

Case Report

Surgical excision of filum terminale arteriovenous fistulae after lumbar fusion: Value of indocyanine green and theory on origins (a technical note and report of two cases)

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Abstract

Background: Intradural filum terminale arteriovenous fistulas (AVFs) are uncommon. We report two cases of this rare entity in which we used indocyanine green (ICG) videoangiography to identify the fistulous connection of each lesion.

Case Description: Two male patients presented with unresolved lower extremity weakness and paresthesias following lumbar fusion surgery. In each case, angiography showed an AVF between the filum terminale artery (FTA), the distal segment of the anterior spinal artery (ASA), and an accompanying vein of the filum terminale. A magnetic resonance image (MRI) obtained before lumbar fusion was available in one of these cases and demonstrated evidence of the preexisting vascular malformation. Surgical obliteration of each fistulous connection was facilitated by the use of ICG videoangiography. This emerging technology was instrumental in pinpointing fistula anatomy and in choosing the exact segment of the filum for disconnection.

Conclusion: Our findings indicate that intradural filum terminale AVFs may have a congenital origin and that ICG is a useful tool in their successful surgical management. As these cases demonstrate, spine surgeons should remain vigilant in evaluating patients based on their clinical symptomatology, even in the presence of obvious lumbar pathology.

Key Words: Arteriovenous fistula, filum terminale, indocyanine green videoangiography, intradural, perimedullary

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INTRODUCTION

Intradural filum terminale arteriovenous fistulas (AVFs) are exceedingly rare. Only three reports have focused specifically on an AVF arising from the filum terminale artery (FTA).^[6,13,24] Endovascular embolization and surgical ligation of these fistulae are the main treatment options. Interruption of the fistulous connection is

the goal of surgical treatment and remains the gold standard;^[23] multi-level decompressive laminectomy and stripping of dilated draining veins appears unnecessary.^[17] Difficulty often arises, however, in identifying the precise location of fistulization intraoperatively.

We present two cases of filum terminale AVE, which were treated surgically with the use of indocyanine

green (ICG) videoangiography. Interestingly, both of our patients underwent lumbar fusion years prior, and reported progressive symptoms after their surgeries. Pre-lumbar fusion imaging in one of our patients (unavailable in the other) showed evidence of an unrecognized AVF, indicating that the lesion was present prior to lumbar surgery, and possibly congenital, rather than iatrogenic.

CASE REPORTS

Case 1

History

A 57-year-old, left-handed gentleman, with a history of back pain and leg weakness presented with complaints of bilateral lower extremity numbness and difficulty walking. He underwent an L4–S1 postero-lateral fusion 2 years prior, without getting any relief from the symptoms [Figure 1]. For 3 months prior to presentation, he had experienced acute deterioration in his lower extremity strength, intermittent sensations of numbness and tingling bilaterally, bowel and bladder dysfunction, and had sustained multiple falls. He complained of having “two logs” attached to his body.

Examination

A neurological examination revealed bilateral proximal and distal lower extremity weakness, decreased lower extremity pinprick sensation and proprioception, decreased anal sphincter tone, and perineal hypoesthesia. Digital subtraction angiography demonstrated filling of the anterior spinal artery (ASA) extending past the conus and appearing to fistulize directly into a spinal vein in the region of the patient’s previous surgery [Figure 2].

Case 2

Presentation

A 63-year-old, right-handed gentleman, with a history of chronic back pain presented to an outside facility in 2007 with numbness and weakness in his right foot. He



Figure 1: X-rays of L4–S1 instrumentation and fusion

underwent an L4–L5 interbody fusion and pedicle screw-rod fixation. Following surgery, his back pain improved, but he complained of slowly worsening numbness and weakness involving his lower extremities. He sustained multiple falls postoperatively, including a fall that resulted in fractures of his femur and wrist.

Workup

An electromyography (EMG) suggested bilateral chronic L4 to S3 radiculopathy. A magnetic resonance image (MRI) obtained from after his lumbar fusion demonstrated flow voids in the lumbar cistern and increased T2 signal in the thoracic cord and conus medullaris. A spinal angiogram showed an AVF located around L4–L5. However, the precise location of artery–vein transition was not apparent despite sophisticated neurovascular imaging [Figure 3].

Operation

Under general anesthesia, each patient’s old incision was opened and dissection carried down to his respective spinal instrumentation [Video 1]. Extensive scarring from previous surgery obscured normal tissue planes. Once the thecal sac was opened and the arachnoid adhesions were taken down, it was noted in each case that the filum terminale harbored several enlarged blood vessels carrying arterialized blood [Figure 4]. Using a Zeiss Pentero IR 800 microscope (Carl Zeiss Co., Oberkochen, Germany), ICG videoangiography was performed, which demonstrated the exact point of fistulization in each case. This occurred on the terminal filum at its junction with the apex of the thecal sac in case one, and at the inferior extent of our exposure at L5 in case 2. Visualizing the filum with ICG videoangiography – and with the filum pinched with a bipolar forceps above the presumed fistulization site – intermittent brief release

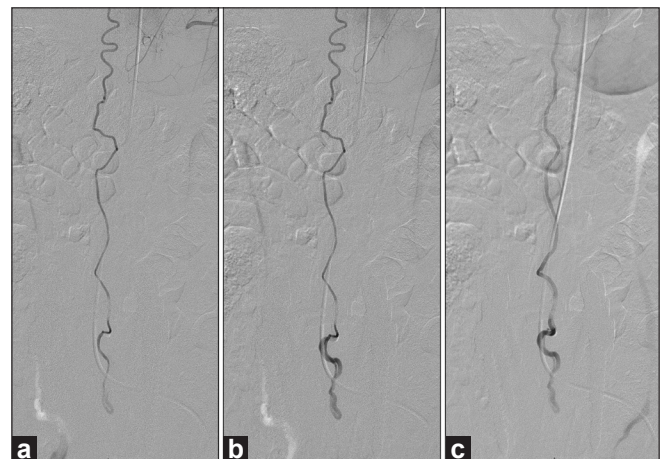


Figure 2: (a) Early phase spinal angiogram of the left T-10 intercostal artery injection, showing filling of the anterior spinal artery and the site of the filum terminale arteriovenous fistulas. (b) Later phase spinal angiogram showing initial fistulization. (c) Venous phase of spinal angiogram showing dilated and tortuous draining veins of the malformation



Figure 3: DynaCT demonstrating serpentine vessel opacification, unable to localize exact fistula site from this study. Lumbar fusion instrumentation (pedicle screws) is visible

of the forceps pressure caused progressive opacification of distal artery, fistula, and eventually the draining veins [Figure 5]. Repeat ICG videoangiograms showed that pressure directly on the identified fistula sites (as opposed to above them) completely eliminated the opacification of the draining veins. 3-0 silk was used to doubly ligate the filum a few millimeters above and below these sites, and then the filum was coagulated and divided [Figure 6]. The resected AVF was sent for pathology [Figure 7]. Final ICG videoangiograms demonstrated the absence of any abnormal vessel opacification.

Complications and postoperative course

Case 1 had two complications associated with his surgery. Intraoperatively, there was cauterization injury to his S3–S4 nerve roots due in part to obscured tissue planes from extensive epidural scarring. Impaired perineal sensation,

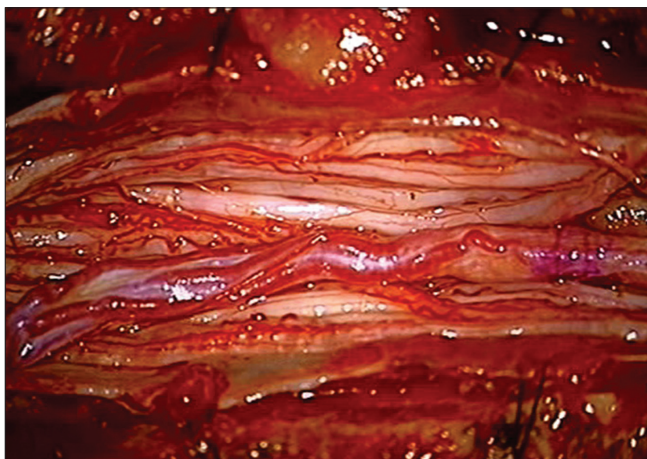


Figure 4: Intraoperative photograph showing the enlarged filum terminale artery connected to the arterialized dilated venous plexus

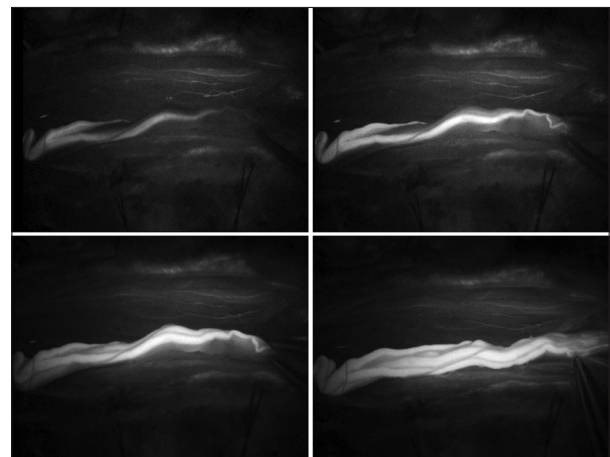


Figure 5: Indocyanine green videography showing enhancement with progressive filling of the fistula and draining vein with intermittent release of the feeding artery

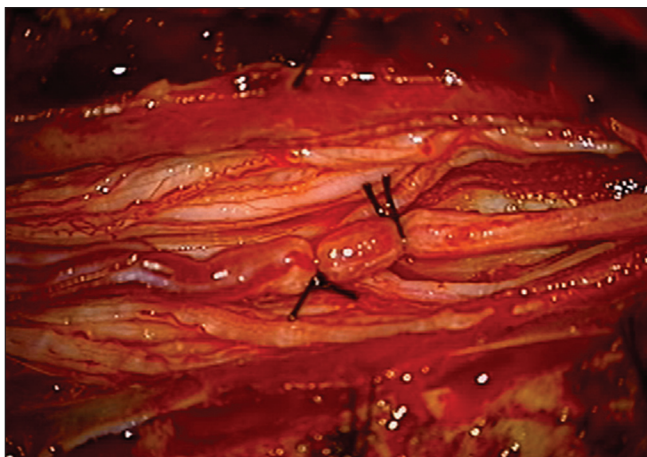


Figure 6: Intraoperative photograph showing ligated filum terminale fistula

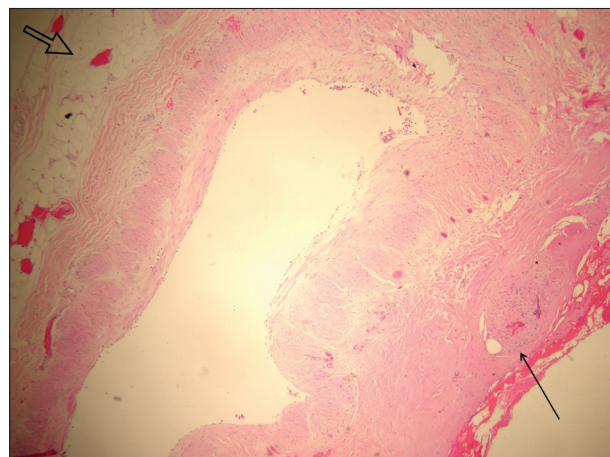


Figure 7: Artery and vein penetrating the thick, fibrous dura at the filum terminale with fibrofatty changes (bold arrow). Abrupt transition occurs between the feeding arterial vessel and venous vessel. Filum terminale vein at the center of the image is enlarged and filum terminale artery is distorted (black arrow)

rectal tone, and sphincter control improved during the course of admission and during his follow-up. He gained full control of his bowel and bladder within 90 days of his surgery. Scarring also made creating a hermetic seal of the thecal sac difficult, and a symptomatic pseudomeningocele formed and was repaired 1 month after his original surgery. Case 2 had no perioperative complications.

Both patients experienced dramatic improvement in lower extremity function immediately, which continued in the weeks and months following surgery. The patients reported increased and new sensations in the legs, and both were able to discard their walkers and ambulate independently.

DISCUSSION

Intradural ventral AVFs are characterized by a fistulous connection between the ASA and enlarged venous channels in the subarachnoid space.^[14] Also known as type 4 spinal arteriovenous malformations (AVMs)^[5] and direct AVFs,^[20] these lesions account for only 10% of spinal AVMs.^[7,26]

AVFs of the terminal filum are rare. A review of the literature yields only four reported cases of intradural terminal filum AVF fed by the artery of the filum terminale.^[6,13,24] Similar but disparate lesions include three cases of dural or intramedullary AVFs in the conus medullaris fed by the lateral sacral artery.^[15,16,22] In this report, our two cases had remarkably similar and intriguing clinical presentations and pathologies.

Treatment modality: Surgical obliteration

For the treatment of each of these filum terminale AVF, we chose surgical disconnection rather than endovascular embolization to provide definitive treatment. Multiple studies have concluded that surgery has advantages over endovascular embolization in terms of cure rate and complication rates.^[3,8,12,17,23] Results from a long-term retrospective study of 29 spinal dural AVF patients indicated that surgery had a high success rate and low morbidity.^[3] Of the few previously reported cases of filum terminale AVF, two were treated surgically^[24] and one was managed with a “multidisciplinary approach”. Jin *et al.*^[13] performed a transarterial embolization of the lateral sacral artery; however, because of the technical difficulty, microsurgical resection of arterial terminale fistula and draining vein was also performed.

In our cases, embolization was precluded given the anatomic relationship of the fistula to the ASA. Embolization of filum terminale AVF is complicated by the structural anatomy of the FTA; not only is the FTA serpiginous, risking penetration, but also its vascular integrity diminishes as it descends distally.^[6]

Use of indocyanine green

Surgical management of filum terminale AVF requires accurate visualization of the vascular anatomy and precise localization of the fistula. Failure to identify the site of fistulization can result in occlusion of normal vessels, leading to spinal cord or nerve root infarction.^[4] The use of ICG in our patients eliminated any uncertainty associated with identifying the fistula site, and allowed rapid surgical disconnection and confirmation.

ICG was first introduced for neurosurgical vascular application by Raabe *et al.*,^[18] and has since been used for all varieties of cerebrovascular surgery, including aneurysms, AVMs, and extracranial–intracranial bypass surgery.^[4,9-10,19,25] A modified digital camcorder (which is integrated with the surgical microscope; in our case, the Zeiss Pentero IR 800) detects the fluorescent light emitted by IC-green dye (Akorn, Buffalo Grove, IL, USA) binding to globulins and records video images of the field of view.^[9]

As highlighted by our operative technique, ICG allows tracking of the sequential arterial, capillary, and venous angiographic changes in real time. Temporary occlusion of the vasculature allowed us to start, stop, and pause the progression of vessel filling. The videoangiogram delineated how the ASA split into two vessels and how a third branch (the FTA) descended to fistulize exactly where the filum met the tip of the apex of the thecal sac (case 1) or at the level of L5 (case 2). In both cases, the arterial phase demonstrated abnormal early filling of the fistula and the filum terminale veins. After surgical extirpation of the fistula, ICG videoangiography demonstrated physiologic filling of the filum terminale veins and preservation of normal vasculature.

Compared with the use of radiological contrast agents, intravenous ICG is not associated with risk from radiation exposure or renal failure.^[10] Intravenous ICG is non-invasive, making it advantageous versus intraoperative angiography. In fact, for spinal AVF surgery, an angiogram is especially arduous as it requires the insertion of a long armored femoral sheath catheter before the patient is positioned prone.^[9-10] ICG may eliminate the need for a postoperative angiogram.^[9]

Theory on origins

Venous drainage of the filum has been at the center of previous theories on the origin of filum terminale AVFs. Tender *et al.*^[24] reported two cases of filum terminale AVF and speculated that this lesion may be acquired in origin. They noted that a deficiency in medullary spinal venous drainage and a predominance of rostral drainage, combined with arterialized venous input from the AVF, may predispose to the development of venous congestion and myelopathy. This concern about lateral drainage may be overemphasized. The lateral medullary venous system may have a more variable drainage system

than the longitudinal system,^[21] and the vein of the filum terminale only drains vertically in two directions.^[6] Ambiguity of lateral drainage on angiogram may also be due to the diminutive size of the veins, arteries, and capillaries in comparison to the FTA and vein.^[6]

Rosenblum *et al.*^[20] reviewed 81 cases of spinal AVMs and deduced that intradural AVMs are congenital in origin, whereas dural AVMs are acquired lesions. Jin *et al.*^[13] presented one case of perimedullary AVF of the terminale filum and theorized that the fistula was the result of a congenital lesion. There has been one report of iatrogenic perimedullary fistula in a patient who had undergone resection of a conus ependymoma.^[2] We present two cases of intradural terminale filum AVF and we have reason to believe at least one case was congenital in origin. Our two patients were diagnosed with lumbar degenerative disease for which they underwent lumbar fusion. However, the surgery failed to resolve their radiculomedullary symptoms. Review of a lumbar MRI obtained before surgery showed evidence of a preexisting intradural vascular malformation in case 1 [Figure 8]. It is likely that these filum terminale AVFs were the original perpetrators of many of each patient's symptoms.

Vascular injury is a well-known complication following lumbar disc surgery.^[1,11] Most complications from lumbar laminectomy occur at the L4–L5 region; however, a review of literature yields only two acquired cases of dural AVMs following lumbar disc surgery: 1) a 60-year-old man with S1 dural AVF 3 years after lumbar discectomy^[1] and 2) a 27-year-old man with a L5 dural AVM 7 years after lumbar disc herniation surgery.^[27] In the case of the L5 dural AVM,^[27] the vascular malformation developed contralateral to the previous surgical site, confounding the concept that this was an iatrogenic lesion.^[1,27]



Figure 8: Sagittal T2-weighted MRI of thoracolumbar spine revealing flow voids (arrow) in the low thoracic spinal levels, indicative of a spinal arteriovenous malformation. This MRI was obtained prior to lumbar fusion surgery and demonstrates evidence of arteriovenous fistulas in the presence of degenerating L4 disc

There have been no identified iatrogenic cases of filum terminale intradural AVF. Based on the clinical course of our patients and intraoperative observations, we do not believe the previous lumbar fusion operations led to the formation of an AVF. There was neither any evidence of dural tears noted at the time of the original surgery nor were they seen upon reoperation. It is difficult to envision how an extradural spine procedure with no evidence of penetration through the dura could have caused these intradural vascular defects.

CONCLUSION

We report the use of ICG videoangiography in the surgical treatment of two rare cases of filum terminale AVF. ICG videoangiography allowed intraoperative localization of the fistulae sites and confirmation of their disconnection from the venous drainage system. The unique anatomy of the filum terminale presents navigational challenges for endovascular treatment, and microsurgery, especially with the aid of ICG, may be the treatment of choice for most of these lesions. Evidence of arteriovenous fistula on pre-lumbar disc surgery MRI in case 1 and resolution of myelopathy following surgical extirpation of both fistulas suggest that 1) the filum terminale arteriovenous fistulas were the original cause for symptoms and 2) intradural filum terminale AVF may be congenital in origin.

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