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Case Report

A Case of De Novo Basilar Artery Aneurysm Associated with Proximal Stenosis Treated by Coil Embolization

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Objective: This report highlights a case of a de novo aneurysm assumed to be caused by hemodynamic stress resulting from proximal basilar artery stenosis.
Case Presentation: A 76-year-old woman presented at our hospital with tinnitus. Although MRI did not reveal the cause of her tinnitus, it did uncover an incidental finding of basilar artery stenosis. The patient reported a history of cerebral infarction, diabetes, and hypertension. Six years following the initial discovery of basilar artery stenosis, a saccular aneurysm was detected at the bifurcation of the basilar artery and the right anterior inferior cerebellar artery, corresponding to the distal portion of the basilar artery stenosis. Upon revelation of an enlarged aneurysm on the subsequent two-year follow-up MRI, the patient received coil embolization treatment. No signs of recurrence were observed on the next two-year follow-up MRI.
Conclusion: It was assumed that proximal basilar artery arteriosclerotic stenosis had caused hemodynamic stress on the distal vessel wall, and that this was responsible for the formation and growth of a de novo aneurysm. This case suggests that cerebrovascular arteriosclerotic changes may be associated with de novo aneurysm formation and therefore requires careful follow-up.

Keywords ▶ de novo aneurysm, hemodynamics, wall shear stress, arteriosclerotic stenosis

Introduction

Hemodynamic factors are considered to be involved in the development of de novo cerebral aneurysms; however, cases establishing this clear causal relationship are rare. We report the case of a patient with a de novo cerebral aneurysm associated with proximal stenosis that was enlarged and treated with endovascular coil embolization. We have also presented a literature review on the subject.

Case Presentation

Patient: A 76-year-old woman.

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Medical history: The patient has taken ticlopidine (100 mg) orally since she had medullary infarction at the age of 50 years. The patient is being treated for diabetes and hypertension.
Social history: No history of cigarette smoking.
Present illness: Basilar artery stenosis was noted incidentally on MRI performed for examination of tinnitus in year X (**Fig. 1A**), and the course was followed thereafter. After a period of four years, aggravation of stenosis was suspected (**Fig. 1B**), but course observation continued because the condition was asymptomatic. After six years, a saccular bulge was noted at the bifurcation of the basilar artery–right anterior inferior cerebellar artery (BA–AICA), located distal to the basilar artery stenosis (**Fig. 1C**). Moreover, this lesion enlarged gradually during the subsequent two-year course (**Fig. 1D**) and was thus treated with coil embolization to prevent rupture.

Angiography findings

On vertebral angiogram, stenosis and tortuosity of the basilar artery were observed in the proximal region of the right anterior inferior cerebellar artery bifurcation of the basilar artery, and an aneurysm projected toward the right-side wall of the basilar artery, which was the distal region of the stenosis. The neck length of the aneurysm was 3.2 mm and

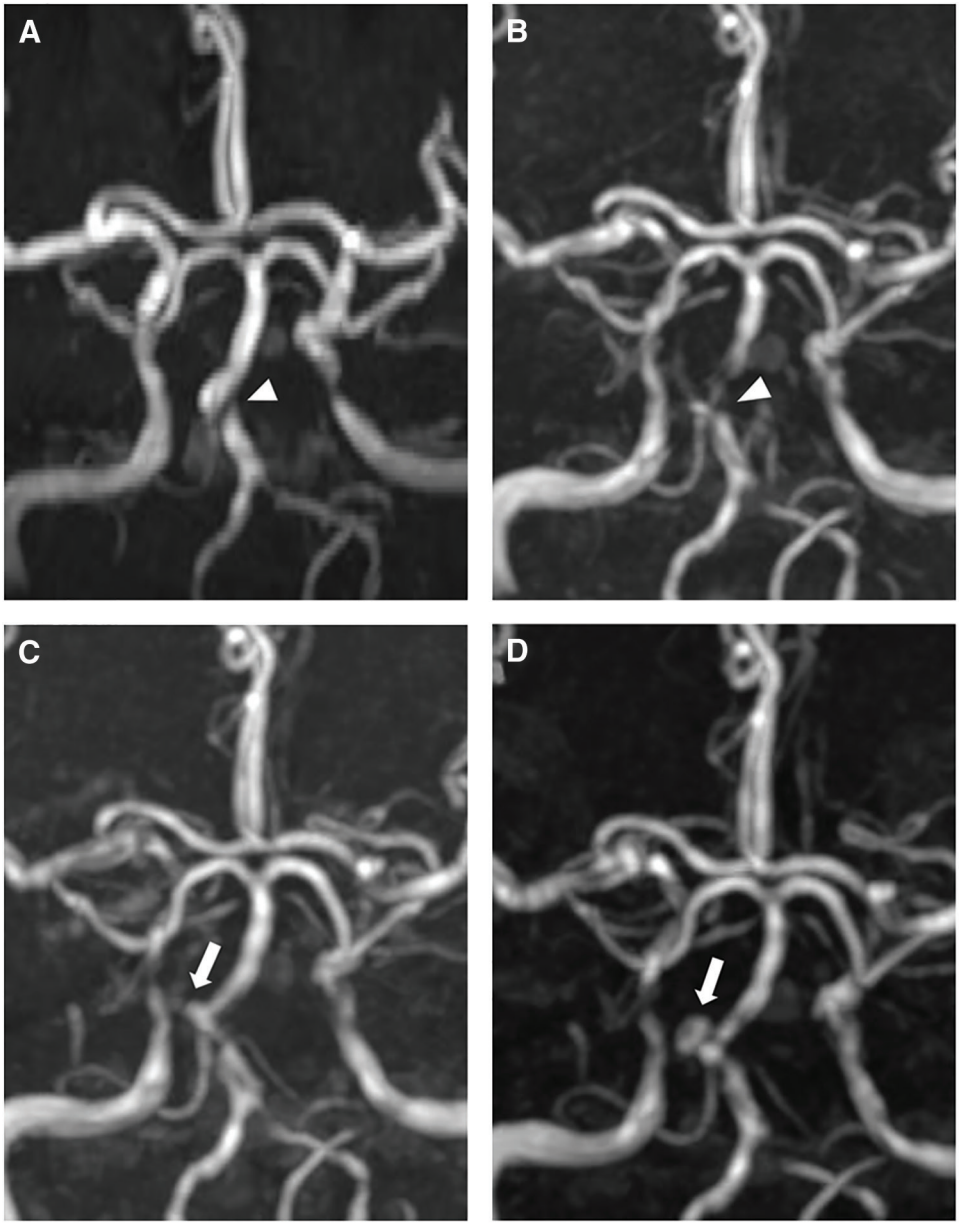

Fig. 1 Time-course changes in MRA findings. (**A**) In year X, stenosis of the basilar artery is noted (arrowhead). (**B**) In year X+4, aggravation of stenosis, a change in the direction of blood flow in the basilar artery in the region of distal stenosis, or Venetian blind artifact-induced intensity reduction was noted (arrowhead). (**C**) In year X+6, a saccular protrusion was noted in the distal stenosis (arrow). (**D**) In year X+8, the protrusion increased in size and clearly became an aneurysm (arrow).

its size was 4.2 × 5.4 × 4.1 mm. The degree of basilar artery stenosis was 56% based on the warfarin-aspirin symptomatic intracranial disease method, and the distance from the stenosis to the neck of the aneurysm was 3.1 mm.

During the observation period, a de novo aneurysm developed and gradually increased in size, which needed therapeutic intervention. As the involvement of proximal stenosis as a cause was considered, percutaneous angioplasty for stenosis and conversion of the bloodstream by concomitant use of stents in aneurysm treatment were considered. However, there was no evidence of ischemic change attributed directly to the stenosis during the eight years, and angioplasty was considered to have a risk of perforating branch occlusion. Moreover, thromboembolic complications during the postprocedural period were of concern in view of the diffuse arteriosclerotic changes in the parent vessel. As such, we decided to perform only intra-aneurysmal embolization without stenosis treatment. In the case of recurrence during the postoperative course, stenosis treatment as well as simultaneous aneurysmal re-coil embolization was performed. Ticlopidine

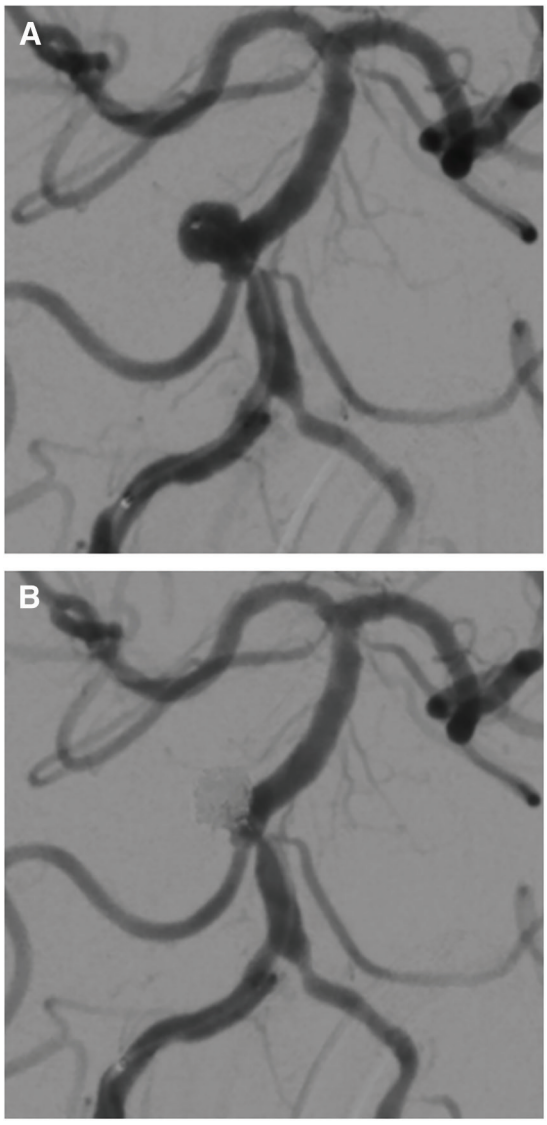

Fig. 2 Intraoperative angiography findings. (**A**) Before coil embolization. (**B**) After coil embolization. The aneurysm was completely occluded.

(100 mg) was changed to clopidogrel (75 mg) one week before embolization.

Surgical findings

After the induction of general anesthesia, a 6-Fr sheath was inserted from the right femoral artery, heparin was administered to adjust the activated clotting time (ACT) to 250–300 seconds, and a 6-Fr Envoy (Johnson & Johnson, New Brunswick, NJ, USA) was placed in the right vertebral artery. An Excelsior SL-10 STR (Stryker, Kalamazoo, MI, USA) was guided into the aneurysm using Traxcess (Terumo, Tokyo, Japan). After placement of the Axium Prime Frame 5 mm × 10 cm (Medtronic, Minneapolis, MN, USA) as the first coil, we decided to use the double catheter technique as the first coil mass was unstable. An Excelsior XT-17 Pre-Shaped

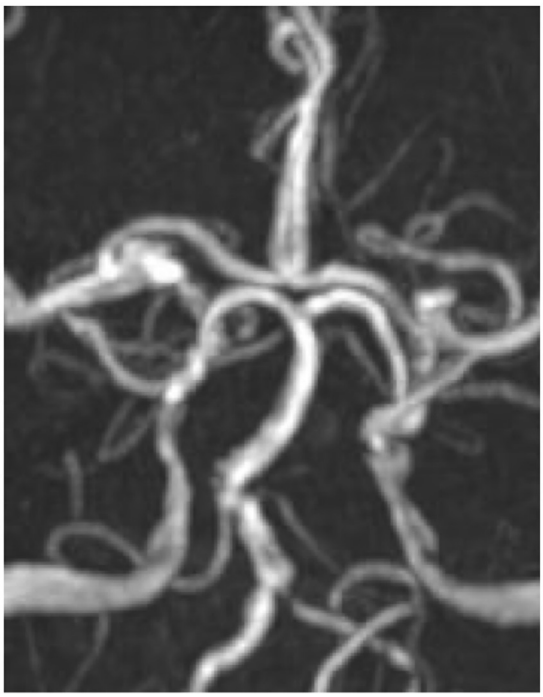

Fig. 3 MRA findings at two years after treatment. No recurrence of the aneurysm.

45 (Stryker) was guided into the aneurysm using a CHIKAI14 (Asahi Intecc, Aichi, Japan) and a Target 360 Ultra 4 mm × 8 cm (Stryker) was placed. At this time point, the stabilization of coil mass was achieved. After that, a total of five coils (30 cm) were inserted, and finally, the aneurysm was obliterated completely (**Fig. 2**).

Postoperative course

There were no postoperative neurological deficits, and the patient was discharged on the fifth postoperative day. During the postoperative period of two years, there was no recurrence on follow-up MRA (**Fig. 3**), and the course was favorable.

Discussion

De novo cerebral aneurysm was defined as a newly identified aneurysm at the second or later examination after the exclusion of a cerebral aneurysm on the first imaging. It is necessary to exclude the possibility of being not visualized or overlooked, even though the aneurysm was present during the first examination. Regarding the mechanisms responsible for being not visualized, microaneurysms, thrombosed aneurysms, poor visualization of the aneurysm due to blood flow, and technical reasons due to the imaging apparatus have been considered.[1] In the present case, the first MRI imaging was done through Intera 1.5T (Philips Medical

Systems, Best, The Netherlands); however, Vantage 1.5T (Toshiba Medical Systems, Tochigi, Japan) was used for subsequent imaging evaluation from year X+4 to X+8 with similar acquisition conditions: 3D time-of-flight method; repetition time, 23 ms; and echo time, 6.8 ms. However, strict matching of the slab angle is difficult. Although the signal intensity may appear different due to the slab surface, vascular angle, and blood flow direction, the possibility of intensity reduction (Venetian blind artifact) at the joint between slabs in multiple overlapping thin-slab acquisitions cannot be excluded. The aneurysm in the present patient must be judged as a de novo cerebral aneurysm as its development and enlargement could be observed. On MRA, poor visualization of the basilar artery distal to the stenosis was evident for the first time in year X+4 (**Fig. 1B**). This poor visualization might be caused by the Venetian blind artifact because intensity reduction was noted in the internal carotid artery on the same cross-section as well as in the same region on the original MRA image. On the contrary, this poor visualization on MRA might be caused by a jet flow produced by the proximal stenosis. The impingement of the jet flow at the distal vascular wall might induce turbulence, resulting in poor visualization. The reason for absence of intensity reduction in this region in year X+6 or later was presumed to be stabilization of blood flow by the rectifying effect resulting from de novo aneurysm formation.

The incidence of de novo cerebral aneurysms is approximately 0.3%–1.8%/year, and females, cigarette smokers, hypertension, and multiple cerebral aneurysms are included among the risk factors according to the previous reports.[2,3] However, patients with multiple aneurysms or a medical history of subarachnoid hemorrhage were evaluated in many studies, suggesting that the true incidence of de novo aneurysms might be lower than that of previous reports.

In addition to the structural vulnerability of the blood vessel wall, hemodynamic factors play an important role in the development of de novo aneurysms. Vascular vulnerability includes the presence of defects of the tunica intima, destruction of the elastic lamina, and normal variations, such as persistent carotid-basilar anastomosis and fenestration, as congenital factors, and vascular wall degeneration, arteriosclerosis, and hypertension as acquired factors.[4,5] The present patient had a medical history of diabetes and hypertension, which were the main factors for cerebrovascular arteriosclerotic changes, suggesting an important role of acquired factors.

Several clinical reports on hemodynamic factors have been previously described. De novo aneurysm formation at the contralateral internal carotid artery–posterior communicating artery bifurcation after therapeutic carotid artery occlusion has been reported.[6] In addition, cases of de novo aneurysm formation on the wall opposite to the middle cerebral artery T-shape anastomosed with the donor and on the greater curvature side of the curved middle cerebral artery and superficial temporal artery distal to the anastomosed region after external carotid artery–internal carotid artery (EC–IC) bypass have been reported,[7] suggesting the involvement of hemodynamic stress. Regarding the pathophysiology of cerebral aneurysms using computational fluid dynamics (CFD), many studies focusing on wall shear stress (WSS), which is the force of blood flow rubbing against the vascular endothelium, have been published. High WSS promotes angiogenesis, which is involved in vasodilation, vascular tortuosity, and the development of aneurysms.[8,9] In contrast, low WSS induces degeneration of the vascular wall (angioregression), leading to arteriosclerosis and apoptosis.[10–12] When a high WSS-induced aneurysm grows to a certain size, the blood flow in the aneurysm decelerates and the WSS decreases. When there is a reduction of WSS, degeneration occurs in the aneurysm wall, suggesting the possibility of rupture.[13] However, there are opposing opinions that both high WSS[14] and low WSS[15] are involved as factors of rupture; thus, the integrated conclusion remains controversial.

Previous studies on cerebral de novo aneurysm formation have mainly investigated vascular bifurcation aneurysms; however, there are few studies on proximal vascular stenosis-induced de novo aneurysms. Kono et al. reported four cases of cerebral aneurysms associated with proximal stenosis, in which they performed CFD analysis in two.[16] Focusing on WSS and the shear stress gradient (wall shear stress gradient [WSSG]), which is a quantitative parameter evaluating the homogeneity of the distribution of WSS, they stated that jet blood flow induced at the proximal stenosis generates high WSS and high positive WSSG at the collision site, leading to aneurysm formation. According to their analysis, when the degree of stenosis is higher than 40% or the distal vascular wall is located within 10 mm of the stenosis, WSS and WSSG can drastically increase in parallel with an increase in the stenotic ratio or shortening of the distance from the stenosis. In the present patient, although CFD analysis was not performed, the degree of stenosis was 56% and the distance from the stenosis to the aneurysmal neck was 3.1 mm, fulfilling the conditions to produce high WSS and high positive WSSG.

Generally, BA–AICA aneurysms are relatively rare. Several studies have reported that aneurysms arising from

the basilar artery account for 5.9%–7.4% of all cerebral aneurysms.[17,18] Moreover, according to a summary of ruptured basilar artery aneurysms reported by Nukui et al., three (3.1%) of 94 cases were BA–AICA aneurysms.[19] In addition to the low frequency of BA–AICA aneurysms, it might be considered that the aneurysm in the present case was produced and enlarged by strong hemodynamic stress due to the proximal atherosclerotic stenosis, based on the anatomical characteristics such as the aneurysm formation site and running direction of the basilar artery. In cases of intracranial atherosclerotic stenosis, a careful and close follow-up is needed to look for de novo aneurysm formation as well as ischemic changes in the long term.

Conclusion

We report a patient in whom aggravation of arteriosclerotic changes of the basilar artery and de novo aneurysm formation and enlargement were observed and treated during the eight-year course. Arteriosclerotic changes may be associated with de novo aneurysm formation due to hemodynamic stress, for which careful long-term observation is necessary.

Disclosure Statement

The authors declare no conflicts of interest.

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