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Case Report

Contrast extravasation from basilar artery without aneurysm formation on digital subtraction angiography in computed tomography angiogram-negative subarachnoid hemorrhage: A case report

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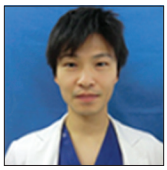

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ABSTRACT

Background: The causes of angiogram-negative subarachnoid hemorrhage (SAH) on initial angiography, which accounts for 10–30% of spontaneous SAH, are heterogeneous and still unclear. We report a case of nonaneurysmal SAH, in which initial computed tomographic angiography (CTA) showed no source of bleeding, but the subsequent digital subtraction angiography (DSA) revealed contrast extravasation from the basilar artery without aneurysms.

Case Description: A 67-year-old woman with a medical history of hypertension presented as SAH of World Federation of Neurological Surgeons Grade II. CTA on admission did not show any cause of bleeding and DSA was subsequently performed to show contrast extravasation from a perforator of the middle third of the basilar artery without aneurysms during the subsequent DSA, resulting in profound deterioration SAH and neurological status. The patient was conservatively treated. Follow-up DSAs on days 2 and 16 showed no source of bleeding as well.

Conclusion: Although the precise cause of bleeding in this case is uncertain, SAH might be caused by local dissection of the basilar artery perforator, and the bleeding site might heal spontaneously without forming of a pseudoaneurysm.

Keywords: Basilar artery perforator aneurysm, Contrast extravasation, Subarachnoid hemorrhage, Unknown etiology

INTRODUCTION

Approximately 10–30% of spontaneous subarachnoid hemorrhage (SAH) patients have unknown origin on initial angiographic imaging.[5,8,15] Because computed tomographic angiography (CTA) is less invasive and more rapid than digital subtraction angiography (DSA), CTA is frequently used to detect a cause of SAH.[3] DSA is commonly performed after CTA to confirm or rule out the source of bleeding.[18] The authors report a rare case of spontaneous SAH in which the cause of bleeding was not identified on initial CTA, but the subsequent DSA showed contract extravasation from a perforator of the middle third of the basilar artery without any aneurysm.

CASE PRESENTATION

A 67-year-old woman with a medical history of hypertension presented to our hospital after sudden onset of headache with vomiting. On admission, she was drowsy, but there were no neurological deficits. The blood pressure and pulse rate were 197/111 mmHg and 84 beats/min, respectively. She was clinically judged World Federation of Neurological Surgeons Grade II. CT of the head showed SAH of Fisher Group 3, especially in the prepontine cistern, and CTA showed no source of bleeding [Figure 1]. DSA also revealed no vascular lesion such as an aneurysm, but showed contrast extravasation from a perforator of the middle third of the basilar artery [Figure 2], although blood pressure was well controlled. The patient fell in a coma, and CT demonstrated an increase in the amount of SAH mainly in the posterior fossa [Figure 3]. Her brainstem reflexes and spontaneous breathing were preserved. Because there was neither treatable saccular nor dissecting aneurysms, the patient was treated conservatively under general anesthesia. On the following day (day 1), CT showed the development of acute hydrocephalus, which was treated by ventricular drainage. The second DSA performed on day 2 showed no source of SAH and the wall of the basilar artery was smooth [Figure 4a and b]. General anesthesia was discontinued on day 7, but the patient remained in a coma associated with cerebral infarction occurring in bilateral frontal lobes due to cerebral vasospasm. DSA performed on day 16 also showed no source of bleeding [Figure 4c and d]. Tracheostomy and ventriculoperitoneal shunting were performed on days 17 and 59, respectively. She then developed shunt dysfunction and underwent shunt reconstruction on day 112. There was no rebleeding during her stay in our hospital. The patient became an apallic state with quadriplegia and was transferred to a long-term hospital with modified Rankin scale of 5.

DISCUSSION

Spontaneous SAH is caused by rupture of a cerebral aneurysm in 80–90% of patients.[10] On the other hand, 10–30% of spontaneous SAH patients have negative findings on initial angiography.[5,8,15] Angiogram-negative SAH patients have a more favorable and a lower rebleeding rate than aneurysmal SAH patients.[5,19] Thus, some refers angiogram-negative

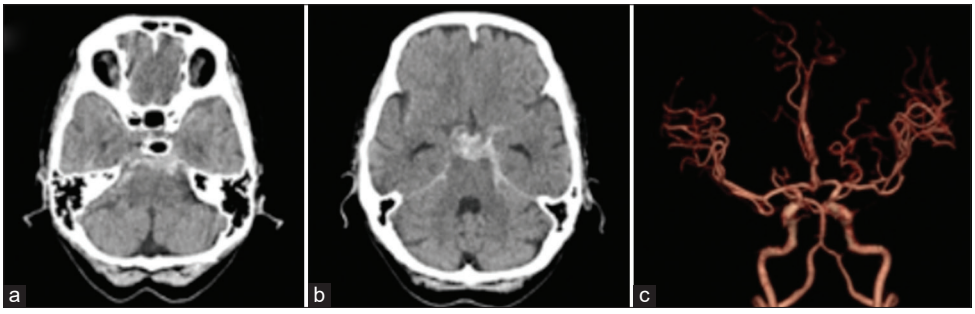

Figure 1: Computed tomography (CT) and CT angiography (CTA) on admission. (a and b) CT shows subarachnoid hemorrhage (SAH) predominantly in the posterior fossa. (c) CTA shows no cause of SAH.

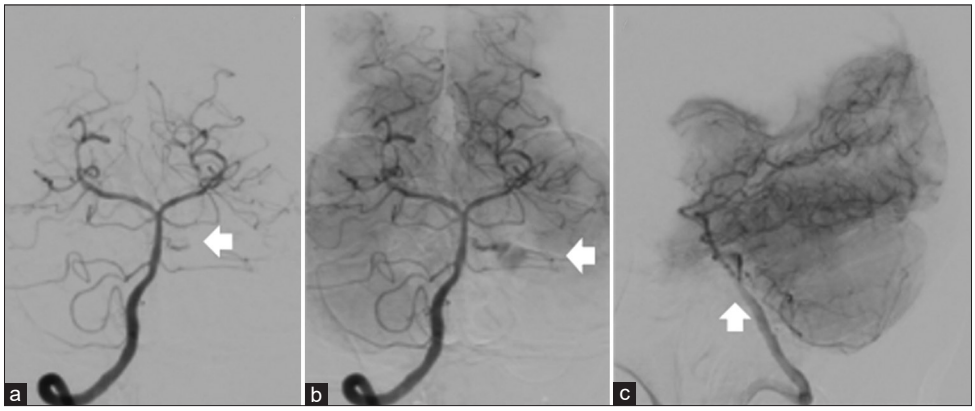

Figure 2: Digital subtraction angiography (DSA) on admission. DSA in an arterial phase (a, anteroposterior view) shows contrast extravasation (arrow) from a perforator of the middle third of the basilar artery, which is increased in a capillary phase (b, anteroposterior view; c, lateral view).

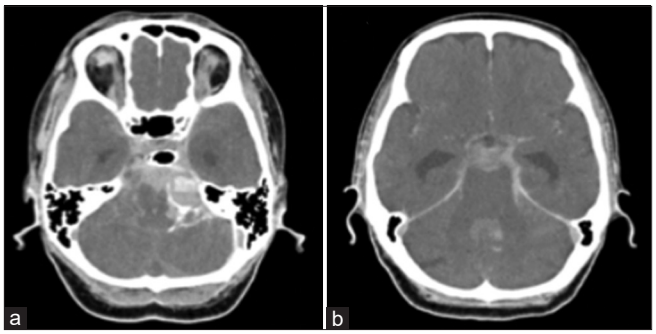

Figure 3: (a and b) Noncontrast computed tomography performed just after the initial digital subtraction angiography showing contrast extravasation with increasing amount of subarachnoid hemorrhage predominantly in the left cerebellopontine cistern.

SAH as benign SAH. However, angiogram-negative SAH is heterogeneous and can be divided into perimesencephalic SAH (PSAH) and non-PSAH.[14] PSAH is found in 50–75% of patients with angiogram-negative SAH and has a more favorable outcome than non-PSAH.[16] Mohan *et al*. reported that rebleeding, cerebral vasospasm, and acute hydrocephalus occur more frequently in a case of non-PSAH than PSAH.[13]

Possible sources of angiogram-negative SAH include vascular malformations, tumors, vasculitides, infections, venous thromboses, and nonvisualized aneurysms.[9] However, accurate causes of most of angiogram-negative SAH remain unclear despite improvements in diagnostic imaging techniques. In this case, CT on admission showed non-PSAH, and the initial CTA did not detect any bleeding source. However, DSA showed contrast extravasation from a perforator of the middle third basilar artery without the visualization of saccular and dissecting aneurysms. If we had not encountered contrast extravasation timely on DSA, the patient would be judged angiogram-negative SAH.

DSA is the gold standard in terms of diagnosing the source of bleeding. However, CTA is more rapid and less invasive than DSA.[3] Goddard *et al.* reported the sensitivity of CTA for the detection of intracranial aneurysms between 83.6% and 92.7%, and the specificity between 77.2% and 98.9%.[7] Therefore, CTA is the first choice for investigating a cause of SAH in most hospitals. The rate at which DSA detects the cause of bleeding in cases of CTA-negative SAH is 4–8%.[1,20] Although there are few patients with SAH who have negative findings on CTA and positive findings on DSA, it is difficult to distinguish small perforating arteries of less than 1 mm in diameter or to detect very small aneurysms on CTA.[3] Therefore, even if CTA findings are negative for the cause of SAH, DSA is necessary to investigate the cause of bleeding.

Because there was no treatable aneurysm and it is difficult to explore the middle third basilar artery in a direct surgery, we performed conservative therapy in this case. DSA on days 2 and 16 showed neither contrast extravasation nor

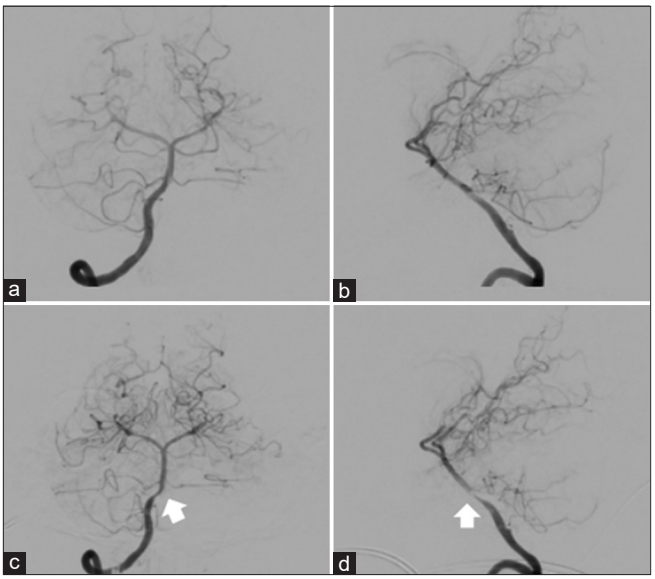

Figure 4: Right vertebral artery angiography on days 2 (a, anteroposterior view; b, lateral view) and 16 (c, anteroposterior view; d, lateral view). (a and b) No source of hemorrhage is shown, and the wall of the basilar artery is smooth. (c and d) No cause of hemorrhage is revealed, but vasospasm of the basilar artery (arrow) is shown.

the source of bleeding, and no rebleeding occurred during the hospitalization. To the best of our knowledge, there were no cases of SAH in which there was only contrast extravasation without aneurysm shown on DSA following negative findings on CTA as in the present case. However, we found four cases of SAH in which there were negative findings on DSA following contrast extravasation without aneurysm on CTA.[3,17] One of the four cases had rebleeding, and the DSA at that time found a pseudoaneurysm, which was endovascularly treated.[3] The other three cases had no rebleeding, and no cause of bleeding was identified.[3,17] Cho *et al*. considered the cause of bleeding to be the rupture of a small perforator from the superior cerebellar artery followed by spontaneous thrombosis.[3] Stentson *et al*. suggested that the cause of bleeding was a tiny dissection of the basilar artery or a rupture of a perforator of the basilar artery.[17]

Although the precise cause of bleeding in this case is uncertain, the possibility of basilar artery perforator aneurysms (BAPAs) should be considered: BAPAs can cause angiogram-negative SAH and spontaneously disappear.[4] BAPAs were first reported by Ghogawala *et al*. and are defined as an aneurysms in which the neck is located entirely on a perforating artery and the basilar trunk is not directly involved.[6] Chavent *et al*. reported that BAPAs are of dissecting origin because the shape of the aneurysm is often described as fusiform and the size of aneurysm varies between closely repeated angiograms.[2] In addition, Mizutani *et al*. reported a dissecting lenticulostriate artery (LSA) aneurysm caused by a hemodynamic stress due to hypertension, in

which disrupted internal elastic lamina was histologically confirmed.[12] Although any histological report of BAPAs has not been found, dissection may cause BAPA formation as with the hypertension-induced dissecting aneurysm of LSA reported by Mizutani *et al*.[4,12] Enomoto *et al*. considered that BAPAs are associated with the middle or distal perforators from the basilar artery and that the angle of the perforator upward or horizontally from the basilar trunk is affected by a hemodynamic stress and is related to hemorrhage.[4,11] In addition, Enomoto *et al*. reported that BAPAs may disappear spontaneously by conservative treatment.[4] No aneurysm was observed on a series of DSAs in this case, however, it is conceivable that the cause of bleeding may be similar to that of BAPAs. In this case, thrombosis occurred immediately after the initial bleeding, resulting in no findings on CTA, whereas DSA showed contrast extravasation from a perforator of the basilar artery due to rebleeding. Although the number of directly observable cases is few and the concept of the disease has not been established, it is thought that there may be a group of diseases that cause SAH due to localized dissection of a perforator of the basilar artery without aneurysmal formation.

CONCLUSION

SAH with no visualization of bleeding sources on CTA and contrast extravasation from a perforator of the basilar artery on DSA is extremely rare. Although thrombosis developed immediately after the initial bleeding, rebleeding occurred during the subsequent DSA and contrast extravasation from a perforator of the middle third of the basilar artery was observed, suggesting that the mechanism of bleeding might include a localized dissection like BAPAs. The present case suggests that there may be a group of diseases which cause the localized dissection of a perforator of the basilar artery and result in SAH without forming aneurysms.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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