

Sudden severe chest pain: thoracic dural arteriovenous fistula aneurysm rupture with intracranial subarachnoid haemorrhage

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ABSTRACT

Spinal perimedullary arteriovenous fistula (AVF) or dural arteriovenous fistula (DAVF) presenting as intracranial subarachnoid haemorrhage (SAH) is uncommon. A total of 16 cases have been reported to date. A majority of the reports described cervical spinal DAVF, while two other case reports described intracranial SAH secondary to lumbar and thoracic DAVF, respectively. We report a 61-year-old Chinese man with intracranial SAH secondary to thoracic DAVF aneurysm, who presented with sudden, severe chest pain, initially suggestive of aortic dissection/ acute myocardial infarction. However, a careful examination of the history and physical signs, followed by appropriate and timely investigations enabled effective treatment to be administered promptly with a good outcome. This serves to illustrate the importance of investigating the entire cerebrospinal system when neurological symptoms and clinical signs suggest extracranial primary pathology.

Keywords: secondary cardiac injury, spinal arteriovenous malformation, spinal haemorrhage, subarachnoid haemorrhage

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INTRODUCTION

Non-traumatic subarachnoid haemorrhage (SAH), while only accounting for 3% of all stroke,⁽¹⁾ contributes to 10% of stroke mortality across a wide range of age groups. The current literature reports that 85% of non-traumatic SAHs are caused by intracranial vascular abnormalities, with the majority being aneurysms. Approximately 10% can be attributed to perimesencephalic/venous bleeding, otherwise

known as perimesencephalic nonaneurysmal SAH. The remaining 5% of SAH are due to rare conditions, which include spinal arteriovenous malformation (AVM), arteriovenous fistula (AVF) with association to aneurysm and spinal tumour haemorrhage.^(2,3)

We present a case of non-traumatic intracranial SAH secondary to a ruptured dural arteriovenous fistula (DAVF) at the level of the thoracic spine. Our patient presented with an unusually severe acute chest pain (analogue pain score of 10/10, with a score of 0 indicating no pain and a score of 10 indicating the most severe pain experienced), which at initial investigations pointed to cardiac or thoracic aorta aetiologies.⁽⁴⁾ Successful treatment was accomplished with endovascular embolisation of the spinal DAVF.

CASE REPORT

A 61-year-old Chinese man with a history of chronic lower back pain and hyperlipidaemia presented with acute, severe left-sided chest pain while he was shopping. The pain was described as a stabbing pain that radiated to the back and was associated with acute dyspnoea and diaphoresis. The patient also complained of pain radiating down both his arms and legs, and this was associated with paraesthesia. The symptoms occurred in pulses, with a 2–3 seconds interval. Bystanders who witnessed his distress helped him to a wheelchair. Instead of relieving his symptoms, the patient experienced sharp pain and tightness over the occiput and down the back of his neck. He only felt symptomatic relief on lying down supine in the security room while awaiting the arrival of paramedics.

On arrival at the emergency room, the patient was noted to be slightly drowsy and had slurred speech. Both his pupils were equal and reactive at 3 mm. He was afebrile, with a regular heart rate of 82/min and blood pressure of 151/48 mm Hg. He had no focal weakness except for numbness over the right side

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Fig. 1 Axial CT brain image shows subarachnoid haemorrhage centred at the basal cistern.



Fig. 2 Contrast-enhanced sagittal T1-W MR image shows intradural arteriovenous fistula.

of his chest and arm. The examination of the other systems was unremarkable. Electrocardiography (ECG) demonstrated a right bundle branch block with minimal ST elevation at the V2 and V3 leads. Chest radiograph was normal, with no widening of the mediastinum.

Computed tomography (CT) of the brain was performed in view of the slurring of speech and drowsiness, which showed SAH centred about the basal cisterns (Fig. 1). A focus of dense haemorrhage was observed at the left cerebellomedullary cistern, but no hydrocephalus was evident. CT angiogram of the brain was performed, which was negative for any aneurysms or AVMs. A subsequent digital subtraction angiography of the cerebral vessels was performed 24 hours later, which also confirmed the absence of any abnormality.

The patient's clinical condition was stable over the next 48 hours, except for the persistent chest pain which radiated to the back at the mid thoracic level and down the upper limbs despite regular oral opiates analgesia. Some pain relief was achieved with the addition of gabapentin. The recorded pain score indicated an improvement from 8/10 to 3/10. Magnetic resonance (MR) imaging of the spine was performed in view of the SAH and back pain. It showed intradural serpiginous T1 and T2 hypointense structures with enhancement extending from T5 to T9 (Fig. 2). The slight displacement of the spinal cord raises the suspicion of an underlying AVM. Subsequent spinal angiogram confirmed a right T7 spinal AVF with enlarged venous drainage channels (Fig. 3). This was associated with an arterial aneurysm at the apex and venous pouches. A cutaneous/subcutaneous AVM was also identified, with

feeders from the left T8, T9 and right T11 intercostal arteries. The anterior spinal artery was observed to be supplied from the right T9 intercostal artery. As the right T7 feeding vessel of the spinal AVF was located anterior to the spinal cord, a decision was made for embolisation rather than operative ligation.⁽⁶⁾

Glue embolisation was performed under general anaesthesia, with 25% glue to the lipiodol mixture. The intercostal vessel at the T7 level was identified and cannulated with an ultraflow micro-catheter. The feeding of the spinal DAF artery was entered into just before the fistulous point, showing the aneurysmal vessels. Post embolisation checks did not show any residual filling of the fistula. Post procedure, the patient experienced transient pain and numbness radiating down his legs, especially when he stood up to ambulate, which was eventually resolved. He was discharged well with full recovery of his limb powers.

DISCUSSION

From our literature review, there are approximately 16 reports on spinal DAVF with intracranial SAH,⁽⁷⁾ mostly located in the cervical region. Only one case each was reported in the thoracic⁽⁸⁾ and lumbar regions.⁽⁹⁾ Ohmori et al reported a patient with thoracic DAVF located at T8, who underwent surgical ligation instead of endovascular embolisation,⁽⁸⁾ unlike our patient. The majority of spinal DAVF occurs at the thoracolumbar region of the spine,^(10,11) and the more common presentation is that of spinal compression symptoms secondary to engorgement of the venous drainage vessels rather than spinal haemorrhage.⁽¹²⁾ This case report illustrates

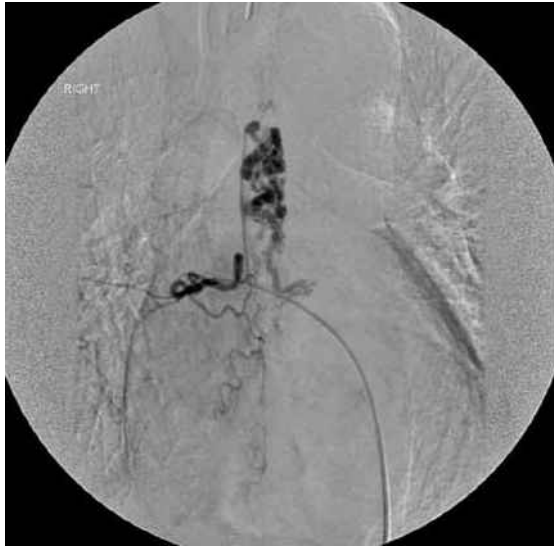


Fig. 3 Spinal angiogram shows the spinal arteriovenous fistula at T7 level.

the importance of good history-taking and targeted investigation so as to identify this rare entity.

This patient initially presented with symptoms suggestive of a cardiac event. In retrospect, this may have been related to the sympathetic outflow surge secondary to SAH, which is an occurrence that is well described in the literature.⁽¹³⁾ The associated ECG changes and even early raised cardiac enzymes results indicate cardiac injury.⁽¹⁴⁾ However, on more careful analysis of the history, we noted that the pain described by the patient was unusual. The initial sudden, severe pain on the left side of the chest which radiated from the front to back was more characteristic of a dissecting aortic aneurysm. This could be explained by the acute rupturing of the aneurysmal part of the thoracic DAVF at the start of the attack. Also, the patient described the pain as spreading across his chest and down his upper as well as lower limbs. This was likely due to the effect of subarachnoid space free blood irritation of the nerve roots as it spreads cephalad toward the cervical region and caudally toward the lumbar region. Moreover, the most durable pain sensation was a localising pain around the T8 level of the back, likely from a local ischaemic effect of the spinal cord secondary to rupture and subsequent transient vasospasms of the affected feeding vessels. Even more interesting to note in the presenting history was the patient's complaint of worsening neck pain and lower occipital pain when he was being assisted onto a wheelchair after the attack. The flexing of the patient's hips to sit down and the subsequent straightening of his legs was similar to the performance of Kernig's sign for meningismus. When

Dr Vladimir Kernig first demonstrated this sign, he also performed the examination with the patient seated upright.⁽¹⁵⁾

While most cases of spontaneous SAH are worked up initially with CT or digital subtraction angiography of the cerebral vessels, it is prudent to keep in mind the rare causes of SAH such as spinal DAVF, especially when the presenting signs and symptoms are atypical. We should not be too quick to brand a diagnosis of "angio-negative SAH". It was only after careful re-taking of the history that an MR imaging of the spine was ordered, which successfully identified the primary pathology and allowed definitive treatment to be administered. This case report illustrates the importance and utility of careful history-taking in a patient who presents with spontaneous SAH, especially when the cause is not readily apparent on initial investigations.

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