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Case Report

Spinal subarachnoid hemorrhage as a consequence of dissection with pseudoaneurysm in a cervical radiculomedullary branch of the anterior spinal artery ☆

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ABSTRACT

Spinal subarachnoid hemorrhage is a rare condition, and it generally arises as a consequence of arteriovenous malformation, although more rarely can be caused by aneurysm, dissection, or pseudoaneurysm. In the following, we present a case of a 58-year-old male who while undergoing treatment for nephrolithiasis, developed persistent hypertension, refractory to his home medications, along with headache, neck pain, and unilateral ptosis and upper extremity ataxia. Initial CT scan demonstrated acute subarachnoid hemorrhage in the posterior fossa extending to the C7 level, Angiography ultimately revealed a focal irregularity compatible with dissection and 1mm pseudoaneurysm within the left anterior spinal artery radiculomedullary feeder at the C5-6 level. The patient was managed conservatively with 81mg ASA and repeat angiography revealed resolution of the lesion, in concordance with management of dissection and pseudoaneurysm of the carotid and vertebral arteries. Subarachnoid hemorrhage as a consequence of dissection and pseudoaneurysm of a cervical radiculomedullary feeder has been previously unreported in the literature.

Introduction

Subarachnoid hemorrhage (SAH) is a major cause of morbidity and mortality worldwide. Spinal SAH is a rare condition, constituting less than 1% of SAH. The most common cause for SAH of spinal origin is from a spinal cord arteriovenous malformation (AVM). SAH as a consequence of aneurysm, dissection, or pseudoaneurysm of the spinal vessels is exceedingly rare. A review by McGuire et al found a total of 137 patients

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with isolated spinal artery aneurysms (SAA), and described 5 cases with their respective management and outcome, all of which presented in the context of hemorrhage [1]. Berlis et al. described a case series of 3 patients who were found to have SAH of spinal origin in 1 patient due to presumed dissection of a segmental artery, although mycoses were also likely, and in the other 2 patients, ruptured fusiform aneurysms of the artery of Adamkiewicz proximal to the anterior spinal artery [2]. The presentation of these patients is varied but generally involves headache, back/neck pain, and motor/sensory deficits corresponding to the level of the lesion. No consensus exists regarding the optimal management and treatment for SAA, however, in some cases, there was hematoma evacuation, or embolization of the aneurysms, whereas in others, treatment was conservative [1,2]. In both of these case series, the patients did well, with all but one having complete resolution of their neurological systems. Two cases of spontaneous SAH in the thoracic spine as a consequence of pseudoaneurysm have been reported. In 1 patient, clot evacuation with surgical ligation was performed, and well tolerated, while in the other, conservative management led to resolution of symptoms [3,4].

Below, we describe the management of a 58-year-old male who was ultimately found to have dissection with pseudoaneurysm, in the context of SAH, of a C5-6 cervical radiculomedullary artery supplying the anterior spinal artery.

Case discussion

A 58-year-old male with a past medical history of thoracic and abdominal aortic aneurysms, 40 pack-year smoking history (quit 15 years ago) obstructive sleep apnea, chronic back pain, hypertension, and coronary artery disease presented to the emergency department with back pain, difficulty voiding, and hematuria, ultimately found to be due to obstructive nephrolithiasis. He had a urethral stent placed which resolved the nephrolithiasis. During this hospitalization, he was found to be persistently hypertensive despite home medications, hydralazine, and labetalol administration, up to the 190-200s systolic. He began to complain of neck pain and headache, which he described as “musculoskeletal.” Neurological examination revealed a mild left ptosis and a mild upper extremity ataxia, but no other focal neurological deficits. Due to concern for acute stroke, he underwent CT and CTA of the head and neck, which demonstrated acute subarachnoid hemorrhage, centered in the posterior fossa (Fig. 1), and extending to the C7 level with an irregular contour of the right PICA, suspicious for a small aneurysm.

The patient underwent cerebral angiography the following day which initially was interpreted as negative with no clear explanation for the patient’s hemorrhage. Our institutional protocol for angio-negative SAH is to perform an MRI of the brain and cervical spine. The cervical MRI surprisingly revealed a small cord infarct or contusion at the C5-6 level that was associated with a ventral epidural focal vessel irregularity (Fig. 2). Upon further analysis of angiography, there was a corresponding focal irregularity suspicious for a dissecting pseudoaneurysm of the radiculomedullary artery supplying the anterior spinal artery at the level of C5-6. MRI brain also revealed scattered posterior circulation infarcts predominantly in the left PICA distribution, which was concordant with the preoperative symptoms. Retrospective review of the initial CTA showed a small left vertebral V2 dissection with a tiny pseudo-aneurysm that did not correlate with angiography.

On bleed day 8, the patient underwent repeat angiography to further investigate the suspicious lesions in the cervical vasculature. The focal irregularity became more conspicuous and was compatible with dissection and 1 mm pseudoaneurysm within the left anterior spinal artery radiculomedullary feeder at the C5-6 level (Fig. 3). In conjunction with the MRI findings in the cervical spine, it was ultimately deduced that this lesion was the cause of his hemorrhage. We decided to treat this conservatively with ASA 81 mg daily as we would with most other carotid or vertebral artery dissections. The patient’s tremulousness gradually improved and he showed no signs of vasospasm on daily trans-cranial doppler. He required aggressive BP management with nicardipine drip, which was then transitioned to oral. He remained in the ICU for a total of 14 days.

Outpatient follow-up ensued. The patient endorsed some balance difficulties, however, these were relatively mild. Repeat MRI as an outpatient 1 month after discharge showed increased prominence of the pseudoaneurysm, however only showing perianeurysmal enhancement (Fig. 4A). Surveillance angiography followed, and the aneurysm was unable to be identified, and the segmental artery showed poor filling, suggesting interval thrombosis (Fig. 4B). Nonetheless, the patient underwent close follow-up with MRI imaging at 3 months to re-evaluate the aneurysm which showed no lesion (Fig. 5A). He underwent repeat angiography at 6 months, which showed complete resolution of the pseudoaneurysm and a normal angiographic appearance of the radiculomedullary artery anastomosing with the ASA (Fig. 5B). Further follow-up was not recommended, and the patient has not had any new stroke-like symptoms at the time of writing, 2 years after his initial presentation. He will remain on lifelong ASA 81 mg.

Discussion

SAH of spinal origin is a rare condition. Even rarer are those that arise as a consequence of aneurysm. As stated above, spinal SAH due to dissection and pseudoaneurysm of a cervical radiculomedullary artery has not been reported in the literature. Arterial dissection occurs when there is a tear in the lining of blood vessels, creating a second false lumen and potentially causing complete compromise of the artery. Dissection of various arteries can occur, many of which are potentially fatal. Dissection is frequently seen in young and middle-aged males and is associated with hypertension or migraine. The pathophysiology generally involves intimal tear or vasa vasorum dysfunction. Pathways involved include TGF-β signaling, as well as collagen and fibrillin genes. Treatment of dissection is highly dependent on the etiology and location of the dissection [5].

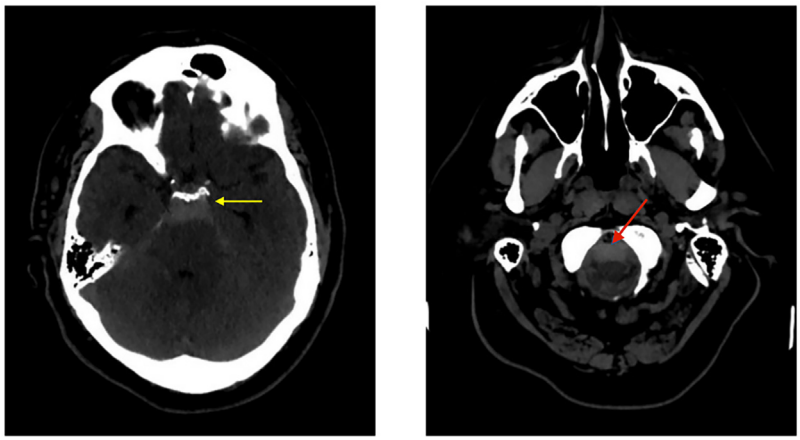

Fig. 1 – (A, B) Nonenhanced CT head. Acute thick blood is seen in the subarachnoid space at the prepontine cistern (yellow arrow) and at the cervicomedullary junction.

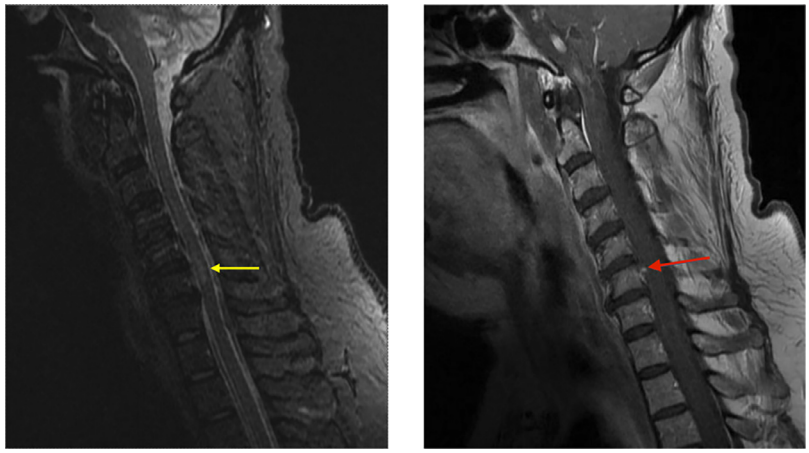

Fig. 2 – (A) Sagittal T2/STIR reveals focal cord hyperintensity at the C5-6 (yellow arrow). (B) Sagittal T1 postcontrast image reveals focal enhancing structure.

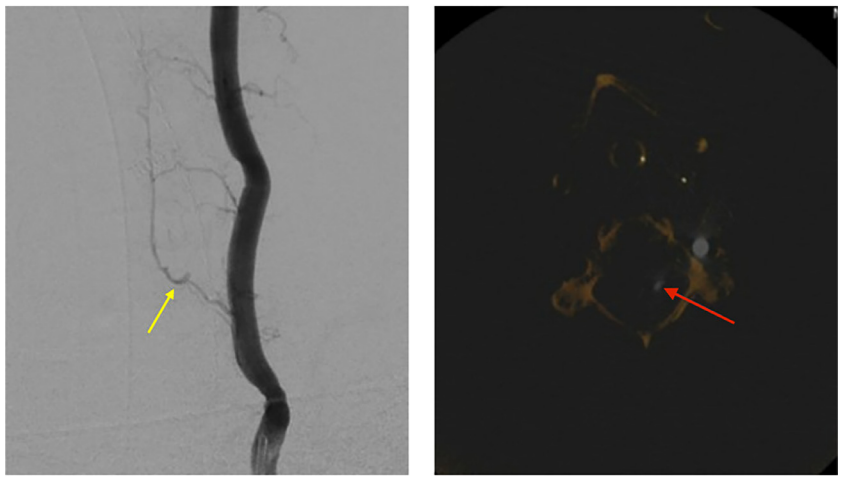

Fig. 3 – (A) DSA frontal view of a left vertebral artery injection. The C5-6 radiculomedullary artery proximal to the ASA connection is irregular with a superimposed aneurysm compatible with a dissecting pseudoaneurysm (yellow arrow). (B) Axial reconstruction following 3D rotational angiography demonstrates the aneurysm best seen and corresponds to the focus of enhancement seen on prior MRI.

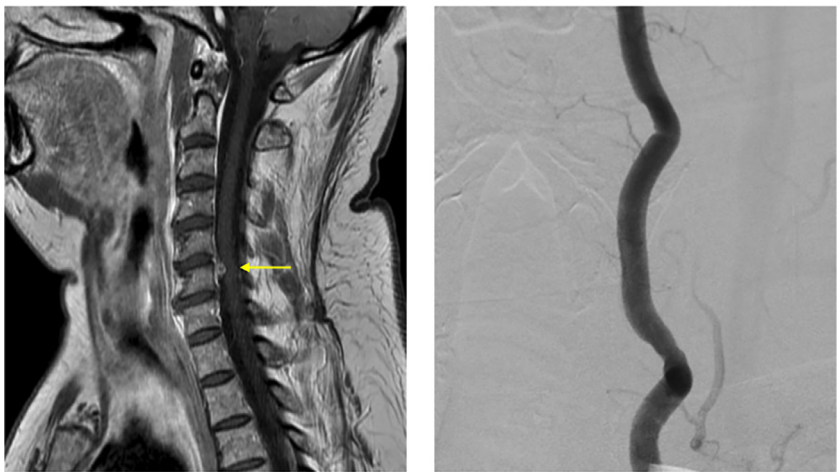

Fig. 4 – (A) Sagittal T1 post contrast image reveals interval enlargement of the pseudoaneurysm however now with a focal filling defect and peri-aneurysmal enhancement. (B) Left vertebral artery injection, frontal view, no longer demonstrates filling of the C5-6 radiculomedullary artery. While this could be related to injection technique, the finding in conjunction with the recent MRI suggests interval thrombosis.

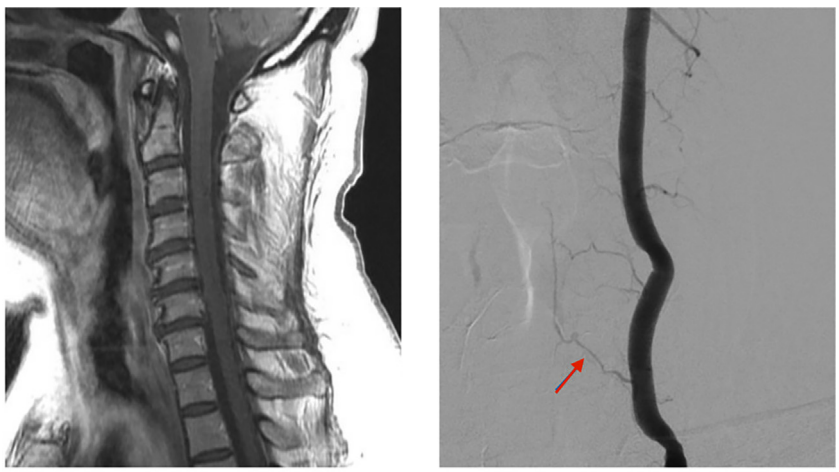

Fig. 5 – (A) Sagittal T1 post contrast image 3 months after initial hemorrhage no longer reveals any abnormal enhancement. (B) Left vertebral artery injection on DSA 6 months after initial hemorrhage, demonstrates a normal angiographic appearance of the left C5-6 radiculomedullary artery (blue arrow) without evidence of dissection or pseudoaneurysm.

Pseudoaneurysm, as well, is a well-recognized and described entity. They do not contain any layers of the vessel wall, and the wall of the pseudoaneurysm is composed of products of the coagulation cascade. They can arise as a consequence of arterial access, trauma, infection, and atherosclerosis [6].

Dissecting pseudoaneurysms (DP) of the carotid and vertebral arteries is well described. They may arise spontaneously, as a consequence of trauma, fibromuscular dysplasia, or due to underlying connective tissue diseases such as Marfan or Ehlers-Danlos syndromes. Dissection of these arteries is a major contributor to stroke in young patients. Symptomatology and treatment of DP depends on the size, location, and presence of DP enlargement. Risk factors for enlargement include smoking, hyperlipidemia, and larger initial size. Surgical or endovascular approaches are often used to treat symptomatic DP, with stenting being the most common modality in 1 study. Medical management alone is often sufficient for initial treatment as DP typically has a benign course [7].

In the case of our patient, identifying the cause of this SAH proved difficult, requiring multiple radiographic studies, both invasive and noninvasive, due to the small size of the lesion as well as the fact that there was initial concern for an aneurysm of the intracranial posterior circulation. Based on the small size of the lesion, supply to the ASA, and the resolution of symptoms, intervention was not pursued. This is consistent with the management suggested by the literature for dissecting pseudoaneurysms in other locations. Since the patient demonstrated resolution of the lesion, further imaging surveillance will not be pursued unless the patient becomes symptomatic.

Conclusion

Spinal SAH is a rare condition. In the above, we describe a patient who presented with a dissecting pseudoaneurysm of the cervical radiculomedullary artery leading to SAH, previously unidentified in the literature. In this case, conservative management was pursued, deduced from the literature regarding both spinal SAH, as well as DP. More research is needed to properly characterize this disease and to provide evidence-based guidelines for its management.

Patient consent

Complete written informed consent was obtained from the patient for the publication of this study and accompanying images.

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