Case Report

Rupture of a Left Posterior Inferior Cerebellar Artery Aneurysm with a Vertebral Artery Originating from the Aorta

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Objective: Cerebral aneurysms (ANs) in the cortical segment (CS) of the distal posterior inferior cerebellar artery (PICA) with a vertebral artery (VA) of aortic origin are markedly rare. Endovascular therapy was performed to treat subarachnoid hemorrhage caused by a ruptured cerebral AN.

Case Presentation: The patient was a 68-year-old female who was transported to emergency care for headache. Detailed examination revealed an AN in the CS of the PICA with a left VA of distal aortic origin from the left subclavian artery (LT. SA). Endovascular therapy using n-butyl-2-cyanoacrylate (NBCA) was performed to treat the cerebral AN, resulting in a favorable outcome.

Conclusion: Endovascular therapy for cerebral ANs is an effective treatment method.

Introduction

Posterior inferior cerebellar artery aneurysms (PICA ANs) are relatively rare. Distal PICA ANs peripheral to vertebral artery-posterior inferior cerebellar artery aneurysms (VA-PICA ANs) are markedly rare. In particular, among distal PICA ANs, the incidence of cortical segment (CS) aneurysms (CSANs) is low.

Furthermore, few studies have reported CSANs with a VA originating from the aorta, especially that originating from an area distal to the left subclavian artery (Lt. SA).

We report a patient in whom endovascular treatment for subarachnoid hemorrhage related to CSAN rupture with a VA of aortic origin, originating from an area distal to the Lt. SA, led to a favorable course.

Case Presentation

A 68-year-old female. Headache developed at 21:00 the day before arrival. It exacerbated the following morning, and she was transported to her previous hospital by ambulance. Head computed tomography (CT) revealed subarachnoid hemorrhage, and she was referred to out hospital. On arrival, the Japan Coma Scale (JCS) score was 1 and there was no paralysis of the limbs. Vomiting was noted. Concerning physical findings, the body temperature, blood pressure, and pulse rate were 36.5°C, 170/88 mmHg, and 88/min, respectively. She had a history of hypertension. Head CT demonstrated subarachnoid hemorrhage (Fisher group 3, World Federation of Neurosurgical Societies (WFNS) grade II, Hunt & Kosnik grade II) and intraventricular hemorrhage (**Fig. 1A** and **1B**). Head computed tomography angiography (CTA) revealed an AN-like finding in the CS of the left PICA (**Fig. 1C**). Subsequent cerebral angiography other than left vertebral arteriography did not reveal any abnormalities as the source of hemorrhage. The left VA was not
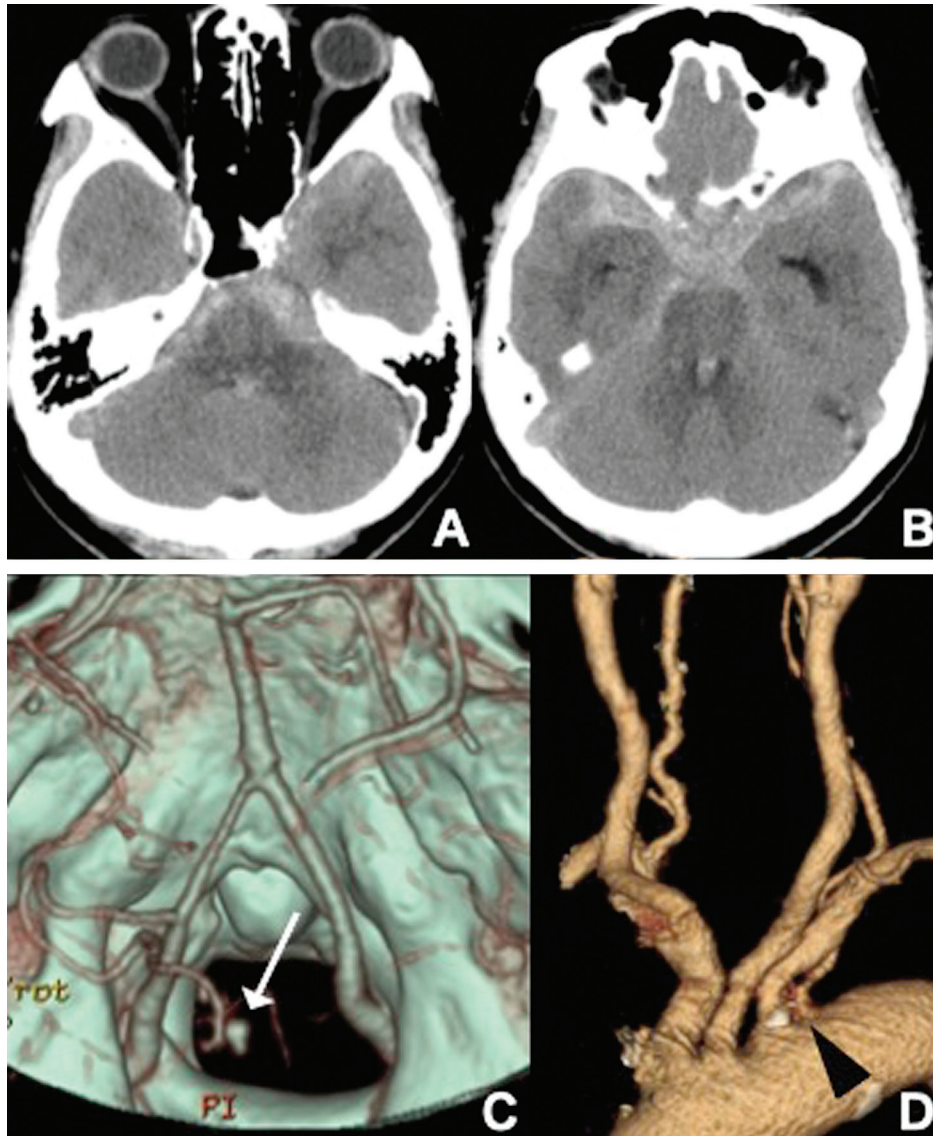
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Fig. 1 Head CT revealed subarachnoid hemorrhage (Fisher group 3, WFNS grade II, Hunt & Kosnik grade II) and intraventricular hemorrhage (**A** and **B**). Head CTA showed an AN-like finding (white arrow) in the CS of the left PICA (**C**). CTA of the head and neck revealed a VA of aortic origin, originating from an area distal to the Lt. SA (black arrow head: origin of the left VA) (**D**). AN: aneurysm; CS: cortical segment; CTA: computed tomography angiography; Lt. SA: left subclavian artery; PICA: posterior inferior cerebellar artery; VA: vertebral artery

visualized on left subclavian arteriography and a VA originating from the aorta was detected. It was difficult to select the left VA using a catheter, and left vertebral angiography was unable to be performed. For confirmation, three-dimensional (3D) angiography of all vessels, excluding the left VA, was performed; therefore, at this point, a large volume (300 cc) of contrast medium had been used and aortography was not conducted. Head CT demonstrated enlargement of the ventricle and emergency ventricular drainage was performed. The patient was managed in the stroke care unit (SCU). Although a large volume of contrast medium was used, there was no deterioration of renal function. On Day 3, CTA of the head and neck revealed a VA of aortic origin, originating from an area distal to the Lt. SA (**Fig. 1D**). Cerebral angiography confirmed a "pearl and string" sign in the CS of the left PICA. On the same day, endovascular treatment was performed (**Fig. 2**).

Under general anesthesia, a 6-Fr Roadmaster (GOODMAN CO., LTD., Seto, Aichi, Japan) was guided to the V1 segment of the left VA and a 4-Fr Cerulean (Medikit Co., Ltd., Tokyo, Japan) was guided to the V2 segment of the left VA. Using a Traxcess (Terumo Corporation, Tokyo, Japan),
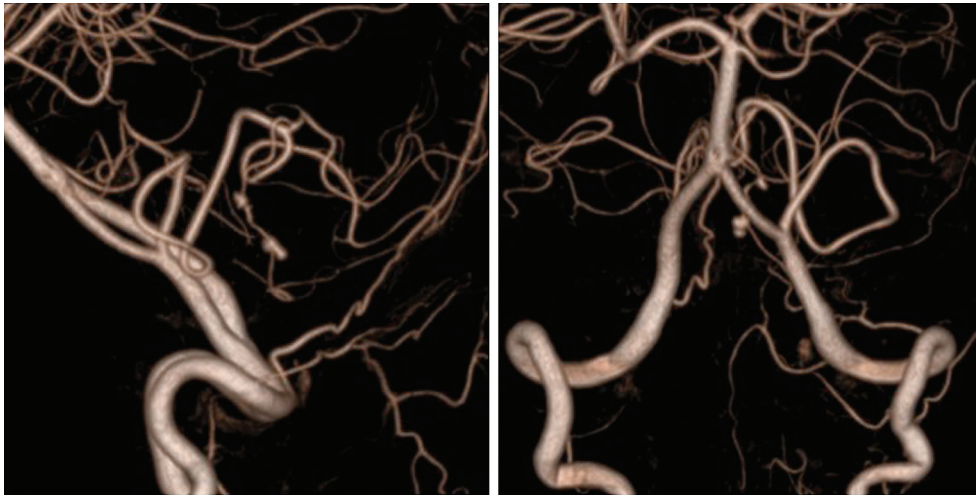
Fig. 2 Cerebral angiography showed a fusiform cerebral AN in the CS of the left PICA (volume rendering, left: lateral view, right: frontal view). AN: aneurysm; CS: cortical segment; PICA: posterior inferior cerebellar artery

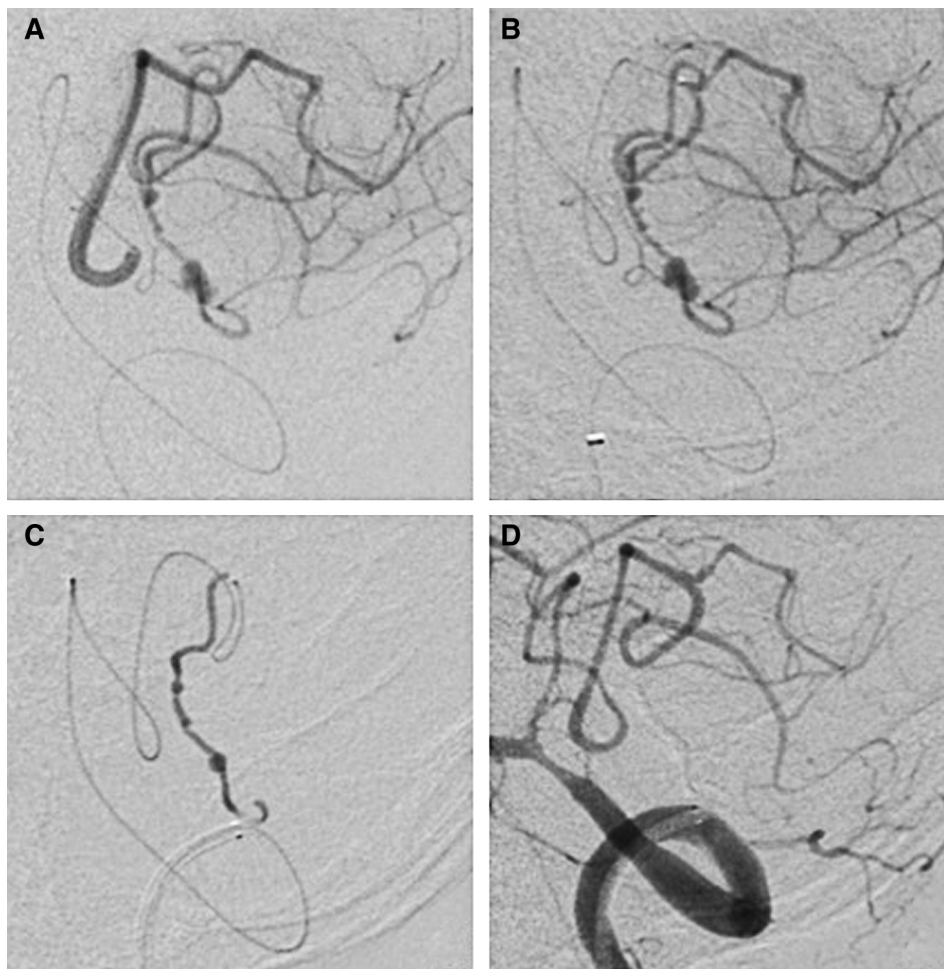**Fig. 3** Angiography of the PICA through the lateral medullary segment (lateral view) (**A**). Angiography of the PICA through the telovelotonsillar segment (lateral view) (**B**). An SL-10 was guided into the parent blood vessel of the cerebral AN, and 0.01 cc of 13% NBCA was administered (**C**). There was no migration of NBCA to a distal area or reflux (**D**). AN: aneurysm; PICA: posterior inferior cerebellar artery
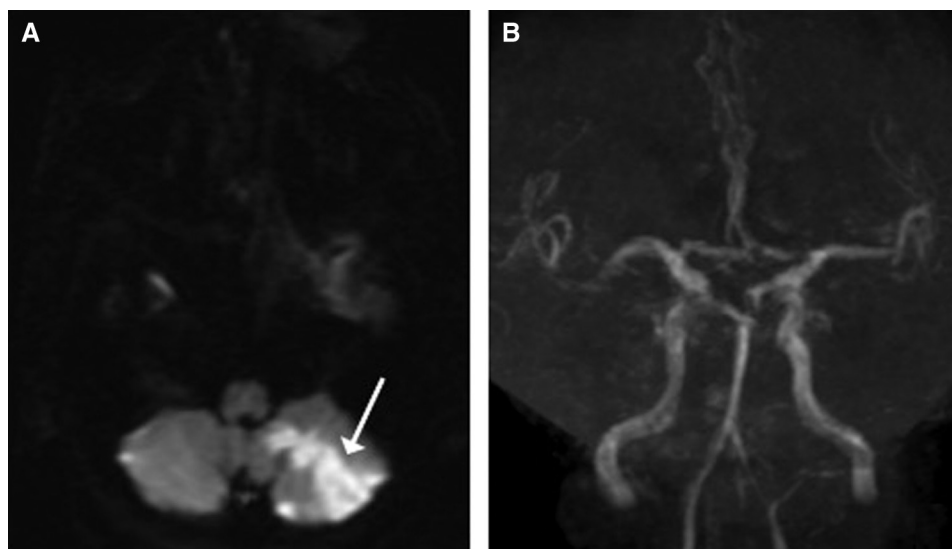
Fig. 4 Cross-section on diffusion-weighted head MRI the day after surgery. In the CS of the left PICA, acute-phase cerebral infarction (white arrow) was observed (**A**). Frontal view on head TOF-MRA MIP the day after surgery. Visualization of the left PICA trunk was maintained (**B**). CS: cortical segment; PICA: posterior inferior cerebellar artery; TOF-MRA MIP: time-of-flight magnetic resonance angiography maximum intensity projection

we attempted to guide an SL-10 (Stryker, Kalamazoo, MI, USA) into the parent blood vessel of the AN, the CS of the left PICA (**Fig. 3A** and **3B**). The ledge of the SL-10 was caught in the bifurcation of the parent blood vessel, making guidance into the blood vessel to be treated impossible. However, a target blood vessel was able to be selected by guiding the Roadmaster and Cerulean as highly as possible, improving their supportability. After administering 0.01 cc of 13% NBCA, the AN and parent blood vessel were embolized (**Fig. 3C**). There was no migration of NBCA to a distal area or reflux (**Fig. 3D**). Head magnetic resonance imaging (MRI) the day after surgery demonstrated fresh cerebral infarction involving a portion of the left cerebellar hemisphere (**Fig. 4**). There was no cerebral vasospasm. On Day 64, the patient was referred to a recovery-phase hospital with a modified Rankin Scale (mRS) score of 3.

Discussion

PICA ANs account for 0.49%–3% of all cerebral ANs, being relatively rare.[1] Approximately two-thirds of PICA ANs comprise VA-PICA ANs. Distal PICA ANs, which are peripheral to VA-PICA ANs, account for 0.28%–0.7% of all cerebral ANs, being markedly rare.[2] Many distal PICA ANs develop in the telovelotonsillar segment, whereas a small number of such ANs develop in the CS.[2] Distal PICA ANs are small and the rate of saccular-type ANs is high.

That of fusiform-type ANs is low, but most lesions are dissecting ANs.[3] Yamamoto et al.[4] reported that the aneurysmal type was evaluated as fusiform in one of six patients with distal PICA ANs. Ishikawa et al.[5] noted such findings in 1 of 12 patients with distal PICA ANs. Another study suggested that this characterizes distal PICA ANs.[6] Fusiform ANs are associated with arteriosclerotic lesions,[7] and rarely develop in the presence of congenital factors such as internal elastic lamina disorder and media defect.[8] Nishino et al.[6] reported a patient with a distal PICA AN in whom there was no arteriosclerotic change of a parent artery during surgery, and hypoplasia of the media and internal elastic lamina was histologically observed, with no history of head trauma, infectious disease, or heart disease, and suggested the involvement of congenital-factor-related arterial wall fragility in the pathogenesis.

It was previously reported that distal PICA ANs are associated with variations in the process of development. As an etiological factor, hemodynamic stress was proposed. As relevant vascular anomalies, arteriovenous malformation (AVM),[9] primitive trigeminal artery,[10] primitive hypoglossal artery,[11] direct aortic origin of the VA,[11] AICA-PICA anastomotic channels,[12] hypoplasty of the PICA,[13] megadolicho- basilar artery,[14] telangiectasia,[15] and different branches feeding the bilateral hemispheres[16] have been reported. A VA originating from the aorta is the most frequent variant of the VA (morbidity rate: 2.4%–5.8%; 2.4%–2.5% of patients who underwent cerebral angiography,

5.25% of patients with a tentative diagnosis of extracranial cerebrovascular disease).[17] The VA normally consists of longitudinal anastomosis connecting seven intersegmental arteries. These intersegmental arteries disappear, excluding the 7th intersegmental artery, which grows to the SA. However, if anastomosis between the 6th and 7th intersegmental arteries does not develop on the left side, the 6th intersegmental artery may remain, and the left VA may originate from the aortic arch between the common carotid artery (CCA) and Lt. SA. On the dorsal side of the cervix, the left VA may be formed as longitudinal anastomosis between the C1 and C7 cervical vertebrae and postcostal longitudinal anastomosis in the cervical intercostal obliteration zone. If postcostal longitudinal anastomosis remains following the development of the cervical vertebrae, the left VA may appear in an area distal to the Lt. SA; the 8th or 9th intersegmental artery may have remained.[18] Among left VAs of aortic origin, those originating from an area between the CCA and Lt. SA account for the highest percentage (81.9%), and those originating from an area distal to the Lt. SA account for 3.9%. In the present case, the left VA was not visualized on left subclavian arteriography; therefore, a VA originating from the aorta was assumed, but it may have been necessary to consider the possibility that this blood vessel originated from an area other than between the CCA and Lt. SA. Komiyama et al.[19] reported that the incidence of left VA dissection in patients with a left VA of aortic origin was 19.0%, and that it was significantly higher than that of dissection of a VA originating from the SA. To our knowledge, only Gács et al.[20] have reported a case of distal PICA AN with a VA originating from the aorta (an AN in the posterior medullary segment of the PICA). There was no description of the concrete origin of the left VA in their patient, but our CSAN patient with a left VA of aortic origin may be markedly rare, considering that the frequency of a left VA originating from an area distal to the Lt. SA is markedly lower than that originating from an area between the CCA and Lt. SA.

For our patient, endovascular treatment was selected and pathological examination was not conducted. However, two fusiform ANs connected with the same blood vessel were present, and a VA originating from the aorta was observed, suggesting a dissecting lesion related to structural hemodynamic stress. However, if hemodynamic stress alone had been etiologically involved, upstream VA dissection may have been induced rather than dissection of the distal PICA. Nishino et al. reported dissecting ANs related to hypoplasia of the media/internal elastic lamina, that is, congenital arterial wall fragility.[8] Hiscott et al.[13]

noted that the incidence of PICA anomalies was high. In the present case, the fusiform AN of the distal PICA may have been associated with arterial wall fragility in addition to hemodynamic stress.

Treatment methods include direct surgery (trapping) and endovascular treatment. As advantages of the former, treatment can be accurately performed under direct vision, the procedure is minimally invasive due to superficial lesions despite craniotomy, and OA-PICA bypass can be additionally conducted in some cases. The disadvantages of the former include invasiveness and intraoperative-prone position. As advantages of the latter, it is less invasive than craniotomy and treatment can be promptly performed following cerebral angiography. As its disadvantages, it is sometimes difficult to guide a microcatheter into branches, and when selecting a liquid embolic material, such as NBCA, it may migrate to the periphery or other blood vessels. At our hospital, there are several specialists in endovascular treatment and a system to promptly perform endovascular treatment was present; therefore, endovascular treatment as a minimally invasive treatment method was initially selected. As it was difficult to select a blood vessel to be treated by a microcatheter, endovascular treatment should be promptly switched to direct surgery when treatment is evaluated as difficult. The microcatheter was able to be guided and treatment was performed as scheduled. The peripheral fusiform AN was considered to be a dissecting AN and NBCA was used as an embolic material to occlude the parent blood vessel. Occlusion of the parent blood vessel led to postoperative cerebral infarction, but a peripheral blood vessel was affected and there was no marked cerebral swelling, resulting in a favorable course. To our knowledge, this is the first report of endovascular treatment for CSANs. Endovascular treatment using NBCA for CSAN patients, such as the present patient, is not covered by health insurance, but it may be useful as a treatment method.

Conclusion

We treated a patient with subarachnoid hemorrhage related to CSAN rupture with a VA of aortic origin, originating from an area distal to the Lt. SA, and performed endovascular treatment, leading to a favorable course. In the present case, dissection of the distal PICA was suspected. This may have been associated with hemodynamic stress related to the VA of aortic origin and congenital wall-structure fragility. Endovascular treatment using NBCA for CSANs may be a useful option.

Disclosure Statement

We declare no conflict of interest.

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