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Ruptured Basilar Artery Perforator Aneurysm Definitely Diagnosed with Intraoperative Microsurgical Findings: Case Report and Literature Review

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Abstract

Initial three-dimensional computed tomography and cerebral angiography fail to identify any aneurysm in 20% of cases of subarachnoid hemorrhage. Basilar artery (BA) perforator aneurysms are rare, and approximately 30%-60% were not identified by initial angiography. A 71-year-old male was transferred with a sudden onset of headache and loss of consciousness. Computed tomography demonstrated subarachnoid hemorrhage, but no ruptured aneurysm was detected. Repeat preoperative cerebral angiography indicated a bifurcation aneurysm of the circumflex branch of the superior cerebellar artery perforator, but microsurgical observation identified the BA perforator aneurysm. If the location of the BA perforator aneurysm cannot be clearly identified, as in this case, repeat angiography should be considered, and the treatment strategy should be decided based on a detailed consideration of the site of the aneurysm.

Keywords: subarachnoid hemorrhage, repeat angiography, basilar artery perforator aneurysm, surgical intraoperative findings

Introduction

Subarachnoid hemorrhage (SAH) of unknown etiology is classified into perimesencephalic and non-perimesencephalic types based on the findings of computed tomography (CT). As nonperimesencephalic SAH is often caused by the rupture of a cerebral aneurysm and carries a poor prognosis due to rebleeding, identifying the rupture point is important. No aneurysm is detected in approximately 20% of SAH cases by the initial three-dimensional (3D) CT angiography or cerebral angiography.[1)] In particular, aneurysms of the perforator of the basilar artery (BA) are difficult to diagnose because of their small size, and their shape may undergo changes caused by thrombosis.[2)] We experienced a rare case of SAH in which repeat preoperative angiography showed an aneurysm of the superior cerebellar artery (SCA) perforator, but the actual aneurysm location was on the BA perforator in the intraoperative findings.

Case Report

A 71-year-old male was transferred to our hospital after experiencing a sudden onset of headache and loss of consciousness. The patient's consciousness level was Glasgow Coma Scale (GCS) 12 (E3V4M5) and his blood pressure was 210/120 mmHg on admission. Laboratory evaluations did not show any abnormalities. He had a medical history of hypertension and angina after percutaneous coronary intervention. Head CT demonstrated SAH (Hunt and Kosnik Grade IV; World Federation of Neurosurgical Societies Grade IV) extending from the premedullary cisterns superiorly into the bilateral sylvian fissures and interhemispheric fissure (Fisher Group 3) (Fig. 1a). However, 3D CT angiography of the cerebral vessels demonstrated no clear evidence of a ruptured cerebral aneurysm.

A digital subtraction angiography (DSA) performed on the day after admission found no cerebral aneurysm (Fig. 1 b). Repeat conventional angiography showed no obvious aneurysm on day 5, but multiplanar reconstruction (MPR)

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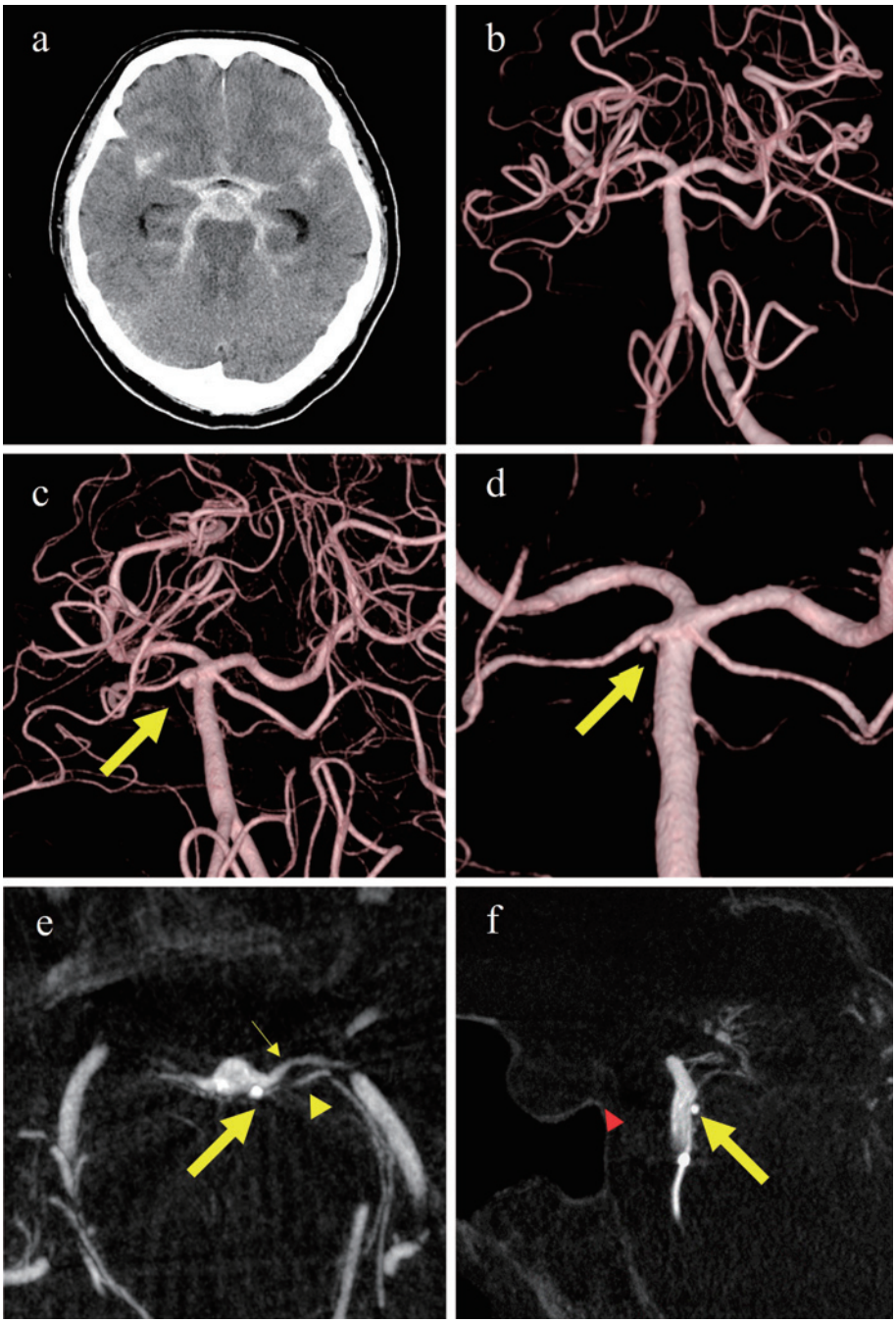

Fig. 1 a: Head CT scan demonstrated diffuse SAH in the perimesencephalic cistern (Fisher group 3). b: DSA showing no cerebral aneurysm on day 1. c: MPR angiogram on day 5 showed a tiny aneurysm (arrow) between the BA and the left SCA. d: MPR angiogram on day 14 showed an aneurysm (arrow) with a neck on the SCA, which was slightly different from the finding on day 5. e: Selective angiogram (MIP axial view) using a microcatheter on day 21 revealed the aneurysm (arrow) between the SCA (small arrow) and the circumflex branch of the SCA perforator (arrowhead). f: The aneurysm (arrow) was below the posterior clinoid process (red arrowhead).

angiography showed a tiny aneurysm between the BA and the left SCA (Fig. 1c). However, since the origin of the aneurysm was not clear, no treatment strategy could be established. MPR angiography on day 14 showed an aneurysm with a neck on the SCA, which was slightly different from the findings on day 5 (Fig. 1d). Sedative and analgesic care was performed for 14 days after the onset. The patient's level of consciousness improved to GCS 14 (E4V4M 6) on day 18. On day 25, selective angiography of the distal side of the BA was performed using a microcatheter to examine the origin of the aneurysm in more detail. These findings suggested a diagnosis of a bifurcation aneurysm

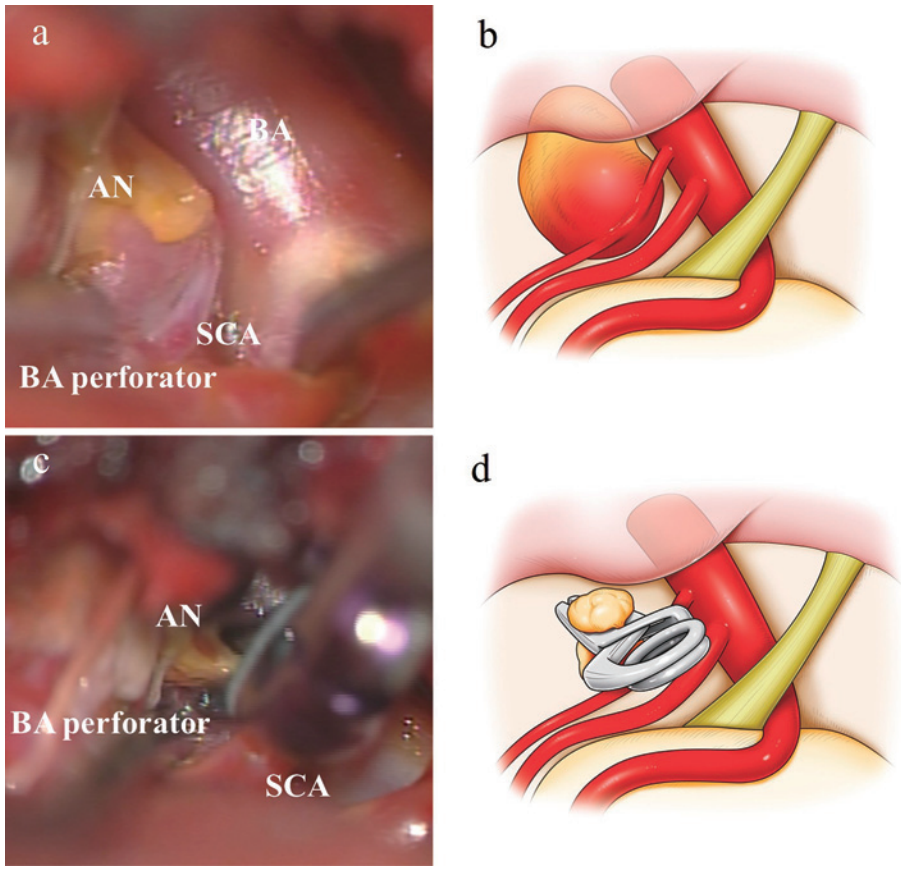

Fig. 2 a, b: Pre-clipping surgical views showing that the neck of the aneurysm was located on the basilar artery perforator. c, d: Post-clipping surgical views showing that only the rupture point was clipped.

between the SCA and the circumflex branch of the SCA perforator (Fig. 1e). The aneurysm of the SCA perforator was judged difficult for endovascular intervention, so neurosurgical treatment was planned.

A subtemporal transtentorial approach was planned based on the height relationship between the aneurysm and the posterior clinoid process (Fig. 1f). Cerebral aneurysm clipping was performed on day 42. The intraoperative findings showed that the aneurysm neck originated from the BA perforator. The location of the aneurysm was different from that of the bifurcation aneurysm of the circumflex branch of the SCA perforator detected by preoperative cerebral angiography (Fig. 2a, b). However, clipping of the neck of the aneurysm was not possible due to its location on the BA perforator and the need to preserve this vessel. Consequently, the rupture point was clipped (Fig. 2 c, d). The postoperative course was uneventful, but the patient's level of consciousness deteriorated to GCS 11 (E3V4 M4) and a follow-up CT demonstrated hydrocephalus. Ventriculoperitoneal shunting was performed on day 63. The patient was transferred to another hospital to continue rehabilitation on day 85 with a modified Rankin Scale (mRS) score of 2. His final consciousness level was GCS 14 (E4V4 M6). Three months after rehabilitation, his mRS had improved to 0.

Discussion

BA perforator aneurysms are rare, and the pathophysiology is unknown.[2,3] BA perforator aneurysms tend to be small with fusiform type, and the angiographical findings change over time.[2] The size of BA perforator aneurysm is often ≤3-7 mm in diameter.[3] Most BA perforator aneurysms are located on the BA perforator and not on the BA trunk. The first case was reported in 1996,[4] and the number of cases may have increased over the last few years possibly due to advances in diagnostic technologies, such as 3D CT angiography, magnetic resonance angiography, and conventional angiography. BA perforator aneurysm may have been treated as SAH of unknown etiology before 1996. Our review identified 19 papers and 54 cases and analyzed 55 cases, including this case. Initial angiography failed to identify about 30%-60% of BA perforator aneurysm.[3] Our review found that the initial angiography failed in 34 cases (62%) (Table 1).[2,4-15] Therefore, repeat angiography is recommended within 7 days.[14] In our case, repeat angiography was performed on day 5, and MPR angiography demonstrated the aneurysm. Since the origin of the

Table 1 Clinical characteristics of 34 patients with ruptured BA perforator aneurysms diagnosed as unverified aneurysm on initial angiography

Case no.	Author/year	Age (yrs)	Location of aneurysm origin on BA[a]	Time until aneurysm detection
1	Ghogawala et al./1996[4]	56	Distal	9 days
2	Hamel et al./2005[5]	51	Middle	N/A
3	Sanchez-Mejia and Lawton/2007[6]	27	Distal	2 months
4		68	Proximal	2 months
5	Mathieson et al./2010[7]	51	Middle	N/A
6	Nyberg et al./2013[8]	45	Middle	2 months
7		65	Middle	9 weeks
8		62	Distal	2 weeks
9		55	Distal	7 days
10	Apok et al./2013[9]	65	Distal	5 days
11	Chavent et al./2014[2]	55	Distal	8 days
12		39	Distal	8 days
13		56	Distal	8 days
14	Chalouhi et al./2014[10]	N/A	Middle	3 days
15	Sivakanthan et al./2015[11]	45	Distal	N/A
16		N/A	Distal	N/A
17		N/A	Distal	N/A
18	Forbrig et al./2016[12]	72	Distal	18 days
19		59	Distal	13 days
20		65	Middle	8 days
21		53	Distal	47 days
22	Satti et al./2017[13]	52	Middle	8 days
23	Buell et al./2018[14]	N/A	Middle	6 days
24		N/A	Middle	5 days
25		N/A	Distal	7 days
26		N/A	Distal	5 days
27		N/A	Middle	5 days
28		59	Distal	5 days
29		69	Distal	2 months
30		65	N/A	N/A
31		57	Distal	N/A
32		62	N/A	N/A
33	Enomoto et al./2020[15]	60	Distal	39 days
34	Present case	71	Distal	5 days

[a]Location of aneurysm origin on BA defined as proximal from vertebral artery union to anterior inferior cerebellar artery, middle from anterior inferior cerebellar artery to SCA, and distal from SCA to BA top.
N/A, not available

aneurysm was unclear and the form of the aneurysm could change, treatment was not performed at this point, and angiography was repeated in this case. However, the aneurysm was identified within 7 days in only 12 of the 26 cases (46%), excluding 8 of the 34 cases in which the time to identify the aneurysm was unknown in the papers. Aneurysms were identified within 1 month in 18 cases (69%). No significant difference in outcome was observed between the groups in which the aneurysm was identified and those in which it was not identified within 1 month.

No optimal treatment strategy has been accepted for BA perforator aneurysms. Conservative treatment was given in 23 of the 55 cases (42%) and resulted in rebleeding in 2 cases (8%). Endovascular treatment (EVT) was performed

Table 2 Previous and present cases of surgical treatment and EVT for BA perforator aneurysms

Case no.	Author/year	Age (yrs)	Location of aneurysm origin on BA[a]	Detection on initial angiogram	Time until aneurysm detection	Treatment	Rebleeding	Perforator preserved	Ischemic complication	Outcome
1	Ghogawala et al./1996[4]	56	Distal	N	9 days	Surgery	N	Y	N	GOS 5
2	Hamel et al./2005[5]	51	Middle	N	N/A	Surgery	N	N	N	N/A
3	Sanchez-Mejia and Lawton/2007[6]	27	Distal	N	2 months	Surgery	Y	N	N	mRS 0
4		68	Proximal	N	2 months	Surgery	Y	N	N	N/A
5		2	Middle	N/A	N/A	Surgery	N	N/A	N	mRS 0
6	Mathieson et al./2010[7]	51	Middle	N	N/A	Surgery	N	N	N	mRS 0
7	Chen et al./2012[16]	66	Middle	Y	N/A	EVT	N	N	N	GOS 4
8	Nyberg et al./2013[8]	45	Middle	N	2 months	EVT	N	Y	N	GOS 5
9		65	Middle	N	9 weeks	EVT	N	N	N	GOS 5
10	Ding et al./2013[17]	58	Middle	Y	N/A	EVT	N	N	Y	GOS 3
11		62	Distal	N	2 weeks	EVT	N	N	Y	GOS 3
12	Gross et al./2013[18]	52	Distal	Y	N/A	Surgery	N	N	N	mRS 1
13	Apok et al./2013[9]	65	Distal	N	5 days	Surgery	N	N	Y	mRS 4
14	Chavent et al./2014[2]	N/A	Middle	N	3 days	EVT	N	Y	N	mRS 0
15	Sivakanthan et al./2015[11]	45	Distal	N	N/A	Surgery	N	Y	N	mRS 1
16	Peschillo et al./2016[19]	N/A	Distal	Y	N/A	EVT	N	N	Y	mRS 2
17		N/A	Distal	N	N/A	EVT	N	Y	N	mRS 0
18		N/A	Distal	N	N/A	EVT	N	N/A	N	mRS 2
19	Forbrig et al./2016[12]	72	Distal	N	18 days	EVT	N	N	N	mRS 2
20	Satti et al./2017[13]	52	Middle	N	8 days	EVT	N	N	Y	mRS 0
21	Buell et al./2018[14]	N/A	Middle	N	6 days	EVT	N	Y	N	mRS 1
22		N/A	Middle	N	5 days	EVT	N	Y	N	mRS 1
23		N/A	Distal	Y	N/A	EVT	Y	N/A	N	mRS 6
24	Chau et al./2018[20]	53	Distal	Y	N/A	EVT	N	N	Y	mRS 0
25		59	Distal	N	5 days	EVT	N	Y	N	mRS 0
26	Bhogal et al./2019[3]	55	Distal	Y	N/A	EVT	N	N	N	mRS 1
27		65	N/A	N	N/A	EVT	N	N	N	mRS 2
28		66	Distal	Y	N/A	EVT	N	N	N	mRS 3
29		41	Middle	Y	N/A	EVT	N	N	Y	mRS 0
30		52	Distal	Y	N/A	EVT	N	N	N	mRS 0
31		39	Middle	Y	N/A	EVT	N	N	N	mRS 2
32	Present case	71	Distal	N	5 days	Surgery	N	Y	N	mRS 0

[a]Location of aneurysm origin on BA defined as proximal from vertebral artery union to anterior inferior cerebellar artery, middle from anterior inferior cerebellar artery to SCA, and distal from SCA to BA top.
GOS, Glasgow Outcome Scale; N, no; N/A, not available; Y, yes

in both of these cases after rebleeding. One case with EVT had a good outcome (mRS 0), but the other case had a poor outcome (mRS 5). This poor outcome case presented with initial World Federation of Neurosurgical Societies grade V.

The shape of BA perforator aneurysm changes over time, so the pathology is considered to be pseudoaneurysm.[3] The present case appeared to be pseudoaneurysm in the intraoperative view. Aneurysms observed in the late arterial phase of angiography are said to have a high possibility of thrombosis, so such findings may indicate conservative treatment.[15] Our analysis found that 21 cases (91%) with conservative treatment had good outcomes (mRS 0 or 1), so conservative treatment should be one of the treatment options.

Surgical treatment was performed in 10 cases and EVT in 22 cases (Table 2).[3-14,16-20] EVT may use coil, stent, Onyx, and flow diverter techniques. The perforator was preserved

in 2 cases (20%) after surgical treatment and in 6 cases (27%) after EVT. Ischemic complications due to perforator disruption occurred in 1 case (10%) after surgical treatment and 6 cases (27%) after EVT. Ischemic complication occurred after EVT using any of the coil, stent, flow diverter, and Onyx, so the specific device was not considered as contributory. Good outcomes for mRS scores 0 and 1 were obtained in 6 cases (66%) after surgical treatment and 13 cases (60%) after EVT.

In this case, the diagnosis was made on day 25, and the surgical treatment was performed on day 42 after SAH. There is no reference to the period from diagnosis to treatment in the literature from the past. Out of 55 reported cases, 4 had rebleeding in the untreated state on days 10, 10, 13, and 20 after SAH onset. Since these instances of rebleeding occurred later than the rebleeding in our case and few cases of rebleeding have been reported after conservative treatment, we also considered conservative treatment. However, the risk of rebleeding after conservative treatment was not negligible, so we decided to continue with surgical treatment.

In our present case, preoperative angiography initially indicated SCA-SCA perforator aneurysm, but the actual aneurysm was located on a more proximal middle BA perforator than the SCA. As mentioned above, BA perforator aneurysms are small and not easy to diagnose, even by angiography. In our case, the definitive diagnosis was obtained based on the microsurgical observation, but the true nature of the aneurysm was unknown when EVT was selected. Therefore, such a difference between the actual location of the aneurysm and the angiographical findings may have occurred among the previously reported cases of EVT, implying that EVT using the flow diverter or Onyx may have impaired normal reflux and increased the risk of ischemic complication. Our review found no difference in outcomes between surgical treatment and EVT, so we cannot conclude which treatment is better. If the aneurysm can be clearly identified by angiography, EVT should be considered, but if the origin of the aneurysm cannot be clearly identified or if change over time occurs, surgical treatment should also be considered.

Conclusion

The present case of BA perforator aneurysm was identified by repeated DSA but was definitely diagnosed under intraoperative microsurgical observation. The accuracy of angiography has definitely improved, but accurate diagnosis for small, shape-changing aneurysms such as BA perforator aneurysm is difficult to establish. If the location of the aneurysm cannot be clearly identified, repeat angiography should be considered, and surgical treatment should also be considered to identify the actual location in the surgical view.

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Abbreviations

BA: Basilar artery
CT: Computed tomography
3D: Three-dimensional
DSA: Digital subtraction angiography
EVT: Endovascular treatment
GCS: Glasgow Coma Scale
MPR: Multi-planar reconstruction
mRS: Modified Rankin Scale
SAH: Subarachnoid hemorrhage
SCA: Superior cerebellar artery

Author Contributions

TK and NO contributed to the manuscript concept and drafted the manuscript. AY and TS revised the manuscript and contributed the manuscript concept. YK and HN have contributed to the acquisition and interpretation of clinical information. All authors have read and approved the final version of the manuscript.

Informed Consent

We have signed informed consent from patients and guardians before submitting to NMC Case Reports.

Conflicts of Interest Disclosure

The authors and all co-authors are no conflicts of disclosure. Authors who are members of the Japan Neurosurgical Society have registered online for self-reported COI Disclosure Statement Forms.

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