



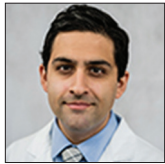
Case Report

Incidentally discovered dural arteriovenous fistula during middle meningeal artery embolization for the treatment of chronic subdural hematoma

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ABSTRACT

Background: The incidence of chronic subdural hematoma (CSDH) is increasing with population age and anti-coagulant use. Embolization of the middle meningeal artery (MMA) is an emerging, less invasive alternative to open surgery in treating this condition. Dural arteriovenous fistula (DAVF) is a rare condition whose association with CSDH is not well understood. We present three cases with incidentally discovered DAVFs during MMA embolization for the treatment of CSDH that necessitated adjustments to initial treatment strategy.

Case Descriptions: We retrospectively reviewed all MMA embolizations performed for the treatment of CSDH beginning in 9/2019 to 11/2020. Imaging and hospital course of three cases of incidentally discovered DAVF, including patient demographics, clinical presentation, methods of treatment, imaging and outcome were assessed. Thirty MMA embolizations were performed as primary or adjunct treatment of CSDH. DAVF was discovered angiographically in 3 (10%) cases. All patients reported a history of prior closed head injury, although the timing of injury and subdural blood product age did not correlate in 2 of the 3 cases. All subjects experienced complete symptomatic and radiographic resolution of the subdural hematoma and DAVF following intervention.

Conclusion: As MMA embolization for CSDH becomes more frequent, so may the incidental diagnosis of DAVF. Awareness of this potential association is critical to diagnosing DAVF with angiography and altering treatment strategies as needed.

Keywords: Dural arteriovenous fistula, Embolization, Endovascular, Middle meningeal artery, Subdural hematoma

INTRODUCTION

Chronic subdural hematoma (CSDH) is a common disease among elderly population, and the prevalence continues to rise with trends toward increased life expectancy of the general population.^[2,5] Although surgical evacuation through burr holes or conventional craniotomy remains the treatment of choice for large symptomatic CSDH, embolization of the middle meningeal artery (MMA) is emerging as an increasingly common treatment strategy.^[3,12] Dural arteriovenous fistulas (DAVFs) are abnormal shunts between dural arteries and venous sinuses.

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Venous sinus thrombosis, prior surgery, and trauma^[7,9,10] have all been implicated in DAVF formation, presumably through inflammatory and neoangiogenic-mediated recanalization of extant arteriovenous anastomoses.^[11]

To date, no study has addressed the association between DAVF and CSDH; with increased adoption of MMA embolization, however, this topic now warrants further investigation and awareness. We present three cases in which DAVF was discovered incidentally during MMA embolization for CSDH and required adjustments to the treatment plan.

MATERIALS AND METHODS

Among 65 patients evaluated in consultation or admitted to the neurosurgery service from September 2019 to November 2020, we performed 30 MMA embolizations (46%) as primary or adjunct treatment for CSDH. All cases were performed at a single institution by a fellowship-trained neurointerventionalist (JGAJ). Among all cases, 3 (10%) were found to harbor ipsilateral DAVF on angiography. The patient specific characteristics, presenting symptoms, hospital course, procedural details, operative reports, images, and outcomes were reviewed. IRB approval was obtained based on institutional protocol.

ILLUSTRATIVE CASES

Case 1

A 66-year-old male with a recent syncope-related fall and closed head injury presented with progressively worsening gait instability, dizziness, and headaches. The fall occurred 1 week before presentation. On examination, no focal neurologic deficits were found. Noncontrast head CT demonstrated a 7 mm right convexity mixed density subdural hematoma [Figures 1a and b]. Given his significant headaches and progressive functional decline, MMA embolization was performed as primary treatment. Intraprocedural digital subtraction angiogram revealed a right parietal Cognard Grade I DAVF [Figures 1c and d]. Given the absence of cortical venous reflux and benign natural history of the fistula, the remainder of the procedure did not deviate from routine MMA embolization. The right temporal and right frontoparietal branches of the MMA were embolized with n-BCA glue with indirect penetration of liquid embolic into the fistulous point. Following embolization, there was no further opacification of the fistula. The patient experienced no procedure-related complication and was discharged the subsequent day. At 1-month follow-up, there was complete resolution of his presenting symptoms and surveillance CT demonstrated no residual SDH.

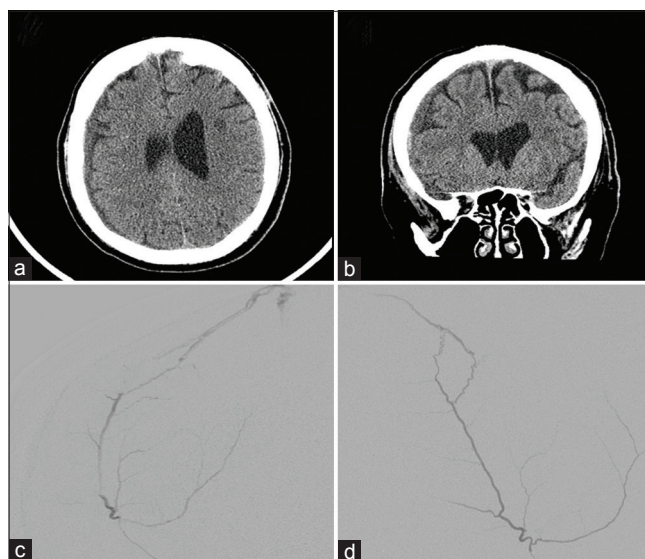


Figure 1: (a and b) Axial and coronal noncontrast CT demonstrates a 7 mm right convexity mixed density subdural hematoma. (c and d) Anteroposterior and lateral projections, superselective catheter angiogram, and right middle meningeal artery frontoparietal branch injection demonstrating a right parietal dural arteriovenous fistula draining into the superior sagittal sinus.

Case 2

A 77-year-old male with a recent syncope-related fall and closed head injury presented with gradually worsening isolated right upper extremity monoparesis. The fall occurred 1 month before presentation. On examination, the patient demonstrated 4/5 strength in his right upper extremity, with no additional neurologic deficits. Noncontrast head CT demonstrated bilateral mixed density convexity subdural hematomas [22 mm on the left and 14 mm on the right; Figures 2a and b]. The patient underwent a left craniotomy for evacuation of the larger hematoma, in conjunction with bilateral MMA embolization as primary treatment for the right convexity subdural hematoma in addition to limiting potential recurrence along the left convexity. Intraprocedural digital subtraction angiogram revealed two Cognard Grade III DAVFs, one arising from the parietal branch of the right MMA draining into an occipital cortical vein, and the second arising from the temporal branch of the left MMA draining into the vein of Labbe [Figures 2c-f]. Given the presence of cortical venous reflux and associated risk of future intracranial hemorrhage, treatment of the fistulae was indicated. This was approached in two staged procedures. Following an uncomplicated left MMA embolization (frontoparietal branch), the incidental findings were discussed with the patient and he underwent a second stage embolization for the DAVF on a later date. During the second stage, a more prolonged infusion of liquid embolic was employed to ensure penetration into the recipient cortical venous outflow, resulting in nonopacification of



Figure 2: (a and b) Axial and coronal noncontrast CT demonstrates bilateral mixed density convexity subdural hematomas. (c and d) Anteroposterior and lateral views, superselective catheter angiogram, and right middle meningeal artery parietal branch injection demonstrating a dural arteriovenous fistula (DAVF) draining into an occipital cortical vein. (e and f) Oblique anteroposterior and lateral views, superselective catheter angiogram, and left middle meningeal artery temporal branch injection demonstrating a DAVF draining into vein of Labbe.

the aforementioned fistulae. The postoperative course was complicated by seizures which resolved with uptitration in antiepileptic medications. At 6-month follow-up, there was complete resolution of the patient's presenting symptoms and surveillance CT demonstrated no residual SDH.

Case 3

A 59-year-old male with a recent fall and closed head injury presented with progressively worsening headaches. The fall occurred 9 days before presentation. On examination, no neurologic deficits were found. Noncontrast head CT demonstrated a 5 mm left convexity CSDH [Figure 3a]. He underwent burr hole evacuation and was discharged on the subsequent day. He presented again on postoperative

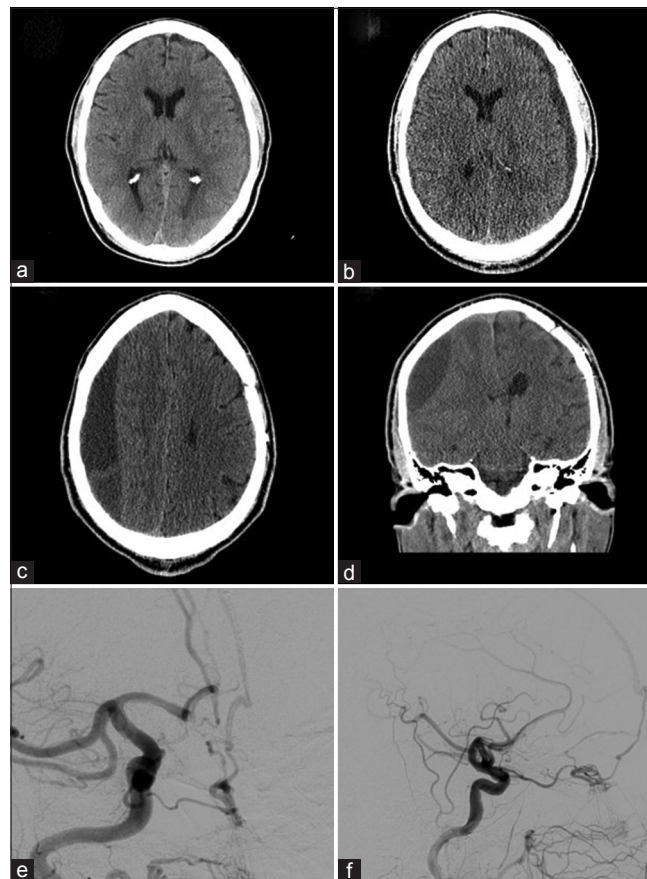


Figure 3: (a) Axial noncontrast CT demonstrates 5 mm left convexity chronic subdural hematoma corresponding to the patient's initial presentation. (b) Axial noncontrast CT demonstrates interval increase in size of the left convexity subdural hematoma. (c and d) Axial and coronal noncontrast CT performed 1 month later demonstrates a new large right subacute on chronic subdural hematoma. (e and f) Anteroposterior and lateral views, selective catheter angiogram, and right common carotid artery injection demonstrating a cribriform plate dural arteriovenous fistula with ophthalmic artery feeders draining into a right frontal cortical vein.

day 6 with recurrence of his headaches. Noncontrast head CT demonstrated recurrence of the left convexity subdural hematoma [Figure 3b]. He subsequently underwent a left craniotomy for evacuation. He was discharged on postoperative day 2 with no postprocedural complications. At 1-month follow-up, he continued to complain of severe headaches. Surveillance CT demonstrated a new 27 mm right convexity mixed density subdural hematoma [Figures 3c and d]. He subsequently underwent right MMA embolization as primary treatment. Intraprocedural digital subtraction angiogram revealed a Cognard Grade III cribriform plate DAVF supplied by the right ophthalmic artery draining into a right frontal cortical vein [Figures 3e and f]. The right MMA temporoparietal and frontal branches were embolized with n-BCA glue. Follow-up was arranged

to treat the DAVF at a later date and he was discharged the following day. The patient was subsequently lost to follow-up, but presented to the emergency department 4 months later with diplopia and a right abducens nerve palsy. Noncontrast head CT demonstrated significant reduction in size of the previously embolized right convexity subdural hematoma. He was admitted and underwent attempted endovascular treatment of the DAVF. Catheterization of the arterial and venous sides of the fistula achieved suboptimal positioning for liquid embolic embolization and so he underwent a right craniotomy for surgical ligation of the fistula. The patient experienced no postprocedural complications. At 2-week follow-up, there was complete resolution of his presenting symptoms and surveillance CT demonstrated no residual SDH.

DISCUSSION

Although it may often be assumed that an initial trauma incites both DAVF and CSDH, as in the patients described herein, the causal relationship between these entities remains unclear. Patients 1 and 3 endorsed recent head trauma but their subdural hematoma appeared chronic; patient 3 developed a new contralateral CSDH without intervening head trauma in the setting of a known DAVF. Nevertheless, this report underscores the importance of angiographically assessing for DAVF before MMA embolization. DAVF is often identified during angiography performed after recent head trauma.^[6] One case series associated CSDH with lower grade DAVFs, which are traditionally perceived to carry a

more benign natural history.^[8] Yet two-thirds of patients in this series harbored high-grade fistulae which required adjustments to the initial embolization plan [Table 1]. Microparticle sizes typically used in MMA embolization may travel through DAVF to the lungs, whereas liquid agents can occlude functional venous outlets when infused too distal or prevent future access to the fistula in prematurely terminated injections.

Further, investigation into whether CSDH predisposes to DAVF, or vice versa, represents an important research topic that has potential to increase our understanding of these diseases. DAVFs represent a minority of intracranial vascular malformations. Although historically difficult to quantify, one study determined the detection rate of DAVF among adults to be 0.16/100,000 adults/year.^[1] However, noninvasive imaging is less sensitive in detecting DAVF compared to other vascular pathologies, making the true prevalence difficult to ascertain.^[4] Digital subtraction angiography is rarely utilized as a diagnostic tool for the workup of CSDH. However, factors such as young age, spontaneous occurrence of SDH, absence of coagulopathy or antiplatelet medication use, recurrent, or refractory SDH may all be predictors of an underlying vascular malformation such as DAVF.^[8]

The present series is the first to report multiple cases of DAVFs found incidentally during MMA embolization for the treatment of CSDH (10% of all comers in our series). As MMA embolization emerges as a more common treatment option, there is a growing need to recognize the presence of associated DAVF and adjust treatment strategies as needed.

Table 1: Clinical data of patients with incidentally found DAVF during MMA embolization.

	Case 1	Case 2	Case 3
Age	66	77	59
Gender	Male	Male	Male
Presenting symptom	Headache, gait instability	R upper extremity weakness	Headache, diplopia
Mechanism of injury	Fall	Fall	Fall
SDH (L/R)	7 mm	22 mm/14 mm	5 mm/27 mm
SDH Location	R convexity	Bilateral	Bilateral
SDH treatment indication	Headache, functional decline	Neurologic deficit	Headache, mass effect
Recurrent SDH	No	No	Yes
DAVF vascular supply/venous drainage	R MMA/superior sagittal sinus	R MMA/occipital vein; L MMA/Vein of Labbe	R ophthalmic artery/Right frontal cortical vein
DAVF grade (Cognard)	I	III	III
DAVF treatment indication	None	Hemorrhage risk prevention	Hemorrhage risk prevention
Treatment of SDH	Primary embolization	Craniotomy+embolization	Burr hole+craniotomy+embolization
Treatment of DAVF	Primary embolization (nBCA)	Primary embolization (nBCA)	Craniotomy
Outcome (clinical/radiographic)	Complete resolution	Complete resolution	Complete resolution

SDH: Subdural hematoma, DAVF: Dural arteriovenous fistula, MMA: Middle meningeal artery, R: Right, L: Left

CONCLUSION

We present three cases of incidentally discovered DAVFs during MMA embolization for the treatment CSDH. This series underscores the importance of evaluating for DAVF during initial angiography and making appropriate alterations in treatment strategy. Additional studies are warranted to further investigate the relationship between CSDH and DAVF, as the prevalence of high-grade DAVF might be higher than previously thought among select patients presenting with isolated subdural hematoma.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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