

Ruptured Basilar Artery Perforator Aneurysm: A Case Report and Review of the Literature

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Basilar artery perforator aneurysms (BAPAs) are a rare cause of subarachnoid hemorrhage (SAH), and the natural history is still unknown. Herein, we report a case of ruptured BAPA that appeared during the observation period and then spontaneously disappeared; we have also conducted a review of the literature and performed an analysis based on the type of management. This case of BAPA had a unique course, and our observations may help establish a treatment strategy. A 60-year-old man presented with acute diffuse SAH, World Federation of Neurosurgical Societies (WFNS) Grade II and Fisher Grade 3. Initial three-dimensional digital subtraction angiography (DSA) did not show the source of the hemorrhage. DSA performed on day 39 showed a BAPA with a diameter of 3 mm at the posterior surface of the upper third of the basilar artery. Conservative treatment was chosen. DSA performed on day 64 showed complete resolution of the aneurysm. *BAPAs are likely pseudoaneurysms, and not saccular aneurysms, caused due to dissection of basilar perforator arteries.* BAPAs are often not recognized on initial imaging, and hence, it is necessary to repeat the DSA examination. Considering the relatively high rate of spontaneous resolution, we chose conservative management. When BAPAs enlarge or do not disappear after conservative treatment, additional therapy such as multiple stents should be considered.

Keywords: basilar artery perforator aneurysm, subarachnoid hemorrhage

Introduction

Basilar artery perforator aneurysms (BAPAs), first described by Ghogawala et al.¹⁾ in 1996, are a rare cause of subarachnoid hemorrhage (SAH). BAPA is an aneurysm with the neck located entirely on a perforating artery without directly involving the basilar trunk.²⁾ Although the number of cases being reported in recent times is increasing, the natural history is still unknown. We report a specific clinical case in which the source of bleeding was not identified at the onset of SAH; a BAPA appeared during the observation period and then spontaneously disappeared. This case of BAPA had a unique clinical course, and our observations may help establish a treatment

strategy. We have also conducted a review of the literature and performed an analysis based on the type of management.

Case Report

A 60-year-old man with a history of hypertension and type 2 diabetes mellitus was admitted to the hospital for sudden headache in the occipital area. He was drowsy on admission, but there were no other neurologic deficits (World Federation of Neurosurgical Societies [WFNS] Grade II). A head computed tomography (CT) scan showed diffuse SAH mainly in the posterior fossa with hydrocephalus (Fisher Grade 3) (Fig. 1). Computed tomographic angiography (CTA) and three-dimensional digital subtraction angiography (DSA) did not show the source of bleeding (Fig. 2A). We only performed ventricular drainage at first, and decided to re-examine the bleeding source later. A second CTA performed on day 9 also did not show the source of bleeding. Cerebral infarction due to vasospasm occurred in the bilateral frontal lobes on day 9, resulting in disturbance in consciousness and quadriplegia. A ventriculoperitoneal (VP) shunt was performed for hydrocephalus on day 31. DSA performed on day 39 showed an aneurysm with a diameter of 3 mm at the posterior surface of the upper third of the basilar artery. The blood flow into the aneurysm was very slow; therefore, it was observed in the late arterial phase. Although the aneurysm was close to the basilar artery, the artery was not directly involved (Figs. 2B and 2C). We diagnosed this aneurysm as BAPA and chose conservative management. DSA performed on day 64 showed complete resolution of the aneurysm (Fig. 2D). Although the patient was able to talk and was conscious, quadriplegia was still present (modified Rankin Scale [mRS] score = 5), and he was transferred to a rehabilitation hospital.

Discussion

Although BAPAs are a rare cause of SAH, increasing number of cases are being reported due to the evolution in imaging technology. A review of 36 cases concluded that BAPAs are very small with a diameter ranging from 0.5 to 7 mm (mean 2.5 mm).³⁾ BAPAs are thought to be of dissecting origin for two reasons: (1) the shape of the aneurysm is often described as dolichoectatic (fusiform), or with a large base and (2) variation is observed in the size of the aneurysm between closely repeated angiograms.⁴⁾ The blood flow into the aneurysm is quite slow, which was also observed in our case; further, intraluminal thrombus was found in a previous report.⁵⁾ A hypertension-induced

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Table 1 Characteristics of previously reported basilar artery perforator aneurysms managed with conservative treatment

Study	Age(years)/Sex	WFNS/H&H Grade	Fisher Grade	Bleed pattern	Detection on initial angiogram	Time until aneurysm detection	Size(mm)	Origin of the perforator on the basilar artery
Park et al. (2009) ⁹⁾	54/F	WFNS I	2	pm	Yes		1	Distal 1/3
Park et al. (2009) ⁹⁾	67/M	WFNS I	2	pm	Yes		1	Distal 1/3
Park et al. (2009) ⁹⁾	53/M	H&H 1	2	pm	Yes		1	Distal 1/3
Ding et al. (2013) ¹⁰⁾	55/NA	H&H 2	3	Diffuse	No	1 week	1.8	Distal 1/3
Chavent et al. (2014) ⁴⁾	55/M	WFNS I	3	Diffuse	No	8 days	1.7	Distal 1/3
Chavent et al. (2014) ⁴⁾	39/F	WFNS I	2	pm	No	8 days	1.5	Distal 1/3
Chavent et al. (2014) ⁴⁾	56/M	WFNS I	3	Diffuse	No	8 days	1	Distal 1/3
Forbrig et al. (2016) ¹¹⁾	71/F	WFNS V	4	Diffuse	Yes		7	Middle 1/3
Forbrig et al. (2016) ¹¹⁾	65/M	WFNS I	4	Diffuse	No	8 days	1	Middle 1/3
Forbrig et al. (2016) ¹¹⁾	82/M	WFNS V	4	Diffuse	Yes		2	Distal 1/3
Forbrig et al. (2016) ¹¹⁾	60/F	WFNS I	3	Diffuse	Yes		2.5	Middle 1/3
Forbrig et al. (2016) ¹¹⁾	53/M	WFNS I	3	Diffuse	No	47 days	1	Distal 1/3
Aboukais et al. (2016) ²⁾	67/M	WFNS I	2	pm	No	6 days	3	Middle 1/3
Daruwalla et al. (2016) ¹²⁾	76/M	H&H 4	4	pm	Yes		2.5	Middle 1/3
Finitsis et al. (2017) ³⁾	59/M	WFNS I	3	Diffuse	No	9 days	0.5	Distal 1/3
Finitsis et al. (2017) ³⁾	62/F	WFNS II	4	Diffuse	No	4 days	1	Middle 1/3
Finitsis et al. (2017) ³⁾	78/M	WFNS IV	4	Diffuse	Yes	16 days	3	Distal 1/3
Finitsis et al. (2017) ³⁾	53/F	WFNS II	3	NA	No	7 days	1.2	Middle 1/3
Buell et al. (2018) ⁸⁾	NA/NA	H&H 2	NA	NA	No	7 days	1	Distal 1/3
Buell et al. (2018) ⁸⁾	NA/NA	H&H 3	4	Diffuse	No	5 days	2	Distal 1/3
Buell et al. (2018) ⁸⁾	NA/NA	H&H 3	NA	NA	No	5 days	1.7	Middle 1/3
Chau et al. (2018) ¹³⁾	69/M	WFNS IV	4	Diffuse	No	2 months	2.5	Distal 1/3
Current study	60/M	WFNS II	3	Diffuse	No	39 days	3	Distal 1/3

End: endovascular, EVD: extraventricular drainage, F: female, FD, flow diverter, FU: follow-up, GOS: Glasgow Outcome Scale, H&H: Hunt and Hess, M: male, mRS: modified Rankin Scale, NA: not available, pm: perimesencephalic, SAH: subarachnoid hemorrhage, VP: ventriculoperitoneal, WFNS: World Federation of Neurosurgical Societies.

Location of aneurysm from the origin of the perforator artery	Treatment	Pontine stroke	Complications	FU(months)	Time until aneurysm disappearance	GOS/mRS Score
Proximal	Conservative	No	Third nerve palsy, hemiparesis, vasospasm	16	16 months	GOS 5
Proximal	Conservative	No	None	15	16 months	GOS 5
Proximal	Conservative	No	None	1	1 month	GOS 5
Proximal	Conservative	No	None	19	6 months	GOS 5
Proximal	Conservative	No	None	6	3 months	mRS 0
Proximal	Conservative	No	None	12	3 months	mRS 0
Proximal	Conservative	No	None	12	3 months	mRS 0
Proximal	Conservative(failed end)	Yes	Mild hemiparesis, hydrocephalus, VP shunt	11	7 days	mRS 1
Proximal	Conservative	Yes	Mild hemiparesis, hydrocephalus, VP shunt	15	Unknown	mRS 1
Proximal	Conservative(failed end)	Yes	Rebleeding 20 days after SAH, severe hemiparesis, dysarthria	6	Unknown	mRS 5
Proximal	conservative(failed End)	Yes	Vasospasm	78	2 months	mRS 0
Proximal	Conservative	No	None	6	3 months	mRS 0
Proximal	Conservative	No	None	1.5	6 weeks	mRS 0
Proximal	Conservative	No	Died on day 16 because of poor general condition, acute hydrocephalus, EVD	NA	4 days	mRS 6
Proximal	Conservative	No	None	2	6 weeks	mRS 0
Proximal	Conservative/end (FD)	No	Rebleeding day 10 post-SAH, ptosis, hemiparesis, hypoacusis	3	3 months	mRS 0
Proximal	Conservative	Yes	Quadripareisis	14	unknown	mRS 5
Proximal	Conservative	No	None	2	6 weeks	mRS 0
NA	Conservative	No	None	2	NA	mRS 1
Proximal	Conservative	No	Acute hydrocephalus, EVD, VP shunt	42	1 week	mRS 1
NA	Conservative	No	None	62	NA	mRS 0
Proximal	Conservative	No	Acute hydrocephalus, EVD	12	12 months	mRS 0
Proximal	Conservative	No	Acute hydrocephalus, EVD, VP shunt, quadripareisis from vasospasm	19	9 weeks	mRS 5

Table 2 Characteristics of previously reported basilar artery perforator aneurysms managed with endovascular treatment

Study	Age(years)/sex	WFNS/H&H Grade	Fisher Grade	Bleed pattern	Detection on initial angiogram	Time until aneurysm detection	Size(mm)	Origin of the perforator on the basilar artery
Chen et al. (2012) ¹⁴	66/M	H&H 3	3	pp	yes		7	Middle 1/3
Nyberg et al. (2013) ¹⁵	45/M	NA	NA	pm	no	2 months	NA	Middle 1/3
Nyberg et al. (2013) ¹⁵	65/F	NA	NA	pm	no	9 weeks	2	Middle 1/3
Ding et al. (2013) ¹⁰	58/NA	H&H 2	3	Diffuse	yes		2	Middle 1/3
Ding et al. (2013) ¹⁰	62/NA	H&H 3	4	Diffuse	no	2 weeks	1.9	Distal 1/3
Chalouhi. (2014) ¹⁶	NA/F	WFNS I	4	Diffuse	no	3 days	1.5	Middle 1/3
Peschillo et al. (2016) ¹⁷	NA/M	WFNS II	4	pm	yes		1.5	Distal 1/3
Peschillo et al. (2016) ¹⁷	NA/M	WFNS II	4	Diffuse	no	NA	1.2	Distal 1/3
Peschillo et al. (2016) ¹⁷	NA/NA	WFNS IV	4	pm	no	NA	1.5	Distal 1/3
Forbrig et al. (2016) ¹¹	72/M	WFNS II	4	Diffuse	no	18 days	2	Distal 1/3
Forbrig et al. (2016) ¹¹	59/M	WFNS II	3	pp	no	13 day	2.5	Distal 1/3
Finitsis et al. (2017) ³	62/F	WFNS II	4	Diffuse	no	4 days	1	Middle 1/3
Satti et al. (2017) ¹⁸	52/M	H&H III	4	Diffuse	no	8 days	1.8	Middle 1/3
Buell et al. (2018) ⁸	NA/NA	H&H 3	NA	NA	no	6 days	1.5	Middle 1/3
Buell et al. (2018) ⁸	NA/NA	H&H 1	3	Diffuse	no	5 days	2.5	Middle 1/3
Buell et al. (2018) ⁸	NA/NA	H&H 4	NA	NA	yes		2	Distal 1/3
Chau et al. (2018) ¹³	53/NA	WFNS I	4	Diffuse	yes		1.8	Distal 1/3
Chau et al. (2018) ¹³	59/NA	WFNS I	3	Diffuse	No	5 days	1.5	Distal 1/3

End: endovascular, EVD: extraventricular drainage, F: female, FD: flow diverter, FU: follow-up, GOS: Glasgow Outcome Scale, H&H: Hunt and Hess, ICH: intracranial hemorrhage, M: male, mRS: modified Rankin Scale, NA: not available, pm: perimesencephalic, pp: prepontine, SAH: subarachnoid hemorrhage, VA: ventriculoatrial, VP: ventriculoperitoneal, WFNS: World Federation of Neurosurgical Societies.

Location of the aneurysm from the origin of perforator artery	Treatment	Pontine stroke	Complications	FU(months)	Time until aneurysm disappearance	GOS/mRS Score
Distal	End (coiling)	No	None	24	Immediate	GOS 4
Proximal	End (stents)	No	None	14	2 months	GOS 5
Proximal	End (stents)	No	None	4	1 month	GOS 5
Proximal	End (failed)	Yes	Quadriplegia, facial nerve palsy, dysarthria	NA	Unknown	GOS 3
Proximal	End (Onyx)	Yes	Hemiparesis, dysarthria, acute hydrocephalus, EVD, VP shunt	22	Immediate	GOS 3
Proximal	End (FD)	No	None	6	2 weeks	mRS 0
Proximal	End (FD)	Yes	In-stent thrombosis reversed with abciximab, monoparesis	6	13 days	mRS 2
Proximal	End (stent + FD)	No	In-stent thrombosis reversed with tirofiban	34	1 day	mRS 0
NA	End (FD)	No	Vasospasm, acute hydrocephalus, ICH post-EVD meningitis	6	1 day	mRS 2
Proximal	End (failed)	No	Mild cognitive impairment, gait disorder, vasospasm, VP shunt	5	Immediate	mRS 2
Proximal	End (coiling)	Yes	Rebleeding 13 days after SAH, hemiparesis, VA shunt	23	Immediate	mRS 2
Proximal	Conservative/End (FD)	No	Rebleeding day 10 post-SAH, ptosis, hemiparesis, hypoacusis	3	3 months	mRS 0
Proximal	End (stents/stent)	Yes	Acute hydrocephalus, EVD, vasospasm, growth of aneurysm, hemiparesis, abducens nerve palsy, dissection	7	Immediate	mRS 0
NA	End (stents)	No	None	11	NA	mRS 1
Proximal	End (stents)	No	None	12	4 months	mRS 1
NA	End (failed)	NA	Rupture of perforator artery during microguidewire manipulation	NA	NA	mRS 6
Proximal	End (stents/coiling)	Yes	Vasospasm, recurrence of aneurysm	6	6 months	mRS 0
NA	End (stents)	No	None	6	6 months	mRS 0

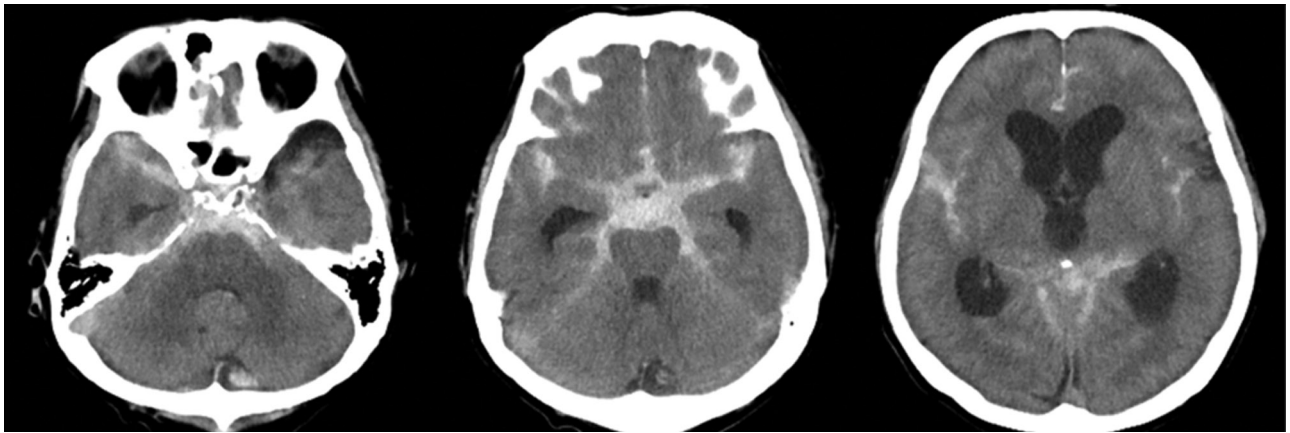


Fig. 1 Non-contrast CT of the head demonstrated ventriculomegaly and diffuse subarachnoid hemorrhage predominantly in the perimesencephalic cistern (Fisher Grade 3). CT: computed tomography.

dissecting aneurysm has been reported in a lenticulostriate artery, and the disrupted internal elastic lamina caused by hemodynamic stress has been confirmed histologically.⁶⁾ Although we did not find any histological report of BAPA, hypertension may play a role in BAPA formation. In our case, the aneurysm was not visualized on the initial DSA, but appeared on day 39, and disappeared on day 61. The shape of the aneurysm had changed, and the aneurysm was separated from the basilar artery. The aneurysm may not have been a saccular aneurysm but a pseudoaneurysm caused due to dissection of a basilar perforator artery. The aneurysm that was observed in the late arterial phase seemed to have a high possibility of causing occlusion by thrombus, and conservative treatment would be appropriate. Basilar perforator arteries are divided into 3 groups: proximal, middle, and distal. All BAPAs are associated with the middle or distal perforators. Moreover, in contrast to proximal perforating artery aneurysms, distal perforating artery aneurysms are extremely rare.^{3,5)} The average diameter of the middle and distal perforators is thicker than that of proximal perforators. The branch angle from the basilar artery is upward to horizontal in the middle and distal perforators, but in the proximal perforators the angle is downward.⁷⁾ These may affect hemodynamic stress and define the site of BAPAs.

BAPAs are often not recognized on the initial imaging due to the elevation of intracranial pressure caused by bleeding. In a single-center study, it was observed that only 22% of ruptured aneurysms were visualized on the initial imaging.⁸⁾ In 45% of cases, the hemorrhage was located in the perimesencephalic or prepontine region.⁴⁾ In cases of SAH around the brainstem with no aneurysm on the initial imaging, it is necessary to repeat the DSA, keeping the possibility of BAPAs in mind. In SAH due to ruptured BAPAs, rebleeding or vasospasm has been reported, and the clinical course is clearly different from that of benign perimesencephalic SAH; distinguishing between the two is important especially in mild SAH.

We performed an analysis of previous reports and our own case based on the type of treatment provided; 23 patients who received conservative management (Table 1)^{2-4,8-13)} and

18 who underwent endovascular treatment (Table 2)^{3,8,10,11,13-18)} were analyzed. Three patients were excluded because they were described in two different articles.^{8,10)} Although there have been cases wherein direct surgery was performed, many perioperative complications have been reported.^{1,5,19-21)} As BAPAs are often located on the anterior surface of the brainstem, performing direct surgery is very difficult. Furthermore, the fact that BAPAs are perforator pseudoaneurysms may be one of the reasons for numerous complications in direct surgery.

In the conservative group, the aneurysm was occluded spontaneously without complications in 12 patients (52.2%). Two patients (8.7%) experienced rebleeding, five (21.7%) presented with a pontine stroke, three (13.0%) had symptomatic vasospasm, and four (17.4%) underwent VP shunt for hydrocephalus. The outcomes of 19 patients (82.6%) were excellent, with mRS scores of 0–1 (Glasgow Outcome Scale [GOS] score 5). The outcomes of 4 patients (17.4%) were poor, with mRS scores of 5–6; the poor prognostic factors in these patients were severe brain stem infarction in two cases, infarction due to vasospasm in one, and poor general condition in one. When conservative therapy was chosen for the usual ruptured saccular aneurysms, the rebleeding rate was 20%–30% within a month.²²⁾ Moreover, in ruptured dissecting aneurysms of the vertebrobasilar trunk, the possibility of rebleeding in the acute phase was much higher.²³⁾ On the other hand, BAPAs follow a more benign clinical course compared to these.

Endovascular treatment by coil or onyx was attempted in six cases and accomplished in three cases. In the failed cases, perioperative complications occurred and became unfavorable prognostic factors. Although aneurysms immediately disappeared in all three cases where endovascular treatment was accomplished, brain stem infarction developed in two (66.7%). Anastomoses among various perforating arteries of the basilar artery were found in 41.6%–66.6% of cases.⁷⁾ Considering that BAPA is a dissecting aneurysm in the proximal part of the perforator arteries, treatments such as clipping or coil embolization that occlude the BAPA may pose a risk for brainstem ischemia.

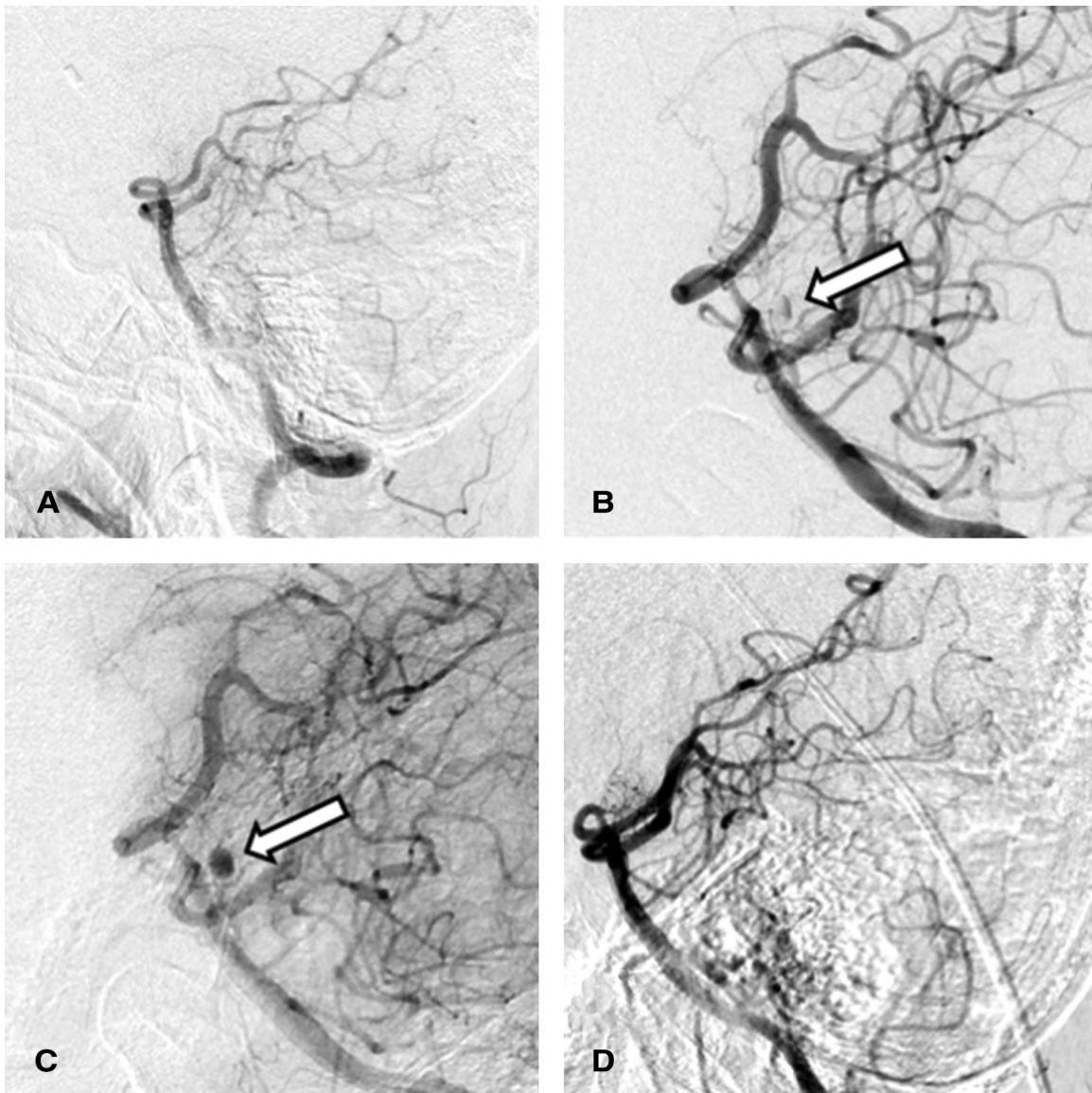


Fig. 2 (A) Initial DSA was negative for the source of hemorrhage. (B) DSA performed on day 39 showed a 3 mm aneurysm (arrow) at the posterior surface of the upper third of the basilar artery. (C) The aneurysm (arrow) was observed in the late arterial phase of the same DSA, as shown in (B), without directly involving the basilar trunk. (D) DSA performed on day 64 showed complete resolution of the aneurysm. DSA: digital subtraction angiography.

Deployment of multiple stents or flow diverter (FD) in the basilar artery is a treatment that reduces blood flow into the BAPAs and promotes thrombosis while preserving the blood flow of the perforating arteries. In the Stents/FD group, the aneurysms disappeared without complications in seven patients (58.3%). In one case, the FD was placed after rebleeding during conservative treatment and there were no complications after FD deployment.³⁾ Two cases (16.7%) required additional treatment due to recurrence or growth of the aneurysm.^{13,18)} Other complications were intracranial hemorrhage other than rebleeding in one patient (8.3%), pontine infarction in three (25.0%), and symptomatic vasospasm in three (25.0%). No patient experienced rebleeding or underwent VP shunt placement. Ten patients (83.3%) had

a mRS score of 0–1 (GOS score 5). Two patients (16.7%) had a mRS score of 2; the unfavorable prognostic factors in these patients were brainstem infarction due to in-stent thrombosis and intracranial hemorrhage around the ventricular drain. Thus, when comparing the conservative group and the Stents/FD group, the ratio of disappearance of the aneurysm without complications was 12/23 (52.2%) and 7/12 (58.3%), respectively. The ratio of good prognosis (mRS score 0–1 [GOS score 5]) in the two groups was 19/23 (82.6%) and 10/12 (83.3%), respectively. Therefore, conservative treatment for BAPAs is acceptable, and additional therapy such as multiple stents or FD should be considered when BAPAs increase in size or do not disappear after conservative treatment of the aneurysms. The possible

interventional treatment may be desirable only for a mild grade case in young individuals because of a small risk of rebleeding in the conservative group. For the endovascular treatment to be considered as first-line therapy, it seems necessary to administer perioperative antithrombotic therapy for safety.

In our analysis, no aneurysm was detected on the initial angiogram in 27 cases. The median time until aneurysm detection was 8 days (interquartile range, 6–14). Therefore, DSA should be repeated within 8 days when the initial DSA is negative. In the conservative group, the rebleeding occurred 6 days and 20 days after the detection of the aneurysm. Although the number of cases used to conclude the above indication is limited, it seems necessary to repeat the DSA within a week after the detection.

Conclusion

SAH caused by ruptured BAPA is rare, and there may be many cases where BAPAs are not recognized in the acute phase. BAPAs may spontaneously disappear by conservative treatment. When BAPAs enlarge or do not disappear, additional therapy such as multiple stents or FD should be considered.

Conflicts of Interest Disclosure

All authors have no conflicts of interest regarding this article. All authors who are members of The Japan Neurosurgical Society (JNS) have registered with the online Self-reported COI Disclosure Statement Forms through the website for JNS members.

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