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Case Report

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

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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)] 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 (Iomeron300, Eisai, Tokyo, Japan) at a 4-mL/s was completed by automated antecubital venous injection. The study protocol contained non-helical scanning method, 80 mV, 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 vasospastic 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

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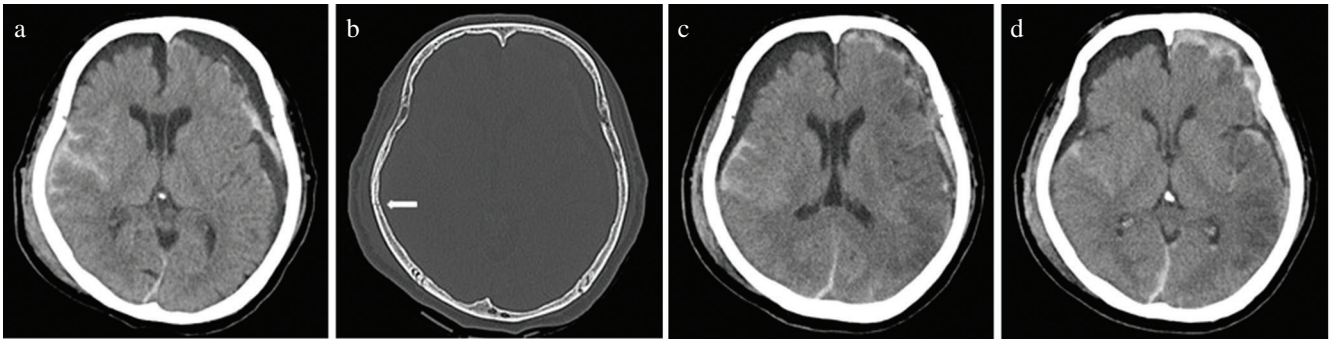

Fig. 1 (a) Admission computed tomography (CT) scan showing diffuse traumatic subarachnoid hemorrhage of the whole cerebrum and a left subdural hematoma within a hygroma. (b) Bone-window CT scan shows a linear skull fracture (arrow) in the right temporoparietal region. (c and d) Five days after admission,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

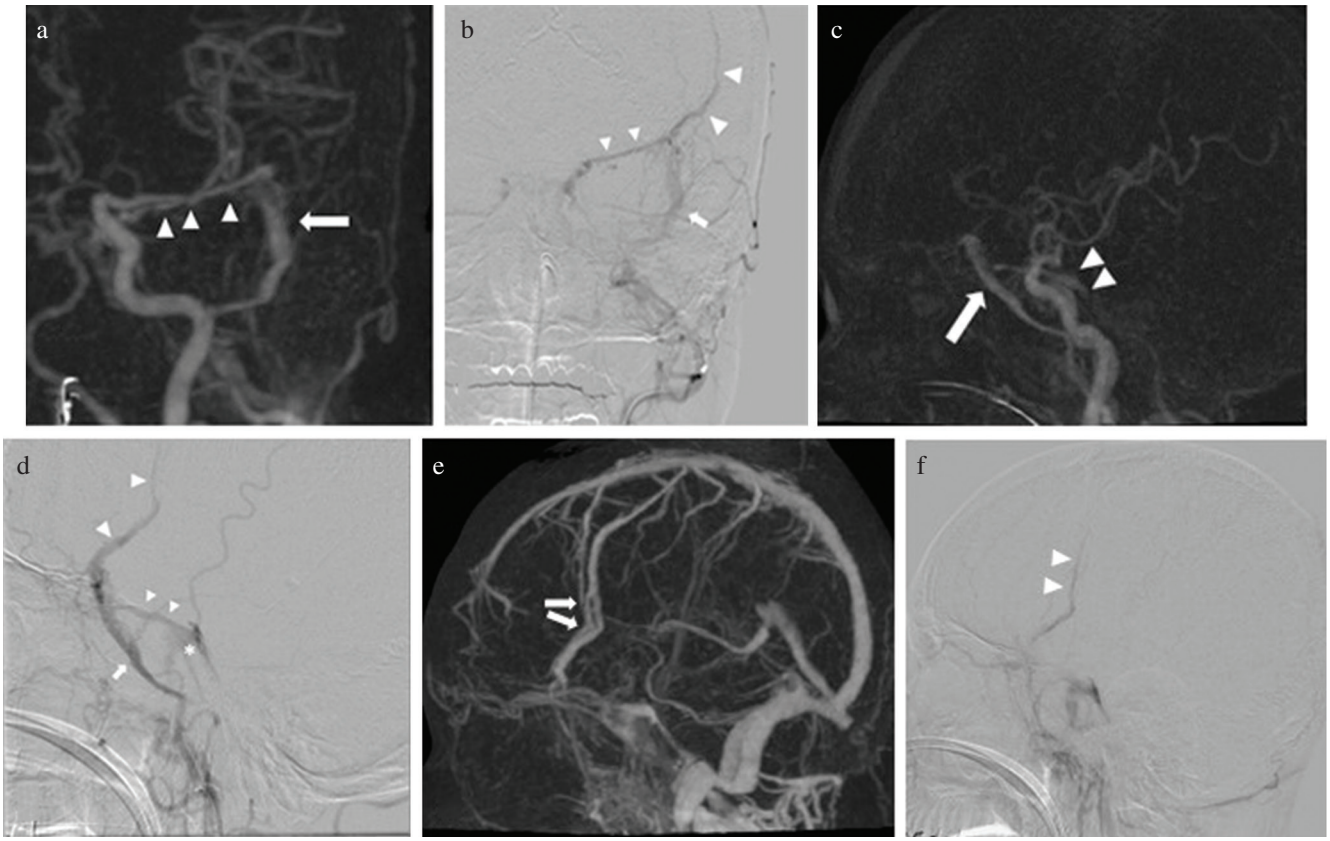

Fig. 2 (a) Four-dimensional anteroposterior computed tomography angiography (4D-CTA) revealed abnormal blood flow in the middle meningeal artery (MMA) (arrow) with a continuous stream toward the median (arrowheads). (c) Lateral CT angiography also revealed a dilated MMA (arrow) and cavernous sinus detected 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.

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

facial nerve palsy, we avoided liquid embolic materials such as like *n*-butyl-2-cyanoacrylate. We performed transarterial embolization through the transfemoral route. A microcatheter (Excelsior SL1018; Stryker Neurovascular, Fremont, CA, USA) was delivered to the site immediately distal to the fistulous point, and a detachable coil (Target XL; Stryker Neurovascular) was used to occlude the main trunk of the MMA near the foramen spinosum (Fig. 3). We confirmed disappearance of the shunt. Postoperative MRI showed that the fistula was obliterated. After the operation, the patient recovered well from the aphasia and motor weakness (right upper and lower limb, grade 4/5); however, the low-density area was still present on follow-up CT. Three weeks after the operation, he was transferred to a rehabilitation institute.

Discussion

We have herein described a rare case of traumatic MMAVF that caused delayed venous infarction. Intracranial venous congestion caused the venous infarction, which was detected by 4D-CTA. This is the first case report of a traumatic MMAVF detected by 4D-CTA based on flow dynamics.

Tearing of the middle meningeal vessels with a linear fracture crossing them usually causes traumatic MMAVFs.[1)] Traumatic MMAVFs of non-fractured sites are rare. This pathophysiology is associated with dura mater–bone separation, in which external forces cause detachment of the dura mater from the inner layer of the skull.[2)] In childhood, fibrous tissue is generally replaced by bone tissue in the bone structure region and skulls are more elastic in children. Deformation causing detachment without fracture more easily occurs in children than adults. The MMA runs between the two middle meningeal veins (sphenobregmatic sinuses).[3)] The outer membrane of these sinuses is adhesive to the skull on the vascular groove, where dura mater–bone separation triggers vascular injuries. Histologically, a media defect is often seen on the bifurcation of the MMA, resulting in fragility against traumatic impact.[4)]

The clinical features, natural history, and ideal treatment of traumatic MMAVFs without skull fracture remain unclear. We reviewed PubMed to identify previous reports 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.

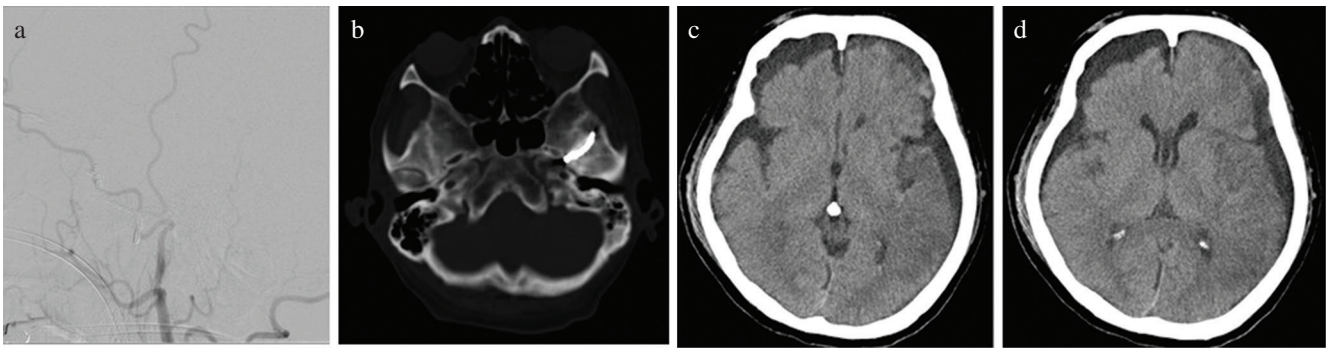

Fig. 3 (a) Transarterial coil embolization of the fistulous point and main trunk of the middle meningeal artery (MMA) was performed uneventfully. Left external angiography demonstrated good obliteration of the AVF. (b) Postoperative computed tomography (CT) (bone window) showed coil materials lining 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	Lesion side	Symptom	Draining route	Interval between trauma and symptom onset	Traumatic pseudoaneurysm	Treatment	Embolic materials	Obliteration
Pakarinen et al.[5]	1965	25/M	N/A	Right	Tinnitus headache	SPS CS	14 hours	None	Ligation of ECA		Yes
Satoh et al.[6]	1982	75/F	Right	Right	Headache nausea	SSS CS	7 days	None	Spontaneous Closure		Yes
Touho et al.[7]	1995	27/M	N/A	Right	Tinnitus	CS	A few days	None	TAE	Coil	Yes
Tsutsumi et al.[8]	2002	23/M	Right	Left	Tinnitus chemosis	CS	16 days	Accompanied	TAE	Coil	Yes
Liu et al.[9]	2008	22/M	N/A	Left	Blurred vision exophthalmos diplopia blepharoptosis	SPS CS CVs	1 month	None	TAE	Detachable balloon	Yes
Takeuchi et al.[10]	2009	21/M	Right	Left	Tinnitus exophthalmos	SPS CS OVs	3 months	None	TAE	Coil	Yes
Abla et al.[11]	2011	9/M	Left	Left	IPH	OVs	8 days	None	Craniotomy TAE	NBCA	Yes
Ko et al.[12]	2014	24/M	Right	Left	Tinnitus	PVP	25 days	Accompanied	TAE	Coil	Yes
Park et al.[13]	2017	69/M	Left	Left	N/A	OVs	N/A	Accompanied	TAE	NBCA	Yes
Yu et al.[14]	2017	8/M	N/A	Left	Tinnitus exophthalmos chemosis	PVP	3 months	None	TAE	Coil Onyx	Yes
Tokairin et al.[15]	2019	24/M	Left	Right	Tinnitus	SPS CS	1 day	None	TAE	Coil NBCA	Yes
Present case	2019	69/M	Right	Left	Venous infarction	SPS CS DVs	5 days	None	TAE	Coil	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.[17] 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 MMAVFs.

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Conflicts of Interest Disclosure

All authors declare that they have no conflicts of interest.

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