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Pathological examination of a ruptured fusiform aneurysm of the middle cerebral artery

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Abstract

Background: Little is known about the pathogenesis and clinical course of fusiform compared with saccular aneurysms. The case of a ruptured fusiform aneurysm accompanied by dissection at the M2 portion of the middle cerebral artery (MCA) is reported, along with pathological findings.

Case Description: A 41-year-old female presenting with subarachnoid hemorrhage was revealed to have a ruptured fusiform aneurysm at the M2 portion of the right MCA on angiography. She was treated with superficial temporal artery-MCA anastomosis and trapping of the aneurysm. The aneurysm consisted of a whitish fusiform dilatation with a thickened wall of the MCA and two red protrusions on it. Pathological examinations revealed disruption and fragmentation of the internal elastic lamina and intimal thickening in the fusiform lesion. There were two aneurysmal protrusions on the main fusiform dilatation. In one protruded lesion, a dissection of the intima was observed.

Conclusion: We propose that a dissection and saccular aneurysm additionally developed on the wall of a preexisting segmental ectasia of the MCA in our case. In this report, we discuss the etiology of fusiform aneurysms of the MCA.

Key Words: Dissecting aneurysm, fusiform aneurysm, internal elastic lamina, intimal thickening, middle cerebral artery

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INTRODUCTION

Fusiform aneurysms of the middle cerebral artery (MCA) have different pathological features, hemodynamics, and natural histories than saccular aneurysms.[2,15] Little is known about the etiology and pathology of fusiform aneurysms arising from the arterial trunk unrelated to the arterial branches in contrast to saccular aneurysms.[7,13] Only a few papers regarding the etiology and classification of fusiform aneurysms of the MCA have been published.[2,13] In addition to their pathogenesis, their clinical course has not yet been fully clarified.[12] We encountered the case of a fusiform aneurysm of the MCA presenting with subarachnoid hemorrhage (SAH). In this paper, unique pathological findings are reported, and the pathogenesis of fusiform aneurysms of the MCA is discussed.

CASE REPORT

A 41-year-old female suddenly developed headache and nausea. She visited a local physician, and was referred to one of our hospitals. On admission, neither consciousness

disturbance nor neurological deficit was observed. Her past history included scoliosis since birth; however, other diseases such as infection or connective tissue disease were not noted. A routine blood examination was not remarkable. Further, the examination of antigens and antibodies for infectious diseases also revealed no abnormality. Computed tomography (CT) demonstrated SAH [Figure 1a], and three-dimensional CT angiography (3D-CTA) showed a dilated M2 portion of the right MCA accompanied by two small notches [Figure 1b]. Magnetic resonance imaging (MRI) performed 4 months prior to the episode had already revealed the irregular wall of the right M2 [Figure 1c]. MRI performed at admission showed the same findings as those of 3D-CTA [Figure 1d]. Cerebral angiography revealed the dilated M2 of the right MCA [Figure 2]. The wall of the dilated portion was relatively smooth. Angiography neither showed a double lumen, pearl and string sign nor the retention of contrast medium. Based on these radiological findings, the lesion was diagnosed as a fusiform aneurysm, and delayed surgical intervention was planned. Repeated angiography was performed on the 23rd day after onset. The radiological findings showed no marked change of the lesion. The third MRI 33 days after the onset showed the same findings as those of the first and second MRI.

She underwent surgery on the 45th day after onset. Right fronto-temporal craniotomy and superficial temporal artery (STA)-MCA anastomosis were performed. Then, the lesion was trapped and resected. The lesion consisted of a dilated arterial trunk with a whitish, thickened wall and two protrusions with red walls on the main lesion [Figure 3]. There was no organized thrombus around the aneurysm. Pathological examination of the resected lesion showed a disrupted internal elastic lamina and intimal thickening of the main trunk [Figure 4e]. The large protrusion on the main trunk had a dissected intima without smooth muscle cells stained with anti-α-smooth muscle actin antibody [Figure 4a, b]. In contrast, the small protrusion had no intimal flap, and no internal elastic lamina was observed in the dome [Figure 4c and d]. Postoperative angiography showed the disappearance of the aneurysm and patency of the bypass. Her postoperative course was uneventful. She was discharged without neurological deficit on the 62nd day after onset.

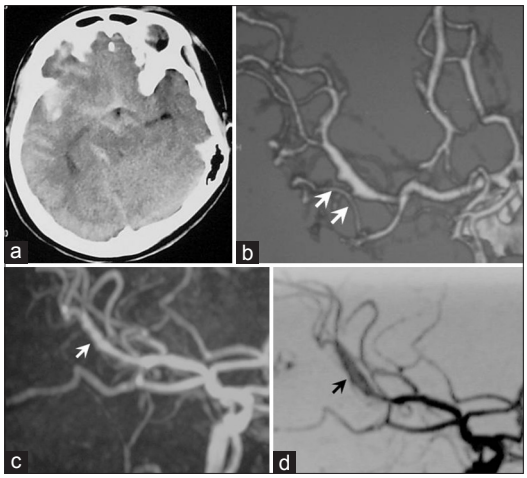

Figure 1: (a) CT showing SAH dominantly on the right side. (b) 3D-CTA showing a dilatation of M2 of the right MCA. Two small notches can be seen on the main lesion (arrows). (c) MRI 4 months before SAH showing an abnormality on the right M2 portion (arrow). (d) MRI on admission showing a dilatation of M2 (arrow)

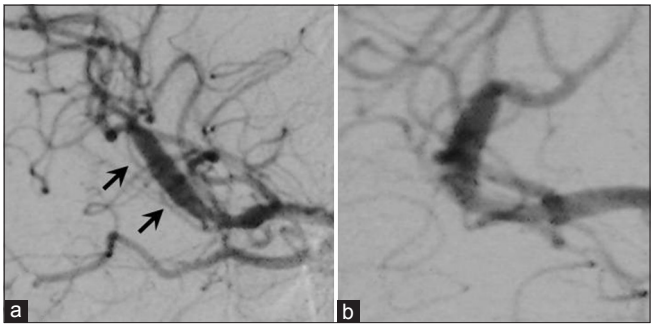

Figure 2: Angiograms with right (a) and left (b) anterior oblique views, showing an aneurysmal dilatation of the right M2 (arrows)

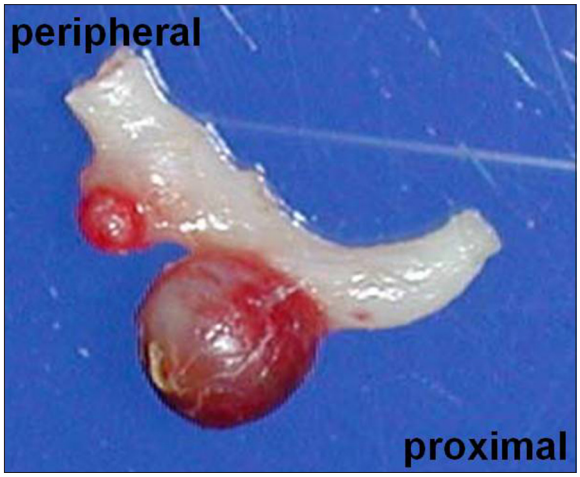

Figure 3: Photograph of the resected lesion showing a dilated white MCA trunk with two red protrusions

DISCUSSION

Fusiform aneurysms, including dissections in the anterior circulation, are rare compared with those in the vertebrobasilar system.[8] Many factors have been considered in the development of arterial dissection, such as infections, connective tissue diseases, and arteriosclerosis.[9] Previously, some authors suggested that fusiform aneurysms originated from atherosclerotic changes.[1] However, reports that fusiform aneurysms had no relation to atherosclerosis in their origin have been published.[12] Thus, the etiology and natural course of this condition remain to be elucidated.

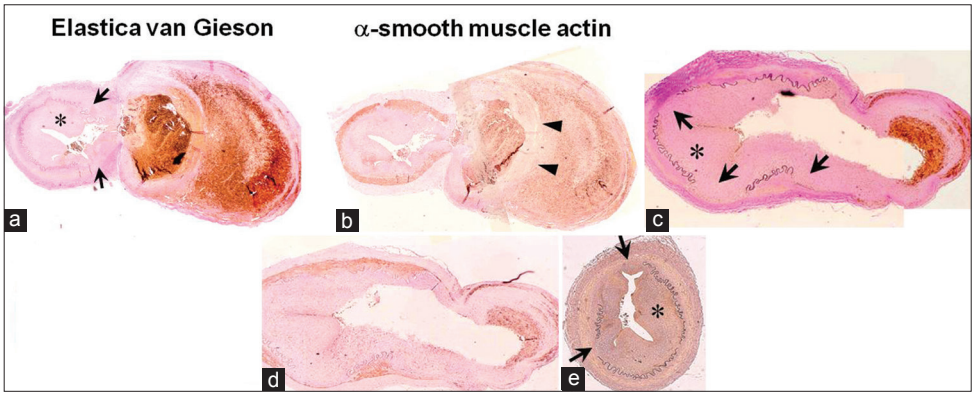

Figure 4: Microphotographs of samples stained with Elastica van Gieson stain (a, c, e) and immunostained with anti-α-smooth muscle actin antibody (b, d). Cross-section of large (a, b) and small (c, d) protrusions, and a dilated arterial trunk between the two protrusions (e) are presented. Figures (a, c, e) demonstrating disruption of the internal elastic lamina (arrows) and intimal thickening (asterisk). The cross-section at the site of the large protrusion (b) showing that the lesion has an intimal flap (arrowheads) without the existence of smooth muscle cells positive for anti-α-smooth muscle actin. The small protrusion (d) has no intimal flap ×40

In our case, MRI 4 months prior to the episode already showed the lesion with an irregular wall. It was reported that, if a dissecting aneurysm is suspected, serial radiological examinations should be conducted, because the radiological characteristics of lesion might change dynamically over a short period of time.[9] The angiographic findings of dissecting aneurysms in MCA are reportedly similar to those of vertebrobasilar arteries.[16] In our case, angiography showed a dilated lesion of the MCA with a relatively smooth wall. Two small protrusions observed on 3D-CTA were not apparent on angiography. Neither a pearl and string sign, double lumen, nor retention of contrast medium was observed. These findings did not change over three consequtive angiograms.

The macroscopic features of our case were very unique. The lesion had a dilated arterial trunk and two small protrusions on it. The main lesion of the MCA trunk was whitish, and the cross-section of the sample showed a thickened wall structure and narrowed cavity. In contrast, the two protruded portions were reddish in appearance. There was no connection between the two protrusions. The size of the protrusions observed on 3D-CTA was much smaller than the total size of those observed macroscopically. These findings indicated that these two protrusions contained a thrombus.

Mizutani *et al.*[13] classified dissecting lesions into four types, based on the condition of the internal elastic lamina and intima. Type 1 has acute, widespread disruption of the internal elastic lamina without intimal thickening. Type 2 shows stretched and/or fragmented internal elastic lamina with intimal thickening. Type 3 possesses fragmented internal elastic lamina, multiple dissections of a thickened intima, and an organized thrombus in the lumen. Type 4 shows minimal disruption of the internal elastic lamina without intimal thickening. The pathological mechanism by which a cerebral dissecting aneurysm is created was reported to be the sudden widespread disruption of the internal elastic laminae,[3,14] and these disruptions are irreversible.[11] The internal elastic lamina is the most important layer of the cerebral arterial wall for the protection of the artery against hemodynamic stress.[14] Factors that weaken the structure of the internal elastic lamina, such as inflammation, infection, trauma, and congenital factors, may be associated with the etiology of fusiform and dissecting aneurysms.[13] Frösen *et al.*[4] described four basic types of saccular aneurysm wall that associated with rupture. They mentioned that apoptosis, de-endothelialization, luminal thrombosis, smooth muscle cell proliferation, and T-cell and macrophage infiltration associated with rupture. Further, inflammation[17] and apoptosis[10] have been reported to result in degenerative changes of aneurysm wall and subsequent rupture in saccular aneurysms. In our case, disruption of the internal elastic lamina and intimal thickening were observed. These findings indicated that degeneration and repair of arterial wall resulted in formation of fusiform aneurysms as saccular ones. However, no history of infection, congenital connective tissue diseases, or trauma was noted. In addition, our patient had no external risk factors for cerebral aneurysm development and rupture, such as smoking, hypertension, and family history.[4] Further investigation about the effect of inflammation and/or apoptosis on pathogenesis and rupture of fusiform aneurysms are mandatory.

The majority of disrupted internal elastic laminae may be covered with local intimal thickening and may not develop into aneurysms.[14] The adaptive intimal thickening compensates for the weakening of the arterial wall caused by a damaged internal elastic lamina.[5,6] Mizutani *et al.*[13] reported three cases of saccular aneurysms located on segmental ectasia in the anterior circulation. They could not detect the presence of a pseudolumen or organized thrombus. Type 4 aneurysms lacking the

internal elastic lamina in the dome may rupture before they have a chance to grow into giant aneurysms. The pattern of lesional internal elastic lamina of Type 4 aneurysms is more similar to Type 1 aneurysms than to common saccular aneurysms.[13] In our case, the internal elastic lamina was disrupted and intimal thickening was observed in the main lesion of the aneurysm. At the site of the two protrusions, the internal elastic lamina and smooth muscle cell layer were absent. The dome wall was comprised of a fragile intima and adventitia. In the large protrusion, immunohistochemical analysis revealed a dissected flap in the thickened intima. These findings suggested that the large protrusion developed as a result of dissection, and then a thrombus formed in the dissected cavity. A part of the thrombus was organized and new vessel formation was observed. In contrast, in the small protrusion, no intimal flap was detected. We speculate that the two protrusions were newly developed dissecting and saccular aneurysms on the preexisting arterial ectasia of the MCA.

CONCLUSIONS

Pathological examination revealed that our case might be a segmental ectasia of the MCA accompanied with newly developed dissecting and saccular aneurysms. Even a simple ectasia of the arterial trunk might have the potential to develop a dissecting or saccular aneurysm. Therefore, for such cases, serial radiological examinations and close observation are necessary.

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