Traumatic Middle Meningeal Arteriovenous Fistula of Non-fractured Site Detected by Four-dimensional Computed Tomography Angiography: A Case Report

Yusuke Iki,1 Yoichi Morofuji,1 Tadashi Kanamoto,2 Nobutaka Horie,1 Tsuyoshi Izumo,1 Takeo Anda,1 and Takayuki Matsuo1

Middle meningeal arteriovenous fistula (MMAVF) of a non-fractured site is extremely rare, and the clinical characteristics are still unclear. We report a case of delayed onset of venous infarction due to an MMAVF following a fall accident. A 69-year-old man sustained multiple trauma due to a fall accident. Head computed tomography (CT) showed traumatic subarachnoid hemorrhage, a left subdural hematoma, and skull fracture in his right temporal bone, all of which were managed conservatively. Five days after his admission, he suddenly exhibited total aphasia and right hemiparesis. Emergent CT revealed sporadic low-density areas in his left cerebral hemisphere, and four-dimensional CT angiography (4D-CTA) showed dilatation of the left middle meningeal artery and early venous drainage in the cavernous sinus and anterior temporal diploic vein (ATDV). A series of hemodynamics of 4D-CTA revealed early venous filling of ATDV interrelated with retrograde upward flow to high convexity in the venous phase. The MMAVF was successfully obliterated by transarterial coil embolization. We herein describe this case of MMAVF in which 4D-CTA was useful for the diagnosis.

Keywords: four-dimensional computed tomography angiography, middle meningeal arteriovenous fistula, venous infarction

Introduction

Middle meningeal arteriovenous fistulas (MMAVFs) are rare structures that create a communication between the middle meningeal artery (MMA) and dural venous sinuses as well as the middle meningeal, diploic, or cortical veins. MMAVFs have mainly been described in single case reports. Although these lesions occur in 1.8% of patients with head trauma,1,2 their natural history remains uncertain, especially in asymptomatic cases. In the present case, an MMAVF led to symptomatic intracranial venous congestion at a non-fractured site. This report describes the unique etiology and angiographic findings in this patient. We herein report a case in which four-dimensional computed tomography angiography (4D-CTA) was useful for diagnosis of an MMAVF.

Case Report

A 69-year-old man was transferred to our hospital with multiple trauma. He had fallen from a height of 5 m. The patient had head trauma, hemopneumothorax, pulmonary contusion, and fractures of the ribs, scapula, clavicle, and toe. Head computed tomography (CT) showed diffuse subarachnoid hemorrhage with a linear skull fracture in his right temporoparietal area and a left subdural hematoma. Chest drainage was immediately established for the right hemopneumothorax. He was managed conservatively for the head trauma. The day after admission, he underwent successful internal fixation of the right scapula and clavicle fractures. Five days after admission, he suddenly exhibited mild somnolence, right hemiparesis (grade 3/5), and global aphasia. We suspected vasospasm and performed emergent 4D-CTA instead of magnetic resonance imaging (MRI) because the patient had undergone external fixation of his toe with a device containing a magnetic substance. A 320 detector rows CT system (Aquilion ONE ViSION, Canon Medical Systems, Otawara, Japan) was used, covering a wide scan range of 160 mm per a single rotation without couch movement. Nonionic contrast medium infusion of 50 mL (Iomeran300, Eisai, Tokyo, Japan) at a 4-mL/s was completed by automated antecubital venous injection. The study protocol contained non-helical scanning method, 80 mA, 100 mA, gantry rotation speed of 1 Hz during the continuous phase, i.e., contrast medium arrival from arterial phase to venous vasculature. Beginning 5 s after injection, three intermittent volume scans of 1 s was acquired at 1 s interval. Subsequently, 25 s continuous imaging was performed as detailed above, and thereafter several intermittent volume scans was acquired during venous wash out. 4D-CTA showed no vaso- spastic changes of the intracranial major vessels, but plain CT revealed multiple low-density areas in left hemisphere that corresponded with the neurological deficit (Fig. 1). 4D-CTA revealed abnormal dilatation of the MMA in the arterial phase in the left middle cranial fossa and early venous filling of the sphenoparietal sinus (Fig. 2). In the venous phase, the anterior temporal diploic vein (ATDV) showed retrograde upward slow flow to high convexity. Left external carotid angiography

1Department of Neurosurgery, Nagasaki University Graduate School of Biomedical Sciences, Nagasaki, Nagasaki, Japan
2Department of Neurology and Strokology, Nagasaki University Graduate School of Biomedical Sciences, Nagasaki, Nagasaki, Japan

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demonstrated a MMAVF located immediately distal to the foramen spinosum, fed by the anterior branch of the left MMA. The shunting flow rate was high. Venous drainage flowed into the cavernous sinus through the sphenoparietal sinus and through the ATDV with retrograde upward flow to high convexity. Angiography showed a to-and-fro stream of the ATDV in the late phase, indicating venous congestion and resulting in venous infarction in the left cerebral hemisphere. We subsequently performed coil embolization for obliteration of the fistulous point. To prevent retinal artery occlusion or

Fig. 1  (a) Admission computed tomography (CT) scan showing diffuse traumatic subarachnoid hemorrhage of the whole cerebrum and a left subdural hematoma within a hygroma. (b) Bone-window CT scan shows a linear skull fracture (arrow) in the right temporoparietal region. (c and d) Five days after admission, plain CT showed multiple low-density areas in the left hemispheric cerebrum and slight brain edema with disappearance of the subdural hematoma.

Fig. 2  (a) Four-dimensional anteroposterior computed tomography angiography (4D-CTA) revealed abnormal blood flow in the middle meningeal artery (MMA) (arrow) with a continuous stream toward the median (arrowheads). (c) Lateral CT angiography also revealed a dilated MMA (arrow) and cavernous sinus detected in the late arterial phase (arrowheads). Left (b) anteroposterior and (d) lateral external angiography showed an AVF fed by the anterior branch of the MMA (arrow). The adjacent middle meningeal sinus was caught by arterial blood, shaping the fistulous point into a dilated high-flow tangle. Venous drainage flowed into the cavernous sinus (asterisk) through the sphenoparietal sinus (small arrowheads) and through the anterior temporal diploic vein (large arrowheads) in a retrograde upward manner. (e) 4D-CTA detected late-venous phase anterior temporal diploic vein (ATDV) filling rather than normal venous return (arrow). (f) Left lateral external angiography showed to-and-fro stream of the ATDV in the late venous phase (arrowhead), indicating venous congestion and resulting in venous infarction in the left cerebral hemisphere. AVF: arteriovenous fistula.
of traumatic MMAVFs of non-fractured sites and found 12 cases, including the present (Table 1).5–15) Two patients presented “aggressive type” symptoms such as intraparenchymal hemorrhage and venous infarction.11) The duration of the asymptomatic phase varied widely from 1 day to 3 months. Among three cases involving an accompanying MMA pseudoaneurysm,8,12,13) two cases were detected by 3D-CTA and one was incidentally revealed by digital subtraction angiography (DSA). Most patients were managed by transarterial embolization of the fistula with good obliteration. We performed 4D-CTA because MRI was contraindicated for our patient (an iatrogenic magnetic substance precluded MRI). 4D-CTA is characterized by both the noninvasive nature of CTA and dynamic acquisition of DSA.

Schechter et al.16) proposed prognostic angiographic classifications of epidural hemorrhage. Among four classifications, the good prognostic group was characterized by extravasation passing into adjacent venous channels as an arteriovenous fistula. This hemodynamic feature was proposed as a protective mechanism. Arterial blood from the injured MMA passes into adjacent venous sinuses or diploic veins to minimize hematoma formation within the epidural space. This could work like a brain-preservation system and might partly contribute to the so-called “lucid interval.” However, traumatic MMAVFs, including that in the present case, often cause remote intracranial complications and neurological symptoms of delayed onset depending on the drainage route and drainage flow rate. Flow dynamics of 4D-CTA in our case detected that epidural extravasation of contrast medium from focal injured MMA remained behind focally and drained into venous system, and venous congestion of ATDV appeared as a slow upward flow of ATDV in the late venous phase. A delayed onset of venous infarction has not been previously reported, and its potential risk has not been adequately discussed. Plain CT investigations in the acute phase of head trauma are strongly recommended to confirm bleeding and swelling, but detection of complicated vascular anomalies often requires detailed hemodynamic information. DSA is currently the first choice for diagnosis of vascular anomalies. MRI evaluation of flow dynamics is limited.

**Fig. 3** (a) Transarterial coil embolization of the fistulous point and main trunk of the middle meningeal artery (MMA) was performed uneventfully. Left external angiography demonstrated good obliteration of the AVF. (b) Postoperative computed tomography (CT) (bone window) showed coil materials lining the main trunk of the MMA near the foramen spinosum. (c and d) Follow-up CT showed residual low-density area in left hemisphere. AVF: arteriovenous fistula.
Table 1: Cases of traumatic middle meningeal arteriovenous fistulas of a non-fractured site

| Author                  | Year | Age/Sex | Impact side | Traumatic pseudoaneurysm | Lesion side | Symptom | Draining route | Interval between trauma and symptom onset | Treatment | Obliteration |
|-------------------------|------|---------|-------------|--------------------------|-------------|---------|----------------|------------------------------------------|-----------|--------------|
| Pakarinen et al.         | 1981 | M57     | Right       | None                      | Right       | Tinnitus | CS             | A few days                               | TAE Coil  | Yes          |
| Sakai et al.             | 1982 | M50     | Right       | None                      | CS          | CS       | CS             | 16 days                                  | TAE       | Yes          |
| Touho et al.             | 1995 | M25     | Right       | None                      | Right       | Tinnitus | CS             | None                                     | Spontaneous Closure                  |
| Tsutsumi et al.          | 2002 | M23     | Right       | None                      | Right       | Tinnitus | PVP            | None                                     | TAE       | Yes          |
| Liu et al.               | 2008 | M22     | Right       | None                      | Right       | Tinnitus | PVP            | None                                     | TAE       | Yes          |
| Abla et al.              | 2011 | M45     | Left        | None                      | Left        | Tinnitus | PVP            | 8 days                                   | Craniotomy TAE                        |
| Ko et al.                | 2017 | M50     | Left        | None                      | Left        | Tinnitus | PVP            | None                                     | TAE       | Yes          |
| Yu et al.                | 2017 | M30     | Left        | None                      | Left        | Tinnitus | PVP            | None                                     | TAE       | Yes          |
| Tokairin et al.          | 2019 | M69     | Right       | None                      | Right       | Tumor    | PVP            | 5 days                                   | TAE Coil Onyx                        |
| Present case             | 2019 | M69     | Right       | None                      | Right       | Venous infarction | 5 days                       | TAE       | Yes          |

However, DSA is invasive, relatively expensive, and time-consuming and has a risk of embolic events. 4D-CTA is a novel technique that might serve as an alternative tool for detecting various types of vascular anomalies.13) Compared with DSA, 4D-CTA was congruous with exact feeder, shunting point, and draining veins, and draining flow congestion having potential risk, as with DSA. Some reports have indicated that 4D-CTA could come to the fore as a noninvasive diagnostic tool for cranial dural arteriovenous fistulas.18,19)

After endovascular treatment, most patients have a good outcome.

Conclusion

We have herein reported a traumatic MMAVF of a non-fractured site that led to delayed venous infarction. Rapid assessment of trauma-associated vascular anomalies should be performed in the acute phase. 4D-CTA could be an alternative to DSA for the diagnosis, treatment, and follow-up of MMAFVs.

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Conflicts of Interest Disclosure

All authors declare that they have no conflicts of interest.

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Corresponding author:
Yusuke Iki, MD, Department of Neurosurgery, Nagasaki University Graduate School of Biomedical Sciences, 1-7-1, Sakamoto, Nagasaki, Nagasaki 852-8501, Japan.
✉ yusuke.nagasaki1119@gmail.com