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Case Report

The Histopathological Findings of Two Nonbranching Saccular Cerebral Aneurysms

Satoru Kida, Hiroshi Tenjin, Tsutomu Tokuyama, Osamu Saito

Department of Neurosurgery, Japanese Red Cross Shizuoka Hospital, Shizuoka, Japan

Abstract

Cerebral aneurysms arising from nonbranching sites are different from ordinary branching aneurysms in clinical course and histology. We pathologically examined two cases of saccular aneurysm occurring at nonbranching sites. One was a pseudoaneurysm arising at a branch of the right pericallosal artery. The other had an entirely hyalinized and thickened aneurysmal wall. Despite similar angiographical findings, our two cases had different pathological features as described above. Based on the pathological findings obtained from these cases, we believe that aneurysms in nonbranching sites are caused by injury to the internal elastic lamina. A ruptured aneurysm may be discovered as a blood blister-like aneurysm, whereas an unruptured one may develop into a "nonbranching true aneurysm."

Keywords: *Arterial dissection, internal elastic lamina, nonbranching aneurysm*

Introduction

The majority of cerebral saccular aneurysms arise at arterial bifurcations, and cerebral aneurysms occurring at nonbranching sites are relatively rare. Previous studies have demonstrated that nonbranching site aneurysms are typically associated with arterial dissection and take the form of so-called blood blister-like aneurysms.[1] However, there are some reports of true saccular aneurysms at nonbranching sites of the internal carotid artery (ICA),[2] it is not clear how their formation differs from bifurcation aneurysms. We recently encountered two cases of a saccular aneurysm arising at nonbranching sites and examined the pathological findings. We discuss the pathogenesis and formation mechanism of nonbranching cerebral aneurysms.

Case Reports

Case 1

A 79-year-old male with a history of diabetes mellitus and hypertension presented with sudden headache and leg weakness without any head trauma. He showed confusion and had mild paresis in his left leg. Computed tomography (CT) of the head showed an acute subdural hematoma in the interhemispheric fissure and subarachnoid hemorrhage [Figure 1a]. CT angiography showed a 4-mm saccular aneurysm on a branch of the right pericallosal artery [Figure 1b]. After confirming that the aneurysm was not infectious, we performed trapping of the aneurysm on day 5. The intraoperative findings showed that the aneurysmal neck was torn at its origin [Figure 1c]. H and E staining of the resected aneurysm showed the lesion was composed of mostly thrombi, and no obvious vascular structures, such as the internal elastic lamina (IEL) or adventitia, were observed [Figure 1d]. The pathogenesis of aneurysm formation was considered to be mild arterial wall injury caused by the edge of the falx cerebri.

Case 2

The patient was a 67-year-old male with a history of diabetes mellitus and hypertension. A 15-mm unruptured aneurysm was incidentally found in the left middle cerebral artery (MCA) at a nonbranching site of the M2 inferior trunk, where the MCA sharply curved [Figure 2a and b]. Neck clipping through the pterional approach was performed, and the aneurysmal dome was resected. The intraoperative findings showed a firm aneurysmal neck and severe

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Address for correspondence:
Dr. Satoru Kida,
Department of Neurosurgery,
Japanese Red Cross Shizuoka Hospital, Shizuoka, Japan.
E-mail: we.must.respect.the.will@gmail.com

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atherosclerosis on the dome [Figure 2c]. Pathological findings showed that the intima was entirely hyalinized and thickened [Figure 3a]. Although Elastica van Gieson stain typically shows the IEL as a black-purple membrane, no such staining was observed in the aneurysmal wall [Figure 3b]. Pathologically, this aneurysm looked like an ordinary bifurcation aneurysm on a branching site.

Discussion

We encountered two different types of nonbranching aneurysms. Pathologically, one was a pseudoaneurysm, and the other was a firm saccular aneurysm similar to those seen on branching sites. Anterior wall aneurysms comprise 0.9%–6.5% aneurysms in the ICA,[3] of which a previous study suggested there are two types, differing in shape, histological features, and treatment requirements.[2] One is a blood blister-like type, showing a small hemispherical bulge located at the anterior wall of the supraclinoid portion of ICA. Most patients with this type of aneurysm suffer massive subarachnoid hemorrhaging because the aneurysmal walls are thin and fragile.[3] Pathologically, abrupt termination of the IEL is observed at the area adjacent to the rupture point, which is not composed of collagenous tissue, as seen in an ordinary aneurysm. The other type is the saccular type, which has a saccular dome with an obviously firm neck similar to ordinary aneurysms arising at arterial bifurcations. Unlike with the blood blister-like type, neck clipping is possible for this type of aneurysm as in ordinary bifurcation aneurysms. We believe that this classification system can be applied to cerebral arteries other than the ICA, such as in our two cases.

Mizutani *et al*. reported that saccular-shaped arterial dissections could develop by tearing of the IEL at nonbranching sites.[4] They classified aneurysms unrelated to the branching zones into four types, of which type 4 aneurysms appear saccular shaped. However, pathologically, these have an abruptly

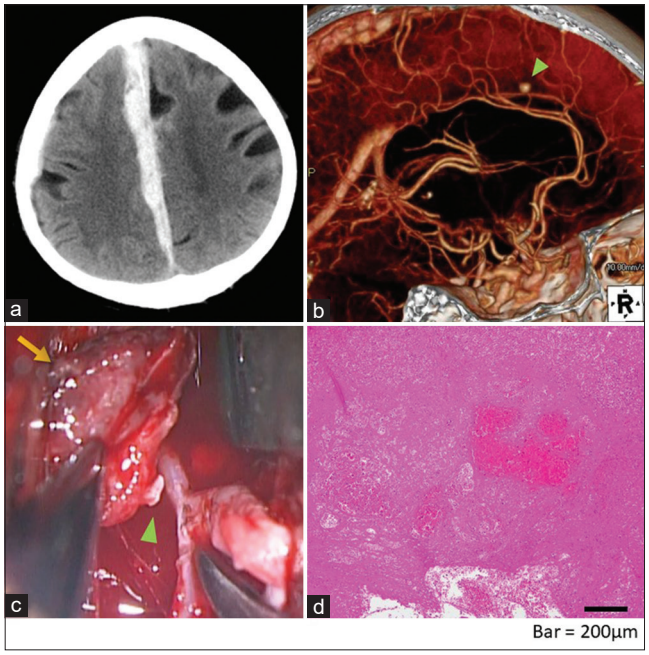

Figure 1: (a) Head computed tomography showing acute subdural hematoma in the interhemispheric fissure. (b) Aneurysm located on a branch of the right pericallosal artery demonstrated by three-dimensional reconstructed computed tomography angiography. (c) Intraoperative image just after removal of the aneurysm (arrow). The aneurysmal neck was torn at its origin (arrowhead). (d) H and E staining of the resected aneurysm (×10). No obvious vascular structures such as internal elastic lamina or adventitia are visible

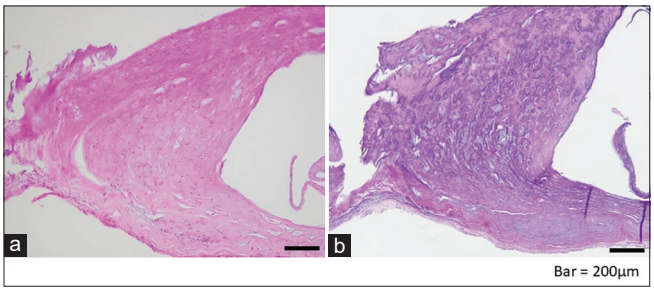

Figure 3: (a) H and E staining of the resected aneurysm (×10) shows a considerably thickened and hyalinized intima. (b) Elastica van Gieson staining section shows lack of internal elastic lamina on the aneurysmal wall

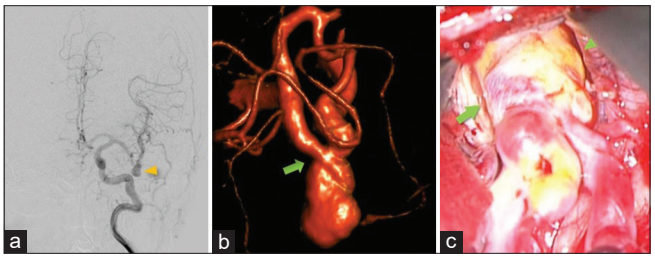

Figure 2: (a) Right internal carotid artery angiography shows a 15 mm saccular aneurysm on the left middle cerebral artery-M2 inferior trunk (arrowhead). (b) Three-dimensional reconstructed image showing the aneurysm originating from the convex portion of the sharply curved vessel (arrow). (c) Intraoperative image showing the aneurysm (arrow) with severe atherosclerosis on the aneurysmal dome (arrowhead)

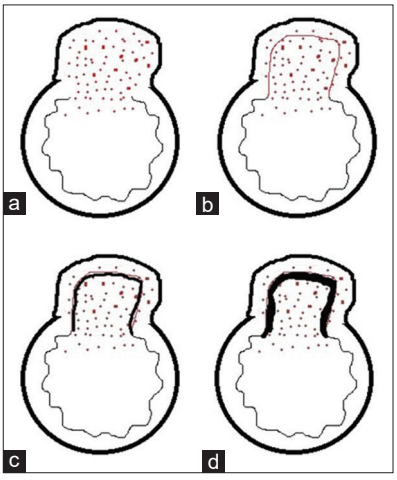

Figure 4: Scheme of the development of nonbranching true aneurysm. When arterial dissection occurs (a), the luminal surface is covered by endothelium in 2–4 days (b). Then, neointima gradually forms over 3 months (c). Finally, fibroblastic tissue thickens the aneurysmal wall and a nonbranching true aneurysm has formed (d)

Table 1: Proposed concept of aneurysm classification

Type	Ordinary aneurysm	Non branching true aneurysm	Arterial dissection blood blister-like aneurysm
Shape	Saccular		Fusiform
			Rapid change in the shape
Pathology	Fibrosis		Thin wall
	Hyalinization		No collagenous tissue
	Fragmentation and disappearance of IEL		Abrupt tearing of IEL
Developmental mechanism	Medial defect at bifurcation	IEL tearing caused by mild arterial wall injury	
Location	Bifurcation	IC anterior wall, nonbranching site	
Time course	Chronic	Acute	

IEL – Internal elastic lamina; IC – Internal carotid

disrupted IEL without intimal thickening, and lack IEL on the dome. Case 1 and blood blister-like aneurysms are consistent with type 4 of this classification system.

The question remains of how an aneurysm like the one in Case 2 is formed. We hypothesize that if the IEL tears but does not rupture, it can become a firm saccular aneurysm or "non-branching true aneurysm." A previous study suggested that when the IEL is injured, the lesion is covered by endothelium in 2–4 days, and neointima is formed in about 3 months.[5] It was also reported that fibroblastic tissue can thicken the aneurysmal wall.[6] Based on these facts, nonbranching true aneurysms can develop, as shown in Figure 4. When the IEL is torn by mild wall injury, such as shear stress or mild trauma, but does not rupture, the inner layer of the thin aneurysmal wall is first covered by endothelium. Neointima is then formed, and finally, the aneurysmal wall becomes thickened by fibroblastic tissue and hyalinization. The fact that blood blister-like aneurysms are only found in cases of arterial rupture, contrary to arterial dissection which has nonhemorrhagic onset, leads us to propose that unruptured blood blister-like aneurysms can develop into nonbranching true aneurysms.

As a pathological feature, ordinary aneurysms show fibrosis and hyalinization of the arterial wall, and fragmented and/or missing IEL.[7] The nonbranching true aneurysm presented in Case 2 had pathological findings similar to ordinary aneurysms, but the difference between these cases is the mode of occurrence. Ordinary aneurysms occur because of a congenital medial defect at the bifurcation,[7] while nonbranching true aneurysms may be due to IEL injury caused by mild arterial wall damage. Table 1 shows a proposed concept of aneurysm classification.

In conclusion, aneurysms arising from nonbranching sites take the form of either pseudoaneurysms or true saccular aneurysms. The latter of which have similar pathological findings to bifurcation aneurysms. This fact and previous studies propose that nonbranching true aneurysms can develop from unruptured blood blister-like aneurysms caused by IEL injury due to mild arterial wall damage. Further reports will help to elucidate the precise mechanism of aneurysm formation at nonbranching sites.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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