Aneurysm at origin of duplicated middle cerebral artery associated with another aneurysm

Yu Iida, Akira Tamase, Tomoya Kamide, Kentaro Mori, Shunsuke Seki, Motohiro Nomura

Department of Neurosurgery, Yokohama Sakae Kyosai Hospital, Sakae-ku, Yokohama 247-8581, Japan

E-mail: Yu Iida - yuiida1204@gmail.com; Akira Tamase - rco55555@gmail.com; Tomoya Kamide - kamide@med.kanazawa-u.ac.jp; Kentaro Mori - squad1979@me.com; Shunsuke Seki - libranaoct@hotmail.com; *Motohiro Nomura - nomura413jp@yahoo.co.jp

*Corresponding author

Received: 28 April 15 Accepted: 09 September 15 Published: 23 October 15

Abstract

Background: A duplicated middle cerebral artery (DMCA) is a rare vessel anomaly. Aneurysms at the origin of DMCA have been reported.

Case Description: We report 2 cases of aneurysms at the origin of DMCA accompanied by aneurysms at different sites. Each case of ruptured and unruptured aneurysm at the DMCA origin was associated with an unruptured aneurysm at the ipsilateral internal carotid artery and a ruptured one at the ipsilateral MCA, respectively. The aneurysms were clipped successfully in both patients.

Conclusion: In cases of DMCA aneurysm associated with an aneurysm at another site, either aneurysm has a high risk of rupture. In such a case, radical treatment is necessary.

Key Words: Aneurysm, anomaly, duplicated, middle cerebral artery

INTRODUCTION

Some anomalies of the middle cerebral artery (MCA) have been reported on autopsy and radiological examinations. Among them, a duplicated MCA (DMCA), accessory MCA, and fenestration are well-known. DMCA arises from the internal carotid artery (ICA) and supplies blood to the MCA territory. The incidence of DMCA has been reported to be 0.7–2.9% on autopsy and 0.24–1.5% on angiography. There are some reports describing an anomalous MCA associated with an aneurysm. An aneurysm at the junction of ICA and DMCA is rare. To our knowledge, only 30 cases of DMCA aneurysms have been reported in the literature. We encountered 2 patients with DMCA aneurysms, ruptured, and unruptured ones. Each ruptured and unruptured DMCA aneurysm was accompanied by an unruptured ICA aneurysm at the bifurcation of the posterior communicating artery (PCoA) and a ruptured MCA aneurysm, respectively.

In this report, we describe the patients with DMCA aneurysms and discuss the clinical course, radiological findings, and management of this rare aneurysm.

CASE PRESENTATION

Case 1

A 41-year-old woman without any significant past history experienced sudden-onset headache and vomiting. Computed tomography (CT) on admission showed subarachnoid hemorrhage (SAH) [Figure 1a]. Angiography [Figure 1b] demonstrated the right DMCA and an aneurysm at its origin. The size of the DMCA aneurysm was 5.5 (width) mm × 5.0 (depth) mm × 6.5 (height) mm. The aneurysm was directed laterally. The angiography also demonstrated a small aneurysm on ICA...
at the bifurcation of PCoA (ICA-PCoA). The aneurysm measured 2.0 mm × 2.0 mm × 1.5 mm in size. Emergency craniotomy was performed. The aneurysm was found at the origin of DMCA [Figure 1c] and PCoA [Figure 1d]. Although the size of DMCA (1.6 mm in diameter) was smaller than the MCA (3.0 mm), it had a somewhat diameter. Both the DMCA and ICA-PCoA aneurysms were successfully clipped [Figure 1e]. Her postoperative course was uneventful. She was discharged without neurological deficit on the 27th day. Angiography performed 8 years after the onset showed no recurrence of the aneurysms [Figure 1f].

Case 2
A 76-year-old woman was revealed to have a right MCA aneurysm at the M1/M2 junction and a right DMCA aneurysm on magnetic resonance imaging 6 years ago [Figure 2a]. The sizes of both aneurysms were small, and the patient was 70 years old. Therefore, radical treatment was not performed at that time. Six years later, she suddenly developed consciousness disturbance. CT on admission revealed SAH and intracerebral hemorrhage [Figure 2b]. Three-dimensional CT angiography showed the right MCA and DMCA aneurysms [Figure 2c]. The diameters of MCA and DMCA were approximately 3.0 and 1.0 mm in diameter, respectively. The size of the MCA aneurysm was 5.0 (width) mm × 4.5 (depth) mm × 8.0 (height) mm, and the size of DMCA aneurysm was 1.8 mm × 1.5 mm × 2.5 mm. Both MCA and DMCA aneurysms were directed laterally. The right MCA ran superior to the common course in the sylvian fissure. Emergency craniotomy was conducted. The DMCA and a small aneurysm at its origin were recognized [Figure 2d]. The DMCA and MCA aneurysms were successfully clipped [Figure 2e and f]. Postoperatively, the patient showed symptoms due to vasospasm. Therefore, the intra-arterial injection of fasudil hydrochloride hydrate and percutaneous transluminal angioplasty were performed. After that, a left ventriculoperitoneal shunt operation was performed for hydrocephalus. Postoperative angiography [Figure 2g] demonstrated that both aneurysms were clipped. She was transferred to another hospital with left hemiparesis for rehabilitation on the 60th day.

DISCUSSION
An aneurysm at the DMCA origin was initially reported by Crompton and Lond in 1962. To date, 30 cases of DMCA aneurysms have been reported in the literature. Therefore, our cases are the 31st and 32nd cases of DMCA aneurysms. Among them, 17 cases including ours were associated with aneurysms at other sites. Hori et al. summarized cases with multiple aneurysms including one at DMCA. They concluded that the risk of rupture of a DMCA aneurysm is high, and aggressive management should be considered, particularly when there are multiple aneurysms. We encountered 2 patients, one with a ruptured DMCA aneurysm and a small unruptured ICA-PCoA aneurysm (Case 1) and another with a ruptured MCA aneurysm and a small unruptured DMCA aneurysm (Case 2). In both cases, the second
aneurysm was located on the same side as the DMCA aneurysm. In Case 1, the ruptured DMCA aneurysm was not small. Although relatively young, the patient had already developed two aneurysms. The DMCA aneurysm was located on the long axis of ICA. In Case 2, the running course of the main trunk of MCA was superior to the common course. This may have been due to the presence of DMCA. It was suggested that hemodynamic stress may contribute to the formation and enlargement of an aneurysm of MCA with an atypical running course.

Kai et al. classified DMCA into two types based on the site of origin. Type A DMCA originates from the top of ICA, and Type B DMCA branches from ICA between the anterior choroidal artery and top of ICA. The diameter of Type A DMCA is comparable with that of the main MCA. On the other hand, the diameter of Type B DMCA is smaller than the main MCA. They also mentioned that the aneurysm is associated with Type B DMCA. In our Case 1, DMCA originated from ICA close to the top, and the diameter was not so small as that of Case 2. This condition may be diagnosed as a carotid trifurcation aneurysm. There is a possibility that an aneurysm may develop on Type A DMCA in cases where the DMCA diameter is not small. Irrespective of this, in cases with DMCA, especially Type B, the possible coexistence of another aneurysm should be considered.

**CONCLUSIONS**

More than half of the cases of DMCA aneurysms have multiple aneurysms. It is suggested that hemodynamic stress might contribute to formation and enlargement of multiple aneurysms. Therefore, the presence of a DMCA aneurysm along with another aneurysm is associated with a high risk of rupture. In such a case, radical treatment is necessary.

**Financial support and sponsorship**
Nil.

**Conflicts of interest**
There are no conflicts of interest.

**REFERENCES**

1. Chang HY, Kim MS. Middle cerebral artery duplication: classification and clinical implications. J Korean Neurosurg Soc 2011;49:102-6.
2. Crompton MR, Lond MB. The pathology of ruptured middle-cerebral aneurysms with special reference to the differences between the sexes. Lancet 1962;2:421-5.
3. Elsharkawy A, Ishii K, Niemelä M, Kivisaari R, Lehto H, Hernesniemi J. Management of aneurysms at the origin of duplicated middle cerebral artery: Series of four patients with review of the literature. World Neurosurg 2013;80:e313-8.
4. Hori E, Kurosaki K, Matsumura N, Yamatani K, Kusunose M, Kuwayama N, et al. Multiple aneurysms arising from the origin of a duplication of the middle cerebral artery. J Clin Neurosci 2005;12:812-5.
5. Kai Y, Hamada J, Morioka M, Yano S, Kudo M, Kuratsu J. Treatment of unruptured duplicated middle cerebral artery aneurysm: Case report. Surg Neurol 2006;65:190-3.

6. Kaliaperumal C, Jain N, McKinstry CS, Choudhari KA. Carotid “trifurcation” aneurysm: Surgical anatomy and management. Clin Neurol Neurosurg 2007;109:538-40.

7. Miyahara K, Fujitsu K, Ichikawa T, Mukaihara S, Okada T, Kaku S. Unruptured saccular aneurysm at the origin of the duplicated middle cerebral artery: Reports of two cases and review of the literature. No Shinkei Geka 2009;37:1241-5.

8. Nomura M, Yamashima T, Kita D, Kida S, Kajinami K, Yamashita J. Duplication of the middle cerebral artery associated with an unruptured aneurysm. Acta Neurochir (Wien) 2000;142:221-2.

9. Toyota S, Kumagai T, Sugano H, Yamamoto S, Mori K, Taki T. Unruptured aneurysm at the origin of the duplicated middle cerebral artery treated by coil embolization: A case report. Open J Mod Neurosurg 2015;5:27-33.

10. Umansky F, Dujovny M, Ausman JI, Diaz FG, Mirchandani HG. Anomalies and variations of the middle cerebral artery: A microanatomical study. Neurosurgery 1988;22 (6 Pt 1):1023-7.