Discrepancy between Preoperative Imaging and Postoperative Pathological Finding of Ruptured Intracranial Dissecting Aneurysm, and Its Surgical Treatment: Case Report

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Abstract

The choice of therapeutic strategy for intracranial dissecting aneurysm is often based on radiographic features, including characteristic geometry (e.g., irregular stenosis, segmental stenosis, aneurysm formation [pearl-and-string sign]), irregular fusiform or aneurysmal dilation, double lumen, and tapering occlusion. However, there is often a discrepancy between preoperative radiographic data and actual dissecting length. The present report describes three cases in which there was a discrepancy between preoperative radiographic data and actual dissecting length in patients undergoing direct trapping with or without revascularization. All three cases experienced good outcomes, but these cases underscore the fact that open surgery is a good option for management of ruptured intracranial dissecting aneurysms for determination of the rupture point, dissecting length, and the relationship between dissecting area and small arteries arising from the associated vessel.

Key words: dissecting aneurysm, preoperative and postoperative findings, trapping, vascular reconstruction, pathology

Introduction

One characteristic feature of intracranial dissecting aneurysms is the communication between the true lumen and the pseudolumen through a disrupted portion of the internal elastic lamina. In some cases, this disruption can advance to the adventitia, resulting in rupture and subarachnoid hemorrhage (SAH), or can be contained within the media, resulting in ischemia or stenosis of the artery.

Surgical treatment for ruptured dissecting aneurysm includes proximal clipping,1-4 or trapping,2-6 and clipping5 with or without revascularization.41 Alternatively, various endovascular strategies can be used, including proximal parent vessel occlusion, internal coil trapping, stent-assisted coil embolization, stent-only therapy, covered stent placement, or any combination thereof.14-15 While recent advances in endovascular approaches have resulted in good outcomes for patients with ruptured dissecting aneurysms,10-12,15 some reports have described recurrence of aneurysmal dilatation or rebleeding after endovascular trapping.10,14,16-21 One important contributor to recurrence and/or rebleeding is that preoperative imaging cannot determine the exact range of the dissecting aneurysm or the location of the entry point in the hyperacute phase.

Computed tomography angiography (CTA) and digital subtraction angiography (DSA) have the advantage of being able to assess the intravascular space. Conversely, magnetic resonance imaging (MRI), such as basi-parallel anatomical scanning (BPAS), can characterize the outer appearance of the artery,22,23 while thin slice T1-weighted imaging and three-dimensional (3-D) spoiled gradient-recalled acquisition (SPGR) can assess the wall of the artery.24 However, it is still difficult to precisely characterize the dissecting area during the hyper-acute phase. In support of this notion, the present report describes three cases in which there was a discrepancy between preoperative radiographic data and postoperative pathological findings in patients undergoing direct trapping with or without revascularization.

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Case 1: Ruptured Right Vertebral Artery Dissecting Aneurysm

I. History
A 56-year-old man presented to an outpatient clinic with sudden onset of left-sided posterior cervical pain. An MRI was performed with many remarkable findings. The next morning, he lost consciousness at his home. Medics were called and arrived to find the patient in cardiopulmonary arrest. Cardiopulmonary resuscitation (CPR) was initiated, and he was transported to our hospital. During CPR, spontaneous breathing and circulation were restored. Upon examination at our hospital, the patient had a Glasgow Coma Score of three.

II. Examination
Computed tomography (CT) revealed subarachnoid hemorrhage with thick subarachnoid clot in the cerebellomedullary cistern (Fig. 1). CT angiography (CTA) revealed a pearl-and-string sign in the V4 portion of the right vertebral artery (Fig. 2, left) and absence of a posterior inferior cerebellar artery (PICA) arising from the left side of the vertebral artery.

III. Operation
Operative management with parent artery trapping was elected based on preservation of brainstem reflexes and the possibility that the patient could recover if our team could evacuate any clot that could have been compressing the medulla oblongata. A lateral suboccipital approach with transcondylar fossa drilling was performed with the patient in the park-bench position. Intraoperatively, no PICA was identified, but there was a penetrating branch arising just distal to the dissection (Fig. 3A). A distal clip was placed on the vertebral artery just proximal to the penetrating branch (Fig. 3B). The proximal end of the dissection was also obliterated by clipping while preserving perforators arising from intact segment of vertebral artery (Fig. 3C). Finally, the dissected artery was trapped and removed.

IV. Pathology
Pathologic examination of the resected specimen showed an entry-only dissecting aneurysm. The entry was contained within the normal segment of the proximal

Fig. 1 Computed tomography reveals subarachnoid hemorrhage with thick subarachnoid clot in the cerebellomedullary cistern. Back streaming of the hemorrhage in the lateral ventricle is also seen.

Fig. 2 Computed tomography angiography (CTA) revealing a pearl-and-string sign in the V4 portion of the right vertebral artery (left). Slices a, b, and c correspond to the pathological specimens (right). Photomicrograph showing serial axial slices of an entry-only dissecting aneurysm. Elastica van Gieson stain, original magnification x40. Panel a shows a slice taken from the narrowed lesion seen on CTA. The media is disrupted, and hematoma is seen just below the adventitia. The intravascular space is narrowed in this slice. Panel b shows a slice taken from the dilated lesion seen on CTA. The internal elastic lamina is disrupted, and the wall adjacent to the rupture site is only composed of fibrin and thin collagen, which forms a pseudoaneurysm. Panel c is a slice taken from the normal-appearing site seen on CTA. The internal elastic lamina is also disrupted in this specimen, while the intravascular space is relatively normal; this means that the angiographically “normal” lesion also includes the entry point.
vertebral artery seen on preoperative CTA (Fig. 2, right).

V. Postoperative course
The patient required tracheostomy in the acute stage of his recovery. He was discharged with mild dysphagia 1 year after the operation with a modified Rankin scale score (mRS) of two.

Case 2: Ruptured C2 Portion of an Internal Carotid Artery Dissecting Aneurysm

I. History
A 30-year-old man suffered sudden onset of severe headache and was transferred to an affiliated hospital.

II. Examination
A CT showed SAH (Fisher group three) (Fig. 4). CTA revealed only an elevated margin proximal to the origin of the posterior communicating artery (PCoA) (Fig. 5). This finding was thought to be consistent with internal carotid artery (ICA) dissecting aneurysm, but the hemorrhagic pattern of the CT strongly suggested bleeding from the right ICA.

III. Operation
Trapping of the right ICA with external carotid (EC)-radial artery (RA)-M2 bypass was elected. Large frontotemporal craniotomy was performed, harvesting the radial artery, and exposing the EC artery, ICA, and common carotid artery in the right neck. Intraoperatively, the rupture point was identified in the C2 portion (Fig. 6, left), and the dissecting aneurysm extended to the distal portion of C1 (Fig. 6, right). The lesion was trapped after confirming that the EC-RA-M2 bypass was functional. The anterior choroidal artery arose from the wall opposite of that of the dissection and was preserved by placing a distal clip in an oblique fashion. An adult-type PCoA, which arose from the middle of the dissecting area, was sacrificed. Finally, the dissected artery was removed.

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Fig. 6 Operative view of the right anterior temporal approach. Dissecting aneurysm of the internal carotid artery is found in C2 to C1. The rupture point is found in the C2 portion (left, arrow head). The dissecting aneurysm extends to the distal portion of the C1 (right, arrow).

Fig. 7 Photomicrograph showing the disrupted internal elastic lamina and hematoma below the adventitia. The intravascular space is maintained within the normal range. Elastica van Gieson stain, original magnification $\times40$.

Fig. 8 Postoperative digital subtraction angiography showing external carotid-radial artery-M2 bypass with good retrograde flow and trapped C2 to C1 portion of the internal carotid artery.

IV. Pathology
Pathologic examination showed a disrupted internal elastic lamina and hematoma below the adventitia. The true lumen was maintained with normal shape and size and without narrowing (Fig. 7).

V. Postoperative course
The patient experienced transient hemiparesis at 1 month after the onset of SAH, but ultimately recovered with no permanent neurologic deficits. Postoperative DSA showed good patency of the EC-RA-M2 bypass with good retrograde flow to the proximal portion of the middle cerebral artery and ICA and disappearance of the dissecting aneurysm of the C1–C2 segment of the ICA (Fig. 8). He was discharged with an mRS of zero.

Case 3: Ruptured Left Vertebral Artery Dissecting Aneurysm

I. History
A 54-year-old woman suffered sudden onset of severe occipital and posterior cervical pain and vomiting. She was transferred to an affiliated hospital.

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III. Operation

Aneurysm trapping with preservation of the occipital artery during craniotomy was planned. A lateral suboccipital transcondylar fossa approach was performed with the patient in the park-bench position.

Intraoperatively, the orifice of PICA was involved by the dissecting aneurysm (Fig. 11). Therefore, we performed occipital artery to PICA bypass prior to the trapping of the dissecting aneurysm, including the obliteration of the origin of the PICA. The dissected artery was removed along with the PICA bifurcation.

IV. Pathology

Pathologic examination showed an entry-only type dissecting aneurysm. The entry was in the angiographically dilated lesion (Fig. 12).

V. Postoperative course

She showed no neurological deficits after the operation and was discharged with an mRS of zero.

Discussion

The diagnosis of intracranial dissecting aneurysm is suggested by characteristic geometry (e.g., irregular stenosis, segmental stenosis, aneurysm formation [pearl-and-string sign]), irregular fusiform or aneurysmal dilation, double lumen, and tapering occlusion. In the present three cases,
irregular fusiform or aneurysmal dilatation (pearl sign) resulted from the presence of the pseudolumen below the adventitia, while segmental stenosis (string sign) resulted from narrowing of the artery lumen and media dissection. However, the findings do not indicate the precise length of the dissecting aneurysm or the entry point.

Mizutani et al. reported that the rupture site of dissecting aneurysms was located just above the orifice of the entrance in all eight human autopsy specimens. But, as demonstrated in the present report, the rupture site is not always just above the orifice of the entrance. For example, in Case 1, the orifice of the entry of dissecting aneurysm was contained within the angiographically normal, pathologically unruptured portion of aneurysm. The dissecting area also extended into the angiographically normal portion of the aneurysm in Case 2 and Case 3.

Many reports have suggested that complete occlusion of the entry point is important for radical treatment. Suboptimal evaluation of the dissecting range and entry point results in a risk of recurrence and/or rebleeding. Accordingly, surgeons must consider that the dissecting aneurysm may extend beyond the angiographic pearl-and-string and into the area characterized as “normal” on imaging. In addition, endovascular trapping carries a potential risk of branch occlusion, which can result in cerebellar or brainstem infarction, because of the difficulty in localizing the perforating branches.

For those reasons, our group utilizes microsurgical trapping, in which the dissecting area can be precisely characterized within a highly magnified operative field, rather than endovascular trapping. This strategy allows rigorous occlusion of the entry point while minimizing the need to sacrifice small branches associated with the dissecting aneurysm. Furthermore, in the case of vertebral artery dissecting aneurysm, the tightly packed clots around the medulla oblongata (as in Case 1) must be removed for decompression and to maintain brainstem function.

However, several reports have described the occurrence of contralateral dissecting aneurysm after ipsilateral vertebral artery occlusion in the context of endovascular trapping or in the context of microsurgical trapping. Investigators in those reports have suggested possible causes, such as hemodynamic stress for the contralateral vertebral artery, angiographically and microscopically normal dissecting aneurysm were still existed and incomplete trapping. Although we have never experienced such recurrence of a dissecting aneurysm, there is the possibility of microscopically normal dissecting aneurysm. Specimens in the present cases had severe media disruption with a dissecting area that could be precisely characterized within a highly magnified operative field, suggesting the possibility that mild media disruption (e.g., a pseudolumen just above the internal elastic lamina) may appear microscopically normal. Furthermore, a dissecting aneurysm in a severely atherosclerotic artery may also appear microscopically normal. Therefore, even if microsurgical trapping seemed to be completely performed, periodic postoperative assessment is still needed. Further, even if rigorous occlusion without sacrifice of the perforating artery (as in Case 1) is successfully performed, there is still a risk of obliteration of the perforating artery, mainly because the vertebral artery becomes a blind end. Fortunately, this did not occur in the present cases, but some investigators have described cases of delayed occlusion of the perforating artery after parent artery occlusion. However, recurrent cases of dissecting aneurysm after open surgery are infrequent, and microsurgical trapping with vascular reconstruction is still effective for obliteration of aneurysms and is associated with a reduced risk of rebleeding from the aneurysm. In addition, if the hemodynamic stress induces formation of a contralateral dissecting aneurysm, endovascular stenting may result in better outcomes if complete obliteration of the aneurysm is possible.
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Conclusion
Current diagnostic imaging cannot precisely characterize the entry point and dissecting length. Open surgery is required for precise characterization of these parameters and also to identify small perforating branches. Trapping with revascularization by open surgery is a good option for management of ruptured intracranial dissecting aneurysms.

Conflicts of Interest Disclosure
I have already submitted the self-reported potential COI disclosure statement of the Japan Neurosurgical Society. And as the corresponding author, I take all responsibility that all coauthors have disclosed all potential COIs concerning this manuscript, in accordance with the policy of the Japan Neurosurgical Society.

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