DOI: 10.5797/jnet.cr.2019-0126

Case Report

A Case of Internal Carotid Artery Dissection with Ischemic Onset, Followed by Subarachnoid Hemorrhage during Diagnostic Angiography

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Objective: Internal carotid artery (ICA) dissection is known to cause binary types of stroke, cerebral infarction, and subarachnoid hemorrhage (SAH). However, it is rare that these two pathologies take place in a clinical scenario. We report a case of ICA dissection with ischemic onset, which was followed by SAH on the same day during diagnostic angiography.

Case Presentation: A 60-year-old woman with chronic hypertension rapidly developed right hemiplegia. She had been suffering from slight headache and abnormal sensation in the right limbs 1 week before the ictus. MRI demonstrated small acute infarctions in the left middle cerebral artery (MCA) territory. The left ICA was not visualized on MRA. Diffusion–perfusion mismatch was indicated by the automated image postprocessing system. Endovascular recanalization was planned to prevent the progression of cerebral infarction. After advancing a 5MAX ACE, initial left ICA angiography was performed, resulting in extravasation of contrast medium from the C2 segment of the left ICA. 3D rotational angiography revealed left ICA dissection of the C2 segment. To secure hemostasis, the patient underwent internal trapping at the C1 and C2 segments of the left ICA. Collateral flow to the left MCA via an anterior communicating artery was observed. On day 28, the patient was transferred to a rehabilitation hospital with right hemiplegia and motor aphasia.

Conclusion: In cases of tandem lesions with preceding neurological symptoms, ICA dissection should be considered as one of the causes. Careful injection of contrast medium may be necessary if ICA dissection is strongly suspected.

Keywords ▶ internal carotid artery dissection, cerebral infarction, subarachnoid hemorrhage, endovascular treatment, parent artery occlusion

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Received: December 28, 2019; Accepted: May 15, 2020
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Introduction

In patients with nontraumatic intracranial artery dissection, preoperative diagnosis of dissection is often difficult when the affected blood vessel is occluded.[1)] Here we report a case of acute cerebral infarction due to occlusion at C2 portion in the internal carotid artery (ICA) followed by subarachnoid hemorrhage (SAH) at the time of diagnostic cerebral angiography, suggesting the dissection that was hardly detectable before angiography.

Case Presentation

Case: A 60-year-old woman
Medical history: Hypertension, uterine myoma, and migraine

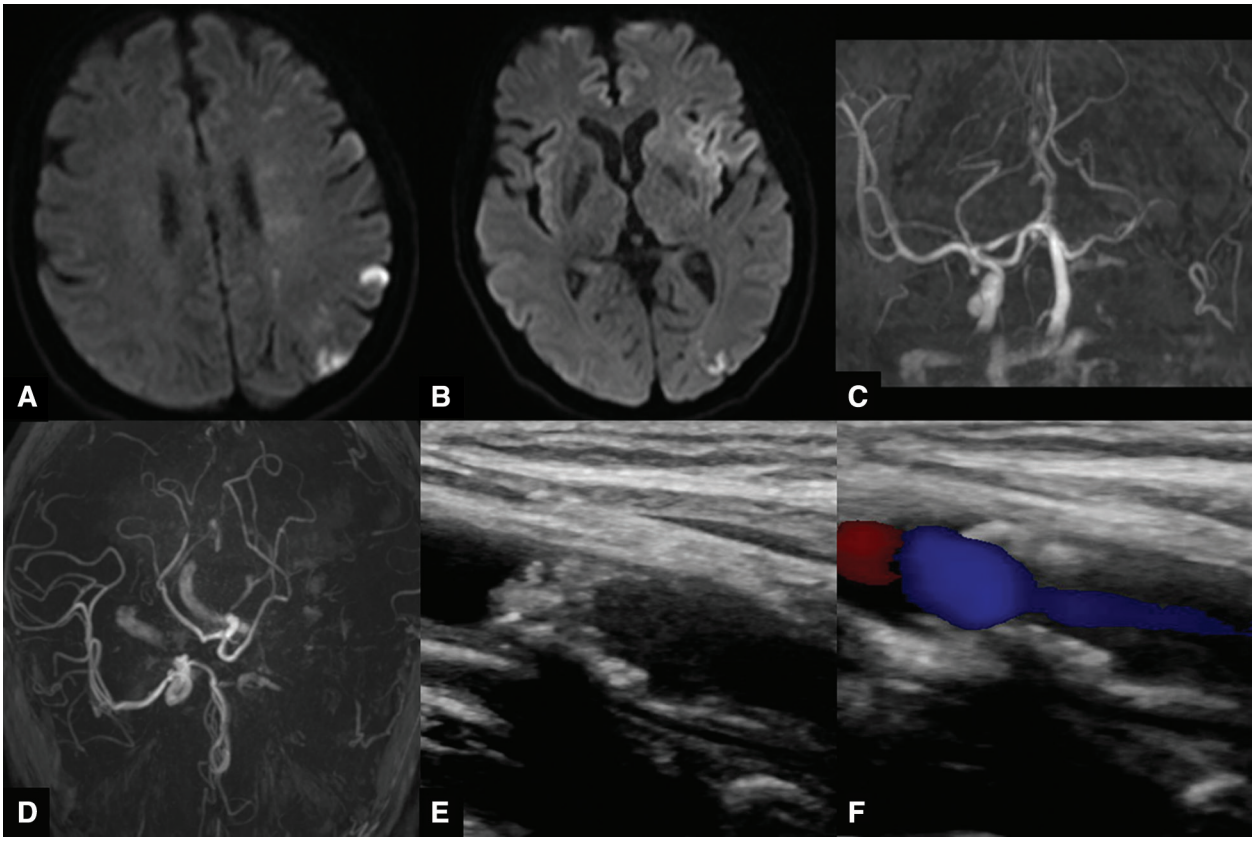

Fig. 1 MRI on admission. (**A** and **B**) DWI showed small acute infarctions in the left MCA territory. (**C** and **D**) The left ICA was not visualized on MRA, and the distal M1 segment of the left MCA was obstructed. (**E** and **F**) Carotid ultrasonography image of the left ICA showed stenosis with high echoic plaques. The peak systolic velocity was 2.3 m/sec. There was no evidence of dissection in the cervical carotid artery. DWI: diffusion-weighted image; ICA: internal carotid artery; MCA: middle cerebral artery

Present illness: She had been suffering from right-sided numbness and headache for 1 week before the ictus. On the day of admission, she developed right hemiplegia and dysarthria in the morning, and was brought to our hospital by ambulance.

Physical examination on admission: The blood pressure and pulse rates were 153/91 mmHg and 65/min (sinus rhythm), respectively. Concerning the neurological status, the Japan Coma Scale score was I-3 and the National Institutes of Health Stroke Scale score was 20/42 (right unilateral spatial neglect, dysarthria, and right incomplete hemiplegia).

Radiological findings: MRI revealed high-signal-intensity area scattered in the left middle cerebral artery (MCA) territory on diffusion-weighted image (DWI) (**Fig. 1A** and **1B**). The DWI-Alberta Stroke Program Early CT Score was 6. On MRA , the left ICA involving its origin was not visualized, but the left anterior cerebral artery (ACA) to the M1 segment of the MCA was visualized from the contralateral side through the anterior communicating artery (AComA) **(Fig. 1C** and **1D**). Furthermore, a small aneurysm was detected at the right ICA–anterior choroidal artery (AChA) bifurcation. According to the automated perfusion analysis software (RAPID [version 4.9.2.2; iSchemaView, Menlo Park, CA, USA]), the ischemic core volume (apparent diffusion coefficient $<620 \times 10^{-6}$ mm^2/sec) was 13 mL and the volume at a T_{max} of >6.0 sec, which represents a 5MAX ACE (Penumbra, Alameda, CA, USA), was 180 mL. The mismatch ratio was 13.1 (**Fig. 2A** and **2B**). Cervical vascular echography demonstrated a stenotic lesion at the origin of the left ICA, with a peak systolic velocity of 2.3 m/sec (**Fig. 1E** and **1F**).

Clinical course: A diagnosis of M1 occlusion of the left MCA related to artery-to-artery embolism involving the stenotic site of the left ICA as the source of embolism was made. Since the onset time was unclear, intravenous thrombolysis with tissue plasminogen activator was not performed. Cerebral angiography and revascularization were planned. The right femoral artery was punctured, and 5000 international units of heparin were systemically administered. Before the stenting procedure, 200 mg of aspirin and 300 mg of clopidogrel were administered as loading of antiplatelets. A 9-Fr Optimo (Tokai Medical Products, Aichi,

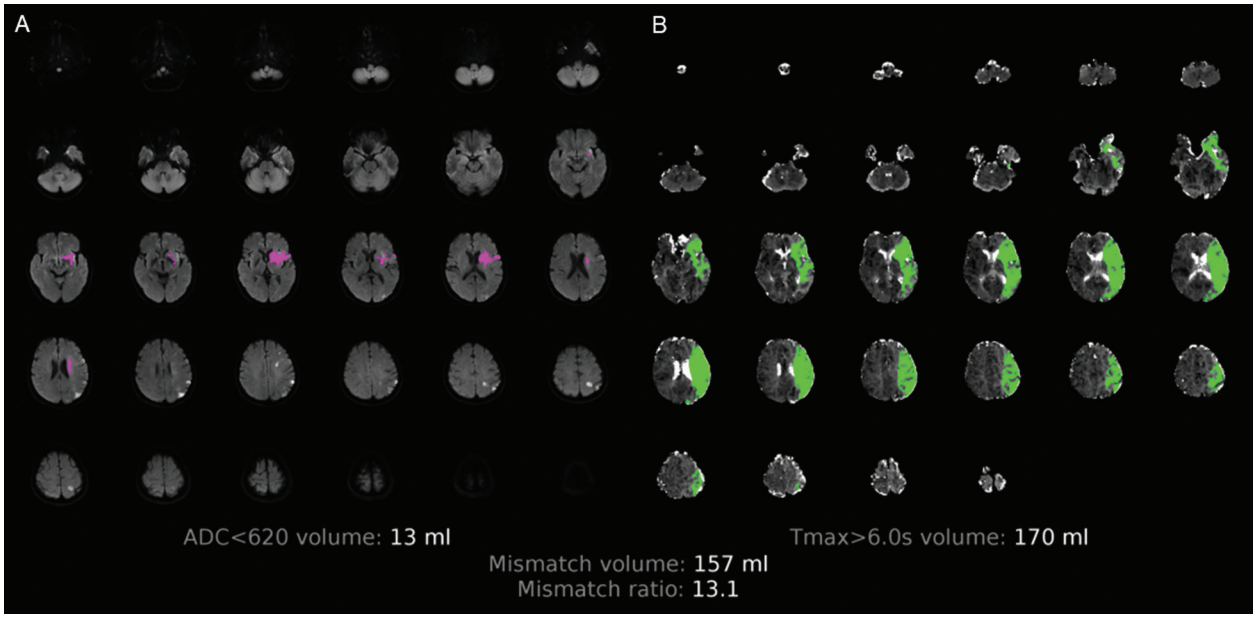

Fig. 2 The automated image postprocessing system (RAPID) on admission. (**A**) DWIs overlaid with the stroke core identified using the apparent diffusion coefficient threshold. The estimated stroke core was 13 mL. (**B**) T_{max} maps with green overlay for regions with abnormal flow (T_{max} >6 sec). The estimated volume of delayed flow was 170 mL. The diffusion–perfusion mismatch ratio was 13.1 . DWI: diffusion-weighted image

Japan) was guided into the left common carotid artery (CCA). Although the left common carotid angiography exhibited marked stenosis at the origin of the left ICA (**Fig. 3A**), a 5MAX ACE catheter was passed through the stenotic site and advanced to the petrous ICA to perform thrombectomy at the occluded ipsilateral MCA. When the contrast was injected through 5MAX ACE, leakage of contrast medium from the C2 segment of the left ICA was observed (**Fig. 3B** and **3C**). Subsequent cone beam CT revealed extensive SAH (**Fig. 3D**). No device, such as a microguidewire, had passed through the C2 lesion site before the extravasation. The left CCA was immediately occluded by inflating the balloon with Optimo, and heparin was reversed with 50 mg of protamine sulfate before the patient was sedated and intubated while lowering her systemic blood pressure.

As a right ICA aneurysm was present, the presence of an ipsilateral ICA aneurysm and its rupture was suspected. However, diagnostic 3D rotational angiography revealed fusiform dilation involving the C2 to C1 segments of the left ICA, leading to a diagnosis of ICA dissection-related cerebral infarction and SAH. To achieve hemostasis and prevent additional hemorrhage, internal trapping (IT) was planned, although occlusion of the AChA was considered unavoidable. A Traxcess (Terumo, Tokyo, Japan) microwire was passed through the C2 ICA lesion site to the M2 segment of the left MCA, followed by an SL-10 (Stryker, Kalamazoo, MI, USA) advancing into the M1 segment of the left MCA. As the dissection was not considered involving left M1, the C1 to C2 segment of the left ICA was embolized using 15 coils in order to preserve the collateral flow via AComA to left MCA (**Fig. 4A** and **4B**). Left internal carotid angiography after IT confirmed complete occlusion including the site of dissection (**Fig. 4C** and **4D**). On right internal carotid angiography, the superior branch of the left MCA was visualized through the AComA, but occlusion of the left A2 segment and inferior branch of the left MCA related to distal embolism of a thrombus at the site of dissection was noted (**Fig. 4E** and **4F**). CT following endovascular treatment revealed acute hydrocephalus. To treat hydrocephalus and control the intracranial pressure after SAH, ventriculostomy was performed.

Postoperative course: DWI the day after procedure demonstrated extensive high-signal-intensity areas in the left ACA and MCA territories (**Fig. 5A** and **5B**). On cerebral angiography 2 weeks after the procedure, the left ICA was not visualized while the superior branch of the left MCA was visualized through the A1 segment of the left ACA and M1 segment of the left MCA via the AComA (**Fig. 5C** and **5D**). An osmotic diuretic was used to minimize brain edema, and intravenous drip of Fasudil hydrochloride to prevent SAH-related cerebral vasospasm was performed during the postprocedural period. However, antithrombotic therapy was not conducted considering the risk of hemorrhagic

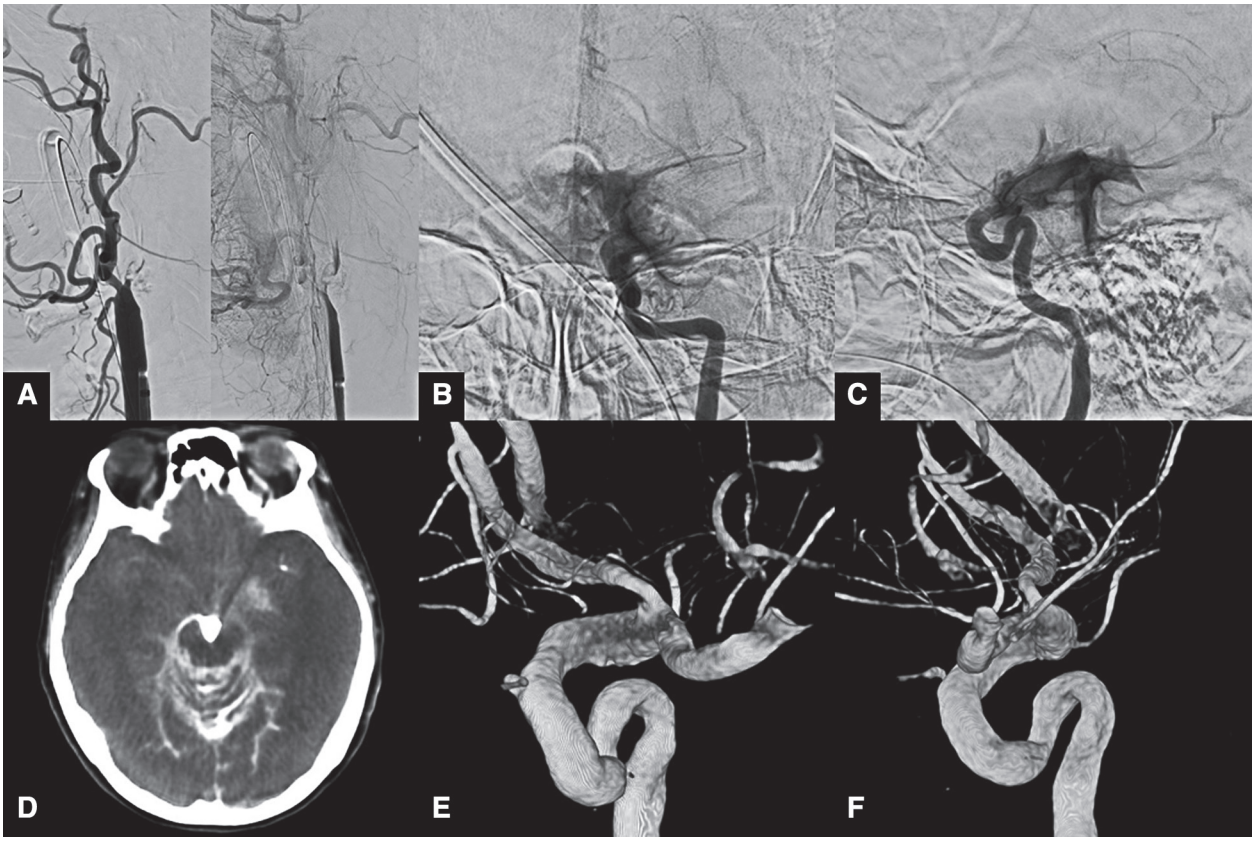

Fig. 3 Pre- and intraoperative angiography. (**A**) Preoperative left carotid angiography showed severe stenosis of the ICA at the level of its bifurcation. (**B** and **C**) Initial left ICA angiography after advancing the 5MAX ACE showed active bleeding into the subarachnoid space from the C2 segment of the left ICA. (**D**) Cone beam CT during the procedure revealed leakage of contrast medium and diffuse SAH. (**E** and **F**) 3D angiography revealed irregular dilatation at the C1 and C2 segments of the left ICA, suggesting dissection. ICA: internal carotid artery; SAH: subarachnoid hemorrhage

infarction. Brain edema was settled down 7 days after the procedure, and extubation was performed. The external ventricular drain was removed in 2 weeks. On discharge, right hemiplegia and motor aphasia remained. The patient was referred to another hospital for rehabilitation with a modified Rankin Scale score of 4.

Discussion

In Japan, nontraumatic intracranial artery dissection accounts for 0.7% of all patients with stroke. Male preponderance was noted with the incidence of 76.1%.[2)] The age of ischemic onset peaks at 40 to 49 years, while hemorrhagic onset peaks at 50 to 59 years.[2)] The frequent site of dissection is the vertebrobasilar arterial system. ICA dissection is relatively rare in Japan,[1,2)] differing from that in Europe and the United States. According to Yamaura et al.,[1)] the vertebrobasilar artery dissection was observed in 299 (93%) of 322 patients with nontraumatic intracranial artery dissection and ICA dissection was observed in only 10 (3%) patients. Of these patients, 76.1% had ischemic onset and 28.1% had hemorrhagic onset.[1)] Another study reported that hemorrhage-onset type accounted for a high percentage of patients with vertebrobasilar artery dissection, whereas ischemia-onset type accounted for a high percentage of patients with ICA dissection.[2)] However, a recent study reported that many patients with dissection in the vertebrobasilar system had presented with ischemia.[3)] There were considerable cases with vertebrobasilar dissection whose onset was ischemia followed by SAH during the clinical course,[4–7)] not usual with dissection in the anterior circulation with few exceptions involving C2 segment and ACA.[8–11)] Previous studies regarding cerebral infarction complicated by SAH found that some patients died and that serious sequelae remained in others, demonstrating a poor prognosis.[4–11)] According to a national survey conducted by Yamaura et al.,[1)] the incidence of SAH after the onset of ischemia was 3.4% (4/118), but the mortality rate was 75% (3/4). If a diagnosis of dissection is made in the initial phase, conservative treatment may lead to a

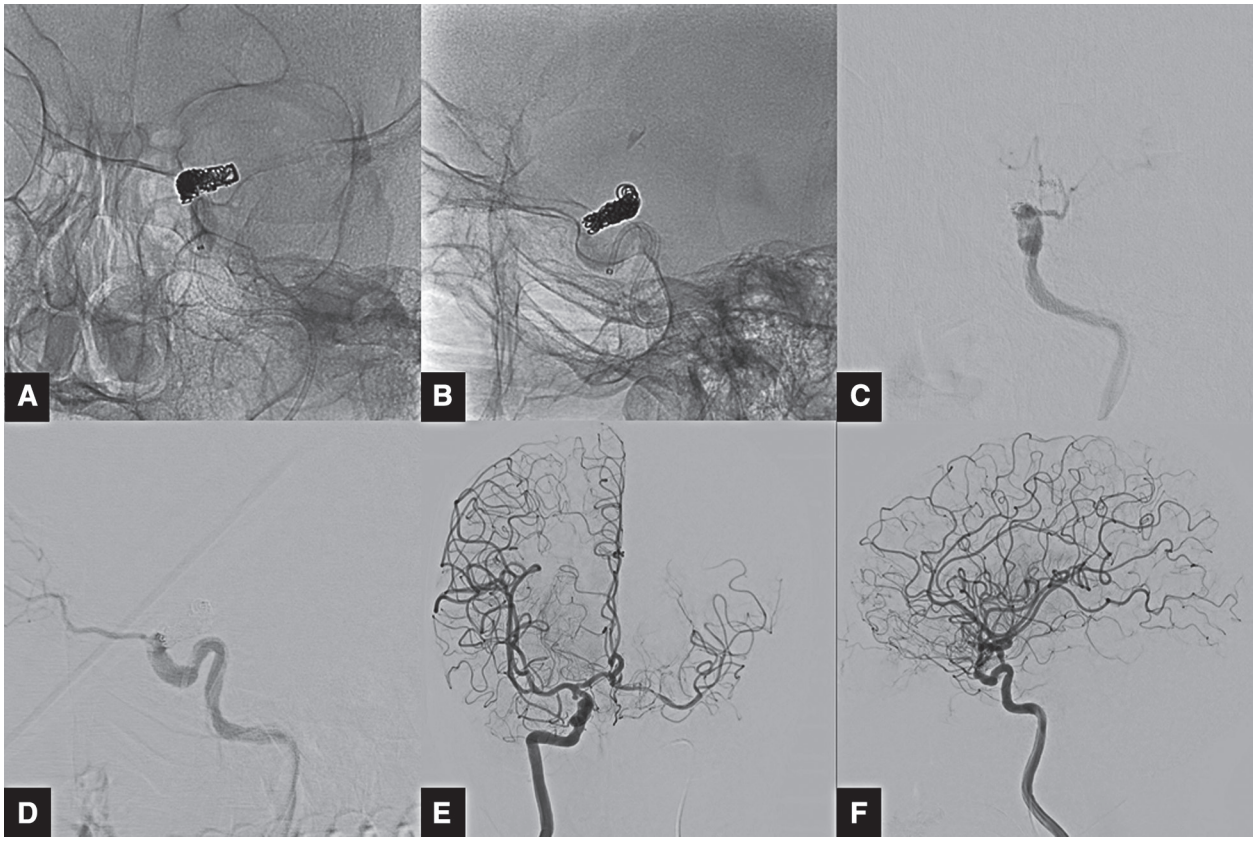

Fig. 4 Postoperative angiography. (**A**–**D**) Left ICA angiography demonstrated obliteration of the C2 segment of the left ICA. (**E** and **F**) Collateral flow to the left MCA via an AComA was observed. The A2 segment of the left ACA and inferior trunk of the left MCA were obstructed due to thrombus migration. AComA: anterior communicating artery; ICA: internal carotid artery; MCA: middle cerebral artery

relatively favorable prognosis in patients with ischemic onset, whereas surgical treatment may result in a poor prognosis in many patients with hemorrhagic onset.[11]

To our knowledge, this is the first case with dissection at the C2 segment in which the ischemic onset due to occlusion of the dissection was followed by SAH at the time of angiography.

Clinical symptoms of ICA dissection include 1) headache, unilateral facial/cervical pain (66%); 2) ipsilateral Horner's sign (<50%); and 3) cerebral ischemia after a few hours to days (50%–95%). If two of these are present, the possibility of this disease is high.[12] In the present case, unilateral headache and cerebral ischemia developed 1 week after right-sided numbness. Arrhythmia, such as atrial fibrillation, was not observed, and the patient's medical history was not consistent with cerebral embolism. Although dissection should be considered as one of differential diagnoses considering her present illness, the history of migraine kept us away from this pathology.

Concerning diagnostic imaging findings, a diagnosis of dissection could be made if either the "pearl and string" or the "string" sign is observed on cerebral angiography, or an intimal flap or double lumen on cerebral angiography or MRI/MRA.[13] When dissection is suspected, dilation of the outer diameter of a blood vessel on time-of-flight (TOF) imaging, 3D-GdT1, or T1-SPACE is also useful for its diagnosis.[14,15] In the present case, it was challenging to suspect dissection based on the symptoms, and only DWI, FLAIR, T2 star, and MRA were performed. Furthermore, due to arterial occlusion, a flow void had disappeared on MRI; thus, it was impossible to evaluate an intimal flap or double lumen on TOF imaging; it was quite challenging to make a diagnosis of dissection before cerebral angiography.

Regarding the pathogenesis of dissection, hemodynamic loading, such as hypertension, may make the internal elastic membrane fragile, forming an entry point through rapid rupture.[16] If dissection between the internal elastic membrane and media progresses, hematoma-related narrowing of the lumen may lead to stenosis or occlusion (subintimal type). If dissection involves the outer membrane, dilation/rupture may occur, inducing SAH (subadventitial type).[17]

In the present case, extensive hypoperfusion related to lumen occlusion at the site of dissection in addition to cerebral infarction related to artery-to-artery embolism in the

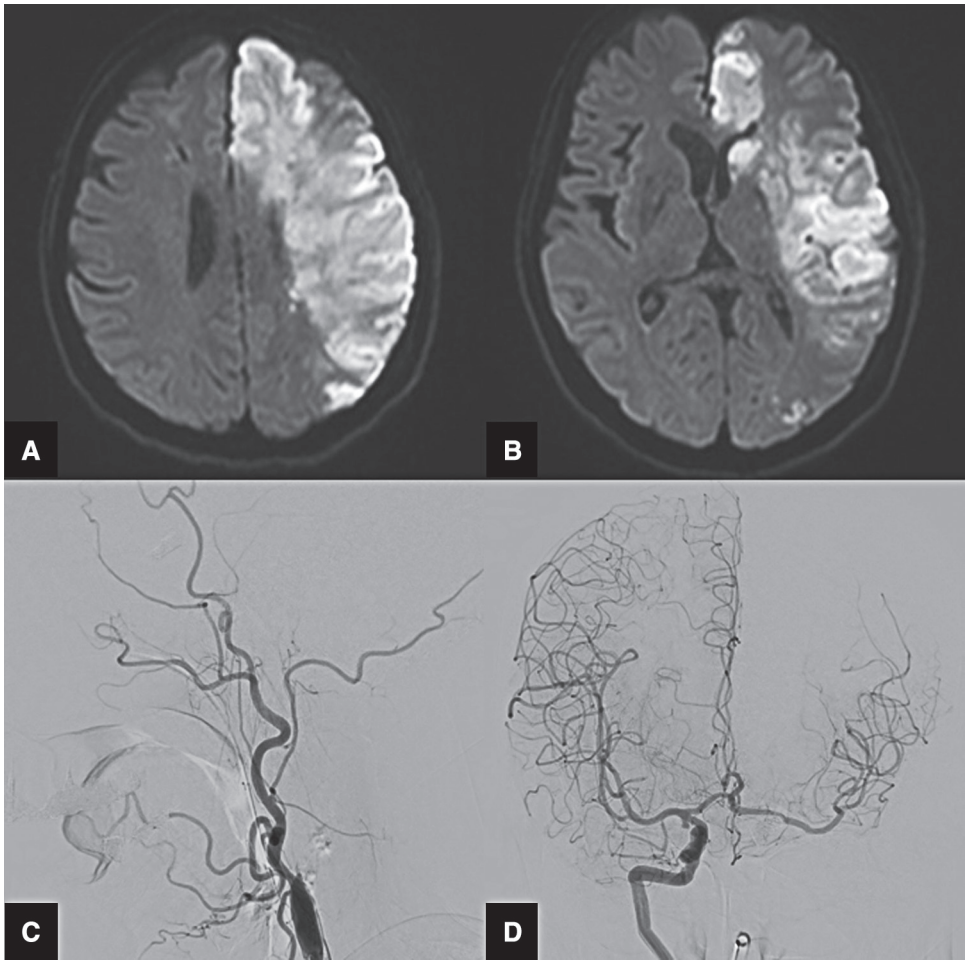

Fig. 5 Follow-up MRI and angiography. (**A** and **B**) Postoperative day 1 DWI showed extensive hemispheric cerebral infarction of the left MCA and ACA territory. (**C**) Postoperative day 14 left carotid angiography revealed obliteration of the ICA. (**D**) Collateral flow to the left MCA via the AComA was still observed. The A2 segment of the left ACA and inferior trunk of the left MCA were obstructed as on postoperative DSA . ACA: anterior cerebral artery; AComA: anterior communicating artery; DWI: diffusion-weighted image; ICA: internal carotid artery; MCA: middle cerebral artery

M1 segment of the left MCA from the site of dissection was noted. SAH developed, and thereafter, a 5MAX ACE was advanced to the left ICA through stenotic lesion, which was present at its origin; we were unable to exclude the possibility that the infusion of contrast medium into the closed or semi-closed space between the stenotic and dissected sites related to wedging of the 5MAX ACE at the ICA origin stenosis increased the intravascular pressure, causing vessel rupture. In another scenario, dissection may have been continued from the left ICA origin to carotid fork. However, cervical echography demonstrated typical plaques of the stenotic lesion at the ICA origin. There was no intimal flap suggestive of dissection or low-echoic lesion suggestive of a thrombus in the false lumen. Furthermore, on left internal carotid angiography at the onset of SAH, no morphological changes suggestive of dissection in the C3 to C5 segments were observed, thus eliminating the assumptions noted earlier. The hypothesis that the pressurized infusion of contrast medium into the closed or semi-closed space between the stenotic lesion at the ICA origin and M1 occlusion of the MCA may have induced rupture of the C2 segment where dissection did not take place. However, in this case, an outlet of contrast medium may have remained in the A1 segment of the left ACA. To our knowledge, there were no reports demonstrating rupture of a normal blood vessel induced by the pressure infusion of contrast medium alone. Therefore, there was little chance of this second scenario.

Previous studies reported hemorrhage during diagnostic cerebral angiography in patients with ruptured aneurysms[18,19]; however, it is unclear whether rupture was related to the procedure or an incidental event. Saitoh et al.[18] found no relationship between rebleeding from ruptured

aneurysms and the volume of contrast medium. Komiyama et al.[19] also reported a similar finding. On the contrary, there is another report depicting an increase in the intravascular pressure around the site of infusion during contrast-medium infusion,[20] suggesting its relationship with rupture during examination. Rebleeding during cerebral angiography within 6 hours after onset was frequently observed in patients with SAH, suggesting that cerebral angiography should be avoided during this period.[19] However, with recent development in the image resolution of digital subtraction angiography, the infusion volume/rate of contrast medium may be lower than previously adopted; the incidence of hemorrhage during angiography may have decreased. There is no similar report on ICA dissection, but slow injection should be particularly carefully performed during cerebral angiography in patients with preceding headache or neurological findings among those with tandem lesions, considering the possibility of dissection. In particular, in patients with extracranial internal carotid artery stenosis (ICS) as in this case, careful attention must be paid for contrast infusion considering the possibility that catheter insertion may create a closed space between the ICS and dissected site.

For surgical treatment in patients with hemorrhage, it is necessary to achieve hemostasis and occlude a ruptured parent artery to prevent rebleeding. Trapping by direct surgery, IT by endovascular treatment, or these procedures in combination with bypass may be effective.[15] Angioplastic clipping or wrapping is inferior to trapping or IT to avoid rebleeding as it is difficult to close an entry point.[8] As to the merit of IT, it can be performed in succession to diagnostic cerebral angiography. In the present case, IT was conducted as prompt as possible. A previous study suggested that bypass should be combined to compensate satisfactory anterograde blood flow, which decreases by IT.[8] However, in the present case, collateral flow via AComA to the left MCA was observed and a thrombus was present in the M2 segment of the left MCA. In addition, the interval from onset was prolonged; therefore, bypass surgery was not performed.

Conclusion

A case with dissection at the C2 segment of ICA initially presented with cerebral infarction due to its occlusion, which resulted in its rupture at the time of angiography, is presented.

In tandem lesions with a preceding headache or neurological symptoms, the possibility of dissection must be considered. Especially in patients with ICS, the careful and slow injection should be conducted to prevent pressurized infusion when performing cerebral angiography.

Disclosure Statement

Dr. Satow reports grants from CANON Medical Systems Corporation, outside of the submitted work. Dr. Inoue reports lecturer's fees from Daiichi Sankyo, Bayer, Bristol-Myers Squibb, and Medtronic outside of the submitted work. The other authors declare no conflicts of interest.

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