We report a unique case of bilateral mirror image M1 aneurysms, one of which was an unruptured aneurysm arising from the proximal end of right middle cerebral artery fenestration with long loop and the other ruptured aneurysm from the contralateral side. We clipped ruptured aneurysm first and unruptured one in three months after the first operation. The difficulties of identifying this unusual vascular anomaly and possible problems during the surgery of an aneurysm at the site of fenestration are discussed with a review of the literature.

KEY WORDS: Cerebral aneurysm • Fenestration • Middle cerebral artery.

INTRODUCTION

The fenestration of middle cerebral artery (MCA) is a rare anatomical variant. It is often asymptomatic. However, with the recent advances of neuroradiologic technique, the reported cases of MCA fenestration associated with cerebral aneurysms are increasing. To date, about 67 cases of MCA fenestration were reported and two thirds of these cases were reported in the later half from the first case report in 1962 to present. We report a rare case of bilateral MCA aneurysms, one of which was developed at the proximal end of right MCA fenestration with long convex loop on the middle portion of M1.

CASE REPORT

A 32-year-old man, previously healthy, presented with sudden onset of severe headache and vomiting. The patient had no history of trauma and his past medical history was unremarkable. He demonstrated meningeal irritation signs with no focal neurological deficit. A computed tomography (CT) scan of the head showed slight subarachnoid hemorrhage in the left insular cistern (Fig. 1). A three-dimensional CT angiography showed bilateral mirror image MCA aneurysms located at the middle portion of M1 segment and a vascular loop enclosing the right MCA aneurysm was suspected (Fig. 2, 3A). Conventional cerebral angiography showed both aneurysms arose at the similar distance, about 1.5 cm, from the internal carotid artery (ICA) bifurcation and directed inferolaterally. The left one measured 4 mm in diameter and the right one measured 6 mm in diameter with...
smooth surface. Despite of conventional angiography, we could not differentiate definitely the MCA fenestration from overlapping of vessels (Fig. 3B). We presumed the left one was ruptured and guglielmi detachable coil embolization was tried but it was discontinued due to intimal damage to the left ICA. A left frontotemporal craniotomy was performed. The surgical exploration revealed that the left MCA aneurysm was ruptured and it was clipped in usual manner without difficulty. After three months, the second operation was performed. Three-dimensional rotational angiography at that time clearly defined the relationship of the fenestration and an aneurysm (Fig. 3C). The fenestration started from the middle portion of the M1 and a saccular aneurysm arose from the proximal end of the fenestration. The size of diagonal loop was measured about 1 cm from proximal to distal end of the fenestration. The aneurysm was also clipped successfully with right frontotemporal craniotomy. Most of characteristics observed in the previous study could be confirmed under the surgical microscope but some unexpected problems were encountered. The aneurysmal sac was directed toward behind the fenestrated branches. The fenestrated branches were densely adhered to the aneurysmal sac all the way. Furthermore, it was hard to distract or retract the branches because the distal end of fenestration was fused (Fig. 4). After dissection of aneurysmal neck, an obliquely angled aneurysmal clip was applied. The postoperative course was uneventful and he was discharged without neurologic deficit.

**DISCUSSION**

In 1962, Crompton reported the first case of MCA fenestration in 347 middle cerebral arteries examined in autopsy\(^4\). The incidence of MCA fenestration varies from 0.02% to 1% according to the methods of investigation (e.g., autopsy or angiographic studies). It was observed 0.17% to 1% in autopsy studies\(^4,33-38\) and 0.02% to 0.43% in angiographic studies\(^3,10,28\). Although it shows similar incidences between anatomical and angiographic studies, the conventional angiography has some diagnostic difficulties. The MCA fenestration that is obscured by combined aneurysm or other arterial branches could be unrecognized. We also could not confirm the presumed MCA fenestration with conventional angiography but definite diagnosis was made with three-dimensional rotational angiography. The stereoscopic images from this study revealed clear relationship of the fenestrated vessels and the aneurysm at the proximal end of fenestration. It is believed that stereoscopic reconstruction technique such as
three-dimensional rotational angiography could be helpful to overcome the limitation of conventional angiography in detecting these hidden anomalies and identifying the relationship of fenestration loop and aneurysms.

To date, about 67 cases of MCA fenestration including our case were reported\(^1-39\). Most of previous articles on the MCA fenestration focused on the association of cerebral aneurysm located at the site of fenestration or distant site from it. In the review of previous reports, 19 cases were coincided with cerebral aneurysms\(^5,12,19,24,29,30,35\) and 6 cases of aneurysm developed at the proximal or distal end of fenestration\(^5,12,18,19,24\). Other data indicated that the MCA fenestration was developed mainly at the M1 portion and there was no ethnic predilection between Asian and Western countries. Although these were sporadic cases, it is considered that arterial fenestration would be a spectrum of congenital anomaly that commonly accompany with cerebral aneurysms and fenestration site would be a preferred site of aneurysm development. Finlay measured stereological data from six segment of brain arteries, each including fenestration (five from vertebrobasilar junction and one from the MCA). He found media defect on the both edge of fenestration and difference of subendothelial layering between proximal and distal end of fenestration. He stated that the propensity of aneurysm development at the site of fenestration could be explained by structural abnormality and hemodynamic stress like the aneurysms arising from any bifurcation. Structural investigation by Finlay supports the assumption that proximal end of fenestration could be a preferred site of aneurysm development\(^4\).

The MCA fenestration is a rare vascular anomaly and an incidental finding in itself. But when saccular aneurysms develop in its proximal end of fenestration, neurosurgeons could be encountered the surgical problems that we had experienced. The closed loop of fenestration enclosing the aneurysmal sac is an unfamiliar surgical environment. It could disturb the dissection between the fenestrated branches and aneurysmal sac to make space for the clip placement. Although the difficulties in dissection and clip placement may be different to the size and relationship of fenestrated loop and aneurysmal sac, it is considerable that the surgical difficulties in the treatment of aneurysm adjacent to this unfamiliar vascular anomaly could be a new clinical significance of arterial fenestration.

**CONCLUSION**

We report a rare case of MCA aneurysm originated from proximal end of fenestration with long loop at the middle portion of M1 segment. Vascular neurosurgeons have to consider such hidden vascular anomaly in the review of preoperative examination and prepare the possible surgical problems during operation.

- Acknowledgements
  This paper was supported by a grant from the Jeju National University Hospital Research Fund in 2008.

**References**

1. Baba M, Shimizu T, Kagawa M, Kitamura K, Kobayashi N: [A case of multiple anomalies of cerebral vessels–fenestration of the middle cerebral artery aneurysm of the anterior communicating artery and arteriovenous malformation on the frontotopar region (author’s transl.).] No Shinkei Geka 5 : 59-64, 1977
2. Barbara H, Marek S, Stawomir B, Dabrowka S: Angiographic and CT-angiographic appearance of middle cerebral artery fenestration in a patient with spontaneous subarachnoid hemorrhage. Pol J Radiol 68 : 87-89, 2003
3. Cizek B: On a rare form of fenestration of the middle cerebral artery. Folia Morphol (Warsz) 54 : 9-13, 1995
4. Crompton MR: The pathology of ruptured middle-cerebral aneurysms with special reference to the differences between the sexes. Lancet 280 : 421-425, 1962
5. Deruty R, Pelissou-Guyotat I, Mottotese C, Bognar L, Laharote JC, Turjman F: Fenestration of the middle cerebral artery and aneurysm at the site of the fenestration. Neurol Res 14 : 421-424, 1992
6. Finlay HM, Canham PB: The layered fabric of cerebral artery fenestration. Stroke 25 : 1799-1806, 1994
7. Fujimoto M, Nakahara I, Tanaka M, Iwamuro Y, Watanabe Y, Harada K: [Multiple intracranial aneurysms and vascular abnormalities associated with neurofibromatosis type 1: a case report.] No Shinkei Geka 32 : 355-359, 2004
8. Fuse T, Niwa Y, Harada S: Local intraarterial fibrinolytic therapy for embolic stroke associated with vascular anomalies--two case reports. Neurol Med Chir (Tokyo) 40 : 641-644, 2000
9. Gailloud P, Albayram S, Fasel JH, Beauchamp NJ, Murphy KJ: Angiographic and embryologic considerations in five cases of middle cerebral artery fenestration. AJNR Am J Neuroradiol 23 : 585-587, 2002
10. Ito J, Maeda H, Inoue K, Onishi Y: Fenestration of the middle cerebral artery. Neuroradiology 13 : 57-39, 1977
11. Jeong SK, Kwak HS, Cho YI: Middle cerebral artery fenestration in patients with cerebral ischemia. J Neurol Sci 275 : 181-184, 2008
12. Kalia KK, Ross DA, Gurin PH: Multiple arterial fenestrations, multiple aneurysms and an arteriovenous malformation in a patient with subarachnoid hemorrhage. Surg Neurol 35 : 45-48, 1991
13. Kim DW, Kang SD: Middle cerebral artery fenestration associated with an aneurysm. Korean J Cerebrovasc Surg 7 : 158-160, 2005
14. Kim MS, Hur JW, Lee JW, Lee HK: Middle cerebral artery aneurysms detected by conventional angiography and magnetic resonance angiography. J Korean Neurosurg Soc 37 : 263-267, 2005
15. Kitami K, Kamiyama H, Yasui N: [Angiographic analysis of the middle cerebral artery in cerebral aneurysms--its branching pattern and so-called vascular anomalies.] No Shinkei Geka 13 : 283-290, 1985
16. Lazar ML, Bland JE, North RR, Bringewald PR: Middle cerebral artery fenestration. Neurosurgery 6 : 297-300, 1980
17. Lee SJ, Hong SK: M1 fenestration in a patient with multiple cerebral arterial aneurysms: case report. J Korean Neurosurg Soc 27 : 536-539, 1998
18. Nakamura H, Takada A, Hida T, Ushio Y: Fenestration of the middle cerebral artery associated with aneurysm--case report. Neurol Med Chir (Tokyo) 34 : 555-557, 1994
19. Nußbaum ES, Delillo A, Janjua TM, Nußbaum LA: Fenestration of the middle cerebral artery with an associated ruptured aneurysm. J Clin...
20. Okudera H, Koike J, Toba Y, Kuroyanagi T, Kyoshima K, Kobayashi S: [Fenestration of the middle cerebral artery associated with cerebral infarction. Report of two cases.] Neurolog Med Chir (Tokyo) 27: 559-563, 1987
21. Osborn RE, Kirk G: Cerebral arterial fenestration. Comput Radiol 11: 141-145, 1987
22. Parmar H, Sitoh YY, Hui F: Normal variants of the intracranial circulation demonstrated by MR angiography at 3T. Eur J Radiol 56: 220-228, 2005
23. Sanders WP, Sorek PA, Mehta BA: Fenestration of intracranial arteries with special attention to associated aneurysms and other anomalies. AJNR Am J Neuroradiol 14: 675-680, 1993
24. Schmieder K, Hardenack M, Harders A: Proximal long fenestration associated with an internal carotid artery aneurysm. Case illustration. J Neurosurg 86: 733, 1997
25. Teal JS, Rumbaugh CL, Bergeron RT, Segall HD: Angiographic demonstration of fenestrations of the intradural intracranial arteries. Radiology 106: 123-126, 1973
26. Uchino A, Kato A, Abe M, Kudo S: Association of cerebral arteriovenous malformation with cerebral arterial fenestration. Eur Radiol 11: 493-496, 2001
27. Uchino A, Kato A, Takase Y, Kudo S: Middle cerebral artery variations detected by magnetic resonance angiography. Eur Radiol 10: 560-563, 2000
28. Uchino A, Takase Y, Nomiya K, Egashira R, Kudo S: Fenestration of the middle cerebral artery detected by MR angiography. Magn Reson Med Sci 5: 51-55, 2006
29. Ueda T, Goya T, Kinoshita K: [Fenestration of the middle cerebral artery associated with an aneurysm.] Neurol Med Chir (Tokyo) 23: 278-281, 1983
30. Ueda T, Goya T, Kinoshita K, Wakuta Y, Mihara K: [Multiple anomalies of cerebral vessels. A case of multiple aneurysms associated with fenestration of the middle cerebral artery and persistent primitive trigeminal artery.] No Shinkei Geka 12: 531-536, 1984
31. Umansky F, Dujovny M, Ausman JI, Diaz FG, Mirchandani HG: Anomalies and variations of the middle cerebral artery: a microanatomical study. Neurosurgery 22: 1023-1027, 1988
32. Wollschlaeger G, Wollschlaeger PB, Lucas FV, Lopez VF: Experience and result with postmortem cerebral angiography performed as routine procedure of the autopsy. Am J Roentgenol Radium Ther Nucl Med 101: 68-87, 1967
33. Yasargil MG: Microneurosurgery, New York: Thieme-Stratton Inc., 1984, pp72-91