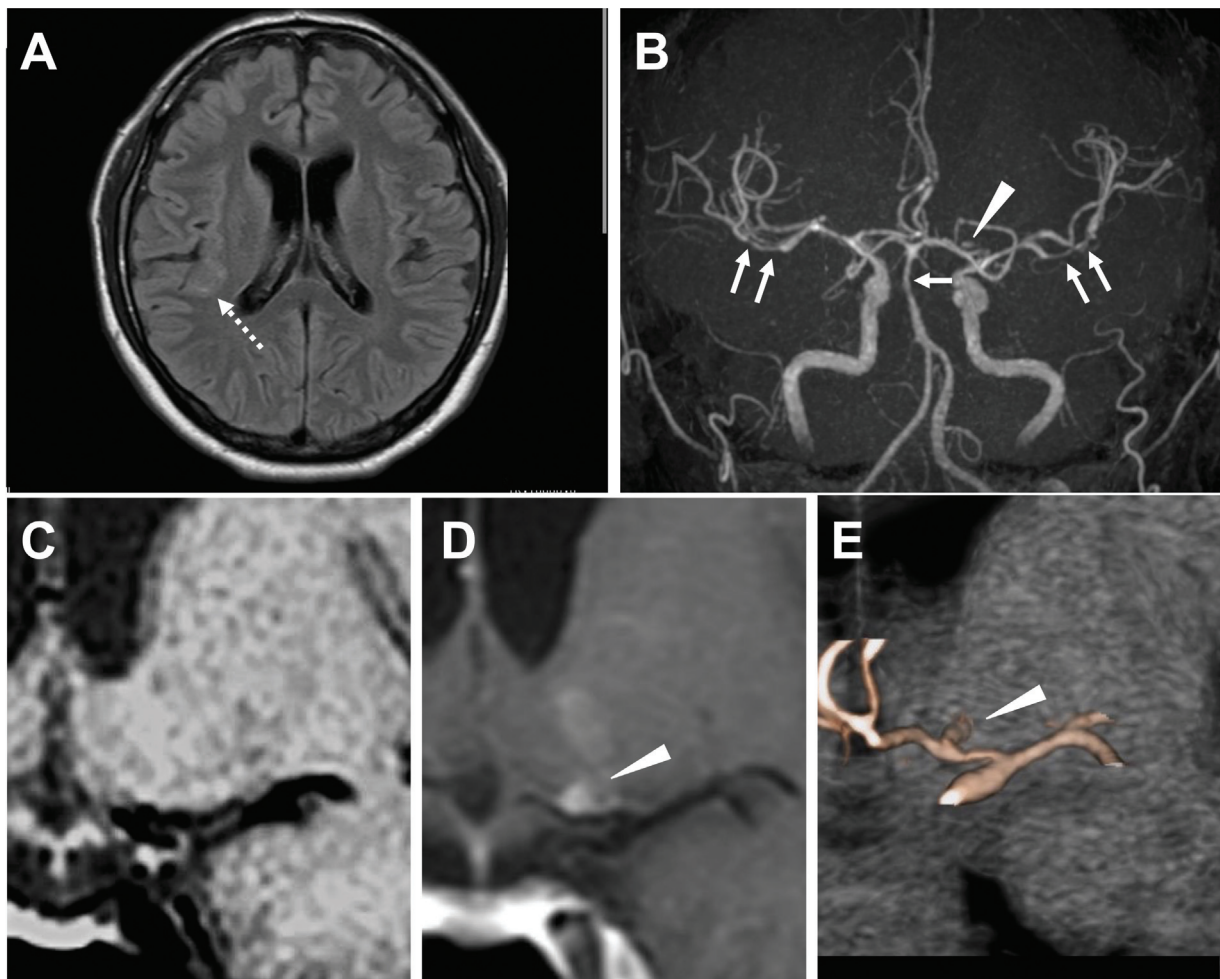


Fig. 1 Magnetic resonance imaging of a 58-year-old man suggesting a dissecting aneurysm of the artery of Heubner. An axial fluid attenuation inversion recovery image (A) shows thin subarachnoid hemorrhage on the right distal Sylvian fissure (*dotted arrow*). The MR angiogram (B) shows diffuse cerebral vasospasm (*arrows*) as well as an aneurysmal lesion on the left A1 (*arrowhead*). Non-contrast-enhanced (C) and post-contrast (D) iMSDE images demonstrate an enhanced aneurysmal lesion on the left A1, which is depicted by the volume rendering of a three-dimensional CT angiogram superimposed on a coronal section of non-contrast-enhanced iMSDE image (E). iMSDE: improved motion-sensitized driven equilibrium.

however, he complained of sustained headache without repeated TIA symptoms (i.e., transient weakness in his left extremities). Brain magnetic resonance (MR) images demonstrated thin subarachnoid hemorrhage (SAH) in the right distal Sylvian fissure (▶Fig. 1A) and MR angiography demonstrated diffuse cerebral vasospasm and a fusiform aneurysm above the horizontal portion of the left anterior cerebral artery (▶Fig. 1B). Notably, MR vessel wall imaging with an improved motion-sensitized driven equilibrium (iMSDE) technique showed marked enhancement of the aneurysm and adjacent brain tissue (▶Fig. 1C–E). Catheter carotid angiography revealed a fusiform aneurysm (8 mm in size) originating from the left RAH, which arose from the junction of the horizontal portion of the left anterior cerebral artery and the anterior communicating artery (▶Fig. 2). There was no evidence of moyamoya-like vessels, vasculitis, or tumor stains in the four vessel observations. Given the “string and pearl” appearance of the aneurysm along with the RAH (▶Fig. 2C) and the clear enhancement of the aneurysm noted during vessel wall MR imaging (▶Fig. 1C–E), a ruptured nature was strongly suspected. Thus, we considered this patient first developed minor subarachnoid bleeding from the dissecting aneurysm of the RAH (presenting with thunderclap headache), followed by TIA as a delayed neurological deficit (DIND) due to diffuse cerebral vasospasm from the thin SAH. Based on these considerations, we decided to start with conservative treatment for cerebral vasospasm with strict blood pressure maintenance and close observation for the rebleeding from the ruptured aneurysm of the RAH. Aspirin therapy was discontinued.

After a month, his headache disappeared, and TIA did not recur. Notably, repeated catheter angiography and MR vessel wall imaging revealed obliteration of the aneurysm and RAH without any neurological sequelae (▶Fig. 3A–D). As expected, MR images showed a clinically silent cerebral infarct in the ipsilateral caudate nucleus (▶Fig. 3E). His clinical outcome was excellent (modified Rankin scale 0 at 3 months after the onset), and daily oral low-dose aspirin was re-started for the essential thrombocytosis. We still continue his follow-up to monitor recurrence of the arterial lesion.

Discussion

We present the first case of spontaneous obliteration of a ruptured dissecting aneurysm of the RAH after a month of conservative treatment. This case showed thunderclap headache as an initial symptom with no evidence of SAH on CT. Consequently, TIA occurred and MR imaging showed thin SAH, suggesting either RCVS or DIND due to diffuse cerebral vasospasm secondary to SAH. Repeat diagnostic work-up revealed a fusiform cerebral aneurysm on the horizontal part of the anterior cerebral artery and diffuse cerebral vasospasm, indicating the patient developed aneurysmal SAH but not RCVS. Catheter angiography clearly demonstrated a fusiform aneurysm located along with the RAH. Moreover, clear contrast enhancement of the aneurysm by the MR vessel wall imaging suggested the ruptured nature of the dissecting aneurysm of the RAH.

There were nine previously reported RAH aneurysms presenting mostly with SAH (seven of nine cases). All were

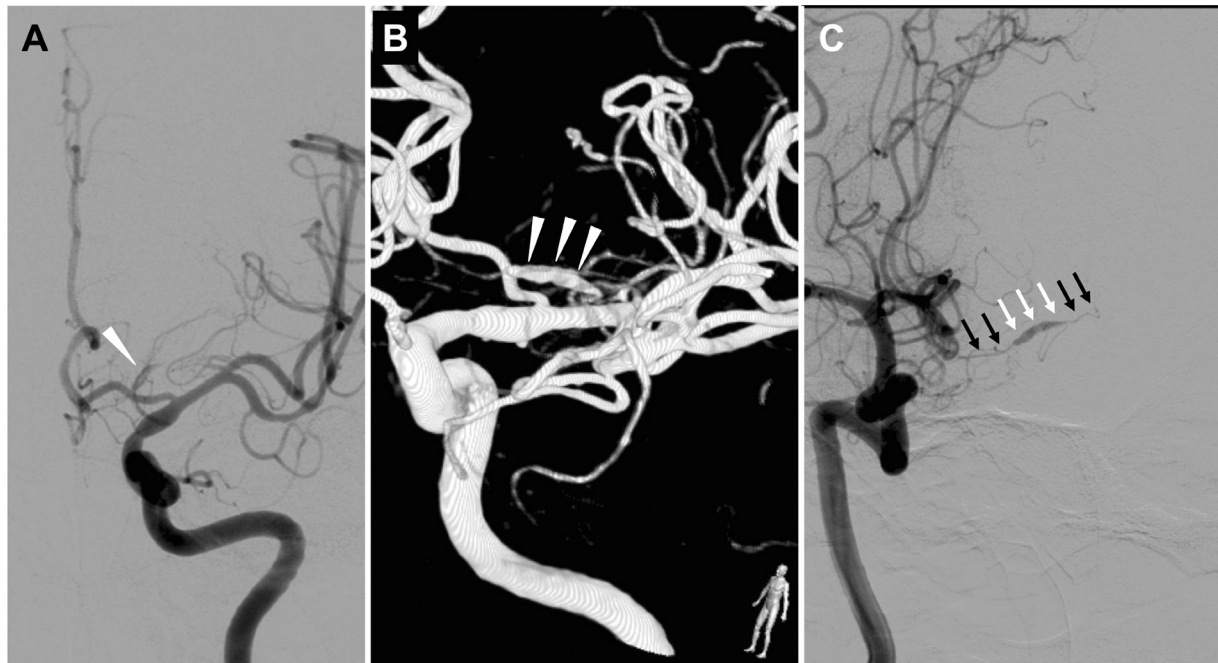


Fig. 2 Catheter angiography showing a recurrent artery of Heubner aneurysm. The anteroposterior view (A) and three-dimensional reconstruction of conventional and rotational angiography (B) of the left carotid artery demonstrates a recurrent artery of Heubner (RAH) fusiform aneurysm (arrow heads) on the left A1. The oblique view of the right carotid artery angiogram (C) shows the fusiform aneurysm (white arrows) more clearly along with the left RAH (black arrows).

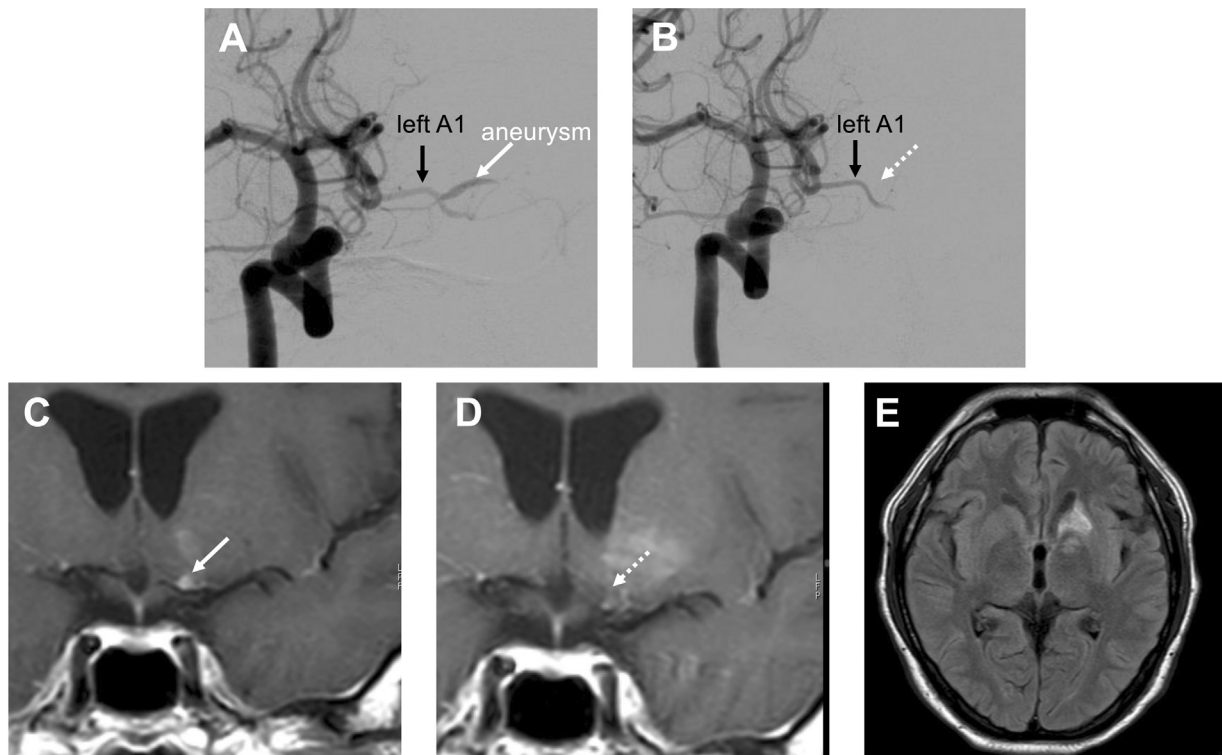


Fig. 3 Serial catheter angiograms and magnetic resonance (MR) vessel wall imaging before and after the conservative treatment showing spontaneous obliteration of the recurrent artery of Heubner aneurysm. Serial catheter angiograms (A, B) and MR vessel wall imaging (C, D) before and after the conservative treatment. The fusiform aneurysm (A) with contrast enhancement on the MR vessel wall imaging (C) was spontaneously obliterated (B), and the contrast enhancement on the affected vessel was reduced. Fluid attenuation inversion recovery image demonstrated cerebral infarct in the ipsilateral caudate head without any neurological symptoms, including cognitive function (E).

treated with open or endovascular surgery, as summarized in **Table 1**.¹⁻⁷

Previous reports all concluded that the RAH aneurysm can be surgically treated with favorable outcome. The postoperative course was uneventful, even if RAH was obliterated after the surgery. However, Ogata et al reported an RAH aneurysm presenting with non-severe SAH, which was resected after trapping of the RAH with postoperative neurological deficit and cerebral infarct in the ipsilateral caudate nucleus.⁷ The deficit was fully recovered; the authors stated that preservation of the RAH might be difficult in surgical treatment of the aneurysm. Mansfield et al reported a dissecting RAH aneurysm in a patient with osteogenesis imperfecta who ultimately died from postoperative severe cerebral vasospasm.⁵ The patient presented with severe SAH (Glasgow coma scale 5); therefore, surgical treatment of the RAH aneurysm was unavoidable and performed successfully with the expected clinically silent cerebral infarct. Our patient presented with TIA due to cerebral vasospasm from non-severe SAH. Presumably, the aneurysmal SAH occurred a week before without any sign of rebleeding. Due to the ischemic symptoms of symptomatic diffuse cerebral vasospasm, we decided to perform conservative management. If rebleeding from the aneurysm occurred or was anticipated, surgical clipping was a treatment option as previously reported.

Our diagnostic effort by catheter angiography and contrast-enhanced vessel wall MR imaging clearly demonstrated the aneurysm of the RAH, suggesting a dissecting nature as the bleeding source of SAH. As mentioned above, RCVS and aneurysmal SAH were suspected first in this case. Khoo et al previously reported a case of RAH aneurysm masquerading as an anterior cerebral artery (A1) aneurysm radiologically and concluded that careful interpretation of the cerebral vasculature shown by angiography was important in this clinical setting.⁶ Although catheter angiography clearly demonstrated the “string and pearl” appearance along with the RAH, we still suspected the possibility of simultaneous RCVS, causing thin SAH and vasospasm, and unruptured RAH aneurysm. Notably, the vessel wall MR imaging was helpful in diagnosing the ruptured nature of this aneurysm, which showed clear contrast enhancement of the aneurysm and adjacent brain tissue. As demonstrated by Omodaka et al previously, the circumferential enhancement of the saccular cerebral aneurysm by vessel wall MR imaging could be useful in differentiating ruptured from unruptured intracranial aneurysms.⁸ Given the fusiform, “string and pearl” appearance and clear contrast-enhancement of the aneurysm, we could diagnose this aneurysm was a dissecting aneurysm as the bleeding source of the SAH in this patient.

Table 1 Summary of past reports and present case for the aneurysm of recurrent artery of Heubner

Author, year	Country	Age	Sex	Clinical Presentation	Underlying condition	DSA findings	Clinical course	Outcome
Bechan, 2014	Netherlands	24	F	SAH	Moyamoya angiopathy	Ruptured right RAH aneurysm	Obliterated (Coiling)	No infarct
Mansfield, 2015	USA	50	F	SAH	Osteogenesis imperfecta	Fusiform left RAH 4mm-aneurysm	Obliterated (Clipping)	Infarct (caudate nucleus/Clinically silent)
Ogata, 2017	Japan	31	M	SAH	None	Fusiform right RAH <5 mm aneurysm	Trapped (Clipping)	Infarct (caudate nucleus/Clinically silent)
Vellore, 2014	Australia	58	F	SAH	Hypertension	Saccular right RAH 2.7 mm aneurysm	Obliterated (Clipping)	No infarct
Wanibuchi, 2001	Japan	56	M	SAH	None	Saccular right RAH 5 mm aneurysm	Obliterated (Clipping)	N.D.
		66	M	Incidental	None	Saccular left RAH 2.5 mm aneurysm	Obliterated (Clipping)	N.D.
		65	M	Incidental	None	Saccular left RAH 2.7 mm aneurysm	Obliterated (Clipping)	N.D.
Khoo, 2019	Australia	71	F	SAH	None	Left RAH 5 mm aneurysm	Obliterated (Clipping)	Infarct (caudate nucleus/Clinically silent)
Hong, 2019	Korea	53	F	SAH	None	Left RAH 8 mm aneurysm	Obliterated (Clipping)	Infarct (caudate nucleus/Clinically silent)
Nakazaki, 2021 (present case)	Japan	58	M	Vasospasm from SAH	Essential thrombocythemia	Fusiform left RAH 8 mm aneurysm	Obliteration (Spontaneous)	Infarct (caudate nucleus/Clinically silent)

Conclusion

In conclusion, we experienced a case of RAH aneurysm presenting with delayed cerebral ischemia due to diffuse cerebral vasospasm with minor SAH who was conservatively treated. Serial MR vessel wall imaging with contrast enhancement is helpful to diagnose the ruptured nature and monitor its chronological change in combination with conventional radiological imaging techniques. Long-term follow-up is mandatory because the disappearance of the vessel wall enhancement does not necessarily secure the permanent cure of the lesion.

Note

This paper was presented at Mt. Fuji workshop on CVD held at Sendai, Japan, on August 28, 2021.

Funding

None.

Conflict of Interest

None declared.

References

- 1 Wanibuchi M, Kurokawa Y, Ishiguro M, Fujishige M, Inaba K. Characteristics of aneurysms arising from the horizontal portion of the anterior cerebral artery. *Surg Neurol* 2001;55(03):148–154, discussion 154–155
- 2 Vellore Y, Madan A, Hwang PYK. Recurrent artery of Heubner aneurysm. *Asian J Neurosurg* 2014;9(04):244
- 3 Hong CE, Chung Y, Won YS, Chung P-W, Rho MH. Ruptured aneurysm of recurrent artery of Heubner: a case report. *Nerve* 2019;5:94–96
- 4 Bechan RS, van Rooij WJ. Endovascular treatment of a ruptured flow aneurysm of the heubner artery as part of a moyamoya collateral network in a young patient with an occluded middle cerebral artery. *Interv Neuroradiol* 2014;20(06):791–795
- 5 Mansfield K, Rahme R. Dissecting aneurysm of the recurrent artery of Heubner in a patient with osteogenesis imperfecta. *Can J Neurol Sci* 2015;42(06):461–465
- 6 Khoo J, Khoo B, Alalade AF, Webster J. Case of recurrent artery of Heubner aneurysm masquerading as partially thrombosed A1 aneurysm radiologically. *World Neurosurg* 2019;130:444–449
- 7 Ogata A, Sakata S, Okamoto H, Abe T. Ruptured dissecting aneurysm of the recurrent artery of Heubner: consideration of pathological findings. *Neurol India* 2017;65(03):623–625
- 8 Omodaka S, Endo H, Niizuma K, et al. Quantitative assessment of circumferential enhancement along the wall of cerebral aneurysms using MR imaging. *AJNR Am J Neuroradiol* 2016;37(07):1262–1266