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## Case report

# Intra-operative emergence of occult dural arteriovenous fistula after middle meningeal artery embolization for chronic subdural hematoma: Case report and literature review ☆,☆☆,★

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## ABSTRACT

Dural arteriovenous fistulae of the middle meningeal artery (MMA-dAVF) are high risk lesions that can lead to intracranial hemorrhage. We describe the case of an adult male that presented with chronic subdural hematomas and was treated with burr hole craniotomy plus middle meningeal artery (MMA) embolization. Although the pre-embolization angiogram showed no signs of a fistula, a fistula arising from the MMA and draining into the superior sagittal sinus emerged intra-operatively. To our knowledge, this is the first case of intra-operative emergence of occult MMA-dAVF with intracranial drainage during MMA embolization for chronic subdural hematoma treatment. This observation supports monitoring for and embolizing spontaneous MMA-dAVF following MMA embolization.

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## Introduction

Dural arteriovenous fistulae arising from the middle meningeal artery (MMA-dAVF) are rare; roughly 41 case reports yielding a total of 61 patients have been described since 1951 [1]. The majority of reported cases are traumatic or

iatrogenic in nature [2,3]. MMA-dAVF have proven to be high-risk lesions that can lead to intracranial hemorrhage [1,2]. Although isolated reports of spontaneous resolution exist [4], treatment is often indicated when MMA-dAVF are identified and is typically achieved by trans-arterial embolization of the fistula.

Embolization of the MMA for treatment of chronic subdural hematomas (cSDH) is being actively investigated as a means

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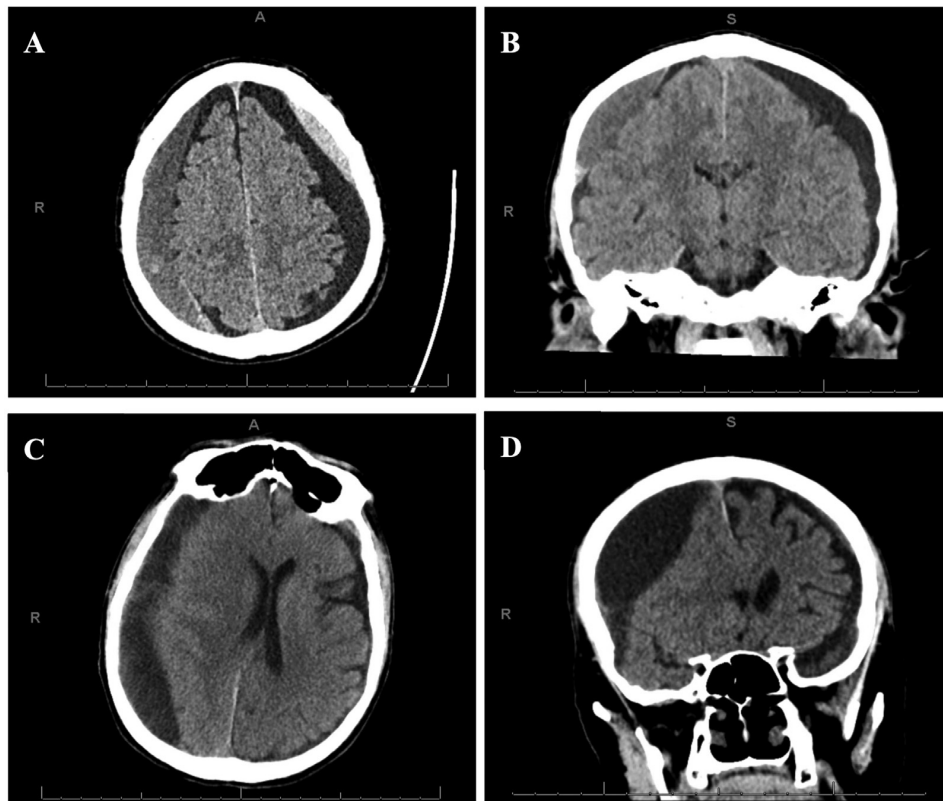
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**Fig. 1 – Head Computed Tomography (CT) on initial and subsequent emergency department presentation. On initial presentation to the ED after sustaining a fall from standing: axial (A) and coronal (B) CT showed bilateral mixed density chronic subdural hematomas (cSDH) with no midline shift. Upon re-presenting to the emergency department a month later after sustaining another fall: axial (C) and coronal (D) CT showed enlargement of right-sided cSDH measuring 3 cm with 1 cm of midline shift.**

of reducing hematoma recurrence and improving clinical outcomes [5]. To our knowledge, only one previous report of 2 cases of sudden MMA-dAVF emergence following MMA embolization exists, both with extracranial drainage [6]. Here, we describe a case of sudden intra-operative emergence of a previously occult MMA-dAVF with drainage to the superior sagittal sinus during MMA embolization.

## Case report

### Presentation

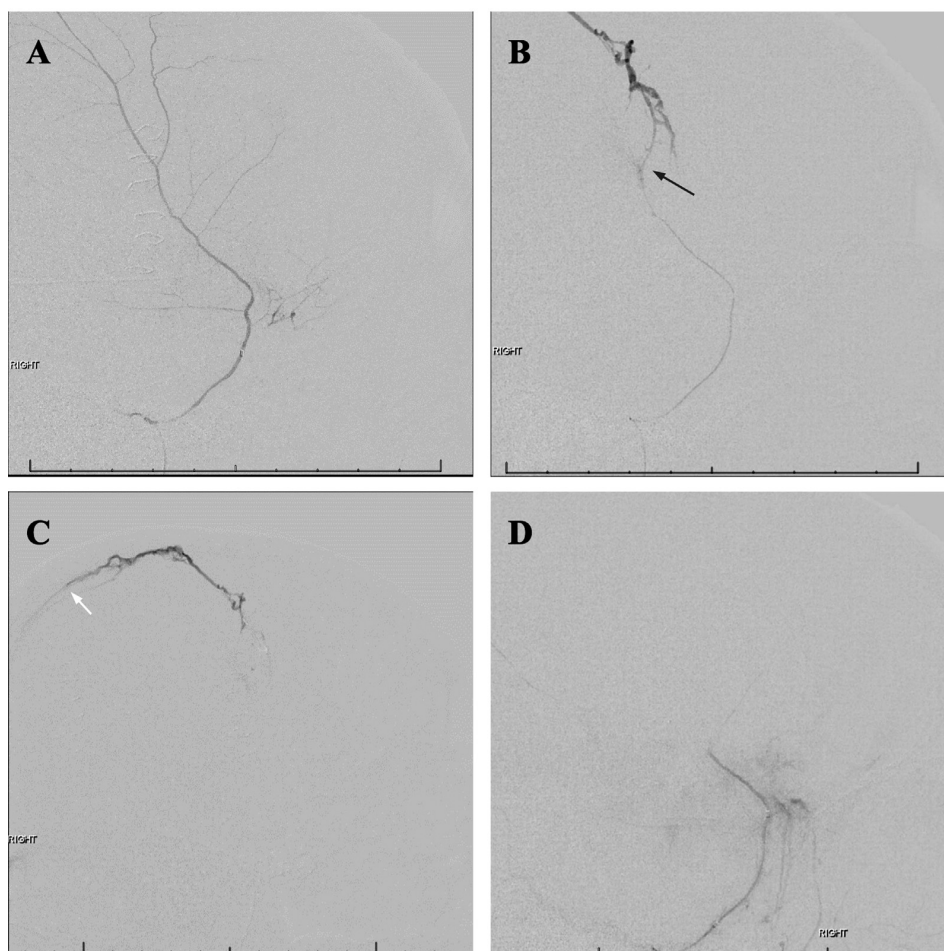
A chronically ill-appearing adult male presented to our emergency department after sustaining a fall from standing. Past medical history was significant for laryngeal cancer status post total laryngectomy. There was no history of antiplatelet or anticoagulation medication. A non-contrast computed tomography scan showed bilateral 15mm mixed density cSDH without midline shift (Figs. 1A and B). The patient was neurologically intact without symptoms of elevated intracranial pressure, and he wished to avoid surgical drainage unless absolutely necessary. Surgical drainage was therefore

deferred and enrollment in our active MMA embolization trial for treatment of cSDH was offered. The patient declined all intervention preferring close outpatient follow up. Unfortunately, a month later the patient re-presented to our emergency department after sustaining another fall. A head computed tomography showed significant interval enlargement of the right cSDH measuring up to 3cm with 1cm of midline shift and a much smaller left cSDH measuring up to 4mm (Figs. 1C and D). He was alert but had mild left sided weakness. Surgical drainage was once again offered and this time he and his family agreed to proceed.

The patient underwent right burr hole craniotomy for evacuation of cSDH with subdural drain placement. He regained full strength in all extremities and the subdural drain was removed on post-operative day 2. Subsequently, the patient was again offered participation in our active MMA embolization trial (clinicaltrials.gov, ID:NCT04270955). He accepted, was randomized to the intervention group, and underwent MMA embolization on post-operative day 5.

### MMA embolization procedure

The patient was placed under general anesthesia and the right internal and external carotid arteries were sequentially



**Fig. 2 – Intraoperative angiograms obtained during MMA embolization. The dural-arteriovenous fistula of the middle meningeal artery (MMA-dAVF) was occult on the pre-embolization injection of the MMA (A). During embolization of the MMA, however, the MMA-dAVF emerged (black arrow, B), draining into the superior sagittal sinus (white arrow, C). Subsequently, the MMA-dAVF was embolized intraoperatively to complete stasis (D).**

accessed via the right radial artery. A biplane angiogram of the external carotid artery and MMA showed no evidence of pre-operative dAVF (Fig. 2A). Following the initial infusions of 250-micron Embosphere particles, a repeat microcatheter angiogram showed new arteriovenous shunting from one of the distal branches of the MMA (Figs. 2B and C). The MMA-dAVF was subsequently embolized with 500-micron particles. Complete stasis of the MMA-dAVF was achieved (Fig. 2D).

#### Outcome

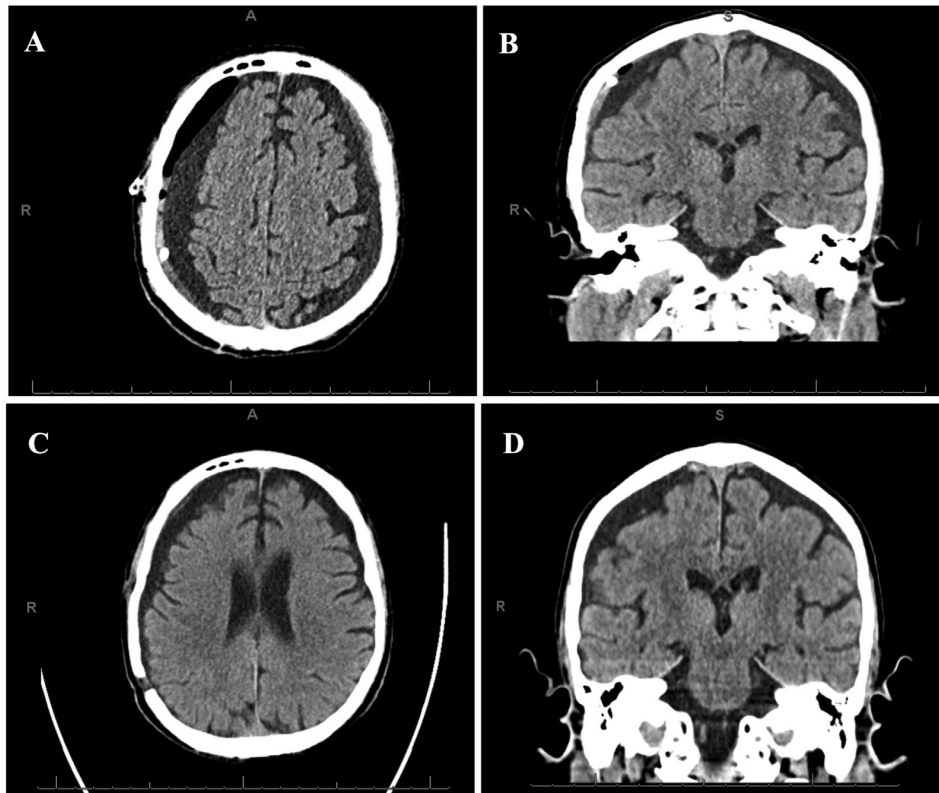
Imaging after cSDH evacuation showed a decrease in size of the right subdural hematoma and resolution of midline shift (Figs. 3A and B). The patient returned to his neurologically intact baseline. The patient was discharged the day after MMA embolization (post operative day 6) in neurologically intact condition. Outpatient follow-up evaluation up to 4 months after surgical drainage and MMA embolization showed further decreased size of the right SDH (Figs. 3C and D). Unfortunately, shortly after his last outpatient follow up the patient suffered

yet another fall down a flight of stairs with resultant critical poly-trauma requiring ventilation and multiple surgeries which eventually culminated in his death.

#### Discussion

We have described the case of an adult male who underwent right-sided MMA embolization for treatment of right cSDH after burr hole craniotomy. Although a pre-embolization selective microcatheter angiogram showed no signs of a MMA-dAVF, the fistula became apparent intraoperatively upon partial MMA embolization. The MMA-dAVF was subsequently successfully embolized intraoperatively. Follow-up at up to 4 months showed significant improvement of right cSDH.

To our knowledge, this is the first report of intraoperative emergence of “spontaneous” MMA-dAVF with intracranial drainage following MMA embolization, and the third reported case of intra-operative MMA-dAVF emergence during MMA



**Fig. 3** – Head CT on postoperative (craniotomy) day one (A&B) and 4 months follow-up (C&D). Axial (A) and coronal (B) CT obtained on postcraniotomy and drain placement day one showed a decrease in size of right subdural hematoma to 1.9 cm (previously 3 cm) with resolution of midline shift (previously 1 cm). A repeat CT (C: axial, D: coronal) at 4 months follow-up showed a decreased 1.2 cm right sided subdural collection.

embolization overall. Unlike the present report, the 2 previous cases of MMA-dAVF emergence reported by Piergallini et al. [6] drained extracranially (see Table 1 for summary). Intracranial drainage of MMA-dAVF is unique and important in that it increases the risk of cortical venous hypertension which in turn is associated with significant risk of hemorrhage and neurologic morbidity [7,8].

We offer 3 competing hypotheses for the apparently “spontaneous” formation of MMA-dAVF subsequent to MMA embolization. First, physiologic arteriovenous shunts have been shown to be present in the dura [9]. These shunts are “physiologic” in that they form and regress or occlude spontaneously. These physiologic fistulae do not typically exceed 12 $\mu$ m, and are thus occult on catheter angiograms. As reported by Piergallini et al. [6] MMA embolization can lead to increased arterial pressure and blood flow to the arteriovenous shunt, hence resulting in what can appear to be a “spontaneous” formation of MMA-dAVF [6]. Second, it is possible that local inflammation and angiogenesis due to the burr hole craniotomy itself resulted in the formation of an MMA-dAVF given that the fistula was in close proximity to the margin of the craniotomy. A fistula formed post operatively might have become apparent secondary either due to true recanalization or due to in-

creased pressure with the intraprocedural contrast injection after partial occlusion of distal MMA branches. A third possibility is that the fistula formation was iatrogenic. A puncture of the artery by the guiding wire or microcatheter could produce a small fistula or frank extravasation, with the latter being a more common event. Since the location of the fistula was not in relative proximity to where the catheter wire was placed it is unlikely that any potential vessel wall trauma caused by the catheter wire could account for the fistula formation [10]. We perform the particle embolization with low pressure infusion of particles and the postembolization angiograms are hand-injected under low pressure. Additionally, although supraphysiologic pressures exerted on the vessel during injection of embolizing particles can result in iatrogenic complications, inadvertently high infusion rates typically manifest as an extravasation of contrast fluid out of the vessel rather than formation of an arteriovenous fistula.

In this case the MMA and MMA-dAVF were embolized to complete stasis with good short term clinical outcome and no recurrence over 4 months of follow-up. It should be noted that long-term efficacy of MMA-dAVF occlusion via particle embolization remains unclear, as does whether or not physiologic MMA-dAVF contribute or predispose to hemorrhages. Never-

**Table 1 – Summary of case reports of MMA-dAVF emergence following MMA embolization.**

	Patient 1	Patient 2*	Patient 3*
Age	67	81	83
Sex	Male	Female	Female
Hematoma Etiology	Bilateral (L = 4 mm, R = 30 mm)	Unilateral (L = 37 mm)	Bilateral (L = 26 mm, R = 10 mm)
Embolization side and type	Right MMA Particle	Left MMA Particle	Left MMA Particle
MMA-dAVF Drainage	Superior Sagittal Sinus	Pterygoid plexus	External temporal fossa
Outcome (Radiographic/Clinical)	<b>Radiographic:</b> Decrease in right sided subdural hematoma size and continued resolution of midline shift at 4 months. <b>Clinical:</b> Regained strength in left extremities and discharged neurologically intact.	<b>Radiographic:</b> No recurrence of subdural hematoma at 3 months. <b>Clinical:</b> Discharged in neurologic state “similar to that before the admission”	<b>Radiographic:</b> No recurrence of subdural hematoma at 3 months. <b>Clinical:</b> Discharged in neurologic state “similar to that before the admission”

MMA: Middle Meningeal Artery; dAVF: Dural Arteriovenous Fistula;  
\* Patients 2 and 3 were reported by Piergallini et al., 2019.<sup>6</sup>

theless, our experience supports monitoring for and embolizing spontaneous MMA-dAVF following MMA embolization.

### Patient consent

The patient is deceased and attempts to reach family/next of kin have been unsuccessful. Institutional IRB authorization to produce this report has been obtained and no personally identifiable information have been included in the report.

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