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Two Cases of Subarachnoid Hemorrhage with Microaneurysmal Changes and Spontaneous Disappearance in the Basilar Artery

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Abstract

A 79-year-old woman presented at our hospital with sudden headache and vomiting. Computed tomography revealed diffuse subarachnoid hemorrhage. Although digital subtraction angiography (DSA) performed on admission and on the following day revealed no vascular abnormalities, DSA on Day 22 revealed microaneurysmal changes in the dorsal basilar artery. However, the aneurysmal changes gradually became smaller during follow-up, and DSA on Day 73 revealed complete disappearance. A 53-year-old man also presented to our hospital with sudden headache and vomiting. Computed tomography revealed perimesencephalic subarachnoid hemorrhage. DSA on Days 9 and 16 revealed microaneurysmal changes in the dorsal basilar artery. Conservative treatment was continued, and DSA on Day 42 revealed spontaneous disappearance of the lesion.
It has been reported that basilar artery perforating aneurysms cause angiogram-negative subarachnoid hemorrhage, which disappears spontaneously. The fact that lesions previously reported as basilar artery perforating aneurysms may include cases of acute dissection of the main trunk or perforating branches of the basilar artery implies that surgical or endovascular treatment may worsen the condition. Therefore, conservative treatment may be an important option.

Keywords: subarachnoid hemorrhage of unknown etiology, basilar artery, basilar artery perforating aneurysm

Introduction

Although subarachnoid hemorrhage of unknown etiology (SAHUE) accounts for approximately 4.7%-20% of all subarachnoid hemorrhage (SAH) cases,[1-4] the causes and mechanisms of SAHUE remain heterogeneous and unclear. We report two cases of SAH in which initial digital subtraction angiography (DSA) revealed no vascular abnormalities; however, the spontaneous appearance and subsequent disappearance of small aneurysm-like changes in the dorsal basilar artery (BA) were observed.

Case Report

Case 1

A 79-year-old woman presented at our hospital with sudden headache and vomiting. Computed tomography (CT) of the head revealed diffuse SAH in the interpeduncular cistern and bilateral Sylvian fissures (Fig. 1a). DSA was performed, and then revealed no obvious vascular anomalies, including an aneurysm (Fig. 1b). DSA on Day 2 revealed similar findings. CT on Day 7 showed washout of the SAH; however, a small hematoma remained around the tip of the BA. DSA on Day 22 revealed a small lesion resembling a saccular aneurysm (approximately 2 mm) in the dorsal BA (Fig. 1c and d). The lesion was observed during the early arterial phase, and after pooling for some time, it completely disappeared before the venous phase. Conservative treatment was continued due to the difficulty of treatment caused by the small size of the lesion. Computed tomography angiography (CTA) on Day 35 confirmed the aneurysmal change without contrast extravasation. CTA on Day 51 showed a slight reduction in aneurysmal changes (Fig. 1e), and DSA on Day 73 revealed the

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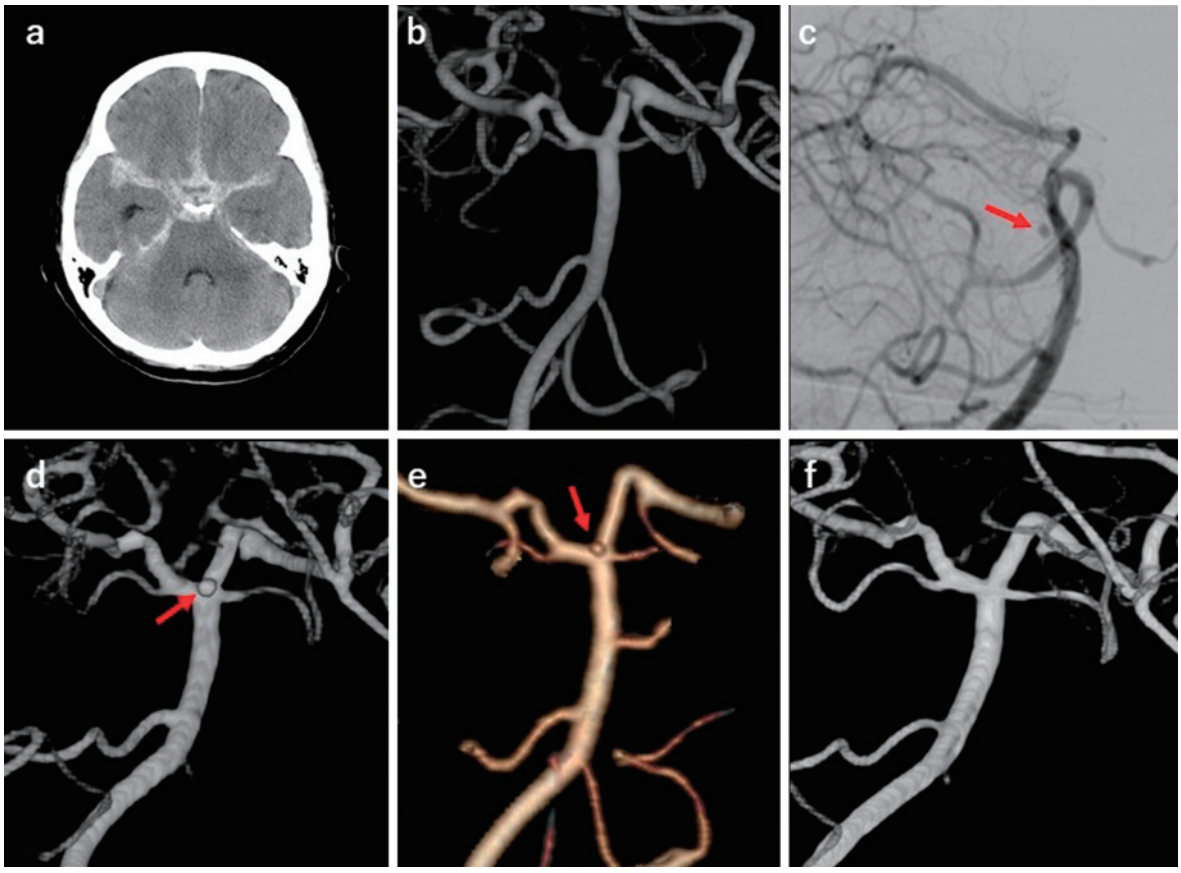

Fig. 1 Initial computed tomography image showing diffuse subarachnoid hemorrhage in the bilateral Sylvian fissure and basal cistern (a). On admission, three-dimensional rotation angiography (3D-RA) of the right vertebral artery revealed no vascular anomalies (b). Right vertebral angiography (c) and 3D-RA (d) on Day 22 revealed a microaneurysmal change (arrow) of approximately 2 mm in the dorsal basilar artery. Computed tomography angiography on Day 35 detected a microaneurysmal change (arrow) (e). However, 3D-RA on Day 73 revealed complete disappearance of the lesion (f).

complete disappearance of aneurysmal changes (Fig. 1f). The patient was discharged from the hospital with a modified Rankin Scale score of 0, and magnetic resonance imaging (MRI) showed no recurrence for 10 years.

Case 2

A 53-year-old man presented at our hospital with headache and vomiting. Head CT revealed SAH localized to the interpeduncular cistern (Fig. 2a). Although DSA on admission revealed no obvious vascular abnormalities including an aneurysm (Fig. 2b), DSA on Days 9 and 16 revealed a small lesion resembling a saccular aneurysm (approximately 1 mm) in the dorsal BA (Fig. 2c). After conservative treatment, as in Case 1, DSA on Day 42 showed complete disappearance of the aneurysmal changes (Fig. 2d). The patient was discharged from the hospital with a modified Rankin Scale score of 0, and CTA revealed no recurrence for 12 months.

Discussion

Perimesencephalic SAH (pm-SAH) accounts for approximately 5% of all SAH cases. It is estimated to account for approximately one-third of non-aneurysmal SAH and 50%-75% of angiogram-negative SAH.[4,5] A total of 84 patients were diagnosed with SAH (excluding traumatic SAH) at our hospital between January 2012 and January 2023. Five patients, including Case 1, had SAH with no identifiable bleeding source, two of whom exhibited typical pm-SAH findings. Frequent CTA, DSA, or MRI was performed in these patients (an average of 4.3 times during the first month); however, there were no vascular abnormalities that could be explained as the source of bleeding in any of the cases, except Case 1. Vessel wall imaging using contrast-enhanced MRI was not performed in Cases 1 and 2.

Rupture of an aneurysm should be suspected in pm-SAH; however, other possible causes include rupture of dilated veins, venous malformations, or subclinical arteriovenous malformation in the interpeduncular cistern.[1,6]

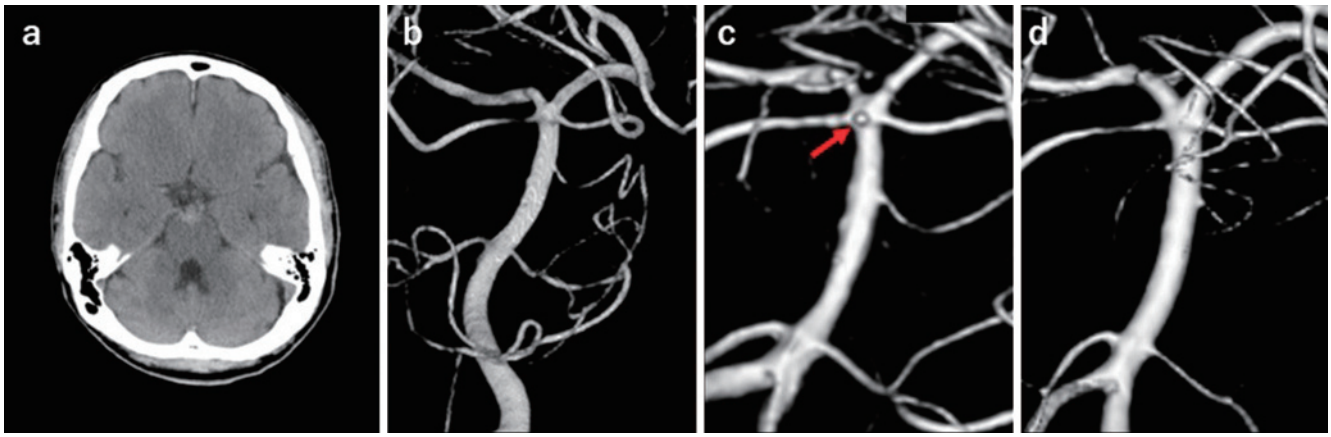

Fig. 2 Initial computed tomography showing subarachnoid hemorrhage localized in the interpeduncular cistern (a). Although three-dimensional rotation angiography (3D-RA) revealed no vascular anomaly on admission (b), 3D-RA on Day 9 revealed a small aneurysmal change (arrow) of approximately 1 mm in the dorsal basilar artery (c). 3D-RA on Day 42 revealed complete disappearance of the lesion (d).

In fact, there has been a case in which a saccular venous aneurysm was found in the transverse pontine vein near the superior cerebellar artery in pm-SAH with typical CT findings, which seemed to be the most likely source of bleeding.[6] Saccular venous aneurysm is revealed in the late arterial or early venous phase. However, our two cases showed aneurysmal changes protruding from the main trunk of the BA in the early arterial phase, which completely disappeared during the venous phase. These findings were inconsistent with the characteristics of venous aneurysm.

Several studies have reported that BA perforating aneurysms (BAPAs) cause angiogram-negative SAH and disappear spontaneously or that tiny dissection of the main trunk of the BA may be the cause of SAH. In a review of 29 conservatively treated BAPAs, including our cases (Table 1),[7-17] the rebleeding rate was 14.3% (4/28). Other studies have reported rebleeding rates ranging from 6.7% to 15%.[17-21] The rebleeding rate was considerably lower than the 20%-30% observed, in which common ruptured saccular aneurysms were conservatively treated.[21] These BAPAs were very small (mean diameter, 1.86 mm); however, many studies have reported on the spontaneous thrombosis of BAPAs, including our cases (Table 1). In general, spontaneous thrombosis of aneurysms more frequently occurs in large or giant aneurysms, with flow of blood being suggested to induce thrombosis.[22] This is contrary to the higher spontaneous disappearance rate of BAPAs compared with that of common saccular aneurysms. Pontine infarction is another reported complication of BAPA, with a frequency of 17.9% (5/28) in the conservative treatment group (Table 1). Therefore, BAPA can cause hemorrhage and infarction. Variations in the size of the aneurysm have been observed between closely repeated angiograms.[17] These features implicate dissection as the origin of BAPA.

Cerebral arteries are composed of the tunica intima, inner elastic lamina, tunica media, and tunica adventitia from the inner side. The main characteristic of dissection in SAH is a "sudden and extensive rupture of the internal elastic lamina," and the degree of rupture can result in various aneurysmal morphologies.[23] Although the ruptured inner elastic lamina does not regenerate, it is repaired over time by neointimal formation, which takes approximately 1-2 months to complete.[24] In our two cases, the aneurysms disappeared on imaging after a similar period.

BAPA is a rare disease with no established treatment. Endovascular treatment using flow diverter stents, direct surgery by proximal occlusion or trapping, and conservative treatment have been reported, with several studies supporting each method.[16-19,21,25,26] In a review of 29 conservatively treated BAPAs, 82.1% (23/28) achieved a favorable outcome (modified Rankin Scale 0-2 or Glasgow Outcome Scale 5). Other studies reported that 77%-91% of cases had a good prognosis by conservative treatment,[17,18,20] whereas only 66% of the surgical group and 60%-90% of the endovascular treatment group had a good prognosis.[17,18,20] Therefore, it cannot be asserted that surgical treatment leads to a significantly better prognosis than conservative treatment.[20] Considering that BAPA is a dissecting aneurysm, clipping or coil embolization has been associated with a high risk of brainstem ischemia. The rebleeding rate is lower in BAPAs than in commonly ruptured saccular aneurysms. Conservative treatment is an important option because 52.2%-70% of BAPAs spontaneously disappear, with the prognosis being favorable in more than 80% of cases.[17,18,20] Forbrig et al. estimated that conservative treatment might be the first-line treatment considering the perioperative risks of endovascular and microsurgical treatment.[10] Finitsis et al. proposed endovascular treatment in the event of increasing size, rebleeding, or lack of regression.[13]

In a review of 29 cases, only 29.6% (8/27) of the aneu-

Table 1 Patients (n = 29) diagnosed with basilar artery perforating aneurysm and treated conservatively

Patient number	Author (year)	Age	Sex	WFNS/ H&H Grade	Fisher grade	Bleed pattern	Aneurysm size (mm)	Time until aneurysm detection	Treatment	Complications	Time until rebleeding	Follow-up duration	Time until aneurysm disappearance	GOS/ mRS score
1	Park et al. (2009)[7]	54	F	WFNS I	2	pm	1	Initial angiogram	Conservative	Vasospasm		16 months	16 months	GOS 5
2	Park et al. (2009)[7]	67	M	WFNS I	2	pm	1	Initial angiogram	Conservative	None		15 months	16 months	GOS 5
3	Park et al. (2009)[7]	53	F	H&H 1	2	pm	1	Initial angiogram	Conservative	None		1 month	1 month	GOS 5
4	Ding et al. (2013)[8]	55	N/A	H&H 2	3	Diffuse	1.8	7 days	Conservative	None		19 months	6 months	GOS 5
5	Chavent et al. (2014)[9]	55	M	WFNS I	3	Diffuse	1.7	8 days	Conservative	None		6 months	3 months	mRS 0
6	Chavent et al. (2014)[9]	39	F	WFNS I	2	pm	1.5	8 days	Conservative	None		12 months	3 months	mRS 0
7	Chavent et al. (2014)[9]	56	M	WFNS I	3	Diffuse	1	8 days	Conservative	None		12 months	3 months	mRS 0
8	Forbrig et al. (2016)[10]	71	F	WFNS V	4	Diffuse	7	Initial angiogram	Conservative (failed End)	Pontine infaction, hydrocephalus		11 months	7 days	mRS 1
9	Forbrig et al. (2016)[10]	65	M	WFNS I	4	Diffuse	1	8 days	Conservative	Pontine infaction, hydrocephalus		15 months	N/A	mRS 1
10	Forbrig et al. (2016)[10]	82	M	WFNS V	4	Diffuse	2	Initial angiogram	Conservative (failed End)	Pontine infaction, rebleeding	20 days	6 months	N/A	mRS 5
11	Forbrig et al. (2016)[10]	59	M	WFNS II	3	pp	2.5	13 days	Conservative, End (coiling)	Rebleeding	13 days	23 months	Immediate (after coiling)	mRS 2
12	Forbrig et al. (2016)[10]	60	F	WFNS I	3	Diffuse	2.5	Initial angiogram	Conservative (failed End)	Pontine infaction, vasospasm		78 months	2 months	mRS 0
13	Forbrig et al. (2016)[10]	53	M	WFNS I	3	Diffuse	1	47 days	Conservative	None		6 months	3 months	mRS 0
14	Aboukais et al. (2016)[11]	67	M	WFNS I	2	pm	3	6 days	Conservative	None		1.5 months	6 weeks	mRS 0
15	Daruwalla et al. (2016)[12]	76	M	H&H 4	4	pm	2.5	Initial angiogram	Conservative	Acute hydrocephalus		N/A	4 days	mRS 6
16	Finitsis et al. (2017)[13]	59	M	WFNS I	3	Diffuse	0.5	9 days	Conservative	None		2 months	1.5 months	mRS 0
17	Finitsis et al. (2017)[13]	62	F	WFNS II	4	Diffuse	1	4 days	Conservative, End (FD)	Rebleeding	10 days	3 months	3 months	mRS 0
18	Finitsis et al. (2017)[13]	78	M	WFNS IV	4	Diffuse	3	16 days	Conservative	Pontine infarction		14 months	N/A	mRS 5

Table 1 Patients (n = 29) diagnosed with basilar artery perforating aneurysm and treated conservatively (continued)

Patient number	Author (year)	Age	Sex	WFNS/ H&H Grade	Fisher grade	Bleed pattern	Aneurysm size (mm)	Time until aneurysm detection	Treatment	Complications	Time until rebleeding	Follow-up duration	Time until aneurysm disappearance	GOS/ mRS score
19	Finitsis et al. (2017)[13]	53	F	WFNS II	3	N/A	1.2	7 days	Conservative	None		2 months	1.5 months	mRS 0
20	Buell et al. (2018)[14]	N/A	N/A	H&H 2	N/A	N/A	1	7 days	Conservative	None		2 months	N/A	mRS 1
21	Buell et al. (2018)[14]	N/A	N/A	H&H 3	4	Diffuse	2	5 days	Conservative	Acute hydrocephalus		42 months	7 days	mRS 1
22	Buell et al. (2018)[14]	N/A	N/A	H&H 3	N/A	N/A	1.7	5 days	Conservative	None		62 months	N/A	mRS 1
23	Chau et al. (2018)[15]	69	M	WFNS IV	4	Diffuse	2.5	2 months	Conservative	Acute hydrocephalus		12 months	12 months	mRS 0
24	Bhogal et al. (2019)[16]	59	M	WFNS I	N/A	N/A	1	Initial angiogram	Conservative	None		3 months	N/A	mRS 0
25	Bhogal et al. (2019)[16]	62	M	WFNS V	N/A	N/A	1.4	N/A	Conservative	Rebleeding	N/A	N/A	N/A	mRS 6
26	Bhogal et al. (2019)[16]	57	M	WFNS I	N/A	N/A	1.2	N/A	Conservative	N/A		N/A	N/A	N/A
27	Enomoto et al. (2020)[17]	60	M	WFNS II	3	Diffuse	3	39 days	Conservative	Acute hydrocephalus, vasospasm		19 months	9 weeks	mRS 5
28	Present Case 1	79	F	WFNS II	3	Diffuse	2	22 days	Conservative	None		120 months	10 weeks	mRS 0
29	Present Case 2	53	M	WFNS I	1	pp	2	9 days	Conservative	None		12 months	6 weeks	mRS 0

End, endovascular treatment; F, female; FD, flow diverter; GOS, Glasgow Outcome Scale; H&H, Hunt and Hess; M, male; mRS, modified Rankin Scale; N/A, not available; pm, perimesencephalic; pp, prepontine; WFNS, World Federation of Neurosurgical Societies

rysms were detected on initial DSA (Table 1), whereas the remaining were detected on average 15.6 days after onset. In other studies, rebleeding occurred 10-20 days after onset.[17,18] Although few studies have reported rebleeding and the factors that predispose patients to rebleeding remain unknown, strict blood pressure control is probably important. Moreover, in many cases, the bleeding pattern might be diffuse SAH and not only perimesencephalic or prepontine. Therefore, DSA should be repeated for at least 2-3 weeks, even if no aneurysm is detected on the initial DSA.

Conclusion

We encountered two cases of dorsal BA aneurysms that were initially treated conservatively as SAHUE and then spontaneously disappeared during follow-up as revealed by DSA. Due to improvements in imaging technology, the number of cases with findings suggestive of microaneurysms or dissections, similar to the present cases, may increase. Careful attention is necessary to prevent even slight changes in the vascular structure of the BA from being missed.

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Informed Consent

Written informed consent was obtained from the patients.

Conflicts of Interest Disclosure

The authors declare no conflicts of interests.

References

1) Inoue A, Sato S, Sekiguchi K, Taniguchi Y, Watanabe T: Subarachnoid hemorrhage of unknown etiology. Operative findings of unclear aneurysms in initial angiography. *Jpn J Stroke* 13: 265-273, 1991
2) Kawano T, Hirata K, Mori K: Long-term follow-up study for subarachnoid hemorrhage of unknown etiology. *Jpn J Stroke* 14: 482-486, 1992
3) Coelho LG, Costa JM, Silva EI: Non-aneurysmal spontaneous subarachnoid hemorrhage: perimesencephalic versus non-perimesencephalic. *Rev Bras Ter Intensiva* 28: 141-146, 2016
4) Flaherty ML, Haverbusch M, Kissela B, et al.: Perimesencephalic subarachnoid hemorrhage: incidence, risk factors, and outcome. *J Stroke Cerebrovasc Dis* 14: 267-271, 2005
5) Goddard AJP, Tan G, Becker J: Computed tomography angiography for the detection and characterization of intra-cranial aneurysms: current status. *Clin Radiol* 60: 1221-1236, 2005
6) Hafez A, Numminen J, Rahul R, Järveläinen J, Niemelä M: Perimesencephalic subarachnoid hemorrhage with a positive angiographic finding: case report and review of the literature. *Acta Neurochir (Wien)* 158: 1045-1049, 2016
7) Park SQ, Kwon OK, Kim SH, Oh CW, Han MH: Premesencephalic subarachnoid hemorrhage: rupture of tiny aneurysms of the basilar artery perforator. *Acta Neurochir (Wien)* 151: 1639-1646, 2009
8) Ding D, Starke RM, Jensen ME, Evans AJ, Kassell NF, Liu KC: Perforator aneurysms of the posterior circulation: case series and review of the literature. *J Neurointerv Surg* 5: 546-551, 2013
9) Chavent A, Lefevre PH, Thouant P, et al.: Spontaneous resolution of perforator aneurysms of the posterior circulation. *J Neurosurg* 121: 1107-1111, 2014
10) Forbrig R, Eckert B, Ertl L, et al.: Ruptured basilar artery perforator aneurysms--treatment regimen and long-term follow-up in eight cases. *Neuroradiology* 58: 285-291, 2016
11) Aboukais R, Zairi F, Estrade L, Quidet M, Leclerc X, Lejeune JP: A dissecting aneurysm of a basilar perforating artery. *Neurochirurgie* 62: 263-265, 2016
12) Daruwalla VJ, Syed FH, Elmokadem AH, Hurley MC, Shaibani A, Ansari SA: Large basilar perforator pseudoaneurysm: a case report. *Interv Neuroradiol* 22: 662-665, 2016
13) Finitsis S, Derelle AL, Tonnelet R, Anxionnat R, Bracard S: Basilar perforator aneurysms: presentation of 4 cases and review of the literature. *World Neurosurg* 97: 366-373, 2017
14) Buell TJ, Ding D, Raper DM, et al.: Posterior circulation perforator aneurysms: a proposed management algorithm. *J Neurointerv Surg* 10: 55-59, 2018
15) Chau Y, Sachet M, Sédat J: Should we treat aneurysms in perforator arteries from the basilar trunk? Review of 49 cases published in the literature and presentation of three personal cases. *Interv Neuroradiol* 24: 22-28, 2018
16) Bhogal P, AlMatter M, Hellstern V, et al.: Basilar artery perforator aneurysms: Report of 9 cases and review of the literature. *J Clin Neurosci* 63: 122-129, 2019
17) Enomoto N, Shinno K, Tamura T, Shikata E, Shono K, Takase K: Ruptured basilar artery perforator aneurysm: A case report and review of the literature. *NMC Case Rep J* 7: 93-100, 2020
18) Kumagawa T, Otani N, Kakei Y, et al.: Ruptured basilar artery perforator aneurysm definitely diagnosed with intraoperative microsurgical findings: case report and literature review. *NMC Case Rep J* 10: 1-7, 2023
19) Sattur MG, Gunasekaran A, Spiotta AM, Lena JR: Basilar artery perforator aneurysms and their contemporary management. *Neurol India* 68: 1301-1306, 2020
20) Elsheikh S, Möhlenbruch M, Seker F, et al.: Flow diverter treatment of ruptured basilar artery perforator aneurysms : A multicenter experience. *Clin Neuroradiol* 32: 783-789, 2022
21) Mayberg MR, Batjer HH, Dacey R, et al.: Guidelines for the management of aneurysmal subarachnoid hemorrhage. A statement for healthcare professionals from a special writing group of the Stroke Council, American Heart Association. *Circulation* 90: 2592-2605, 1994
22) Akimoto Y, Yanaka K, Onuma K, Nakamura K, Takahashi N, Ishikawa E: Spontaneous disappearance of an intracranial small unruptured aneurysm on magnetic resonance angiography: report of two cases. *Asian J Neurosurg* 15: 1055-1058, 2020
23) Mizutani T, Kojima H, Miki Y: Arterial dissections of penetrating cerebral arteries causing hypertension-induced cerebral hemorrhage. *J Neurosurg* 93: 859-862, 2000
24) Mizutani T, Kojima H, Asamoto S: Healing process for cerebral dissecting aneurysms presenting with subarachnoid hemorrhage. *Neurosurgery* 54: 342-347; discussion 347, 2004

25) Gross BA, Puri AS, Du R: Basilar trunk perforator artery aneurysms. Case report and literature review. *Neurosurg Rev* 36: 163-168; discussion 168, 2013
26) Shlobin NA, Cantrell DR, Ansari SA, et al.: Conservative management and natural history of ruptured basilar perforator artery aneurysms: two cases and literature review. *World Neurosurg* 138: 218-222, 2020

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