Arteriovenous fistula and pseudoaneurysm of the anterior spinal artery caused by an epidural needle in a 5-year-old patient

Case report

IBRAHIM ALNAAMI, M.D., M.Sc.,1 FRED C. LAM, M.D., PH.D.,1 GRAHAM STEEL, M.D.,2 BRYAN DICKEN, M.D.,2 CIAN J. O’KELLY, M.D., M.Sc.,1 KEITH ARONYK, M.D.,1,4 AND VIVEK MEHTA, M.D., M.Sc.1,4

Divisions of 1Neurosurgery and 2General Surgery, and 3Department of Anesthesia, University of Alberta Hospital; and 4Stollery Children’s Hospital, Edmonton, Alberta, Canada

Authors present the case of a 5-year-old patient with a spinal arteriovenous fistula (AVF) and pseudoaneurysm of the anterior spinal artery (ASA) caused by a traumatic epidural needle stick injury. A discussion and relevant review of the literature follow.

The boy had a remote history of a liver transplant and required neuraxial blockade for an unrelated abdominal surgical procedure. Initial insertion of the epidural needle at the T9–10 interspace yielded blood. A second attempt at T10–11 was successful. Delayed left leg weakness developed on postoperative Day 8, with an MR image showing a track injury through the cord and a ventral subarachnoid hematoma. Laminectomies from T-9 to T-11 were performed emergently to decompress the spinal cord. The dura mater was opened, the ventral hematoma was evacuated, and brisk venous bleeding was controlled with cauterization. Postoperative spinal angiography demonstrated an AVF and pseudoaneurysm of the ASA. Repeat angiography at postoperative Week 4 demonstrated complete resolution of the AVF and pseudoaneurysm, probably due to intraoperative cauterization of the draining vein. The patient underwent a short course of rehabilitation and had no clinical or electrophysiological evidence of spinal cord damage at the 20-month follow-up.

One should be cognizant of the possibility of a cord injury in a patient with new-onset neurological deficits following an interventional spine procedure. Neuroimaging is essential for prompt diagnosis and treatment. (http://thejns.org/doi/abs/10.3171/2012.12.PEDS12247)

KEY WORDS • thoracic spinal cord injury • epidural needle • subarachnoid hematoma • arteriovenous fistula • anterior spinal artery • spine

Neuraxial blockade is a useful adjunct in anesthesia and may be performed with the patient awake or sedated. In the pediatric patient this technique is difficult to accomplish without placing the child under general anesthesia. The ability to recognize and thus mitigate neurological injury in these patients when performing neuraxial blockade may be limited.3 There have been multiple reports in the literature of traumatic epidural catheterizations causing symptomatic spinal cord compression due to epidural or subdural hematoma collections,1,14,20,35,35 as well as reports of direct spinal cord injury causing immediate or delayed symptoms.3,22,37,43 To the best of our knowledge, however, there have been no reports of delayed neurological deficit in a pediatric patient following an epidural catheterization procedure, resulting from a through-and-through spinal cord lesion and leading to the formation of a ventral subarachnoid hematoma, AVF, and pseudoaneurysm of the ASA.

Case Report

History. This 5-year-old boy had a history of biliary atresia, which was initially treated with a Kasai procedure. Unfortunately, this procedure eventually failed and the child underwent an orthotopic liver transplant. He had a complicated course, requiring retransplant of the liver because of hepatic artery thrombosis. His condition stabilized posttransplantation, but then multiple episodes of upper gastrointestinal bleeding with melena occurred.
A vascular lesion in the small bowel was believed to be the source of bleeding, and a partial small-bowel resection was planned. Insertion of an epidural catheter was arranged for postoperative analgesia. This decision was primarily based on the substantial laparotomy incision planned for surgical access. The primary advantages of this technique are abilities to decrease postoperative opioid consumption and facilitate early postoperative tracheal extubation.

 Epidural catheters can be directly inserted at the level where analgesia is required or threaded up through the sacral hiatus to the desired spinal level within the epidural space. Caudally threaded epidurals are considered to have a lower risk of direct cord injury and hematoma formation, but the technique is only suitable for infants and small children. Placement becomes increasingly difficult after the onset of ambulation, and generally the caudal technique is not possible in children older than 3 or 4 years of age.12,16 This patient was 5 years 2 months of age at the time of surgery, and it was believed that a caudally threaded epidural would not be successful. On the morning of surgery, the patient’s parents, the attending anesthesiologist, and the pediatric anesthesiology fellow discussed the risks and benefits of a direct thoracic epidural insertion. This discussion included the risks of cord injury, CNS infection, hematoma formation, unintentional dural puncture, and failure of technique. Verbal consent was obtained to proceed with epidural placement. The backup analgesic plan was intravenous narcotic infusion and transporting the patient to the pediatric ICU sedated and intubated at the conclusion of the procedure.

After induction of anesthesia and endotracheal intubation, the patient was placed in the lateral decubitus position with standard monitors in place. His spine was palpated for landmark identification, and then the entire back was prepared and draped in sterile fashion. A midline, loss of resistance to air technique was used using an Arrow 17-gauge 8.89-cm Tuohy needle. The first needle pass was conducted at the T9–10 interspace, and no loss of resistance was noted at the expected epidural space depth of 1.5 cm (1 mm/kg). Continued advancement of the needle yielded blood at the 3-cm insertion depth. The needle was withdrawn, and pressure was applied at the site for several minutes. A second needle pass was attempted at the T10–11 interspace and produced a loss of resistance to air at 4 cm with no blood or CSF noted. A 19-gauge FlexTip Plus epidural catheter (Arrow) was threaded to 8 cm at the skin with no noted problems. The catheter was secured to the skin and subsequently loaded with a local anesthetic to good clinical effect intraoperatively.

The patient was extubated in the operating room at the end of the procedure and transported to the pediatric ICU. There was no documentation of any neurological issues during this time, and the catheter was removed on postoperative Day 3 before the patient was transferred out of the ICU and onto the ward. He remained asymptomatic until postoperative Day 8 when a headache developed, which resolved after administering a fluid bolus. The next morning, the patient complained of pain in his left leg and back. The neurosurgical team was consulted immediately and plans were made for urgent MRI.

Examination. Physical examination of the patient demonstrated Grade 1/5 power in his left leg, associated with dysesthesia. In addition, he had bilateral lower limb hyperreflexia. He was rushed urgently to the MRI unit for imaging and diagnosis. Magnetic resonance imaging with STIR sequences of the thoracic spine demonstrated a linear hyperintensity traversing the spinal cord at the T10–11 level, in keeping with a needle tract (Fig. 1A). Axial T2-weighted imaging showed a subarachnoid ventral fluid collection suggestive of a subarachnoid hematoma (Fig. 1B).

Operation. The patient was brought to the operating room following MRI. Thoracic laminectomies from T9 to T11 were performed. The dura mater was opened under the operative microscope, revealing an engorged spinal cord, prominent dorsal vasculature, and the tract injury itself. Ventral to the cord was a subarachnoid hematoma. Access to the ventral aspect of the cord was gained by cutting the dentate ligaments, allowing gentle manipulation of the thoracic cord and visualization of the hematoma. After removing the ventral subarachnoid hematoma, we encountered brisk bleeding from what appeared to be a large caliber vein, which was controlled by cautering the vessel with bipolar cautery.

Postoperative Course. Because of the bleeding encountered intraoperatively, the abnormal dorsal vasculature, and the ventral location of the hematoma, a spinal angiogram was obtained postoperatively. The angiogram demonstrated a dominant supply to the ASA from the T9 segmental artery on the left (artery of Adamkiewicz). The caliber of the ASA was enlarged, suggesting increased flow (Fig. 2). There was early filling of spinal veins. We diagnosed a spinal AVF based on the engorged veins on the dorsal surface of the spinal cord and bleeding from the ventrolateral aspect of the spinal cord, combined with the angiographic findings. In addition, there was filling of an abnormal vascular dilation adjacent to the ASA consistent with a pseudoaneurysm. The degree of arteriovenous shunting was relatively slow, and having coagulated some of the veins at the time of the surgery, we postulated that the lesion might heal spontaneously.

Following the surgical intervention, the patient’s neurological examination improved with increasing motor function in his legs. Conservative management was elected given the slow nature of the arteriovenous shunting and the high risks associated with surgical exposure and repair of the ASA, particularly in the setting of enlarged arterialized veins. An endovascular approach was also believed to be unfeasible given the challenging access and high risk of ASA injury. A repeat angiogram obtained 4 weeks postoperatively demonstrated resolution of both the AVF and pseudoaneurysm along with normalization of the caliber of the ASA (Fig. 3A and B). Follow-up MRI studies at 4 weeks demonstrated associated areas of increased T2 signal consistent with myelomalacia and/or old hemorrhage (Fig. 3C). Imaging studies performed at 1 year demonstrated persistent, slightly smaller T2 signal change in the cord (Fig. 3D). The patient was discharged to a rehabilitation facility where he was admitted for a course of intensive inpatient rehabilitation, and here he regained significant motor function in his left leg. His

Anterior spinal artery AVF caused by epidural spinal needle

neurological injury may have been attributable to direct trauma from the tract of the needle that went unrecognized or to the subarachnoid hemorrhage from the AVF and pseudoaneurysm. Postoperatively, the patient made a full recovery. At 20 months, despite persistent abnormalities of the spinal cord on MRI, motor evoked potentials obtained using a transcranial magnetic stimulator did not demonstrate any evidence of spinal cord damage. We were unable to perform somatosensory evoked potentials in this young patient without sedation.

Discussion

Lower limb weakness is a rare complication of thoracic epidural analgesia. The new onset of neurological symptoms following spinal epidural procedures calls for prompt diagnosis and treatment. The reported incidence of complicated thoracic epidural catheterizations in the literature is unclear, with limited studies comparing the incidence of neurological complications between lumbar and thoracic procedures. Prompt imaging of the spinal cord with unenhanced MRI allows one to visualize the location of the catheter tip; however, the compressive nature of a hematoma may obscure visualization of enlarged veins. Hence, spinal angiography is indicated when bleeding is demonstrated, particularly if the hemorrhage is believed to be subarachnoid. Evidence of cord compression requires urgent surgical evacuation and decompression.

There have been multiple reports of spinal hematomas in the epidural and subdural spaces following spinal anesthesia procedures, as well as reports of pseudoaneurysm and arteriovenous fistula formation following spinal anesthesia.

Fig. 1. Selected sagittal and axial MR images of the thoracic spinal cord (STIR, A; T2-weighted, B; T1-weighted, C and D) demonstrating penetrating cord injury with associated acute ventral subarachnoid hemorrhage. The tract through the spinal cord is best seen on the STIR sequence (white arrow).

Fig. 2. Spinal angiograms depicting consecutive phases of a selective left T-9 injection. Left: There is enhanced flow and increased caliber in the descending portion of the ASA (solid arrowheads) as compared with the ascending portion (open arrowhead). There is a probable pseudoaneurysm (arrow) arising from the ASA. Right: There is early shunting into pial spinal veins, suggesting a traumatic arteriovenous pial fistula (arrow).

Fig. 3. Follow-up arterial (A) and capillary (B) phase angiograms obtained from a selective left T-9 injection, showing no evidence of the previous injury with no pseudoaneurysm and no arteriovenous shunting. The caliber and flow within the descending portion of the ASA (arrow) appears to have normalized at 4 weeks after the surgery. Follow-up sagittal T2-weighted MR image (C) obtained at 4 weeks, demonstrating resolution of the hemorrhage with associated myelomalacia. Sagittal T2-weighted MR image (D) obtained 1 year postinjury, showing reduction in the amount of cord expansion and persistent myelomalacia.
Anterior spinal artery AVF caused by epidural spinal needle

of subarachnoid collections or direct cord injury.\textsuperscript{3,25,37,41,43} All were individual case reports, with patients complaining of new-onset back or neck pain following catheter removal. Subsequent imaging demonstrated small hematoma collections, which did not cause cord compression and did not require surgical intervention. All patients were managed conservatively via repeat imaging to monitor the size of the hematomas. Direct cord injury occurred in patients who had received thoracic spinal epidurals and presented with radiculomyelopathic symptoms at the level of the procedure. None of these cases required surgical intervention, and the patients were managed with observation.

The vascular anatomy of the thoracic cord is unique in that it receives a dominant inferior thoracic radiculopial supply (the artery of Adamkiewicz), which can arise from any level. The current patient’s left-sided symptoms were probably due to a combination of parenchymal injury from the needle and mass effect from the hematoma. The presence of a subarachnoid hemorrhage suggested vascular injury as opposed to ischemic injury to the cord. In cases with significant or progressive neurological deficit, surgical intervention is indicated with the goal of decompressing the spinal cord by evacuating the hematoma. Access to the ventral aspect of the cord can be improved by cutting the adjacent denticulate ligaments, although this maneuver does not necessarily provide clear visualization or control of the ASA. The bleeding that was encountered during removal of the ventral subarachnoid hematoma in our patient was probably attributable to the arterialized veins draining the AVF. Partial coagulation of these veins may have led to gradual resolution of the AVF and pseudoaneurysm. Spontaneous resolution of the traumatic aneurysm and fistula is also a possibility.

Ideal management strategies for traumatic pseudoaneurysms of the spine can be extrapolated from data derived from the natural history of intracranial pseudoaneurysms. Traumatic intracranial pseudoaneurysms compose 1% of all intracranial aneurysms, but their true incidence is probably underestimated given that many patients do not survive penetrating head injuries and thus do not warrant further investigation.\textsuperscript{26,40} Their estimated incidence is reported to be between 5% and 40% in the setting of penetrating head injuries.\textsuperscript{1,2,4,9,24,27} Diagnosis has been made possible since the advent of CT and digital subtraction angiography. The incidence of traumatic subarachnoid hemorrhage due to penetrating head injuries ranges from 31% to 78% based on CT scanning data and significantly correlates with mortality. While there is a paucity of data regarding the natural history and rate of rupture of traumatic aneurysms compared with saccular aneurysms, data do suggest that a smaller aneurysm size (that is, 2 mm) favors lower rates of rupture and spontaneous healing.\textsuperscript{5} For larger lesions with associated subarachnoid hemorrhage, early treatment is recommended once the diagnosis is made. Direct surgical repair of intracranial pseudoaneurysms can be challenging given a high risk of intraoperative rupture and the difficulty in reconstructing the damaged vessel. Proximal occlusion and segmental isolation, with or without bypass, are the most common approaches. Endovascular exclusion of the aneurysmal segments can be a safe alternative to surgical approaches.\textsuperscript{19,21,28,29,44} In the present case, the aneurysm and fistula were believed to be inaccessible to surgical treatment because of their ventral location. Similarly, a transarterial endovascular procedure to eliminate the fistula was believed to be too technically challenging and carried an unacceptable risk to the ASA. Given the risk of intervention in a neurologically improving patient and the probability of spontaneous resolution, a conservative approach was chosen.

Spinal vascular malformations are heterogeneous entities that vary in their pathophysiology and clinical presentation. Traditionally, lesions were categorized into 4 types: Type I, dural AVFs, low-flow fistulas between a radicular artery and medullary vein; Type II, intradural glomus malformations, lesions arising from multiple feeding vessels from the ASA and posterior spinal artery, draining into medullary veins with variable resistance; Type III, juvenile or multimeric arteriovenous malformations, large high-flow lesions with involvement of the paraspinal structures; and Type IV, perimedullary AVFs, an aberrant connection between the spinal arteries and medullary veins with no intervening capillary networks.\textsuperscript{8,37,30} Type IV perimedullary AVFs could be further broken down into Types I, II, and III, using the classification by Gueguen and colleagues,\textsuperscript{23} or Types IVa, IVb, and IVc, as suggested by Anson and Spetzler to avoid confusion.\textsuperscript{3} Type IVa lesions are small, low-flow lesions deriving supply from the ASA; Type IVb lesions have larger pial fistulas composed of distinct shunts fed by the ASA or posterior spinal artery; and Type IVc lesions are giant lesions with multiple dilated feeding arteries and large dilated veins draining from a single fistula.\textsuperscript{30}

A more succinct classification system was suggested by Oldfield and colleagues\textsuperscript{38} in 2002 based on these lesions’ distinctive biological, pathophysiological, and anatomical features: dural AVFs, intradural spinal cord arteriovenous malformations and AVFs, and cavernous angiomas of the spinal cord. Spinal dural AVFs are acquired lesions usually presenting in men in their 5th and 8th decades of life, whereas the others are classically congenital lesions formed by maldevelopment of the embryonic vasculature; however, there have been reports of acquired intradural AVFs both spontaneous\textsuperscript{42} and traumatic in origin.\textsuperscript{32} Our case is most analogous to a traumatic intradural pial AVF. Although there have been reports of successful endovascular treatment of perimedullary AVFs fed by the ASA,\textsuperscript{15,36,39} these small-caliber, slow-flow lesions make selective catheterization difficult, and thus surgery has been widely accepted as the primary treatment when these fistulas are safely accessible. Endovascular treatment has met with success in larger spinal AVFs fed by dilated arteries (Types IVb and IVc).\textsuperscript{30} In our case, surgery was warranted to evacuate the subarachnoid hematoma and decompress the spinal cord. As mentioned above, however, the fistula was not surgically accessible given its ventral location, and the primary goal of surgery should be aimed at the preservation of neurological function, not total obliteration of the fistula.

Conclusions

We describe the case of a 5-year-old boy with a trau-
matic intradural spinal AVF and pseudoaneurysm of the ASA following a spinal epidural procedure. The boy presented with a spinal subarachnoid hemorrhage and progressive neurological deficits following the removal of a spinal epidural catheter. Preserving neurological function is the main goal in the management of spinal cord vascular malformations, and one should recognize the high degree of morbidity associated with aggressive approaches aimed at total obliteration of the malformation. One should also be cognizant of the possibility of cord injury in a patient with new-onset neurological deficits following an interventional spine procedure and should not hesitate to order appropriate imaging studies for prompt diagnosis and treatment.

Disclosure

Dr. O’Kelly is a proctor for a pipeline embolization device case with ev3.

Author contributions to the study and manuscript preparation include the following. Conception and design: Mehta, Dicken. Acquisition of data: Mehta, Alnaami, Lam, O’Kelly. Analysis and interpretation of data: Mehta, Alnaami, Lam, O’Kelly, Aronyk. Drafting the article: Mehta, Alnaami, Lam, Steel, O’Kelly. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Mehta. Study supervision: Mehta.

Acknowledgment

The authors would like to thank Dr. Norton for his clinical neurophysiological assessment of this patient.

References

5. Anson JA, Spetzler RF: Classification of spinal arteriovenous malformations and implications for treatment. BNI Q 8:2–8, 1992
Anterior spinal artery AVF caused by epidural spinal needle

34. Moen V, Irestedt L, Räf L: [Review of claims from the Patient Insurance: spinal anesthesia is not completely without risks.] Lakartidningen 97:5769–5774, 2000 (Swedish)

Accepted December 4, 2012.

Please include this information when citing this paper: published online January 11, 2013; DOI: 10.3171/2012.12.PEDS12247.

Address correspondence to: Vivek Mehta, M.D., M.Sc., Division of Neurosurgery, University of Alberta Hospital, 8440 112 Street NW, Edmonton, Alberta, T6G 2B7, Canada. email: Vivek.Mehta@albertahealthservices.ca.