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Case Report

Ruptured bilateral middle cerebral artery aneurysms diagnosed based on cerebral vasospasm-associated ischemic symptoms: A case report ☆

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ABSTRACT

We report a case of subarachnoid hemorrhage presenting with ischemic symptoms due to cerebral vasospasm. A 64-year-old woman with right facial paralysis was referred to our hospital for treatment because of bilateral middle cerebral artery aneurysms observed using magnetic resonance imaging. She had no headache episodes; however, contrast-enhanced magnetic resonance imaging showed contrast enhancement of the aneurysmal wall only on the left side. Therefore, she was considered to have a ruptured aneurysm and underwent craniotomy and aneurysmal neck clipping. The postoperative course was uneventful; however, she developed aphasia and dysphagia 9 months after the surgery and was readmitted. New cerebral infarction and subarachnoid hemorrhage were observed on the right side, and the patient exhibited marked vasospasm. Because of a headache episode one week earlier, coil embolization was performed after the vasospasm. She was discharged home with a modified Rankin scale score of 2 and planned rehabilitation. Aneurysms that enlarge and rupture in a short time period should be treated with caution. Vessel wall imaging was useful in identifying the ruptured aneurysm in the current case.

Abbreviations: VWI, vessel wall imaging; MRA, magnetic resonance angiography.

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Introduction

Many cases of subarachnoid hemorrhage are diagnosed in the subacute phase. In one reported case, neurological symptoms due to vasospasm-induced cerebral ischemia led to the diagnosis of subarachnoid hemorrhage [1]. In cases with multiple aneurysms, it is difficult to identify the ruptured aneurysm from various perspectives. The main factors are extent of the hematoma; size, shape, and location of the aneurysm; and presence and symptoms of vasospasm [2]. However, vessel wall imaging (VWI) has been occasionally reported to be useful in the objective evaluation of the aneurysmal wall based on its contrast effect [3,4]. In the present report, we describe a rare case of ruptured bilateral middle cerebral artery aneurysms. The patient was diagnosed with subarachnoid hemorrhage and ruptured aneurysm based on vasospasm-induced cerebral ischemic symptoms, and VWI was useful in the diagnosis of the ruptured aneurysm and the administration of appropriate treatment.

Case report

A 64-year-old woman with no medical or life history or family medical history.

The patient presented with right facial paralysis and dysarthria during treatment for herpes zoster at a local dermatologist's office. Cranial magnetic resonance imaging (MRI) showed evidence of cerebral infarction, subarachnoid hemorrhage, and bilateral middle cerebral artery aneurysms, and the patient was referred to our hospital for further examination and treatment.

At initial examination, the patient was fully conscious and exhibited mild dysarthria, mild drooping in the right angle of the mouth, and mild right upper limb paralysis. There were no episodes of headache or loss of consciousness.

MRI showed subacute cerebral infarction in the left frontal lobe (Figs. 1A and B) and hemosiderin deposition around the left Sylvian fissure (Fig. 1C). Magnetic resonance angiography (MRA) showed a 2-mm aneurysm in the right middle cerebral artery and a 4-mm aneurysm in the left middle cerebral artery (Figs. 1D and E). Contrast-enhanced MRI VWI showed contrast enhancement on the wall of the left, but not right, middle cerebral artery aneurysm (Figs. 1F and G), suggesting the rupture of the left middle cerebral artery aneurysm.

Aneurysmal neck clipping was performed using frontotemporal craniotomy under general anesthesia. After the incision of the dura mater, the cerebral surface appeared xanthochromic and the arachnoid membrane was cloudy (Figs. 2A and B), indicating old hemorrhage. An aneurysm was found buried in the temporal lobe (Fig. 2C), and neck clipping was performed using a 5-mm straight clip (Fig. 2D). The pathological findings of the excised aneurysmal wall (Fig. 2E) were consistent with a ruptured aneurysm, which exhibited degeneration in the arterial wall with thrombus formation and failed arterial elastic plate (Fig. 2F).

The neurological symptoms of the patient, who was discharged home with a modified Rankin scale score of 1, remained unchanged, and she was able to maintain activities of daily living. During outpatient consultation, repeat MRI confirmed that there was no change in the findings of old infarction or aneurysm.

However, 9 months after the surgery, the patient was readmitted to the hospital by emergency medical services because of inability to move. On admission, her Glasgow coma scale score was 11 (E4V1[A]M6) and she had aphasia and dysphagia, although there was no gross new paralysis in limbs. She had experienced episodes of headache and vomiting one week prior to admission.

Computed tomography and MRI showed subarachnoid hemorrhage in the right Sylvian fissure (Figs. 3A and B), and diffusion-weighted imaging showed new infarction in the right frontal lobe, which was symmetrical to the old, left-sided cerebral infarction (Fig. 2C). MRA showed an enlarged right middle cerebral artery aneurysm with vasoconstriction and pseudoaneurysm of the right middle cerebral artery (Fig. 2D). The onset was clear; therefore, the patient was treated with coil embolization after the cessation of cerebral vasospasm.

An 8-Fr long sheath was inserted via the right femoral artery, an 8-Fr Optimo EPD balloon catheter (Tokai Medical, Aichi, Japan) was guided through a 4-Fr catheter, and a 6-Fr Cerulean DD6 (Medikit, Tokyo, Japan) was placed as an intermediate catheter into the right internal carotid artery. Right internal carotid angiography identified the aneurysm (Figs. 4A and B). A 45-degree Excelsior SL-10 1.7/2.4-Fr microcatheter (Stryker, Kalamazoo, MI) was guided into the aneurysm, and coil embolization was performed.

A Galaxy G3 microcoil (3 mm × 8 cm; Cerenovus, Johnson & Johnson, New Brunswick, NJ) was framed, with only one loop placed in the pseudoaneurysm (Fig. 4C). The following 4 additional coils were used for embolization (Fig. 4D) and neck remnant (Fig. 4E): Galaxy G3 Xsoft (2.5 mm × 3.5 cm; Cerenovus), Axium Prime 3D (2 mm × 2 cm; Medtronic, Minneapolis, MN), Axium Prime 3D (2 mm × 2 cm; Medtronic), and Axium Prime 3D (1.5 mm × 2 cm; Medtronic).

The patient was awake and well after surgery (Glasgow coma scale score was 15), and rehabilitation for dysphagia and aphasia was started. Dysphagia improved, and she was able to tolerate oral intake; however, aphasia persisted and she was transferred to a rehabilitation center on postintervention day 34 (modified Rankin scale score of 3). On postintervention day 53, she was discharged home because she was able to perform activities of daily living without any issues, although she still had minor aphasia.

The patient's outpatient course was good, with clear consciousness and mild dysarthria and dysarthria. She had no issues in walking and was independent in her daily activities. MRI performed at 3 months after coil embolization showed only the old, known cerebral infarction (Figs. 5A-C), and cerebral angiography at 7 months after surgery showed good embolization of the aneurysm (Fig. 5D).

Consent for submission of this article was obtained from the patient's family.

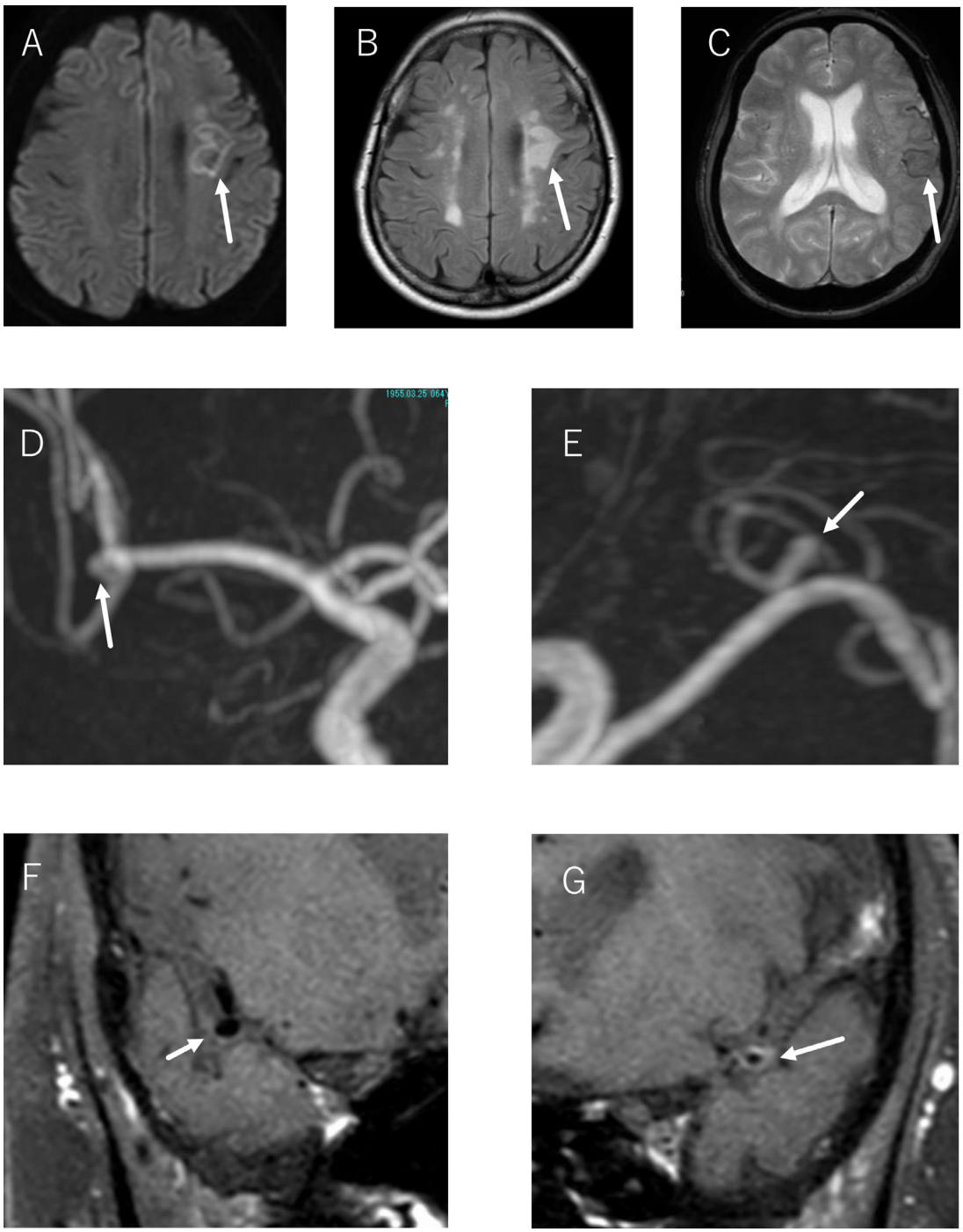

Fig. 1 – Imaging findings at initial examination. (A) Diffusion-weighted and (B) fluid-attenuated inversion-recovery images show an old cerebral infarction in the left frontal lobe (arrow). (C) T2*-weighted image shows an old subarachnoid hemorrhage (arrow). (D, E) Magnetic resonance angiography shows a 2-mm aneurysm in the right middle cerebral artery and a 4-mm aneurysm in the left middle cerebral artery (arrow). (F, G) Vessel wall imaging with contrast-enhanced magnetic resonance images show contrast effect only in the left aneurysm wall (arrow).

Discussion

In the present case, the initial subarachnoid hemorrhage revealed a cerebral aneurysm with cerebral infarction; however, the diagnosis was difficult because the vasospasm had already improved and there were no headache episodes. VWI was useful in the diagnosis of the ruptured aneurysm and the administration of appropriate treatment.

Subarachnoid hemorrhage without headache has been reported in 2%-8% of patients with subarachnoid hemorrhage [5]. Patients may not recognize the headache due to the on-

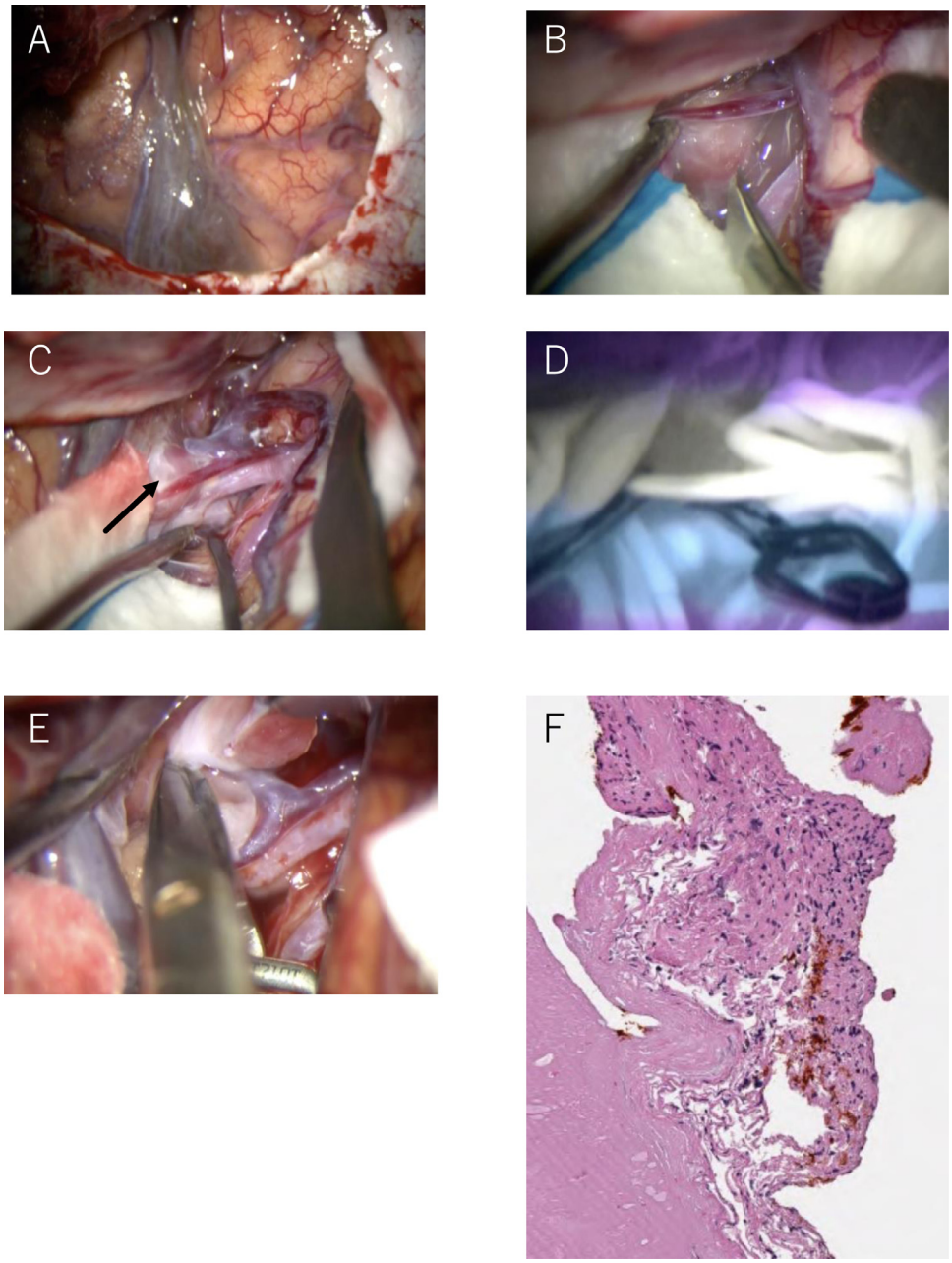

Fig. 2 – Aneurysmal neck clipping. (A) The brain surface appears xanthochromic, and (B) the arachnoid membrane is cloudy. (C) An aneurysm embedded in the left temporal lobe is visible, and (D) indocyanine green fluorescence imaging confirms the blockade of blood flow after clipping. (E) The aneurysmal wall specimen collected for pathological examination, and (F) disruption of the arterial elastic plate is observed (hematoxylin/eosin staining, 100×).

set of aphasia or attention disorder, which is common in left-sided aneurysms [6]. In the present case, the patient did not recall the presence of a headache at initial presentation for the rupture of the left middle cerebral artery aneurysm but recognized an earlier headache episode prior to the rupture of the right middle cerebral artery aneurysm.

Table 1 summarizes the previously reported cases in which subarachnoid hemorrhage was diagnosed based on cerebral infarction symptoms associated with cerebral vasospasm [1,6]. The patients presented with various neurological disorders, such as hemiparesis, dysarthria, aphasia, and confusion, because of cerebral vasospasm, and cerebral infarction was observed in all cases. Additionally, as mentioned above, the patient with the left-sided aneurysm was unaware of previous headaches, and these were present only with the right-sided aneurysm. Hemorrhage was observed in imaging studies in 4 of the 5 cases, but the hemorrhage was minimal in all 5 cases. Of these 5 patients, 3 patients had middle cerebral artery, and 2 patients had posterior communicating artery aneurysms confined to the ipsilateral subarachnoid space. The hematoma

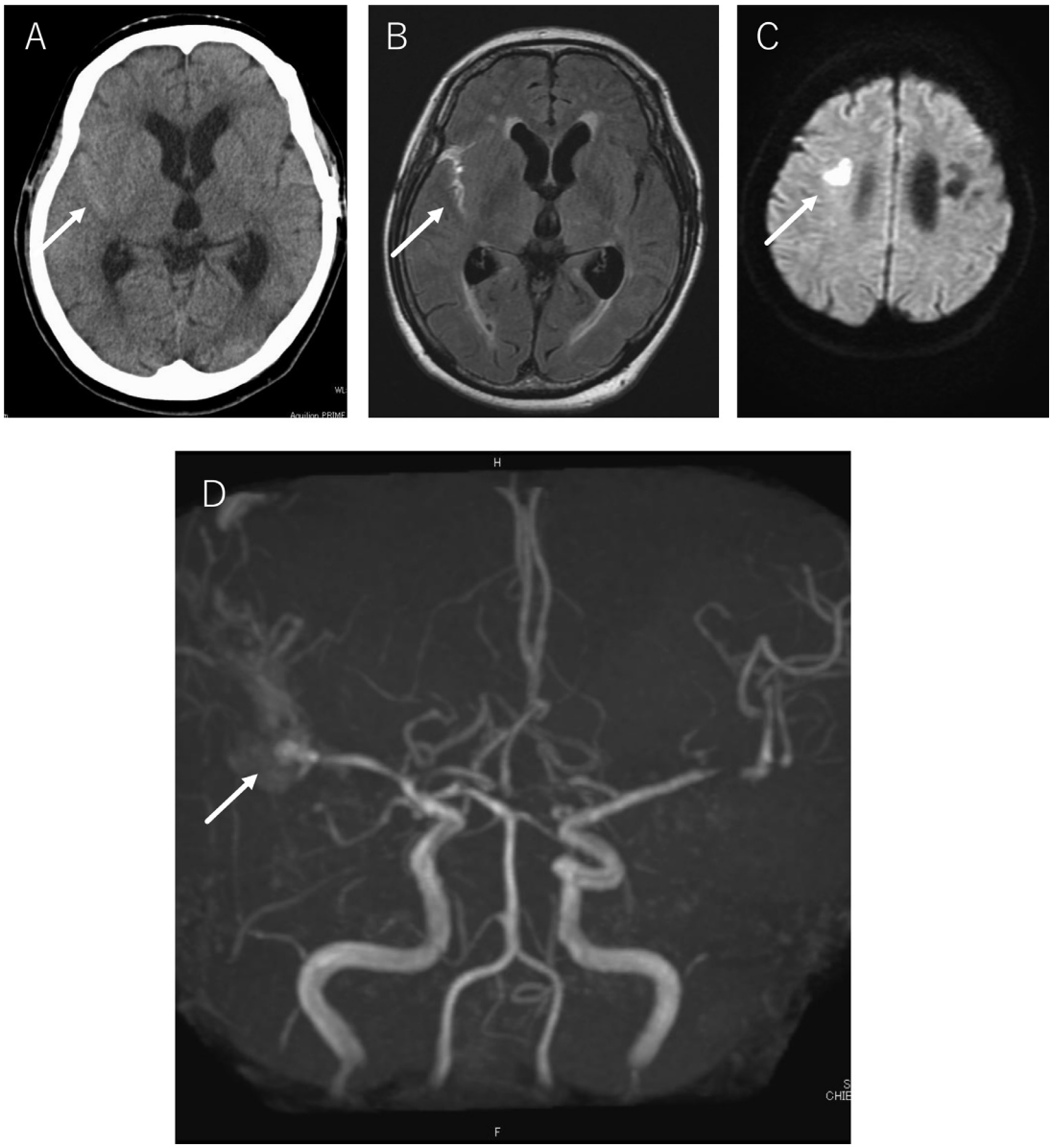

Fig. 3 – Imaging findings on emergency admission. (A) Cranial computed tomography and (B) fluid-attenuated inversion-recovery images show subarachnoid hemorrhage in the right Sylvian fissure (arrow). (C) Diffusion-weighted image shows a new cerebral infarction (arrow). (D) Magnetic resonance angiography shows an enlarged right middle cerebral artery aneurysm (arrow) and poor visualization beyond the M2 segment of the middle cerebral artery due to vasospasm.

was localized to the area where the blood vessels ran; therefore, the vasospasm might have been strong despite the small hematoma volume. The diagnosis of ruptured aneurysm was difficult in all cases, and rebleeding led to the diagnosis of subarachnoid hemorrhage only one case reported by Nussbaum et al. [1]. In patients with subarachnoid hemorrhage diagnosed based on cerebral vasospasm in the subacute stage, it is important to identify the relationship between the ruptured aneurysm and ischemic symptoms.

In the present case, the ruptured cerebral aneurysms were identified using VWI with contrast-enhanced MRI because bilateral middle cerebral artery aneurysms were found in the initial diagnosis of subarachnoid hemorrhage. The number of studies utilizing VWI of intracranial vessels has increased exponentially since the first report describing VWI in 1994. About half of these studies are related to atherosclerotic lesions, but the number of studies on aneurysms is also increasing [7]. In particular, in patients with multiple cerebral aneurysms, VWI with contrast-enhanced MRI is useful in identifying the specific aneurysm that has ruptured based on the contrast enhancement of the aneurysm wall [4,8]. Nagahata et al. [8] reported that VWI showed strong contrast enhancement in 73.8% and 4.8% of the ruptured and unruptured aneurysms, respectively, and that contrast enhancement was

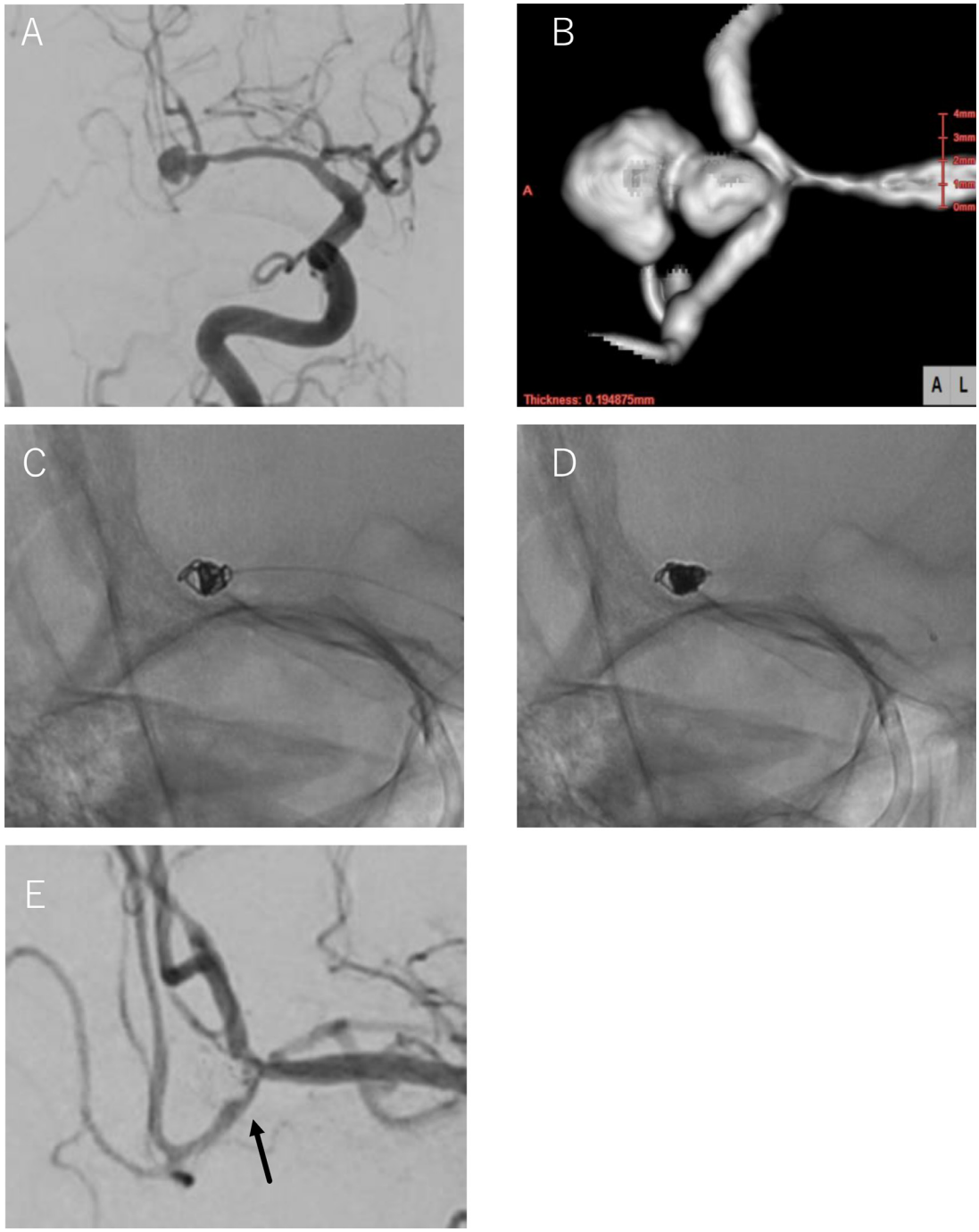

Fig. 4 – Endovascular treatment. Frontal view during preoperative right internal carotid angiography (A) and (B) 3D rotation angiography show an enlarged right middle cerebral artery aneurysm with pseudoaneurysm. (C) Framing and (D) final coil mass are shown. Immediately after treatment, right internal carotid angiography (E) no longer shows the aneurysm in frontal view (arrow).

not observed in 1.6% and 81.9% of the ruptured and unruptured aneurysms, respectively.

The increased use of VWI with contrast-enhanced MRI has revealed that various aneurysm-associated factors correlate with the contrast effect. A high PHASES score, which is used to predict aneurysm rupture, has been reported to be correlated with contrast enhancement in VWI [9,10], and studies have reported that larger aneurysms exhibit stronger contrast enhancement [11]. Additionally, aneurysms can have irregular and asymmetric shapes and the correlation between contrast enhancement and findings of high rupture risk, such as irregular shape and a high aspect ratio, has also been reported [12]. In the present case, despite the passage of time, the aneurysm wall in the left middle cerebral artery was

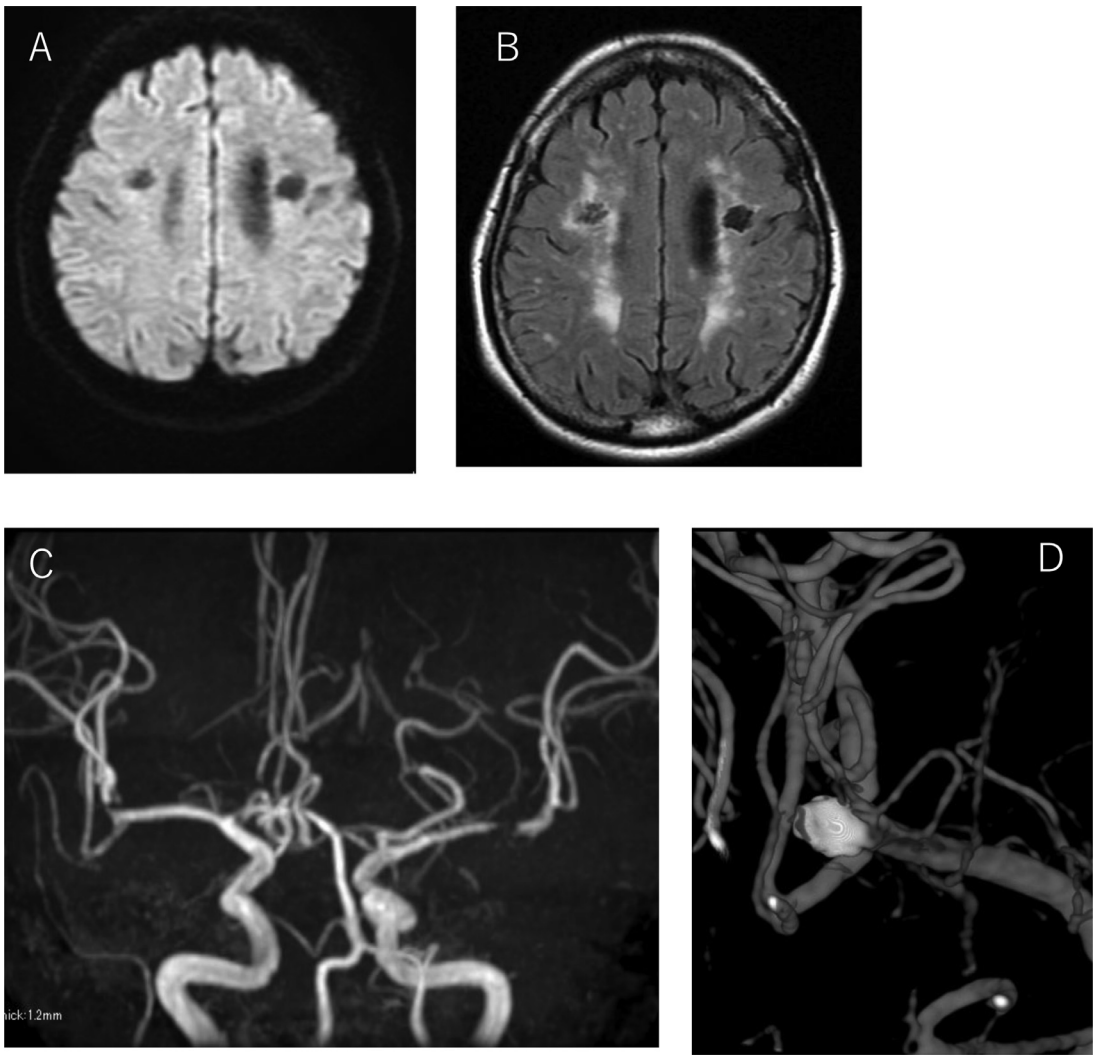

Fig. 5 – Postdischarge imaging findings. (A) Diffusion-weighted and (B) fluid-attenuated inversion-recovery images show known old cerebral infarcts (arrow) in bilateral frontal lobes, and (C) magnetic resonance angiography shows no recurrent aneurysm. (D) 3D rotation angiography of the right internal carotid angiography shows good embolization.

Table 1 – Summary of the cases with subarachnoid hemorrhage diagnosed based on cerebral infarction symptoms associated with vasospasm.

	Age, sex	Symptom	Previous headache	R/L	Diagnostic imaging		
					Infarction	Hemorrhage	Aneurysm
Nussbaum et al.	37, F	Hemiparesis, aphasia	+	R	+	– rebleeding[a]	P-com
	59, F	Hemiparesis, dysarthria	+	R	+	+[b]	MCA
Kim et al.	44, F	Confusion, disorientation	–	L	+	+	P-com
Present case	64, F	Hemiparesis, facial paralysis	–	L	+	+	MCA
		Dysarthria, aphasia	+	R	+	+	MCA

F, female; R, right; L, left; P-com, posterior communicating artery; MCA, middle cerebral artery.

[a] Unable to identify a link between aneurysm and ischemic symptoms, diagnosed after rebleeding during treatment of stroke

[b] Reviewed and identified bleeding.

contrast-enhanced, leading to the diagnosis of ruptured cerebral aneurysm, which successfully treated. On the other hand, the contralateral small aneurysm in the right middle cerebral artery, which did not show contrast enhancement, ruptured 9 months later, suggesting that its shape changed during this period. Horiguchi et al. [13] reported that the time from the confirmation of enlargement to the rupture of an unruptured aneurysm was short, ranging from a few days to several months. In the present case, it is possible that the aneurysm in the right middle cerebral artery enlarged and changed shape during the 3 months after the last examination. As indicated by the PHASES score [14], unruptured aneurysms associated with ruptured aneurysms require careful follow-up. One study previously reported a patient who experienced rebleeding and had a poor outcome, indicating that such cases, in which the bilateral aneurysms in which only one is ruptured, should be treated with caution.

In conclusion, we here presented the case of a patient with ruptured cerebral aneurysm diagnosed based on ischemic symptoms due to cerebral vasospasm following subarachnoid hemorrhage. The diagnosis was difficult because the patient did not have headache and the vasospasm had improved prior to admission; however, VWI with contrast-enhanced MRI was useful for the diagnosis of the ruptured aneurysm.

Patient consent

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

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