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Case Report

Aneurysm of lenticulostriate artery in a patient presenting with hemorrhage in the caudate nucleus and lateral ventricle-delayed appearance and spontaneous resolution

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Abstract

Background: An aneurysm of distal lenticulostriate artery is very rare. The natural course and management of this rare aneurysm are not clear.

Case Description: An 81-year-old woman developed consciousness disturbance. Computed tomography revealed hemorrhage in the right caudate nucleus and lateral ventricles. Three-dimensional computed tomographic angiography demonstrated only an aneurysm at the basilar artery. On angiography, on the sixth day, an aneurysm at the right lenticulostriate artery was demonstrated. Then, the aneurysm disappeared on three-dimensional computed tomographic angiography on the 15th day. Subsequent radiological examinations revealed no vascular anomaly in the right lenticulostriate artery.

Conclusion: An aneurysm at this location can show dynamic changes based on radiological findings. Close radiological observation is necessary.

Key Words: Cerebral aneurysm, delayed appearance, lenticulostriate artery, ruptured, spontaneous obstruction

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INTRODUCTION

An aneurysm originating from the distal lenticulostriate artery is very rare, and it is difficult to treat by direct surgery or endovascular embolization due to its location.[7,18] The natural course and management of this rare aneurysm have not been fully clarified. Recently, we encountered a patient with an aneurysm on the distal lenticulostriate artery presenting with intracerebral and intraventricular hemorrhage. The aneurysm was not detected on the initial radiological examinations. It was first demonstrated on the 6th day on angiography, and had disappeared on the following three-dimensional computed tomographic angiography (3D-CTA) on the 15th day. In this report, we present this case of distal lenticulostriate artery aneurysm, and discuss the clinical course and management of this rare aneurysm.

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CASE REPORT

An 81-year-old woman suddenly developed consciousness disturbance. She was brought to our hospital by ambulance. Computed tomography (CT) revealed hemorrhage in the right caudate nucleus and ventricles, and hydrocephalus [Figure 1a]. 3D-CTA on admission demonstrated an aneurysm on the basilar artery (BA) at the bifurcation of the left superior cerebellar artery, but not on the right lenticulostriate artery [Figure 1b]. Raw 3D-CTA images showed no enhancement adjacent to the hematoma [Figure 1c]. Although the BA aneurysm was detected, it was not considered to be the origin of hemorrhage. A drainage tube was inserted to the left lateral ventricle to control hydrocephalus. On the sixth day, angiography was performed to evaluate the BA aneurysm. The angiography additionally demonstrated an aneurysm located at the right lenticulostriate artery [Figure 2a]. The size of aneurysm was about 3 mm in diameter. Retention of contrast medium in the aneurysm was observed on CT obtained after angiography [Figure 2b]. On retrospective observation of the initial noncontrast and contrast-enhanced (CE) CT, a small low-density region was observed in the hematoma [Figure 1a and c]. This portion appeared to be identical to the enhanced portion on postangiography CT. It was revealed that the aneurysm was surrounded by a hematoma. This aneurysm was diagnosed as the cause of hemorrhage. On magnetic resonance angiography on the 13th day and 3D-CTA on the 15th day, the aneurysm was not opacified on the right lenticulostriate artery [Figure 3a]. Angiography performed on the 23rd day also showed the disappearance of the aneurysm [Figure 3b]. Follow-up 3D-CTA on the 42nd day demonstrated no aneurysm on the right lenticulostriate artery [Figure 3c]. On the 31st day, right ventriculo-peritoneal shunting was performed. After the operation, she gradually regained consciousness. She was transferred to another hospital for rehabilitation for disuse syndrome on the 67th day. 3D-CTA obtained 9 months after onset showed no recurrence of the aneurysm on the right lenticulostriate artery [Figure 3d]. Her activities of daily living normalized at 9 months after onset.

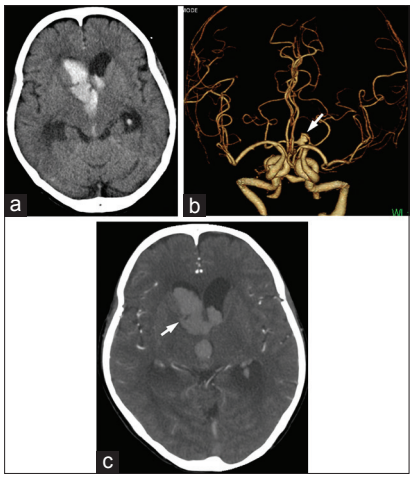

Figure 1: (a) Computed tomography demonstrating hemorrhage in the right caudate nucleus with ventricular rupture. (b) Three-dimensional computed tomographic angiography demonstrating an aneurysm only on basilar artery (arrow). (c) A raw image of three-dimensional computed tomographic angiography on admission showing no abnormal enhancement adjacent to the hematoma. A small low-density area is observed in the hematoma (arrow)

DISCUSSION

An aneurysm arising from the distal lenticulostriate artery is very rare. The natural course and management of an aneurysm on the distal lenticulostriate artery have not been fully clarified. To our knowledge, 62 cases have been reported in the literature.[18,33,35] Among them, 52 cases of distal lenticulostriate artery aneurysms including our case are summarized in Table 1.[1-27,31-45] Patients' ages varied from 2 months to 81 (average 41.9) years. Patients were relatively young compared to those of the cases with common saccular aneurysms. This might be due to difference in aneurysm characteristics. The nature of the lenticulostriate artery aneurysm is likely to be dissection or pseudoaneurysm rather than a true aneurysm. Seventeen cases (32.7%) were associated with cerebrovascular diseases such as moyamoya disease, middle cerebral artery occlusion, and arteriovenous malformation. Association with other vascular anomalies implies that hemodynamic stress might be one of the causes of lenticulostriate artery aneurysm. In fact, almost all the reported cases were ruptured ones, and only 1 case was unruptured.[45] The aneurysm size was described in 35 cases and 29 were less than 5 mm. The only one unruptured aneurysm was as large as 9 × 6 × 6 mm. The ruptured aneurysm size in this location seems to be smaller than that of common aneurysms. These observations suggest that dissection or pseudoaneurysm might be formed and developed due to hemodynamic stress in the lenticulostriate artery.

Regarding the natural course of the aneurysm on the distal lenticulostriate artery, there have been several

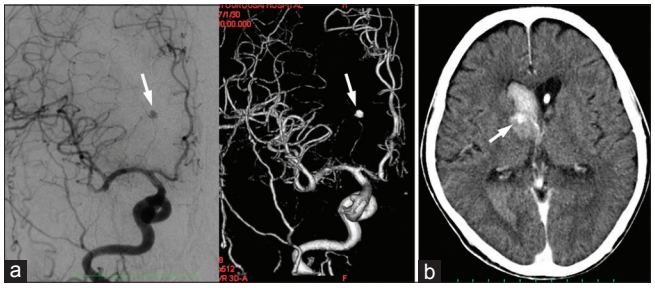

Figure 2: (a) Angiography performed on the sixth day showing an aneurysm originating from the right lenticulostriate artery (arrow). (b) Computed tomography after angiography showing an enhanced lesion in the hematoma (arrow). This portion appears identical to the low-density area indicated in Figure 1a and c

Table 1: Summary of cases of distal lenticulostriate artery aneurysms

Reference	Author, year	Age (years), sex	Associated disease	CT findings	Size of aneurysm (mm)	Treatment	Time of follow-up	Outcome of follow-up radiological examinations	Outcome	Pathology	Others
26	Murakami *et al*. 1984	33, M	Moyamoya, epilepsy	ICH, IVH	Small	VED	NA	NA	Death	True aneurysm	
10	Grabel *et al*. 1989	60, M	Moyamoya	ICH	NA	Hematoma evacuation	3 weeks	Near disap.	GR		Near disap.
11	Gupta *et al*. 1989	36, F	_	ICH	NA	Conservative	2 months	Disap.	GR		
3	Albert *et al*. 1997	8, NA	AVM	ICH	NA	Excision					
16	Kaptain *et al*. 2001	2 months, M	_	ICH, IVH	NA	Excision	_	_	MD	Absence of elastic laminae, no infection	
22	Larrazabal *et al*. 2001	57, F	Moyamoya	ICH, IVH	4	Endovascular, NBCA	NA	Disap.	SD		
42	Vates *et al*. 2001	35, M	Neurocytoma	IVH	7	Excision of aneurysm and tumor	NA	Disap.	SD	True aneurysm	
23	Lehmann *et al*. 2003	26, M	_	ICH	NA	Conservative	3 months	Disap.	GR		
		26, M		ICH, SAH	NA	Conservative	NA	Disap.	GR		
		59, F	Moyamoya	ICH	NA	Conservative	3 weeks	Disap.	MD		
		2.5, F	_	ICH, HCP	NA	Excision	NA	NA	SD		
14	Horn *et al*. 2004	44, F	_	ICH	2	Clipping	NA	Near disap.	GR		
27	Narayan *et al*. 2004	69, F	_	ICH, IVH, HCP	3→4	Clipping	NA	Obliteration	GR		Growth on angiography (day 14)
34	Sakai *et al*. 2005	61, F	Moyamoya-like	ICH	Small	Clipping	NA	Obliteration	SD		
2	Ahn *et al*. 2007	49, M	Moyamoya	ICH, IVH	3	Excision	4 weeks	Obliteration	MD	True aneurysm	Aneurysm detected on angiography on day 14
		24, M	_	ICH, IVH, SAH	4	Conservative	_	_	Death		
25	Matushita *et al*. 2007	5, M	_	ICH, IVH	4	Excision	NA	NA	GR	Thin arterial wall, no inflammation	

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Table 1: Contd...

Reference	Author, year	Age (years), sex	Associated disease	CT findings	Size of aneurysm (mm)	Treatment	Time of follow-up	Outcome of follow-up radiological examinations	Outcome	Pathology	Others
9	Gandhi *et al*. 2008	53, M	HT	ICH, SAH	2	NA	Postoperative	No residual aneurysm	mRS: 2		
		59, M	Moyamoya	ICH, SAH, IVH, HCP	4	Proximal clipping	Postoperative	Obliteration	mRS: 1		
		41, M	Cocaine abuse	ICH	4	Resection	Postoperative	Obliteration	mRS: 1		
		37, F	Moyamoya	ICH, SAH	3	NA	Postoperative	Small residual aneurysm	mRS: 4		
		31, F	Moyamoya	ICH, SAH	3	Proximal clipping	Postoperative	No residual aneurysm	mRS: 2		
38	Takeuchi *et al*. 2009	59, M	HT	ICH, IVH	NA	Conservative	9 weeks	Disap. (all 3 aneurysms)	VS		2 aneurysms (lt) 1 aneurysm (rt LSA)
43	Wong *et al*. 2009	39, M	MCA narrowing	ICH	Small	Conservative	2 years	Disap.	GR		Spontaneous resolution
19	Kochar *et al*. 2010	50, M		SAH	2	Trapping	3D after operative	No residual aneurysm	GR		
8	Ellis *et al*. 2011	71, F	HT, Af on warfarin, HL, FMD	IVH	4 × 2.6	Conservative	5 days	Disap.	GR		Spontaneous resolution (day 5)
12	Harreld and Zomorodi 2011	35, F	Moyamoya, ruptured aneurysm	Clipped rt pericallosal aneurysm	3 → 4.2 x 3.9 × 3.8 (1 month)	Endovascular, NBCA			GR		Enlargement after 1 month
40	Tsai *et al*. 2011	71, F	_	ICH, IVH	4	Embolization, NBCA	1 year	No recurrence	GR		
4	Bhat *et al*. 2012	39, M	_	SAH	NA	Trapping excision	NA	NA	GR		Fusiform aneurysm
21	Lan *et al*. 2012	21, F	_	ICH, IVH	5	Gamma knife radiosurgery	22 months	Disap.	GR		
45	Yasher *et al*. 2012	66, F	HT, Moyamoya, HL	Unruptured	9 × 6 × 6	2 attempts of embolization → proximal clipping	1 day after operative	Complete resolution	GR		
5	Cai *et al*. 2013	41, F	HL	ICH	3	Clipping	3 months	Obliteration	MD		Delayed appearance (day 4)
6	Chalouhi *et al*. 2013	49, M	_	ICH	3 × 3	Embolization with onyx LSA occlusion	NA	Complete resolution	MD		

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Table 1: Contd...

Reference	Author, year	Age (years), sex	Associated disease	CT findings	Size of aneurysm (mm)	Treatment	Time of follow-up	Outcome of follow-up radiological examinations	Outcome	Pathology	Others
17	Kim *et al*. 2013	28, M	Bipolar disorder on lithium	IVH	7.5 x 4.5 → 4.0 x 2.3 → 4.2 x 3.2 (1month) on	VED, clipping → resection, postoperative day 4, residual sac → removed	Postoperative	No evidence of aneurysm	GR	Pseudoaneurysm	Size change (+) Shape change (+)
37	Srivastava *et al*. 2013	14, F	AVM	IVH, HCP	NA	Conservative	NA	NA	GR		CTA: not detected angiography: detected
		45, F	_	IVH	NA	Conservative	NA	NA	GR		
1	Agarwalla *et al*. 2014	41, F	Chronic pain syndrome	ICH	Outpouching → 3	Proximal clipping	Intraoperative	No filling	SD		Delayed enlargement on day 4
13	Heck *et al*. 2014	29, M	_	ICH	1 → 2 (day 80) → slight decrease in size (3 m)	Conservative	NA	Enlarge → decrease in size	mRS: 2		Size change (+)
		63, M	HT, polycystic kidney	ICH, IVH, SAH	2, fusiform aneurysm	Conservative	17 months	Disap.	mRS: 1		Spontaneous obliteration of aneurysm and feeding artery
		32, F	HT, alcoholism	ICH, IVH, SAH	2	Conservative	4 months	Disappeared on CTA	mRS: 3		Spontaneous occlusion
15	Hwang *et al*. 2014	53, F	Moyamoya	ICH, IVH	NA	Embolization, NBCA	1 year	No recurrence	GR		
		44, F	Moyamoya, contralateral intracerebral hemorrhage	ICH, IVH	NA	Embolization, NBCA	1 year	No recurrence	GR		
20	Lama *et al*. 2014	50, M	_	ICH, IVH	3	Conservative	10 days 6 months	Disap. Disap.	GR		LSA dissection and pseudoaneurysm on angiography, spontaneous resolution on day 10
39	Tan *et al*. 2014	81, M	HT	ICH	5.2	Clipping	NA	Cured	NA		
7	Choo *et al*. 2015	15, M	_	ICH	1.94 × 2.03	Conservative	2 weeks	Complete disap.	GR		CTA (day 0): no aneurysm, angiography (day 2): LSA aneurysm complete disappearance
		52, M	Twig-like MCA	ICH, IVH, SAH	2.16-2.27	Clipping	2 weeks	Enlargement	GR		Outpouching → 3 mm aneurysm

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Table 1: Contd...

Reference	Author, year	Age (years), sex	Associated disease	CT findings	Size of aneurysm (mm)	Treatment	Time of follow-up	Outcome of follow-up radiological examinations	Outcome	Pathology	Others
44	Yap *et al*. 2015	8, F		ICH, IVH, HCP	2	VED	15 weeks	Obliteration	MD		Complete resolution (15 weeks)
		9, M	Arnold-Chiari malformation autism spectrum disorder epilepsy	ICH, IVH, HCP	Small	VED	14 weeks	Complete resolution	NA		Complete resolution (14 weeks)
18	Kinoshita *et al*. 2016	59, F	_	IVH	3.8	VED	28 days	Disap.	GR		Spontaneous disap. (day 28)
33	Saito *et al*. 2016	66, F	HT	SAH, IVH	3	VED resection	NA	Disap.	GR	Dissecting aneurysm	
35	Sato *et al*. 2017	61, F	HT	IVH	8.0 × 9.0	Proximal clipping, resection	_	Cured	GR	Partially organized thrombus pseudoaneurysm formation	Detected on angiography (day 22)
	Present case	81, F	_	ICH, IVH	3	VED	15 days	Disap.	GR		Delayed appearance spontaneous resolution

Af: Atrial fibrillation, AVM: Arteriovenous malformation, CTA: Computed tomographic angiography, Disap: Disappeared, F: Female, FMD: Fibromuscular dysplasia, GR: Good recovery, HCP: Hydrocephalus, HL: Hyperlipidemia, HT: Hypertension, ICA: Internal carotid artery, ICH: Intracerebral hemorrhage, IVH: Intraventricular hemorrhage, LSA: Lenticulostriate artery, lt: Left, M: Male, MCA: Middle cerebral artery, MD: Moderately disabled, mRS: modified Rankin Scale, NA: Not available, NBCA: n-butyl-cyanoacrylate, rt: Right, SAH: Subarachnoid hemorrhage, SD: Severely disabled, VED: Ventriculo-external drainage, VS: Vegetative state

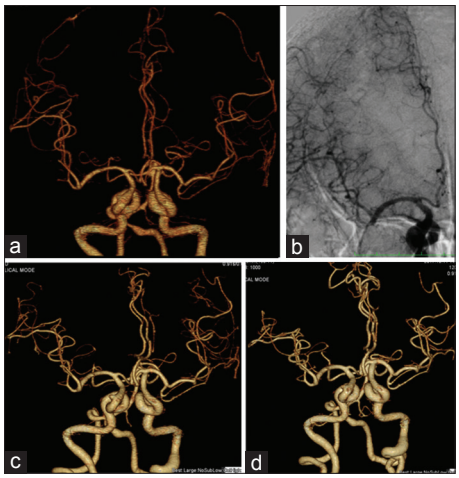

Figure 3: Three-dimensional computed tomographic angiography on the 15th (a) and angiography on the 23rd (b) day showing no aneurysm on the right lenticulostriate artery. (c) Three-dimensional computed tomographic angiography on the 42nd day also showing no lenticulostriate artery aneurysm. (d) Three-dimensional computed tomographic angiography at the 9th month showing no aneurysm on the right lenticulostriate artery

reports describing the spontaneous disappearance of the lesion. Nearly half of the reported cases showed obstruction in their natural courses. Seventeen cases showed spontaneous disappearance or near disappearance in 20 cases of lenticulostriate artery aneurysms which were not radically treated [Table 1]. In our case, the aneurysm disappeared 13 days after onset. Previous reports described that spontaneous obstruction was observed between 5 days and 2 years.[7,8,13,18,43,44] In our case, the aneurysm disappeared in a relatively early period compared with previously reported cases. The aneurysm was located at the distal portion of this thin artery, and blood flow in the artery might be weak compared with that of the main arteries. Therefore, the aneurysm might be compressed by a surrounding hematoma, resulting in thrombosis at onset. After the resolution of compression by hematoma, the aneurysm recanalized and appeared on radiological examinations or cavity mimicking aneurysm was formed in the hematoma. Subsequently, spontaneous disappearance of the aneurysm occurred due to weak blood flow in the affected artery and aneurysm.

This aneurysm may be a dissection or pseudoaneurysm rather than a saccular aneurysm on a main artery in other locations.[17,20,33] Pathological findings were reported in 8 cases [Table 1]. Among them, 3 cases were diagnosed as true aneurysm,[2,26,42] whereas 3 cases were diagnosed as pseudoaneurysm or dissection.[17,33,35] The incidence of pseudoaneurysm or dissection in this artery is higher than that of aneurysm in other locations. These characteristics might also contribute to spontaneous obstruction.[17] A pseudoaneurysm without a vascular wall might sometimes be formed in the hematoma or thick subarachnoid hemorrhage.[28-30] If the blood flow in a pseudoaneurysm is weak, it might show a delayed appearance after pseudoaneurysm formation and then spontaneous obstruction.

If the aneurysm is not obstructed, the lesion is still associated with a risk of rerupture. In such a case, radical treatment should be considered. As for radical treatment, clipping, trapping, or resection was performed in 22 cases, and endovascular embolization in 6. For 1 case, stereotactic radiosurgery was performed, and the lesion disappeared.[21] In our case, we initially planned to clip or trap the aneurysm via the lateral ventricle. As for the treatment of an aneurysm at this location, transcallosal transventricular and transcortical transventricular approaches have been reported as surgical managements.[33] Sato *et al.*[35] reported a case of growing distal medial lenticulostriate artery pseudoaneurysm detected on angiography on day 22. For this case, the lesion was resected via the trans-sulcal transventricular approach. Pathological examination revealed that the main part was fresh clots with partially organized thrombus. The lesion is deeply located, and so an approach to the aneurysm is difficult. The most suitable approach should be selected for each case. There are some reports describing endovascular embolization of the aneurysm.[15,40] However, the lenticulostriate artery is thin, and insertion and advancement of a microcatheter to the parent artery and aneurysm might be difficult. Therefore, endovascular embolization of the aneurysm at this location might be challenging.

In our case, initial radiological examination as 3D-CTA on admission failed to demonstrate the lenticulostriate artery aneurysm. Angiography might not be commonly performed for cases with simple hemorrhage in the caudate nucleus, or intraventricular hemorrhage. We performed angiography for the purpose of evaluating a coincidentally developing BA aneurysm. As a result, the lenticulostriate artery aneurysm was unexpectedly identified. There is a possibility that a distal artery aneurysm such as a lenticulostriate artery aneurysm exists in cases of hemorrhage around the lateral ventricles. In fact, raw images of 3D-CTA obtained on admission showed a small low-density area in the hematoma. It was not clear whether this low-density area represented the obstructed aneurysm. Tan *et al.*[39] reported the spot sign in a case of lenticulostriate artery aneurysm on CE-CT or CE magnetic resonance imaging. The spot sign may be an extravasation of contrast medium into the hematoma. The existence of the contrast medium outside the artery is opacified, revealing a pseudoaneurysm, on radiological examinations such as 3D-CTA and angiography.[28,30] Although hemorrhage in the caudate nucleus due to the rupture of a lenticulostriate artery aneurysm is not common, there is a possibility of the existence of

a lenticulostriate artery aneurysm. Therefore, careful radiological examinations focusing on the presence of a distal artery aneurysm is necessary for cases presenting with simple hemorrhage in a region close to the ventricles.

CONCLUSION

An aneurysm originating from the lenticulostriate artery is rare. This aneurysm may show a delayed appearance and spontaneous resolution. Therefore, serial radiological examinations are mandatory. Also, radiological examinations focusing on a lenticulostriate artery aneurysm are necessary in cases with hemorrhage around the lateral ventricles, although the incidence is low, even though the hemorrhage is considered to be simple.

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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