Cureus

Open Access Case Report

DOI: 10.7759/cureus.201

Spinal Dural Arteriovenous Fistulas Supplied from Multiple Segmental Arteries: Microsurgery Using Indocyanine Green Angiography and Radiosurgery with a Spine Tracking System

Hiroshi K. Inoue [1] , Hiro Sato [2] , Isao Naito [3]

1. Dept of Neurosurgery and Radiation Oncology, Institute of Neural Organization and Cyber Center, Kanto Neurosurgical Hospital 2. Department of Radiation Oncology, Gunma University Graduate School of Medicine 3. Neurosurgery and Endovascular Suergery, Geriatrics Research Institute and Hospital

Corresponding author: Hiroshi K. Inoue, chair.ino@instno-med.org

Disclosures can be found in Additional Information at the end of the article

Abstract

Endovascular occlusion and/or surgical disconnection are the first treatment of choice for spinal dural arteriovenous fistulas (d-AVFs) with venous flow reversal into the perimedullary veins. However, d-AVFs with multiple feeding arteries and fistula points are associated with various difficulties related with treatment. We herein describe three cases of AVFs supplied from multiple segmental arteries treated with surgery using indocyanine green (ICG) angiography or radiosurgery using a spine tracking system. ICG was administered via intravenous injection during and after the surgical disconnection procedures. As double d-AVFs or multi-segmental ventral epidural type AVFs require invasive spinal surgery, less invasive radiosurgery was performed using a CyberKnife with a spine tracking system. ICG angiography revealed changes in venous flow reversal during the surgical maneuvers as well as the disappearance of the AVF after disconnecting all draining veins in Case 1. Among the patients with double AVFs or multi-segmental ventral epidural type AVFs treated with radiosurgery, the fistula points received a marginal dose of 15 to 18 Gy at an isodose of 60% to 67%. The AVFs disappeared or almost disappeared on angiography one year after radiosurgery in Case 2. The signal void on T2-weighted images showing a venous pouch disappeared, the draining veins normalized, and the cord compression was alleviated two years after radiosurgery in Case 3. No episodes of recurrence were noted in any of the patients four to seven years after treatment. ICG angiography plays an important role in obtaining a complete disconnection of the draining veins during complex AVF surgery. Radiosurgery is indicated for spinal d-AVFs with multiple feeding arteries and fistulas in patients without progressive symptoms.

Received 08/27/2014
Review began 08/27/2014
Review ended 09/09/2014
Published 09/12/2014

Categories: Neurosurgery
Keywords: spinal dural avf, multiple feeding arteries, radiosurgery, indocyanine green, intraoperative angiography, residual avf, video, surgery, endovascular treatment, multiple segmental levels

Introduction

Spinal dural arteriovenous fistulas (d-AVFs) are classified into three groups: 1) ventral epidural (previously called "epidural", "osteodural", or "paravertebral" AVF), 2) dorsal epidural, and 3) lateral epidural (most common "classic type" of d-AVF). Symptoms in Group 2 appear suddenly, with epidural hematoma formation, and progressively in Group 3 as a result of abnormal

How to cite this article
Inoue H K, Sato H, Naito I (September 12, 2014) Spinal Dural Arteriovenous Fistulas Supplied from Multiple Segmental Arteries: Microsurgery Using Indocyanine Green Angiography and Radiosurgery with a Spine Tracking System. Cureus 6(9): e201. DOI 10.7759/cureus.201

drainage (flow reversal) into the perimedullary veins. In Group 1, symptoms develop slowly due to compression of the spinal cord and/or nerve roots by enlarged epidural venous pouches [1]. Treatment with endovascular occlusion and/or surgical disconnection of fistulas is required in patients with acute or rapidly progressive symptoms before the spinal cord damage becomes irreversible [2-4]. Benign fistulas are a developing indication for radiosurgery [5].

Endovascular intervention for spinal d-AVFs is a less invasive approach and primary treatment option [6]. A cure rate of 25-77% for endovascular embolization using liquid adhesive materials for classic type d-AVs has been reported [2, 7], and rapid improvements in symptoms are obtained after endovascular treatment [4]. However, it is difficult to treat d-AVFs supplied from multiple segmental arteries or the same pedicle to the anterior spinal artery using endovascular intervention. Moreover, it is necessary to manage residual d-AVFs after embolization with surgery or radiosurgery, as symptoms may remain or recur and new anastomoses may develop.

Surgical disconnection of the draining veins is the first choice of treatment for spinal d-AVFs with venous flow reversal, in addition to endovascular occlusion. Patients with such AVFs experience progressive symptoms due to edema of the spinal cord or the sudden onset of symptoms as a result of subarachnoid hemorrhage (SAH) [8]. Early surgery is recommended in cases involving the rapid progression of symptoms (Figure *1*). The rate of a surgical cure is very high, with a low incidence of complications.

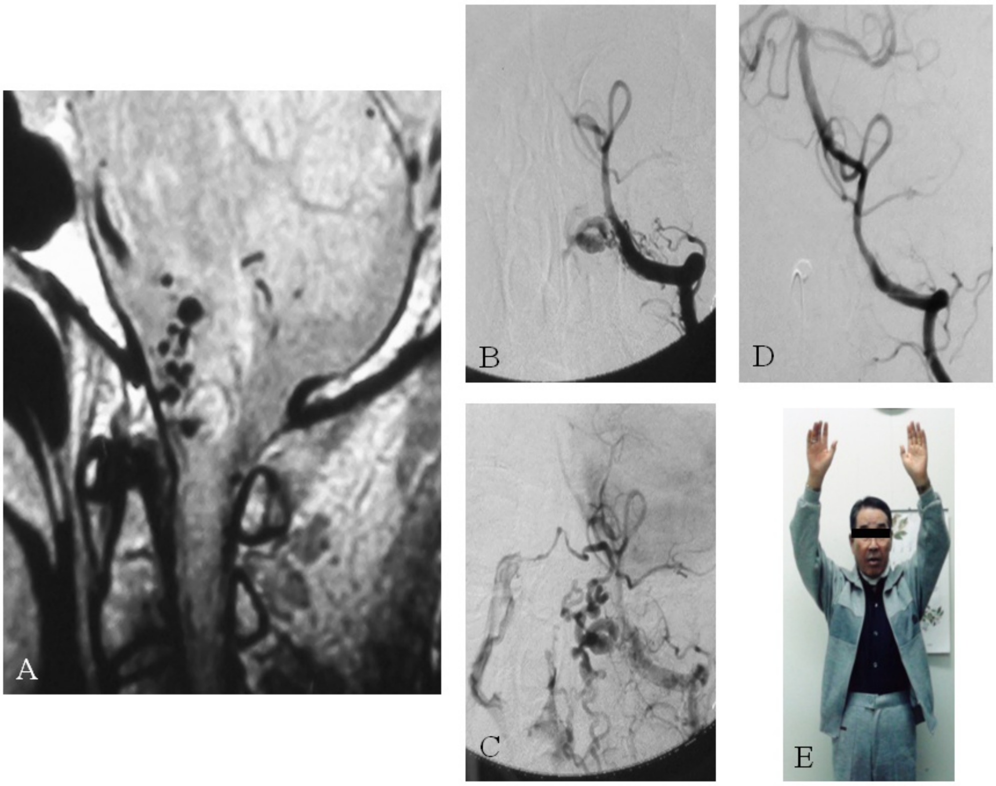


FIGURE 1: A d-AVF at the cervico-medullary junction

A: A MR proton density image showing edema in the medulla with a signal void around the brainstem. B, C: Vertebral angiogram demonstrating a d-AVF at the cervico-medullary junction. D: Following surgical disconnection of the fistula, the AVF disappeared on the angiogram. E: The patient’s symptoms (tetra-paresis, dysphagia, respiratory distress) rapidly improved, with no recurrence for 17 years after surgery.

However, surgical difficulties are often encountered in patients with large d-AVFs supplied from multiple segmental feeding arteries or d-AVFs with feeding arteries originating from a different level than the fistula point [3]. The recently developed indocyanine green (ICG) angiography technique is very useful for detecting feeding arteries or draining veins at different levels [9]. The intra-arterial administration of ICG has also been reported to have benefits with respect to the use of multiple and short interval injections versus intravenous administration, although it is necessary to place a catheter into the target artery prior to surgery [10]. At our facility, we use ICG angiography to detect feeders and drainers as well as confirm the complete disconnection of the fistulas during surgery.

Radiosurgery is a less invasive treatment option for benign cranial d-AVFs [5]. In cases of benign spinal d-AVFs, the ventral epidural type, AVFs with non-progressive symptoms, or non-symptomatic residual AVFs remaining after endovascular intervention are considered indications for radiosurgery, especially those with feeding arteries from multiple spinal segments. AVFs exhibiting flow reversal into the perimedullary veins are not indicated due to risks of bleeding (SAH) and the potential for progressive symptoms during the latent periods prior to total obliteration, similar to cranial d-AVFs with flow reversal into the cortical veins. However, patients with high risks for surgery (general anesthesia) and/or endovascular intervention, such as those with AVFs fed from the same pedicle supplying the anterior spinal artery, are indicated for radiosurgery as a less invasive treatment option.

In this report, we present three cases of spinal d-AVFs treated with microsurgery using ICG angiography and radiosurgery with a spine tracking system. Informed patient consent was obtained or waived for all subjects involved in this report.

Case Presentation

Case 1 (Microsurgery)

A 53-year-old male visited a former hospital due to paraparesis and urinary incontinence and was diagnosed with a spinal d-AVF. Endovascular intervention was intended; however, a provocation test revealed worsening of the paraparesis. The patient then visited our hospital for possible radiosurgery in June 2009. Due to his progressive symptoms, surgical disconnection of the AVF was planned and detailed imaging studies for microsurgery were performed. T2-weighted MR imaging showed edema extending from the lower thoracic spinal cord to conus medullaris and signal void in perimedullary areas (Figure *2*). CT angiography demonstrated perimedullary tortuous vascular structures identical to the void on MR imaging as well as vascular structures (suggesting the presence of feeding arteries) from the right L1 to L3 segment.

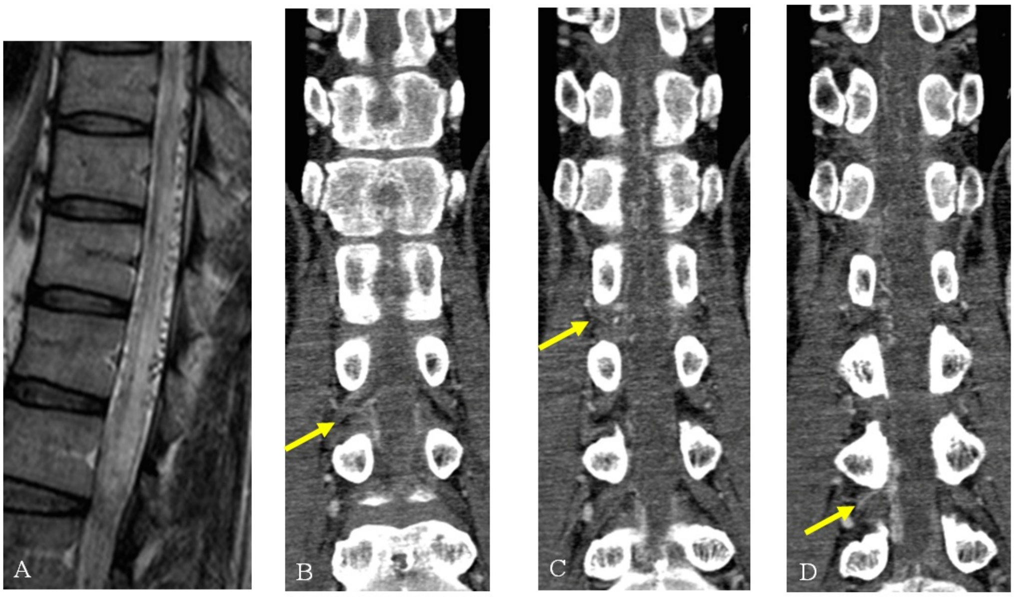


FIGURE 2: A d-AVF at the level of the conus medullaris

A: A T2-weighted, sagittal MR image showing spinal cord edema and a signal void in perimedullary areas expanding from the Th10 to L1 level. B, C, D: Coronal slices of CT angiography showing tortuous vascular structures extending from the Th10 to L4 level. The perimedullary vascular structures were identical to the signal void observed on MR imaging. Segmental arteries extending from the L1 to L3 level (arrows) appeared to be related to the AVF.

Spinal angiography disclosed a d-AVF fed from the right L1 and L2 segmental arteries (Figure 3).

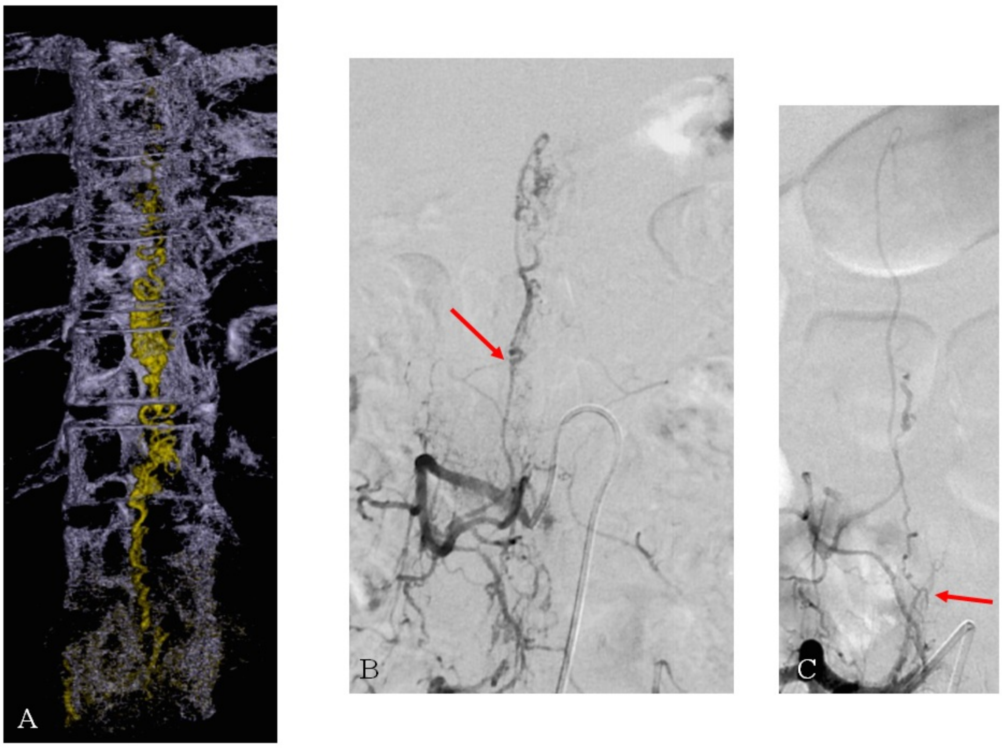
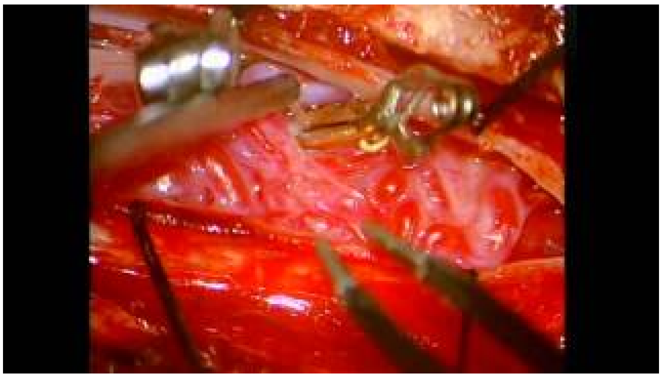


FIGURE 3: A d-AVF at the level of the conus medullaris

A: A 3-D reconstruction image of rotation angiography demonstrating the AVF from the ventral view. B: Spinal angiography (oblique view) of the right L1 segmental artery showing the anterior spinal artery and a feeding artery originating from the same pedicle. The fistula point existed at the Th12 level (arrow) with ascending perimedullary venous drainage. C: Spinal angiography (oblique view) of the right L2 segmental artery showing the other feeding artery and fistula point (arrow). The anterior spinal artery was not related with the AVF.

No feeding arteries were demonstrated from the bilateral Th12 segments. Surgical disconnection was performed in July 2009 at the level of L1-L2 (via hemi-laminectomy), and the intradural draining veins were disconnected by coagulation. The dura, including the fistula point and feeding artery, was coagulated simultaneously in the intra- and extradural spaces. Postoperative angiography showed the disappearance of the lower part of the AVF. However, the upper part of the AVF fed from the different segmental level of Th12 exhibited no changes on follow-up MR images. The residual AVF was subsequently treated in September 2009 at the level of Th12-L1 under guidance with ICG angiography during the procedure. As the right dorsal rootlets of Th12 segment were entirely involved with dilated draining veins and the subarachnoid space around the root exit zone was very difficult to open, the draining veins from the fistula were clipped in order based on the images of ICG angiography. The stepwise disappearance of the perimedullary draining veins was confirmed. Finally, the last residual shunt was coagulated and disconnected. ICG angiography confirmed the lack of flow reversal with a normal venous circulation (video).



VIDEO 1: Spinal dural arteriovenous fistula: ICG angiography

The draining veins from the fistula were clipped in order based on the images of ICG angiography and the stepwise disappearance of the flow reversal was confirmed. Finally, the last residual shunt was coagulated and disconnected. ICG angiography confirmed the lack of flow reversal with a normal venous circulation.

View video here: https://www.youtube.com/watch?v=KqidaW5_roU&feature=youtu.be

The postoperative MR images showed the disappearance of the signal void and spinal cord edema (Figure 4), and postoperative angiography demonstrated the disappearance of the AVF. The patient's paraparesis and urinary incontinence gradually improved, allowing him to walk with a stick. No episodes of recurrence have been observed for five years after the surgery.

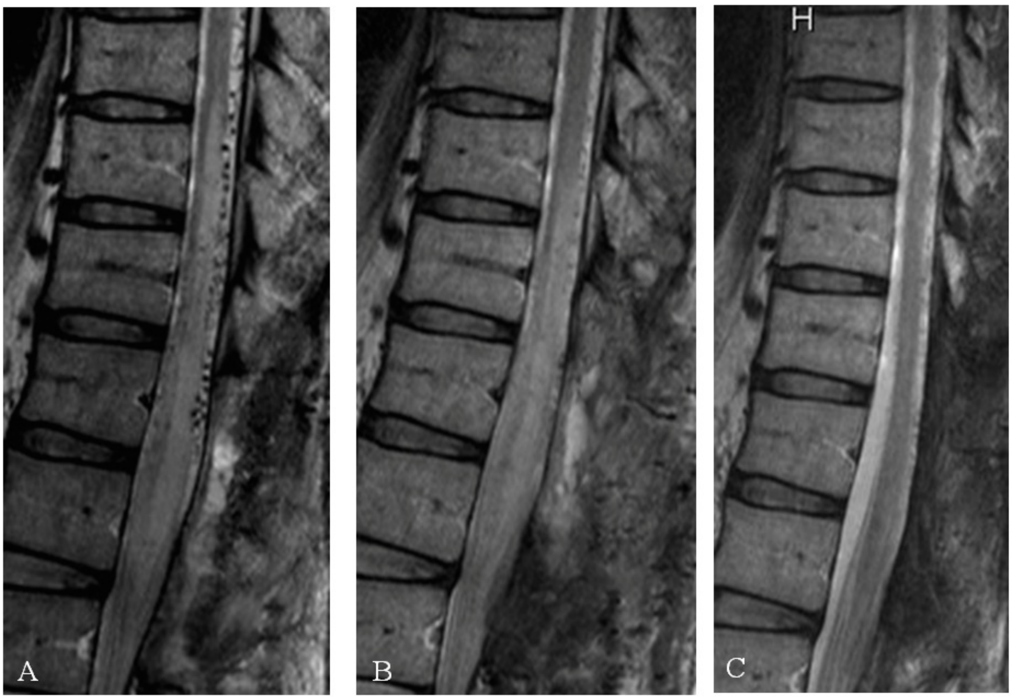


FIGURE 4: A d-AVF at the level of the conus medullaris

A: A T2-weighted, sagittal image showing spinal cord edema and a residual signal void one month after the first operation. B: A T2-weighted, sagittal image demonstrating the disappearance of the signal void one month after the second surgery. C: A T2-weighted, sagittal image revealed the disappearance of the spinal cord edema eight months after the revision surgery.

Case 2 (Radiosurgery)

A 66-year-old female visited a former hospital on July 2006 due to the sudden onset of severe neck pain and was diagnosed with SAH due to d-AVFs in the cervical region. Endovascular occlusion was performed; however, residual AVFs were recognized at the C2, C5, and C6 levels on follow-up angiography after treatment. The patient had no symptoms and was subsequently referred for radiosurgery. She visited our hospital in November 2006. Right vertebral angiography demonstrated spinal d-AVFs fed from the C2-C3 and C5-C6 segmental arteries (Figure *5*). These AVFs were treated with a dose of 18 Gy at an isodose of 60% and 63%, respectively, using fused image of angiography under a spine tracking system.

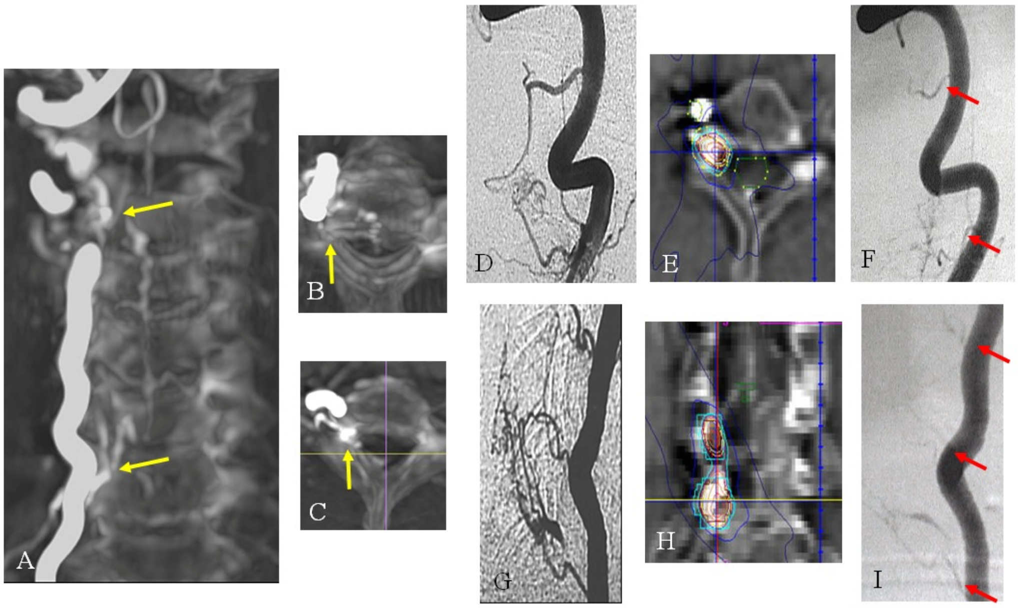


FIGURE 5: Residual d-AVFs after embolization in the cervical region

A-C: The 3-D reconstruction images of rotation angiography demonstrating AVFs on coronal (A) and axial slices (B: C2 level, C: C6 level). Arrows showing fistula points of the AVFs. D: Vertebral angiography (lateral view) presenting a small d-AVF at the C2 level fed from right C2 and C3 segmental arteries. E: An axial reconstruction image of angiography showing the fistula point treated with a dose of 18Gy at an isodose of 60%. F: The AVF disappeared and the dilated feeding arteries normalized (arrows) one year after the radiosurgery procedure. G: Vertebral angiography (lateral view) presenting a d-AVF with fistula points at the C5 and C6 levels fed from multiple segmental arteries. H: A coronal reconstruction image of angiography showing the fistula points treated with a dose of 18Gy at an isodse of 63%. I: The AVF almost disappeared and the dilated feeding arteries normalized or disappeared (arrows) one year after the radiosurgery procedure.

The AVFs disappeared or almost disappeared one year after radiosurgery. Although no follow-up angiography is available yet, the patient has shown no deterioration in symptoms for seven years after treatment.

Case 3 (Radiosurgery)

A 43-year-old male, who had a history of a left leg arteriovenous malformation treated by endovascular intervention, visited a former hospital in April 2007 due to a gait disturbance and was diagnosed with multi-level paravertebral AVFs (ventral epidural type) in the thoraco-lumbar region. Endovascular occlusion was performed, and the symptoms improved. However, he suffered from back pain in June 2009, and the multi-level AVFs (Figure 6) were again treated with endovascular embolization.

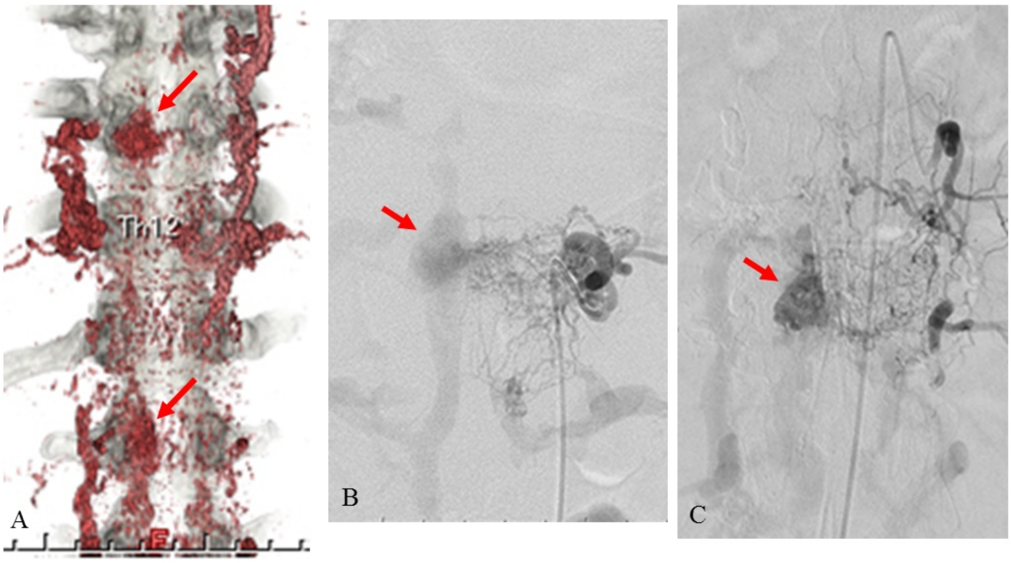


FIGURE 6: Multi-segmental d-AVFs (ventral epidural type) in the thoraco-lumbar region

A: CT angiography demonstrating multi-segmental d-AVFs and venous pouches (arrows). Spinal angiography of the left Th11 segmental artery (B) and L1 segmental artery (C) showing multi-segmental d-AVFs and large venous pouches at the Th11 and L2 levels (arrows).

Unfortunately, he experienced allergic anaphylactic shock to the iodine contrast material during the last procedure in September 2009. Repeated episodes of allergic shock occurred in December 2009 and January 2010, and additional endovascular intervention became impossible thereafter. He therefore visited our hospital for radiosurgery in May 2010. Although no contrast-enhanced studies were available, CT revealed bone erosion of the right Th11 vertebral body, and T2-weighted MR images showed a signal void identical to the enlarged epidural venous pouch compressing the spinal cord and dilated draining veins at the Th11 level seen on the angiogram (Figure 7). The venous pouch, including the fistula points, were treated with a dose of 15 Gy at an isodose of 67%.

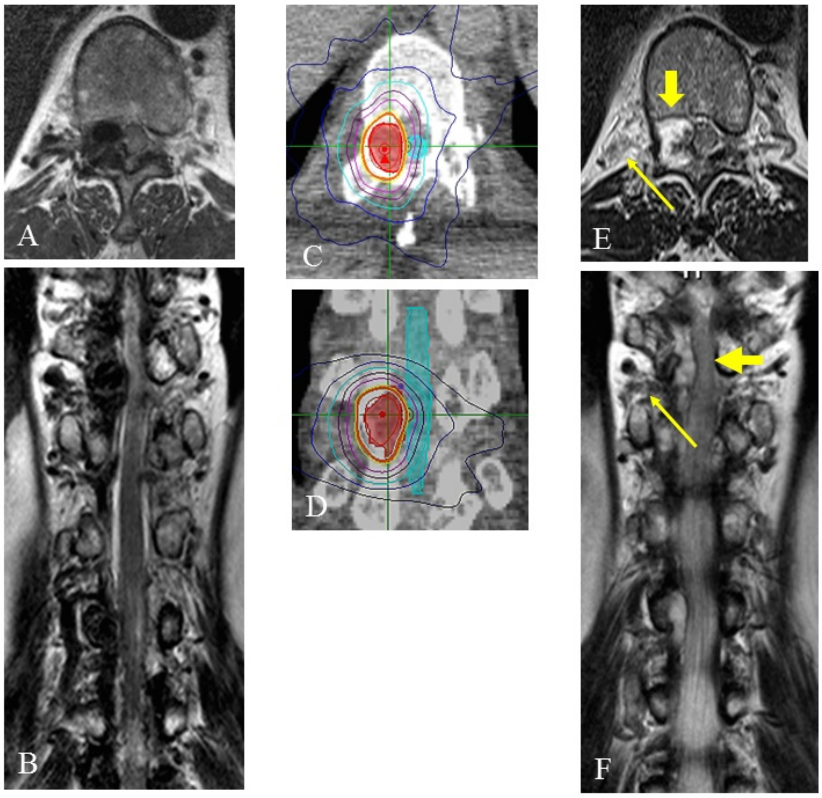


FIGURE 7: Multi-segmental d-AVFs (ventral epidural type) in the thoraco-lumbar region

T2-weighted axial (A) and coronal (B) images demonstrated a signal void in multiple segments extending from the Th10 to L3 level, identical to the dilated veins noted on the angiogram. The spinal cord was compressed at the Th11 level by the large venous pouch. The venous pouch, including the fistula point, was treated with a dose of 15 Gy at an isodose of 67% (C: axial, D: coronal). T2-weighted axial (E) and coronal (F) images revealed the disappearance of the signal void in the venous pouch and an improvement of the spinal cord compression two years after the radiosurgery procedure (large arrows). The dilated vein had also normalized (thin arrows). Angiography was not performed due to the patient's allergic anaphylactic shock reaction to iodine contrast material.

MR images obtained regularly after treatment revealed a gradual decrease in the signal void, while T2-weighted images showed a decrease in the size of the pouch and draining veins two years after radiosurgery. The patient's symptoms of gait disturbance and urinary incontinence gradually improved and almost disappeared. He subsequently returned to his previous work one year after the radiosurgery treatment, with no deterioration in symptoms for four years after.

Discussion

Most patients with spinal d-AVFs develop progressive symptoms comprising sensory abnormalities, motor weakness, and disturbances in urination due to edema of the spinal cord. Surgical disconnection is the gold standard treatment for such patients. However, endovascular intervention is also a first choice of treatment as a less invasive procedure [5-6]. Patients in whom complete disconnection or total obliteration are obtained show improvements in their symptoms immediately after treatment and are permanently cured. Providing an early diagnosis and proper treatment before irreversible changes in the spinal cord occur is therefore important. However, there are treatment difficulties associated with both surgery and endovascular intervention. Additional options are required in such cases, particularly those involving the incomplete obliteration of AVFs after surgery or embolization.

The use of ICG angiography during surgery plays an important role in achieving complete disconnection [11-12], especially in patients with AVFs supplied by multiple feeding arteries and/or fistula points at segmental levels different from that of the feeding artery. We herein demonstrated its usefulness in evaluating changes in the reversal flow during the procedures and confirming the total disappearance of the lesion following disconnection (video). As to disadvantages of ICG angiography, vascular structures lying within or behind the spinal cord can't be evaluated using this modality, in contrast to intraoperative DSA, which can be used to define structures outside of the operative field. The draining veins in the ventral direction should be defined directly under a microscope after cutting the dentate ligament and exposing the ventral roots anteriorly, as shown in the included video. The intra-arterial injection of ICG may help to evaluate the vascular structures in various operative fields, as repeat injections may be given within short times intervals [10], although this technique is difficult to use in cases of AVFs with multiple feeding arteries. The present patient required revision surgery for the residual AVF with a fistula point at the level of Th12, but not the level of L1, where the feeding artery was located. As all fistula points must be defined directly in the operative field for surgical disconnection, we believe that it was better to include the level of Th12 in the first operation in order to avoid the need for a two-step procedure in this case. However, the fistula point at the level of Th12 was atypical, and the dorsal rootlets and root exit zone were entirely involved and occupied by the enlarged perimedullary draining veins. Using a conventional small dural opening for the d-AVFs disconnection around the root exit zone seemed to be impossible in this case.

Spinal d-AVFs present at multiple segmental levels that are fed by multiple feeding arteries are difficult to treat with microsurgery [13]. Endovascular intervention is a first choice of treatment in such cases; however, it is not easy to obtain total obliteration in many cases, as shown in the current Cases 2 and 3. The purpose of embolization is to reduce the shunt flow of AVFs, and to prevent re-bleeding (SAH) and/or to improve symptoms, particularly progressive paraparesis. Residual or benign AVFs, such as ventral epidural type AVFs or AVFs with unrelated clinical symptoms [14], are also a developing indication for radiosurgery [5], and multi-segmental level AVFs are possible targets for radiosurgery using a spine tracking system, as shown in this report. D-AVFs (fistula points) are usually situated extradurally with some distances to the spinal cord. Hence, an effective marginal dose of 18 to 20 Gy is applicable in most cases. Radiosurgery apparently increases the number of treatment indications for d-AVFs, especially in combination with surgery or endovascular intervention. However, the use of radiosurgery requires a latency period of at least six to 12 months before effective changes and/or a desirable level of obliteration are obtained, with one to two years needed for total obliteration. The risks of re-bleeding or symptomatic deterioration must also be considered before and after radiosurgery up to the point of total obliteration, and providing regular follow-up studies after treatment is important. We intend to follow the current patients with d-AVFs using the newly developed spinal MR angiography technique due to its reduced invasiveness and lack of need for contrast materials. MR angiography may thus be used to treat and follow patients with d-AVFs, especially those similar to the patients described in the preset Case 3.

Conclusions

We herein reported three cases of residual AVFs supplied from multiple segmental arteries treated with microsurgery and/or radiosurgery. The use of ICG angiography during surgery is very helpful for obtaining and defining complete disconnection of the AVF. In addition, radiosurgery using a spine tracking system was found to be safe and effective for achieving obliteration of the AVFs with multiple feeding arteries in the present report. Although no episodes of recurrence have been noted four to seven years after treatment in our patients, long-term follow-up studies are required. The application of multidisciplinary and/or combination treatment with endovascular intervention, microsurgery, and radiosurgery increases the treatment indications for complex spinal d-AVFs, which were previously difficult to manage.

Additional Information

Disclosures

Human subjects: Consent was obtained by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

References

1. Krings T, Geibprasert S: Spinal dural arteriovenous fistulas. AJNR Am J Neuroradiol. 2009, 30:639-48. 10.3174/ajnr.A1485
2. Van Dijk JM, TerBrugge KG, Willinsky RA, Farb RI, Wallace MC: Multidisciplinary management of spinal dural arteriovenous fistulas: Clinical presentation and long-term follow-up in 49 patients. Stroke. 2002, 33:1578-83.
3. Schick U, Hassler W: Treatment and outcome of spinal dural arteriovenous fistulas . Eur Spine J. 2003, 12:350-5.
4. Agarwal V, Zomorodi A, Jabbour P, Chalouhi N, Tjoumakaris S, Babu R, Back A, Gonzalez LF: Endovascular treatment of a spinal dural arteriovenous malformation (DAVF) . Neurosurg Focus. 2014, 37:1. 10.3171/2014.V2.FOCUS14180
5. Steiger HJ, Hänggi D, Schmid-Elsaesser R: Cranial and spinal dural arteriovenous malformations and fistulas: An update. Acta Neurochir Suppl. 2005, 94:115-22.
6. Heldner MR, Arnold M, Nedeltchev K, Gralla J, Beck J, Fischer U: Vascular diseases of the spinal cord: A review. Curr Treat Options Neurol. 2012, 14:509-20. 10.1007/s11940-012-0190-9
7. Kirsch M, Berg-Dammer E, Musahl C, Bäzner H, Kühne D, Henkes H: Endovascular management of spinal dural arteriovenous fistulas in 78 patients. Neuroradiology. 2013, 55:337-43. 10.1007/s00234-013-1134-0
8. Aviv RI, Shad A, Tomlinson G, Niemann D, Teddy PJ, Molyneux AJ, Byrne JV: Cervical dural arteriovenous fistulae manifesting as subarachnoid hemorrhage: Report of two cases and literature review. AJNR. 2004, 25:854-858.
9. Choudhri O, Heit JJ, Gupta M, Marks MP, Dodd R: Use of fluorescent indocyanine green angiography during microsurgical resection of a recurrent giant perimedullary cervical spinal arteriovenous fistula in a patient with hereditary hemorrhagic telangiectasia. Cureus. 2014, 6:e185. 10.7759/cureus.185
10. Horie N, So G, Debata A, Hayashi K, Morikawa M, Suyama K, Nagata I: Intra-arterial indocyanine green angiography in the management of spinal arteriovenous fistulae: Technical case reports. Spine (Phila Pa 1976). 2012, 37:E264-7. 10.1097/BRS.0b013e31822ba834
11. Cohen-Gadol A: Surgical correction of a spinal arteriovenous fistula with an unusual presentation. Neurosurg Focus. 2012, 33:video 1. 10.3171/2012.V2.FOCUS12145
12. Fontes RB, Tan LA, O'Toole JE: Minimally invasive treatment of spinal dural arteriovenous fistula with the use of intraoperative indocyanine green angiography. Neurosurg Focus. 2013, 35:video 5. 10.3171/2012.V2.FOCUS13191
13. El-Serwi A, Maubon A, Chapot R: Double spinal dura arteriovenous fistulas . AJNR. 2006, 27:321-323.
14. Van Rooij WJ, Nijenhuis RJ, Peluso JP, Sluzewski M, Beute GN, Van Der Pol B: Spinal dural fistulas without swelling and edema of the cord as incidental findings. AJNR. 2012, 33:1888-

1892.