

CASE REPORT

A rare association of cerebral dural arteriovenous fistula with venous aneurysm and contralateral flow-related middle cerebral artery aneurysm

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SUMMARY

The association of cerebral dural arteriovenous fistula (DAVF) and ipsilateral flow related aneurysm has infrequently been reported. We describe a male patient who presented with an acute haemorrhagic stroke and was found to have a large right fronto-parietal intraparenchymal haemorrhage from the ruptured Borden type II DAVF in addition to a large venous aneurysm and a flow related intraosseous aneurysm of the contralateral middle meningeal artery (MMA) all clearly delineated by CT and DSA. He underwent emergency stereotactic evacuation of the intraparenchymal haemorrhage and successful surgical treatment of all the vascular lesions at the same time with residual neurological deficit. To our knowledge, this is the first such reported case. We discuss the challenging surgical treatment, emphasising the role of CT/DSA in management, and provide a literature review.

BACKGROUND

The association of cerebral dural arteriovenous fistula (DAVF) with cerebral aneurysms is rare; the first case was reported by Kaech *et al*¹ in 1987 and subsequently by a few other case reports and series.^{2–9} A previous series described a 60-year-old man who presented with subarachnoid/intraparenchymal haemorrhage and had right DAVF fed by the right middle meningeal artery (MMA) and associated with aneurysms of the right anterior cerebral and left internal carotid-posterior communicating arteries considered to be remote.³ Hashiguchi *et al*¹⁰ also demonstrated that the development of venous aneurysm over time may be a part of the natural history of DAVF and significantly contributed to the haemorrhagic complications.

In this report, we discuss the rare association of ruptured right DAVF with a venous aneurysm and contralateral flow related MMA aneurysm in a patient presenting with a right haemorrhagic stroke. He underwent emergent stereotactic evacuation of the intraparenchymal haemorrhage and bilateral craniotomies for the obliteration of all the vascular lesions at the same time with residual neurological deficit. We discuss the challenging surgical treatment, emphasising the role of CT/DSA in management, and provide a literature review.

CASE PRESENTATION

The ambulance service was called to attend to our patient who is a man in his 60s, who developed sudden onset of right temporal headaches

associated with an 'electric shock' sensation radiating down his left trunk quickly followed by left-sided hemiplegia. He vomited and also had left facial droop and slurring of speech. He was not a known hypertensive and there was no antecedent trauma or use of anticoagulant.

On arrival to the emergency department, he was found to be confused, with altered sensation and florid paralysis of the left upper and lower limbs. There was right and upward deviation of gaze. He was immediately intubated and sedated for time critical transfer to the intensive care unit (ICU) of our hospital, which is the Regional Neurosurgical Centre.

INVESTIGATIONS

On admission, a CT brain showed a large right fronto-parietal intraparenchymal haemorrhage with a midline shift and extravasation of contrast suggesting an underlying vascular anomaly. Cerebral DSA showed Borden type 2 right frontal dural AV fistula supplied by branches of the bilateral middle meningeal and superficial temporal arteries, and also from the left anterior falx artery. A flow related saccular aneurysm of the left MMA was also demonstrated. Venous drainage was into cortical veins and a large venous aneurysm was seen (figure 1).

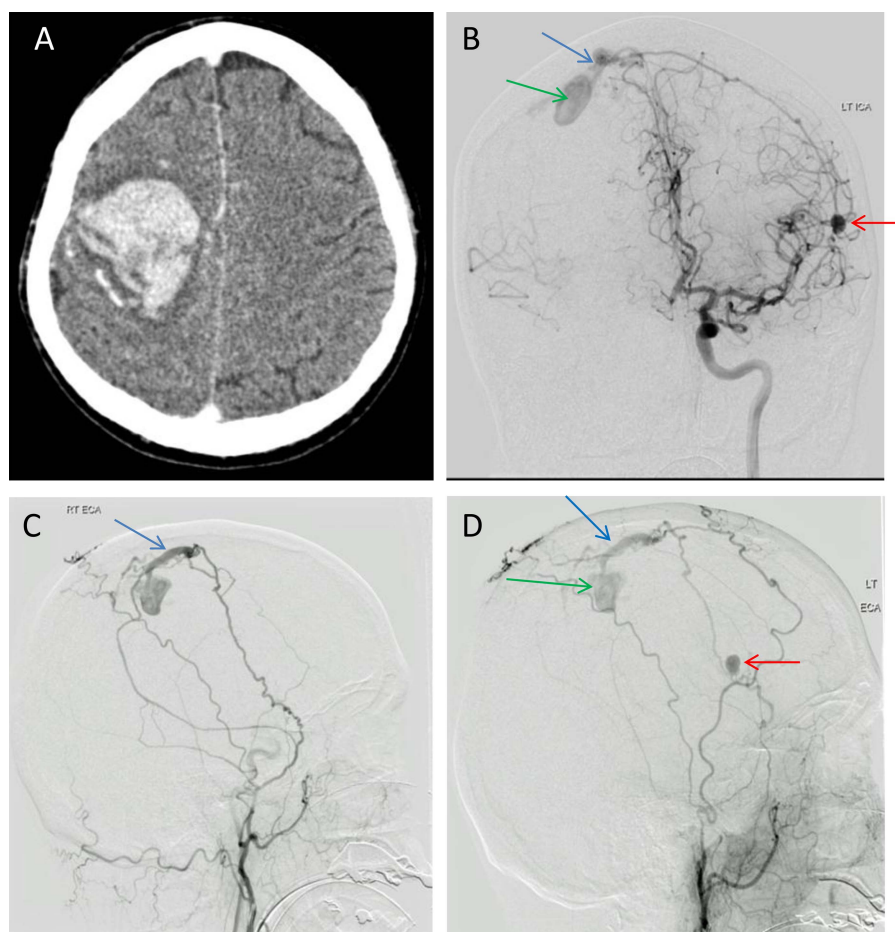
TREATMENT

The vascular lesions described above were considered to be better suited for surgical treatment. After gaining informed consent from the patient's partner, we then proceeded with a lengthy emergency stereotactic right frontoparietal craniotomy for the evacuation of the large right frontoparietal intraparenchymal clots, resection and clipping of the right frontal DAVF and venous aneurysm utilising the BrainLab neuronavigational system. He also had a left pterional craniotomy for clipping of the left intraosseous MMA aneurysm and subsequent insertion of a Codman microsensor intracranial pressure monitor. The inner skull table was seen to be unduly grooved by the rather tortuous middle meningeal vessels. The dura was reinforced with Duraform (Codman & Shurtleff Inc), Duraseal (Confluent Surgical Inc), Tisseel glue (Baxter International Inc), and bone flap was secured with miniplates and screws.

He received blood transfusion for acute perioperative blood loss, anti-seizure, antibiotic and DVT prophylaxis. His postoperative course in the ICU was, however, complicated by ventilator associated *Staphylococcus aureus* pneumonia and vocal

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Figure 1 CT scan and cerebral digital subtraction angiography (DSA) upon admission. (A) CT scan showing right fronto-parietal intraparenchymal haemorrhage with abnormal vessels and midline shift. (B) Arterial-phase DSA of the left internal carotid artery showing right frontal dural arteriovenous fistula (AVF) (green and blue arrows) with filling from the left middle meningeal artery which also has a flow-related saccular aneurysm (red arrow). (C), Arterial-phase DSA of the right external carotid artery showing the dural AVF with supply from the middle meningeal and superficial temporal arteries. There is retrograde filling of the cortical veins (blue arrow), consistent with Borden type II d-AVF. (D), Arterial-phase DSA of the left external carotid artery showing the dural AVF (green arrow) being supplied from the middle meningeal artery, superficial temporal artery as well as the left middle meningeal aneurysm (red arrow). Note the retrograde filling of the cortical veins (blue arrow), consistent with Borden type II d-AVF.



cord oedema requiring prolonged ventilatory support. Steroid therapy used to reduce vocal cord oedema, further compounded his stress-induced hyperglycaemia. Additional complications included transient acute renal insult and left lower limb deep vein thrombosis, resulting in a 3-week stay in the ICU. He was subsequently transferred to the high dependency unit and later to the general ward where upon satisfactory clinical recovery, he resumed very vigorous physical, cognitive and speech rehabilitation for his residual neurological deficits which included dense left hemiplegia, left-sided neglect, visual and speech defects.

OUTCOME AND FOLLOW-UP

On discharge home, he continued to receive community-based allied health support and remains well motivated. He is currently continent, has a normal speech, intact memory and is able to mobilise with assistance on a quad stick through left hip swing.

A follow-up CT showed desorption of the right fronto-parietal haemorrhage with resultant encephalomalacia. Cerebral DSA showed significant occlusion of the right DAVF and venous aneurysm with no retrograde filling of the cortical veins, and the left MMA aneurysm was completely occluded (figure 2).

DISCUSSION

The terms DAVF and dural arteriovenous malformation (DAVM) are often used interchangeably. Intracranial DAVF are acquired arteriovenous shunts confined to the dura, supplied mainly by branches of the external carotid artery and usually drained either anterogradely via the dural venous sinuses or retrogradely into other dural or leptomeningeal venous channels.¹¹ They are frequently idiopathic but can be associated with venous

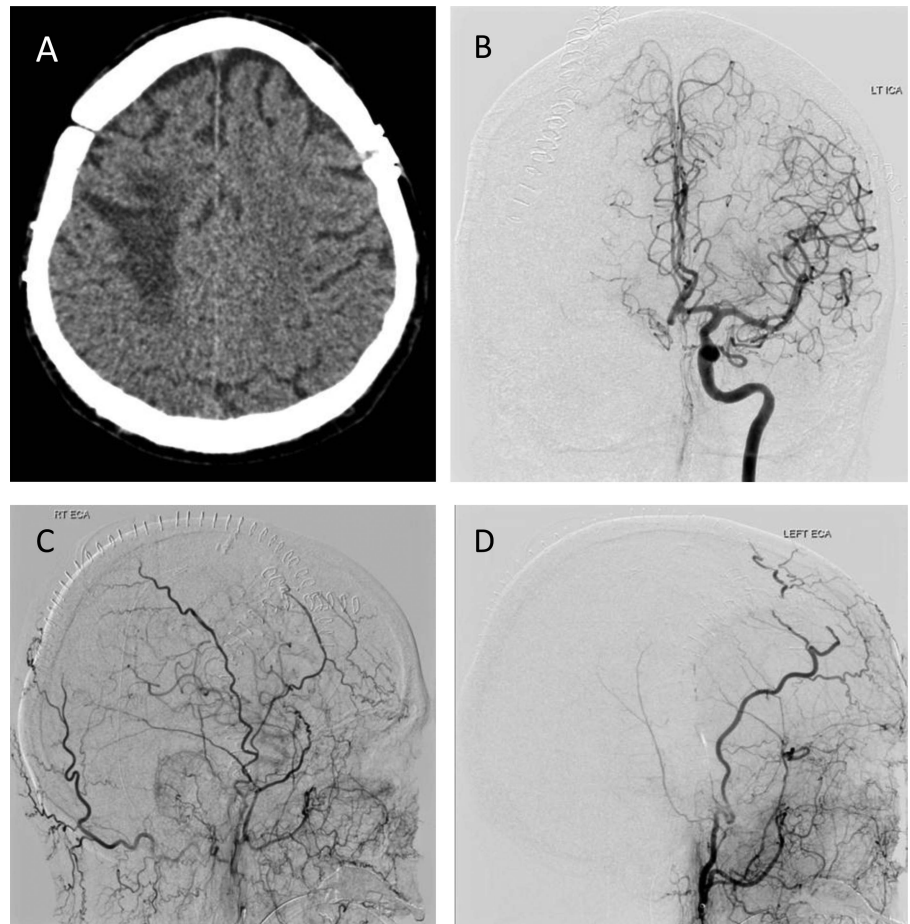
thrombosis, head trauma, tumours, previous neurological surgery, meningitis or a sinus infection.^{11 12}

In our case, the DAVF was supplied mainly by branches of the bilateral external carotid arteries and drained into the superior sagittal sinus via cortical veins/venous aneurysm with venous reflux quite in keeping with the Borden type II category.

An incidence of 13–21% has been cited in cohort studies for the association of pial or dural AVF with cerebral aneurysms. One-third of the aneurysms associated with DAVF are flow related while the rest are remote aneurysms of the circle of Willis.^{2 3} A prior report has found a stronger proclivity for such association in the anterior cranial fossa and cerebral convexity, in men and in middle-aged patients.³ Many postulates have been adduced for the association of DAVF and aneurysms including genetic predisposition, dynamics of blood flow, common inciting event like head injury, and chance phenomenon.² The association of DAVF and contralateral aneurysm in our case is certainly due to flow dynamics as the parent aneurysmal artery was a prominent blood supply of the DAVF.

The commonest presenting feature in these cases is haemorrhagic complication which can involve the parenchymal, sub-arachnoid or rarely the subdural compartment. Haemorrhage results from rupture of the fragile parenchymal veins which had become arterialisated as a result of the cortical venous reflux and hypertension. This is commoner in the presence of venous aneurysm and drainage of DAVF into the deep venous sinuses as in our case.^{10 13} Our patient presented acutely with intraparenchymal haemorrhage and with features of raised intracranial pressure such as sudden headache, visual abnormality, disturbance of consciousness and gross hemiplegia.

Figure 2 CT scan and Cerebral digital subtraction angiography (DSA) after surgery. (A), Follow up CT scan after 6 months showing desorption of the haemorrhage with some encephalomalacia at the site of the right fronto-parietal haemorrhage. (B), Arterial-phase DSA of the left internal carotid artery showing significant occlusion of the right dural arteriovenous fistula (AVF) and complete occlusion of the left meningeal artery aneurysm. (C), Arterial-phase DSA of the right external carotid artery showing significant occlusion of the right dural AVF with no retrograde filling of the cortical veins. (D), Arterial-phase DSA of the left external carotid artery showing a significant occlusion of the right dural AVF and complete occlusion of the left middle meningeal artery aneurysm.



Six-vessel DSA as was performed in our case, has been considered the gold standard in patients with DAVF to search for both flow related and remote aneurysms and to plan treatment options.²

DAVF can be treated by endovascular embolisation or open surgical obliteration. The open surgical technique becomes even more imperative in cases like ours, in which urgent evacuation of a large intraparenchymal haemorrhage is required. Complete disruption of the venous cortical drainage should be conducted for all affected sides of the dural venous sinus, if need be, bilaterally.¹⁴

Learning points

- The association of cerebral dural arteriovenous fistula (DAVF) with contralateral flow-related aneurysm increases the risk of haemorrhagic complications with attendant higher morbidity and/or mortality.
- Six-vessel digital subtraction angiography should be performed in all cases of DAVF to search for the associated aneurysms and for the treatment planning.
- Concurrent surgical treatment should be attempted where feasible to prevent the risk of haemorrhagic complication at a later date.
- The DAVF should be obliterated first before clipping of the aneurysm so as to prevent the rupture of the DAVF if retraction is required to gain access to the associated aneurysm.

Where a DAVF and flow-related aneurysm coexist, concurrent surgical treatment has been strongly advocated by some authors to prevent catastrophic consequences of rupturing of any of the vascular anomalies at a later time.⁷ During surgery, the DAVF should be secured first before clipping of the aneurysm so as to prevent the rupture of the DAVF if retraction is required to gain access to the associated aneurysm.⁷

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Contributors DOO admitted the patient into the hospital and assisted in the surgical procedure. He is also the major contributor in analysing the patient's data and preparing the manuscript. AWH is the senior author who performed the procedure and offered useful comments. RAH is the radiologist who performed and reported the DSA, and provided the relevant images. All the authors read, corrected and approved the final manuscript.

Competing interests None.

Patient consent Obtained.

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