Acute cauda equina syndrome from a ruptured aneurysm in the sacral canal

Case report

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The case is presented of a young woman with acute cauda equina syndrome from a ruptured aneurysm in the sacral canal. The lesion was associated with pathological enlargement of the lateral sacral arteries bilaterally, which presumably occurred to provide cross-pelvic collateral flow in response to the diversion of the right internal iliac artery for renal transplantation. The patient presented with signs and symptoms of spontaneous spinal epidural hemorrhage. The radiographic features of this lesion are described. In addition to angiography and partial embolization of the vascular supply, contrast-enhanced high-resolution computerized tomography was essential in the diagnosis and treatment of this unique aneurysm.

KEY WORDS • spinal epidural hematoma • aneurysm • cauda equina syndrome • sacrum • renal transplantation

SPONTANEOUS spinal epidural hematoma is an uncommon but well-described entity, presenting with painful acute or subacute loss of spinal cord or cauda equina function. These hematomas generally require surgical evacuation, although there are a few examples in the literature of spontaneous resolution of neurological deficits with good outcome.4,7 The etiology of spontaneous spinal epidural hematoma is multifactorial, including presumed vascular malformations; however, in many cases no specific pathology is identified.4,10 The present report describes a young patient with acute cauda equina syndrome caused by a combination of rapid enlargement of and hemorrhage from an aneurysm within the sacral canal, arising from enlarged lateral sacral arteries. The magnetic resonance (MR) imaging findings were distinctive but could easily be misinterpreted as intervertebral disc herniation. Contrast-enhanced computerized tomography (CT) was essential in the elucidation and management of this lesion.

Case Report

This 26-year-old woman, who had undergone a successful cadaveric renal transplant 8 years earlier, presented to a community hospital emergency room with a 1-day history of crampy low-back pain and muscle spasm which began spontaneously while sitting. Symptoms included pain and paresthesiae radiating down both legs and numbness in the left buttock, perineum, upper lateral thigh, and foot. She had been unable to pass urine since the previous evening.

First Examination. On initial assessment, the patient was alert but distressed, afebrile, and without meningismus. She had full strength in her lower extremities but decreased touch sensation in the left S-1 distribution and in the perineal area bilaterally (left side worse than the right). Anal sphincter tone was flaccid, the bulbocavernous reflex was absent, ankle reflexes were absent on the left but present on the right, and all other deep tendon reflexes were intact and symmetrical. Laboratory studies were normal except for a chronic stable creatinine level of 2.3 mg/dl. The patient's medical history included glomerulonephritis-induced renal failure 10 years previously and a congenital arteriovenous fistula between the coronary sinus and the pulmonary vein, repaired via open heart surgery, in 1979. She had no history of back pain.

Diagnostic studies included a CT scan of the pelvic region, which revealed indistinct epidural fat planes in the region of L-5 and S-1, in addition to the trans-
planted kidney and an associated lymphocele. Lumbosacral MR images revealed an extradural mass in the sacral canal behind the body of S-1 and ventral to the dura. The mass had mixed signal intensity on both T₁- and T₂-weighted images (Fig. 1). The rim of the lesion "bloomed" on T₁-weighted and short tau inversion recovery images, consistent with the presence of hemosiderin.

**First Operation.** The lesion was explored via an S-1 laminectomy, and epidural hematoma was encountered surrounding a pulsating mass lying ventral to the dural terminus. As the epidural clot was being evacuated, the surgeons encountered sudden high-flow blood loss from within the sacral canal which was controlled by packing the region with Surgicel. The wound was closed and the patient was transferred to Harborview Medical Center in the immediate postoperative period, with no neurological change.

**Second Examination.** The patient was given a course of dexamethasone (4 mg every 6 hours) and intravenous hydration and immediately underwent investigation with high-resolution CT of the lumbosacral region, both with and without contrast medium infusion. In addition to the expected postoperative changes, these studies revealed a brightly enhancing semispherical mass within the sacral canal at S1-2, enlarged vascular structures in the pelvis lying anterior to the sacrum, and an enlarged sacral artery traversing the right S1–2 sacral foramen and subsequently entering the epidural mass (Fig. 2). There was no evidence of bone erosion or tumor mass.

**Second Operation.** Fresh hematoma superficial to older more organized hematoma was evacuated and profuse arterial bleeding erupted from within the sacral canal. The right-feeding artery was localized emerging through the right foramen and was cross-clipped with metal ligatures, immediately producing cessation of hemorrhage. At surgery, no evidence of vascular malformation and no venous structures apart from apparently normal epidural veins were encountered. Pathological examination of the specimen revealed organizing
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hematoma comprising the wall of the lesion, consistent with a pseudoaneurysm.

Postoperative Course. Postoperative angiography revealed obliteration of the vascular lesion and occlusion of both lateral sacral arteries at the level of the upper sacral foramina. The patient made an uneventful postoperative recovery. To date, she has had no recovery of sphincter control but is fully ambulatory with normal lower-extremity function. Renal function has recovered to its premorbidity baseline level.

Discussion

The vast majority of cases of acute cauda equina syndrome are due to disc rupture, tumor, or infection. Spontaneous spinal epidural hemorrhage is also a known cause of acute cauda equina syndrome; however, it is an uncommon entity. A review of the literature suggests that this is the first reported case of spontaneous spinal epidural hematoma and acute cauda equina syndrome associated with an aneurysm within the spinal canal.

Spontaneous spinal epidural hematoma was recently reviewed in detail by Groen and Ponsen.10 Interestingly, nearly all such cases in the lower spinal region occur in patients over the age of 40 years. In younger patients, this entity occurs nearly exclusively in the cervicothoracic area. While a variety of etiologies may be responsible for spontaneous spinal epidural hematoma, such as coagulopathy, minor trauma, bone disease, and vascular malformations, the majority are cryptic. Younger patients with spontaneous spinal epidural hematoma have a higher incidence of suspected or proven vascular malformation than older patients. Our patient’s clinical presentation was typical of spontaneous hemorrhage in the spinal epidural space, with sudden unprovoked onset of severe localized and radiating pain, paresthesia, and rapidly evolving neurological deficit appropriate to the level of the hematoma.4,6,10

Aneurysms of the spinal vasculature are extremely rare, and the only known examples have been intradural. These are usually, but not always, associated with intramedullary arteriovenous malformations (AVM’s).9,16,17,12 Aneurysms of the aortic vessels may cause neurological symptoms due to mass effect on the lumbosacral plexus; however, this is an extraspinal process.8,13,14

The present lesion was clearly an intraspinal aneurysm, based on both the preoperative MR image characteristics and the initial surgical and angiographic findings. The patient’s neurological symptoms were presumably due to a combination of rapid aneurysmal enlargement and hemorrhage producing mass effect on the cauda equina. At the time of the second operation, the lesion had been converted into a pseudoaneurysm by episodes of rebleeding and thus the absence of a true fundus at that time or on pathological review does not exclude this from being an aneurysm. The aneurysm presumably formed due to enlargement of the lateral sacral arteries to provide a pathway for cross-pelvic collateral blood flow in response to the diversion of the internal iliac artery for the renal allograft. Stewart, et al.,22 described a similar case in which a pseudoaneurysm arose from a pathologically enlarged third lumbar artery 8 years following renal transplantation. In that patient, the aneurysm arose extraspinally and presented with retroperitoneal hemorrhage, and the feeding artery was also apparently involved in the system of cross-pelvic collateral vessels. Aneurysm formation on cross-pelvic collateral vessels may, therefore, represent an unusual complication of renal transplantation.

It is unlikely that the aneurysm in the current case arose in association with a dural or epidural AVM. No draining veins were seen on MR imaging or angiography and no nidus was apparent at surgery. Spinal AVM’s supplied by branches of the internal iliac artery are rare; only 13 cases have been described in the literature.12,15 Spinal dural AVM’s typically exhibit symptoms referable to increased pressure in the medullary veins and capillaries, causing ischemia with a neurological level above that of the nidus;21 spinal epidural AVM’s present either in a similar fashion or with symptoms of radicular compression.11,12,23 This patient had no such pre-existing symptoms. In a recent series of 81 spinal AVM’s, none of the 27 dural AVM’s was associated with spontaneous hemorrhage.21

Fig. 3. Digital subtraction angiogram of the descending aorta revealing the aneurysm (asterisk) within the sacral canal supplied by markedly enlarged lateral sacral arteries. The right internal iliac artery (large arrow) has been diverted for the renal allograft. Several cross-pelvic collateral vessels (small arrows), including the lateral sacral arteries, have hypertrophied to supply left-to-right blood flow from the left internal iliac artery.
Although this patient’s clinical presentation was typical of acute disc herniation causing acute cauda equina syndrome, the initial MR image showed an extradural lesion with features typical of those seen in giant intracerebral aneurysms. These features include heterogeneous signal within the aneurysm on both T1- and T2-weighted images, indicating thrombus (with methemoglobin and deoxyhemoglobin signal) and turbulent blood flow. Moreover, short tau inversion recovery sequence images revealed a dark ring of hemosiderin in the wall of the lesion. No well-defined flow void was seen within the lumen of the aneurysm, presumably due to the slow and turbulent flow. Because of the large size of the aneurysm, MR imaging features of spontaneous spinal epidural hematoma could not be clearly resolved.

Essential to the successful surgical management of this patient was contrast-enhanced CT. The CT scan confirmed the vascular nature of the lesion and also permitted precise localization of the feeding arteries within the sacral canal and sacral foramina. Since embolization or transabdominal occlusion of the right lateral sacral artery was not possible in this case, the artery had to be occluded surgically, which entailed locating the vessel in the presence of copious arterial hemorrhage within a large amount of hematoma and multiple sacral root sleeves. Contrast-enhanced CT should thus be considered in cases of spontaneous spinal epidural hematoma associated with vascular lesions. Such imaging has already been proven of value in the workup of intracranial subarachnoid hemorrhage.

References