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Case Report

Unique image findings around "kissing" distal anterior cerebral artery aneurysms in addition to perianeurysmal edema: A case report

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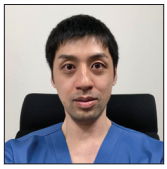

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ABSTRACT

Background: Some aneurysms cause edema formation in the surrounding brain parenchyma and are thought to reflect various phenomena occurring in the aneurysm. Some authors highlighted perianeurysmal edema (PAE) as a finding that indicates higher risk of rupture of the aneurysm. On the other hand, there are no reports of image changes in the surrounding brain parenchyma of aneurysm other than edema formation.

Case Description: We describe a 63-year-old man with unique signal change in the surrounding brain parenchyma of "kissing" distal anterior cerebral artery aneurysms completely different from PAE. The large and partially thrombosed aneurysm presented well-defined signal change surrounding brain parenchyma in addition to PAE. Intraoperative findings revealed the signal change as a space of retaining serous fluid. Drain the fluid and clipping was made for the both anterior cerebral artery aneurysms. The postoperative course was uneventful and his headache was improved the day after the surgery. The perianeurysmal signal change was also disappeared immediately after the surgery except for the PAE.

Conclusion: This case demonstrates a rare phenomenon of signal change around the aneurysm, and there is a possibility that the unique finding exists as an early manifestation of intracerebral hematoma associated with aneurysm rupture.

Keywords: Clipping, Distal anterior cerebral artery, Kissing aneurysm, Perianeurysmal edema, Thrombosed aneurysm

INTRODUCTION

It has been reported that edematous change can occur in the surrounding brain parenchyma of untreated cerebral aneurysms.[5] Edema formation also occurs after coiling and stenting procedures,[3,7,16] but its mechanism is thought to be different and even if it does occur, the risk of the aneurysmal rupture is considered to be low.[13] Several factors such as a pulsatile blood flow,[10] bleeding within an aneurysm wall,[11] and spread of inflammatory cytokines from the partially thrombosed aneurysm[4] have been thought as the candidates of the cause of edema formation. It is understandable that various changes in the aneurysm represented edema formation of the surrounding brain parenchyma and which may be a sign of an increased risk of rupture.[13] On the other hand, there are no reports of changes in the surrounding brain

parenchyma of aneurysm other than edema formation, and the process leading up to the rupture has not been elucidated. We herein report a case of treatment of "kissing" distal anterior cerebral artery (DACA) aneurysms present well-defined signal change in the surroundings in addition to the perianeurysmal edema (PAE) and discuss the clinical implications and mechanism.

CASE REPORT

A 63-year-old man with a history of hypertension presented with a moderate headache lasting 1 week. Fifteen years ago, he had a mild headache and had a computed tomography (CT) of the brain at another hospital and it was evaluated as normal. This time, CT of the brain revealed a high-density mass and surrounding well-defined low density area in the left frontal lobe [Figure 1a]. In addition, edema formation was found in the surrounding brain parenchyma of the high-density mass. CT angiography (CTA) revealed a partially contrasted aneurysm with an intraluminal thrombus arising from the left distal anterior cerebral artery [Figure 1b] and three-dimensional (3D)-CTA revealed a lobulated large aneurysm [Figure 1c]. Magnetic resonance imaging (MRI) also revealed the homogeneous signal change in the left frontal lobe surrounding the aneurysm, as shown by hyperintensity on T2 weighted [Figure 1d] and fluid-attenuated inversion recovery (FLAIR) image [Figure 1e] stronger than the PAE. A part of the surrounding signal change and the aneurysm presented low intensity on T2 star (T2*)-weighted image [Figure 1f]. The major axis of the aneurysm including the thrombotic part was about 20 mm [Figure 1g]. MR angiography also revealed a large lobulated aneurysm [Figure 1h]. Digital subtraction angiography (DSA) and 3D-DSA revealed two saccular aneurysms located at the genu portion of the bilateral anterior cerebral artery (ACA) with domes pressed together [Figures 2a-c], one with a diameter of 14 mm arising from the left ACA and the other of 5mm arising from the right ACA with blebs. Examination of the cerebrospinal fluid (CSF) was normal (clear and colorless, three mononuclear cells per microliter, 47 mg/dL protein). He agreed to undergo an operation clipping both aneurysms to prevent rupture.

Coronal incision and bifrontal craniotomy were made and to dissect the interhemispheric fissure. First, the proximal arteries of bilateral ACA were secured in case of unexpected rupture. Second, an attempt was made to separate the aneurysms, but the complete separation could not be achieved because the aneurysms were tightly adhered and the risk of rupture was assumed to be very high. Most of the left thrombosed aneurysm buried in the frontal lobe and a thin brain parenchyma attached around the neck was incised, a slight yellowish serous fluid was drained [Figure 2d]. There

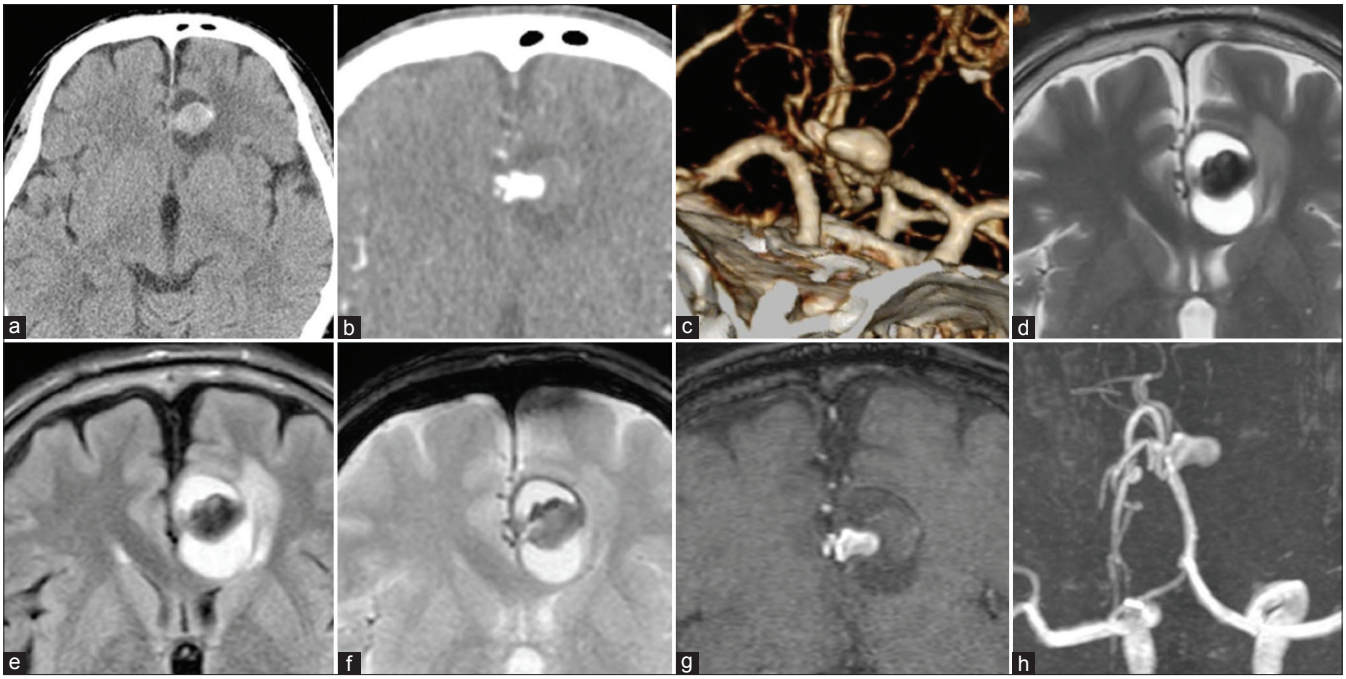

Figure 1: Preoperative computed tomography (CT) and magnetic resonance imaging. (a) Non-contrast CT, (b) CT angiography, (c) three-dimensional-CT angiography, (d) T2 weighted image, (e) fluid-attenuated inversion recovery image, (f) T2*-weighted image. A part of the aneurysm wall and the front rim of the signal change area present low intensity on T2*. (g) Source image of time-of-flight magnetic resonance angiography, (h) Maximum intensity projection image, Blood flow is observed only in a part of the aneurysm, and there is a free space on the outside showing a thrombus. T2* stands for T2 star-weighted image.

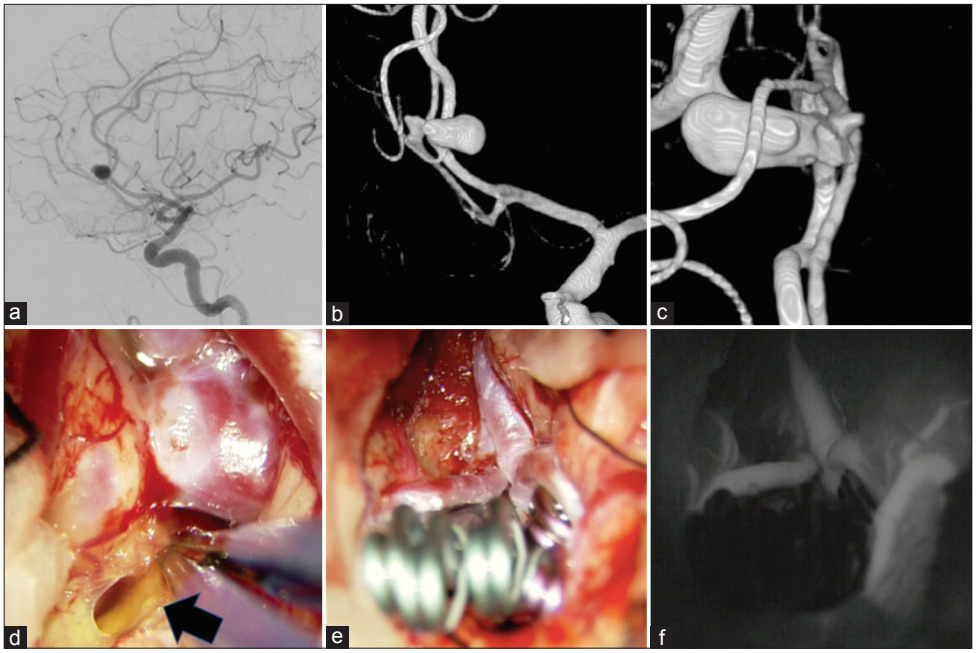

Figure 2: (a) Lateral view of left internal carotid artery angiogram, (b) three-dimensional digital subtraction angiography (3D-DSA) image, (c) operative view of 3D-DSA image, There is a large aneurysm at the genu portion of the left anterior cerebral artery (ACA), and a multilobular aneurysm at the genu portion of the right ACA. Both aneurysm domes are pressed together. (d-f) Intraoperative photographs during clipping of "kissing" distal ACA aneurysms. (d) Opening the space around the left thrombosed aneurysm, yellowish serous fluid was drained (black arrow). The surrounding brain parenchyma was also yellowish to indicate the presence of inflammation. (e) Multiple clips were needed to obliterate both aneurysms. Because of lack of mobility of the aneurysms and limited horizontal space, application of the clips were restricted and should be inserted along the visual axis of the surgical field. (f) Obliteration of the aneurysms and patency of the parent arteries and the branches were confirmed by venous infusion of the indocyanine green.

was no hematoma component and deposition of hemosiderin was observed surrounding brain parenchyma. By draining the serous fluid, the space for inserting clips was secured. There were no findings abnormal thinning and suggestive of rupture in the aneurysm wall that could be observed. Clips were applied to each aneurysm along the visual axis of the surgical field, and the large aneurysm on the left was a broad neck, so multiple clips were applied in parallel [Figure 2e]. Finally, obliteration of the aneurysms and patency of the parent arteries and the branches were confirmed by venous infusion of the indocyanine green [Figure 2f]. Biochemical examination of the serous fluid revealed elevated protein levels (115 mg/dL).

The postoperative course was uneventful. His headache was improved the day after the surgery, and CT scans revealed disappearance of perianeurysmal signal changes except for the PAE [Figure 3a]. Postoperative DSA and 3D-DSA revealed obliteration of both aneurysms [Figures 3b-d]. He was discharged with no neurological deficit 10 days after the surgery. A follow-up MRI performed 6 months after the operation showed improvement of the PAE [Figures 3e and f].

DISCUSSION

In this case, quite unique findings different from PAE were observed around the left aneurysm. Both CT and MRI revealed a well-defined cystic lesion which was more prominent than PAE around the aneurysm. To the best of our knowledge, there have been no reports of the signal changes around cerebral aneurysm as in this case.

In our case, a relative acute enlargement is thought to have occurred in the aneurysm. A part of the thrombus in the aneurysm wall showed a lower signal on T2*-weighted image and these suggested the existence of a fresh intra-mural thrombus and repetitive intra-mural bleeding. Thrombosed aneurysms were characterized by recurrent subacute dissections that resulted in repeated intra-mural hemorrhage lead to the progressive enlargement of the aneurysm.[9,14] The mass effect due to aneurysm enlargement results in decrease perfusion of the surrounding brain parenchyma,[11] and the intra-mural thrombus secrete metabolic factors.[4] These phenomena have manifested as edema formation around the aneurysm.[5,12] Although there are reports of edema

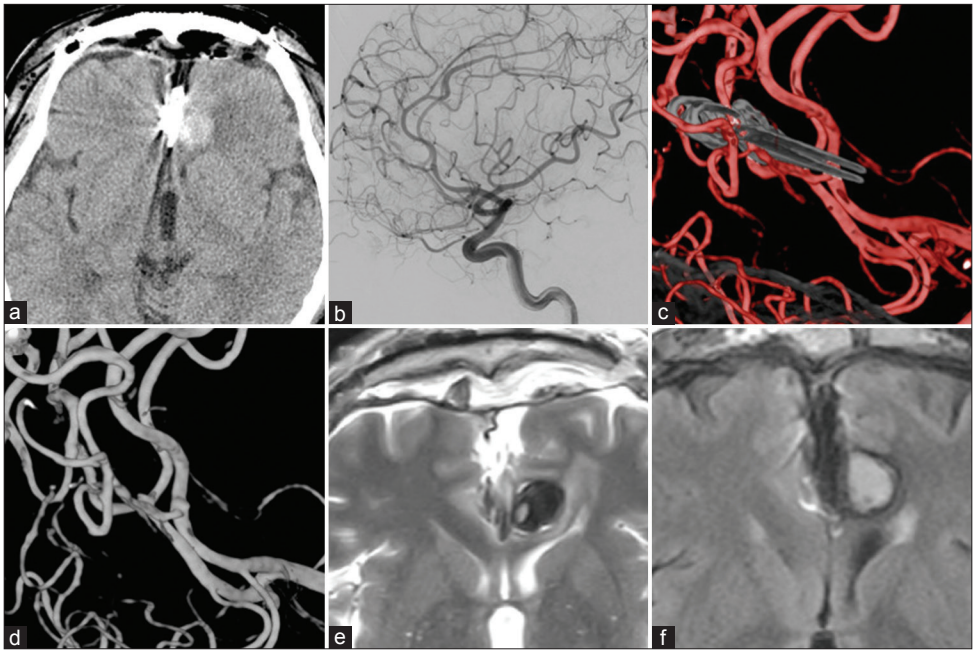

Figure 3: (a) Non-contrast computed tomography the day after the surgery revealed disappearance of perianeurysmal signal changes except for the perianeurysmal edema (PAE) (b) Lateral view of postoperative left internal carotid artery angiogram, (c and d) Postoperative three-dimensional digital subtraction angiography images, obliteration of the both aneurysms were confirmed. (e) T2-weighted image at 6 months after the operation, (f) fluid-attenuated inversion recovery image at 6 months after the operation, Recurrence of the image change around the aneurysm was not observed and both images showed improvement of the PAE.

formation in small aneurysm,[2,6] this is a phenomenon that tends to occur in large, especially partially thrombosed aneurysm,[4,12] so it was not surprising that PAE occurred in this case. Edematous change tends to occur from the thrombosed part of the aneurysm,[9] our case also presented PAE spreading from the thrombosed part of the aneurysm contact with brain parenchyma and the interface of intramural thrombus and the brain parenchyma presented low intensity on T2*-weighted image. These findings supported the concepts that bleeding within an aneurysm wall,[11] and the metabolic factors and/or endothelial growth factor secreted from the thrombosed part of aneurysm induce PAE.[2,4] Moreover, these low intensity on T2*-weighted image continued to the front rim of the cystic lesion. Krings *et al*. described that hemorrhage, as with an edematous reaction, occurring within the thrombosed part which is the periphery of the aneurysm and far from the patent lumen.[9] Pahl *et al*. described that inflammation and consequent weakening of the aneurysm wall and/or exposure of the intra-aneurysmal blood flow to subendothelial tissue leading to the pinpoint leak in the aneurysm and then edema formation was occurring.[13] In our case, intraoperative findings revealed the cystic lesion on MRI was out of the aneurysm wall and yellowish serous effusion was retained. Especially on FLAIR, its signal intensity was higher than CSF and biochemical examination revealed elevated protein levels above CSF, the presence of inflammation was suggested but there was no obvious findings which indicated rupture of the aneurysm. On the other hand, based on the T2*-weighted image findings and the intraoperative findings which revealed the deposition of hemosiderin in the surrounding brain of the aneurysm, the presence of minor leak from the aneurysm could not be denied in our case.

Some of ruptured aneurysms present intracerebral hematoma (ICH), and in rare cases, present without subarachnoid hemorrhage.[15] Occurrence of ICH is associated with the location and size of the aneurysm, and DACA is one of the most frequent sites of rupture with ICH.[1,8] DACA aneurysms commonly form frontal hematoma, and occasionally extend to bilateral frontal lobes through the corpus callosum and form butterfly-like hematoma.[1] The existence of Aneurysmal ICH is strongly associated with poorer functional prognosis.[8] Furthermore, aneurysms which present PAE have a tendency to rupture with ICH.[6,13] The site of the aneurysms form PAE is particularly fragile and buried in the brain parenchyma, so when ruptured, it easily causes ICH. From the above, large DACA aneurysm with PAE as in this case is particularly tend to rupture with ICH, and the unique findings around the aneurysm may have occurred as an early manifestation of ICH. There is a report

of a case that ruptured immediately after PAE was detected, immediate hospitalization and deciding a course of treatment are recommended if this finding is present.[13] Although there was no hematoma component in the cystic lesion, image and intraoperative findings which indicate a minor leak and progression of inflammatory process mean very close to the state of rupture and arising ICH. Consequently, we considered that the perianeurysmal findings observed in this case, including PAE, indicated a higher risk of immediate rupture of the aneurysm and the operation should be performed without waiting.

CONCLUSION

We have described the case of "kissing" DACA aneurysms presented unique findings in addition to PAE. Since DACA is a site that rupture easily associated with ICH, the serous fluid retention around the aneurysm may be a finding that captures the early manifestation of ICH formation. Early treatment should be considered for aneurysms with these surrounding image changes, including PAE.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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