Subarachnoid hemorrhage (SAH) with spinal etiology is present in less than 1% of all cases with SAH. The most frequent cause of spinal SAH is rupture of spinal cord pial arterio-venous malformations (ScAVMs) followed by hemorrhage from highly vascular spinal tumors such as ependymoma or hemangioblastoma. Aneurysms arising from spinal arteries are extremely rare with an incidence of about one in more than 3,000 spinal angiograms. Spinal aneurysms are usually flow related aneurysms associated with ScAVM and less commonly in dural arterio-venous fistulas (DAVF) and in patients with coarctation of the aorta, bilateral vertebral occlusion or Moya-moya disease in whom the anterior spinal artery (ASA) serves as collateral supply. Isolated aneurysms not associated with these conditions are very rare. We report a case of isolated ruptured radiculopial artery aneurysm presenting with spinal SAH.

CASE REPORT

A 72-years-old female presented with sudden onset of the worst back pain of her life while sitting on a chair. The pain was localized to the thoraco-lumbar region with radiation to both lower limbs and exacerbation with motion. She had a positive Lasègue sign with no sensory disturbances, bowel or bladder disturbances or motor weaknesses and no headaches.

Magnetic resonance imaging (MRI) of spine showed T1 and T2 hyper-intensity in the lumbo-sacral thecal sac suggestive of SAH (Figures A, B, C). There was no spinal cord signal abnormality or perimedullary flow voids. There was a focal T1 and T2 hypo-intense lesion in the antero-lateral aspect of the spinal canal at the level of T12-L1 inter-vertebral disc. This lesion showed subtle eccentric nodular enhancement on post contrast T1 images. Spinal MR angiogram was negative for a shunting lesion with no prominent vessels.

The differential diagnoses included: a slow flow or partially thrombosed ScAVM (despite the absence of prominent vessels); a spinal eccentric cavernoma (despite the focal contrast enhancement and the subarachnoid hemorrhage without intramedullary component); a spinal artery aneurysm, and vasculitis. A conventional spinal angiogram was recommended.

Spinal angiography was performed under general anesthesia and included selective catheterization of segmental arteries T8-L4 bilaterally. The left L1 segmental artery angiogram showed the major supply to the ASA axis (radiculomedullary supply, the so-called artery of Adamkiewicz), no venous stagnation, with narrowing of the artery possibly related to vasospasm. The left L2 segmental artery angiogram revealed radiculopial supply via a dorsolateral artery that demonstrated an irregular fusiform

Figure: PSA aneurysm. (a) T1W, (b) T2W sagittal and (c) T2 axial images showing T1 and T2 hyperintensity in thecal sac suggestive of SAH and a focal T1 and T2 hypointense lesion (arrows) in the anterolateral spinal canal at T12-L1 level. (d) Left L2 segmental artery angiogram showing an irregular fusiform aneurismal dilatation (arrow) in the mid-portion of perimedullary ascending segment of radiculopial artery suggestive of dissecting aneurysm. (e) Post treatment, left L2 segmental artery angiogram showing no filling of the radiculopial artery and the coil mass starting from retro-corporeal transverse anastomotic channel (white arrow) back to the segmental artery trunk (black arrow).
aneurysmal dilatation of the mid-portion of the perimedullary ascending segment of the artery suggestive of dissecting aneurysm of the radiculopial artery (Figure 1D). There was evidence of stasis of contrast in the aneurysm. The left L2 segmental artery angiogram also revealed filling of the previously noted radiculomedullary artery via inter-segmental anastomoses to the left L1 segmental artery. Presumably because of the vasospasm on the left L1 radiculomedullary artery, the left L2 radiculopial artery was the predominant supply to the arterial basket at the conus medullaris.

Considering the high incidence of rebleed from dissecting aneurysms of spinal arteries, immediate treatment was considered. The treatment of choice for dissecting aneurysm would be parent vessel sacrifice either via surgery or endovascular route. Given the rich network of anastomoses between ASA and posterior spinal artery (PSA), we expected ASA axis to take over the blood supply to the conus medullaris after the sacrifice of the radiculo-pial artery. A proximal occlusion of the segmental feeding artery was not deemed sufficient given the potential intersegmental anastomoses, so the decision was made to occlude the distal radicular branch including the retro-corporeal anastomosis after an unsuccessful attempt to select the radiculopial artery (given its small size and acute proximal origin from the radicular branch). Since ASA axis was visualized through inter-segmental anastomosis from the left L2 injection, coil embolization was favored over liquid embolic material. To prevent any filling from the retro-corporeal transverse anastomosis, we started coiling from the anastomotic channel and then back to the segmental artery trunk thereby covering the origin of the radiculopial artery. At the end there was no residual filling of the radiculopial artery or the dissecting aneurysm (Figure 1E). The basket of the conus medullaris continued to fill on left L1 segmental artery angiogram as the radiculomedullary artery at this level took over the supply to the basket but no retrograde filling of the dissecting aneurysm occurred.

Post-operatively, the patient had no new neurological deficits, and her lower back pain slowly improved. As there were some worries about the vasospasm, a repeat angiogram was performed after two weeks which again showed no residual filling of the aneurysm and the radiculopial artery. There was normal filling of the ASA axis and the arterial basket at the conus medullaris with minimal residual vasospasm. The patient was discharged from the hospital after two weeks.

**DISCUSSION**

The first case of an isolated ASA aneurysm presumably caused by syphilis was reported by Babonneix and Wediez in 1930. A total of 33 patients with isolated spinal aneurysms have been reported in literature. Of these, only ten (including our patient), were located at the PSA (radiculopial), two were located at a radicular artery, in two cases, the exact location was not specified, and, in the remaining 20 patients, the aneurysm was located on the ASA (radiculomedullary). Association of infectious or inflammatory conditions such as syphilis, pseudoxanthoma elasticum, fibromuscular dysplasia, Behcet’s disease and systemic candidiosis were reported in 5 of 20 patients with aneurysms on the ASA axis. No such association has been reported for aneurysms on the PSA axis. The most common cause of the PSA aneurysms has been described as transmural dissection of the arterial wall and was proven by histopathology in six cases.

Due to their rarity, spinal aneurysms may be missed or their diagnosis can be delayed. In cases of spinal SAH, the possibility of a spinal aneurysm should always be suspected in absence of prominent flow voids or obvious tumor on MRI of spine. Spinal SAH can be diagnosed with T1 hyperintensity in spinal subarachnoid space. Spinal aneurysms usually demonstrate a T2 hypointensity (that can be due to a mural hematoma or stagnating blood flow) with focal contrast enhancement representing the pseudoaneurysm. Dynamic contrast enhanced magnetic resonance angiograms and post contrast scans may help in the diagnosis showing focal nodular enhancement and absence of abnormally prominent vessels. In cases of ambiguity, a spinal computed tomographic (CT) scan with CT angiogram (CTA) of spine is very helpful in both diagnosis and localization of the aneurysm. Spinal aneurysms are often irregular in shape with contrast stagnation within the sac on the late venous phase, suggesting its dissecting nature. A conventional catheter angiogram is always imperative and diagnostic for any suspicious vascular lesion of the spine. This procedure also enables the physician to treat the aneurysm via an endovascular route.

The treatment of PSA aneurysms can be performed either by open surgery or by endovascular embolization. Surgery is an option for PSA aneurysms due to the dorsolateral and superficial location. Endovascular treatment with embolization using either coils or glue is possible. However, this treatment may be technically challenging due to the small size of the radiculopial arteries on which the aneurysms are located. With neither approach is preservation of the parent artery possible nor recommended due to the dissecting nature and absence of true neck. Parent vessel occlusion can be performed with liquid embolic materials. In cases of unstable catheter position and presence of dangerous anastomosis, detachable platinum coils can also serve the purpose.

In conclusion, isolated PSA aneurysms are rare entities caused possibly by arterial wall dissection. Spinal angiographic findings of a saccular out-pouching on a radiculopial artery on the cord surface leads to final diagnosis. Parent vessel sacrifice, either surgically or endovascularly, remains the standard treatment of these lesions.

**REFERENCES**