Aneurysm in a Fenestrated Anterior Cerebral Artery

—Case Report—

Hidenori OGASAWARA, Tetsuji INAGAWA, Mitsuo YAMAMOTO
and Kazuko KAMIYA

Department of Neurosurgery, Shimane Prefectural Central Hospital, Izumo, Shimane

Abstract

An elderly woman suffered rupture of an aneurysm on the anterior cerebral artery, which had developed from the proximal end of a fenestration in the artery. These features were visualized angiographically and confirmed during surgery. Aneurysms rarely develop from fenestrations in the anterior cerebral artery; to date, only eight cases have been reported in the literature. The findings concerning aneurysms associated with fenestration suggest that congenital factors are involved in their pathogenesis.

Key words: anterior cerebral artery, cerebral aneurysm, arterial fenestration

Introduction

There have been numerous reports on fenestration of the vertebrobasilar artery system, for which there is a well-established pathogenetic theory. However, there has been a paucity of reports on fenestration of the internal carotid artery system and, in particular, fenestration of the anterior cerebral artery. Fenestration of the anterior cerebral artery associated with aneurysm is still rarer, with only eight cases having been reported to date.

We treated a patient with a fenestrated anterior cerebral artery associated with an aneurysm. These features were first detected by cerebral angiography and were confirmed by direct surgery. We believe this case to be extremely interesting in terms of the pathogenesis of cerebral artery aneurysms.

Case Report

On May 1, 1986, a 65-year-old female was found lying in the bathroom with severe headache and urinary incontinence. On admission to Shimane Prefectural Central Hospital she was confused and had left preretinal bleeding. Other neurological findings were normal. Computerized tomography (CT) re-

vealed subarachnoid hemorrhage in the suprasellar cistern, bilateral Sylvian fissures, and insular cisterns (Fig. 1). Cerebral angiograms obtained on the day of admission demonstrated fenestration of the left anterior cerebral artery and a cerebral aneurysm (Fig. 2).

Through a left frontotemporal craniotomy, the clot in the subarachnoid space was removed and the neck of the aneurysm was clipped and wrapped. The aneurysm was found to have developed from the proximal end of the fenestration of the anterior cerebral artery. Postoperatively, she remained somnolent. On May 9 she became stuporous due to vasospasm, and left hemiparesis appeared, followed by pulmonary infarction and pneumonia. At present she is vegetative.

Discussion

Since fenestration of the cerebral arteries does not in itself produce symptoms, its clinical significance is unknown. It is often incidentally detected during examination for such central nervous system diseases as cerebral aneurysm, arteriovenous malformation, and brain tumor. The incidence of fenestration of the cerebral arteries discovered at autopsy is reported-

Received November 28, 1986; Accepted September 16, 1987

Author's present address: H. Ogasawara, M.D., Department of Neurosurgery, School of Medicine, Hiroshima University, Hiroshima, Japan.

Fig. 1 CT revealing high-density areas in the suprasellar cistern, bilateral Sylvian fissures, and insular cisterns.

Fig. 2 Angiogram of the left carotid artery showing the fenestration (arrowheads) at the horizontal portion of the anterior cerebral artery and the aneurysm (arrow) arising from the proximal end.

ly 0.1–7.2% in the anterior cerebral artery,⁶⁾ 0.26–0.34% in the middle cerebral artery,¹⁷⁾ and 0.3–0.34% in the vertebrobasilar artery.²¹⁾ The reported incidence detected by cerebral angiography is 0.2% in the anterior cerebral artery,⁵⁾ 0.26–0.34% in the middle cerebral artery,¹⁷⁾ and 0.29–0.9% in the vertebrobasilar artery.¹³⁾ Thus, the incidence of fenestration of the anterior cerebral artery is extremely low.

In evaluating cerebral angiograms, it is important to differentiate between fenestration of the anterior cerebral artery and Heubner's artery, looping of the anterior cerebral artery, looping of the posterior cerebral artery, and duplication of the middle cerebral artery.^{8,10)} In some cases it is necessary to consider the exposure conditions and direction, including such factors as magnification, stereoscopy, and oblique projection.

The most common disease of the central nervous system associated with fenestration of the cerebral arteries is cerebrovascular disease,^{7,12,15)} such as cerebral aneurysm. According to Kwak *et al.*,⁹⁾ the cases of fenestration that have been reported are primarily those that were associated with cerebral aneurysm, and their discovery resulted from carefully interpreted cerebral angiograms for subarachnoid hemorrhage. These authors do not consider the incidence of associated aneurysm to be particularly high.

The development of vertebrobasilar arterial fenestration, which is fairly well understood, is thought to

involve incomplete fusion of a pair of longitudinal neural arteries whose development precedes that of the basilar artery. However, the mechanism of fenestration of the internal carotid artery system has yet to be identified. According to Padget,¹⁴⁾ when the embryo is 4–5 mm in length the primitive internal carotid artery forms the cranial and caudal divisions. When the embryo is 7–12 mm long the cranial division separates into the primitive anterior choroidal artery, primitive middle cerebral artery, and stem of the anterior cerebral artery. At 12–14 mm the stem of the anterior cerebral artery and plexiform anastomoses of the future anterior communicating artery are formed. When the stems of the anterior cerebral, middle cerebral, and anterior communicating arteries are being formed, plexiformity is observed. Thus, it is possible that fenestration of the anterior and middle cerebral arteries occurs when these plexiform anastomoses incompletely fuse and partially remain, but the details are as yet unknown.

Reports of aneurysms accompanying fenestration of the cerebral arteries are scant. Our review of the literature yielded eight cases involving the anterior cerebral artery (Table 1), two the middle cerebral artery, and 10 the vertebrobasilar artery. Among the cases involving the anterior cerebral artery, the patients' ages ranged from 36 to 70 years, with a mean

Table 1 Reported cases of aneurysms associated with anterior cerebral artery fenestration

Case No.	Author (Year)	Age	Sex	Side of fenestration	Site of aneurysm arising from fenestration	Method of demonstration	Direct surgery
1	Crompton (1962) ¹⁾	—	—	—	proximal end	autopsy	—
2	Waga and Morikawa (1979) ¹⁸⁾	36	F	lt.	proximal end	angiography, surgery	+
3	Yamada <i>et al.</i> (1982) ²²⁾	43	M	rt.	proximal end	angiography, surgery	+
4	Inagawa <i>et al.</i> (1983) ⁵⁾	70	F	lt.	proximal end	angiography	—
5	Handa <i>et al.</i> (1984) ³⁾	50	F	rt.	proximal end	angiography, surgery	+
6	Minakawa <i>et al.</i> (1985) ¹¹⁾	56	M	lt.	proximal end	angiography, surgery	+
7	Wakabayashi <i>et al.</i> (1985) ¹⁹⁾	41	M	rt.	proximal end	angiography, surgery	+
8	"	38	M	rt.	proximal end	angiography, surgery	+
9	Present case	65	F	lt.	proximal end	angiography, surgery	+

of 50 years, and there were four males and four females. Among the eight cases diagnosed by cerebral angiography, excluding the autopsy case reported by Crompton,¹⁾ the right side was affected in four and the left side in four.

Anomalies of the arterial wall^{2,4)} and hemodynamic factors^{16,20)} are considered to play an important role in the development of aneurysms. On histological examination, Crompton¹⁾ observed a defect in the media of the cerebral artery at the proximal end of the fenestration. In each of the reported cases of fenestration of the anterior cerebral artery, the aneurysm developed from the proximal end. It is possible that in our cases both the arterial anomaly and local high blood pressure contributed to the development of the aneurysm.

Agreement has not yet been reached as to whether an aneurysm associated with fenestration of a cerebral artery is part of a complex arterial anomaly¹²⁾ or an incidental complication without any special significance.⁹⁾ However, it is assumed that congenital factors are involved in the development of aneurysms in arterial fenestrations. Careful study of additional cases will no doubt help to answer some of these questions.

References

- 1) Crompton MR: The pathology of ruptured middle cerebral aneurysms with special reference to the differences between the sexes. *Lancet* 2: 423-425, 1962
- 2) Forbus WD: On the origin of miliary aneurysms of superficial cerebral arteries. *Bull Hopkins Hosp* 47: 239-284, 1930
- 3) Handa J, Nakasu Y, Matsuda M, Kyoshima K: Aneurysms of the proximal anterior cerebral artery. *Surg Neurol* 22: 486-490, 1984
- 4) Hochstetter F: Über die Entwicklung der A. vertebralis beim Kaninchen, nebst Bemerkungen über die Entstehung der Ansa Viessensii. *Morpholog Jahrbuch* 16: 572, 1890
- 5) Inagawa T, Takeda T, Taguchi H, Yamada T: A case of fenestrated anterior cerebral artery with an associated aneurysm. *Hiroshima J Med Sci* 32: 153-155, 1983
- 6) Ito J, Washiyama K, Hong K, Ibuchi Y: Fenestration of the anterior cerebral artery. *Neuroradiology* 21: 277-280, 1981
- 7) Kawamoto K, Tanaka M, Miki M, Someda K: Fenestration of the vertebrobasilar system. *No To Shinkei* 24: 811-815, 1972 (in Japanese)
- 8) Kraysenbühl HA, Yasargil MG: *Cerebral Angiography*, ed 2. London, Butterworths, 1958, pp 38-40
- 9) Kwak R, Kuwabara K, Niizuma H, Suzuki J: Anomalies of the cerebral artery with intracranial sacular aneurysms. Duplication and fenestration. *No Shinkei Geka* 7: 691-696, 1979 (in Japanese)
- 10) Matsumura M, Nojiri K: Ruptured anterior communicating artery aneurysm associated with fenestration of the anterior cerebral artery. *Surg Neurol* 22: 371-376, 1984
- 11) Minakawa T, Kawamata M, Hayano M, Kawakami K: Aneurysms associated with fenestrated anterior cerebral arteries. *Surg Neurol* 24: 284-288, 1985
- 12) Mizukami M, Mine T, Tomita T: Fenestration of the vertebral artery (vertebral diastematoarteria). Coexistence of cerebral aneurysm and cerebral arteriovenous malformation. *No To Shinkei* 20: 1271-1276, 1968 (in Japanese)

- 13) Nakajima K, Ito Z, Hen R, Uemura K, Matsuoka S: Congenital anomalies of cerebral artery and intracranial aneurysms. *No To Shinkei* 28: 197-201, 1976 (in Japanese)
- 14) Padget DH: The development of the cranial arteries in the human embryo. *Contributions to Embryology* 212: 205-261, 1948
- 15) Shishido T, Watanabe M, Kuramoto S: A case of giant aneurysms associated with vertebral fenestration. *No Shinkei Geka* 7: 79-83, 1979 (in Japanese)
- 16) Suzuki J, Ohara H: Clinicopathological study of cerebral aneurysms. Origin, rupture, repair and growth. *J Neurosurg* 48: 505-514, 1978
- 17) Ueda T, Goya T, Kinoshita K: Fenestration of the middle cerebral artery associated with an aneurysm. *Neurol Med Chir (Tokyo)* 23: 278-281, 1983 (in Japanese)
- 18) Waga S, Morikawa A: Aneurysm developing on the infundibular widening of the posterior communicating artery. *Surg Neurol* 11: 125-127, 1979
- 19) Wakabayashi T, Tamaki N, Yamashita H, Saya H, Suyama T, Matsumoto S: Angiographic classification of aneurysm of the horizontal segment of the anterior cerebral artery. *Surg Neurol* 24: 31-34, 1985
- 20) Wilson G, Riggs HE, Rupp C Jr: The pathological anatomy of ruptured cerebral aneurysm. *J Neurosurg* 11: 128-134, 1954
- 21) Wollschlaeger G, Wollschlaeger PB, Lucas FV, Lopez VF: Experience and result with post mortem cerebral angiography performed as routine procedure of the autopsy. *Amer J Roentgen* 101: 68-87, 1967
- 22) Yamada T, Inagawa T, Takeda T: Ruptured aneurysm at the anterior cerebral artery fenestration. *J Neurosurg* 57: 826-828, 1982

Address reprint requests to: H. Ogasawara, M.D., Department of Neurosurgery, School of Medicine, Hiroshima University, 1-2-3 Kasumi, Minami-ku, Hiroshima 734, Japan.