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Case Report

Multidisciplinary management of multiple spinal dural arteriovenous fistulae

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Abstract: Multiple SDAVFs are quite rare. We present two cases with double synchronous shunts and both were treated during one-stage interventional or surgical procedure. Unique images of the multiple SDAVFs as a PMAVF-like fistula were obtained. These interesting findings suggest the presence of multiple fistulas must be considered in patients being evaluated for SDAVF. A multidisciplinary approach to the management of multiple SDAVFs should depend on the anatomic location and angioarchitecture.

Keywords: Multiple spinal dural arteriovenous fistulae, perimedullary arteriovenous fistula, multidisciplinary management

Introduction

Multiple spinal dural arteriovenous fistulae (SDAVFs) are quite rare and only few cases were reported [1-3]. All instances reported before have a metachronous feature, we describe two cases presented with double synchronous shunts and treated with multidisciplinary management.

We report a surgery confirmed case of multiple SDAVFs which was initially diagnosed as a single SDAVF combined with coexisting perimedullary arteriovenous fistula (PMAVF). Two fistulae were surgically treated at the same time. Case 2 presented first fistula identified by the serpentine veins evident on Magnetic resonance imaging (MRI), and spinal angiogram demonstrated another independence fistula. Both fistulae were successfully obliterated through an exclusively endovascular approach.

Case report

Case 1

The patient is a 30-year-old male with a 10-year history of dysaesthesia in the lower extremity, diagnosed as spinal arteriovenous malformation at that time evident on angiogram from an outside hospital, and he opted for conservative treatment. This time he was admitted to our hospital with the sensory impairment of the legs and sphincter disturbances rapidly progressed (grade 3, Aminoff-Logue Scale, Gait: 2 Micturition: 1) in the last 10 months. Neurological examination showed a sensory level at T10, motor strength decreased (4/5), knee reflex exaggerated on both sides. MRI showed a hypointense flow avoid venous signal on the dorsal surface of the spinal cord extending from T7 to L1, and a mixed signal on T2 suggesting hemorrhage at the level of T11/T12 intervertebral disc (**Figure 1**). There was no clinical evidence of a connective tissue disease and no history of trauma.

A diagnostic digital subtraction angiography (DSA) demonstrated the abnormal venous plexuses fed by branches of the left L1 lumbar artery and left T12 subcostal artery (**Figure 2**). Selective injection of the left subcostal artery revealed tiny branches arose from the radicular artery leading to a fistula at the dural sleeve, and draining into the perimedullary venous system. Selective angiography of the left L1 lumbar artery also demonstrated a dilated vessel

Figure 1. MRI showed a hypointense flow avoid venous signal on the dorsal surface of the spinal cord extending from T7 to L1, and a mixed hemorrhage signal on T2 weighted image.

abnormally communicated with the venous system, but there was no obvious narrowing part from the feeding artery to the venous plexus. We assumed that should be a posterior spinal artery (PSA) travel upward and converge to the venous network. Therefore, our first diagnosis, remained controversial, was a single SDAVF combined with coexisting perimedullary arteriovenous fistula that drained into common venous plexus.

Surgery was undertaken and because of the suspicion of the complex angioarchitecture of the fistulae. The patient underwent electrocoagulation of the fistula through a laminectomy at the T12-L1 level. During surgery the dura was sufficient exposed, unfolded two red and dilated radicular veins on the surface of the spinal cord. Two points of each shunt along dural root sleeve were identified (**Figure 3**). The distended perimedullary veins became dark red immediately after the coagulation. The remaining veins returned to their normal appearance in a few minutes.

His motor weakness was improved to normal in weeks with no recognizable neurological complications. Four months follow-up spinal MR angiography demonstrated no more abnormal vessels.

Case 2

The patient is a 62-year-old male first complained of slight numbness of the legs in 2009, and then suffered a sudden weakness of both lower extremity and unable to walk after drunk in Feb 2012. Physical examination revealed motor weakness (4/5) in both legs and vibratory sense decreased (5/10) in the T-12 dermatome or lower (grade 4, Aminoff-Logue Scale, Gait: 3 Micturition: 1). The patient had a medical history of pelvic fracture in 1991. MRI of the thoracic and lumbar spinal cord demonstrated small flow void signal within the T6-10 level of the spinal cord on T2-weighted image.

Selective spinal angiography of the whole spinal axis was performed that well demonstrated two independence fistulae (**Figures 4**, **5**). One SDAVF with descending drainage veins fed by the left T6 intercostal artery was in accordance with the initial findings on MRI. Selective angiography of the right T3 intercostal artery revealed another fistula. Drainage of this fistula occurred through a slightly dilated radicular vein and into the ascending coronary venous plexus of the cervical cord. Two fistulae were distant and clearly independent from each other.

The patient refused open surgery but was amenable to endovascular therapy. The interventional procedure was performed under local anesthesia. After systemic heparinization, a 5-French (F) Cobra-2 catheter (Terumo, Tokyo, Japan) was placed in the left T6 intercostal artery. We attempt to navigate the Marathon

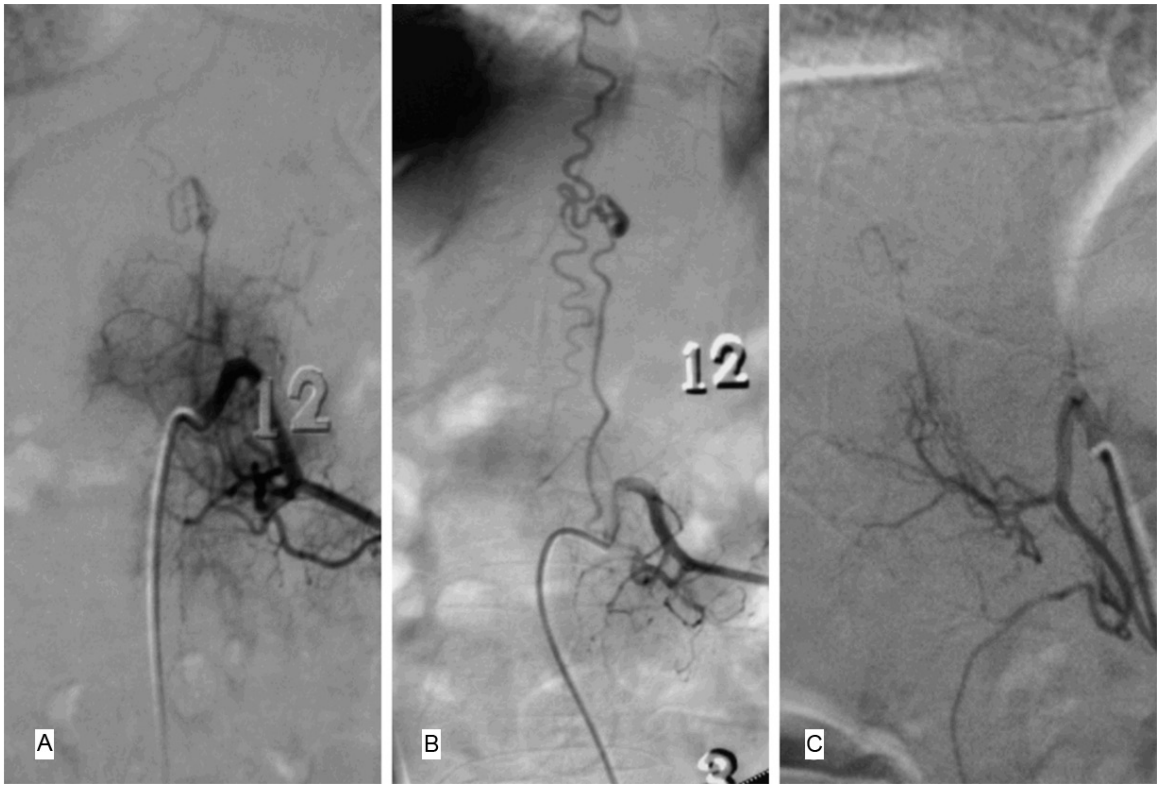

Figure 2. Selective injection of left subcostal artery (A) and left L1 lumbar artery (B) demonstrated two individual fistulae. Compared venous images, two dilated vascular pattern overlapped at T11/T12 level. T12 angiography on the oblique view (C) revealed the glomerular network of tiny branches coalesces at the site of the fistula along the dural root sleeve.

microcatheter (ev3, Irvine, CA, USA) over the 0.008″ diameter Mirage microwire (ev3, Irvine, CA, USA) into the radicular artery. The microcatheter was unable to advance into the tiny branches of feeding artery, and microcatheter angiography showed contrast agent exuded and distal arterial spasm. Protamine sulfate was given immediately to counteract the anticoagulant effect of heparin. Patient had no complaint of chest pain or dyspnea. We gave up superselective catheterization and placed the microcatheter in the intercostal artery, proximal to the origin of the radicular artery. Under subtraction fluoroscopy, 1.2 ml of the glue in a Glubran-2 (GEM S.r.l., Viareggio, Italy): LipiodolUltrafluid (Laboratoire Guerbet, France) mixture of 1:7 was infused through the microcatheter. Glubran penetrated well into the proximal drainage vein and the bleeding artery. Post embolization angiogram demonstrated no filling of the fistula, no signs of contrast exudation, and the distal part of the intercostal artery was also obstructed (**Figure 6**).

A 4-F Cobra-2 catheter was then inserted into the right T3 intercostal artery. Under roadmapping technique, a Marathon microcatheter was navigated over the 0.008″ Mirage microwire into the origin of the radicular artery. The distal fistulous point deemed not accessible by the microcatheter. Super-selection angiography confirmed the position of the microcatheter tip and no evidence of radiculomedullary artery. Under subtraction fluoroscopy, a 1:7 mixture of Glubran-2 diluted in Lipiodol was then slowly infused into this fistula. The Glubran glue adequately penetrated from the proximal drainage vein to the radicular artery. Angiography confirmed no filling of the fistula.

Post-embolization thoracic CT scan showed no hematoma. After coagulation of both fistulae, his motor weakness was improved to normal in both legs in a week, and the dysuria was resolved. Four month follow-up thoracic MRI demonstrated no more hypo-intense flow void signal on the surface of the spinal cord. Six

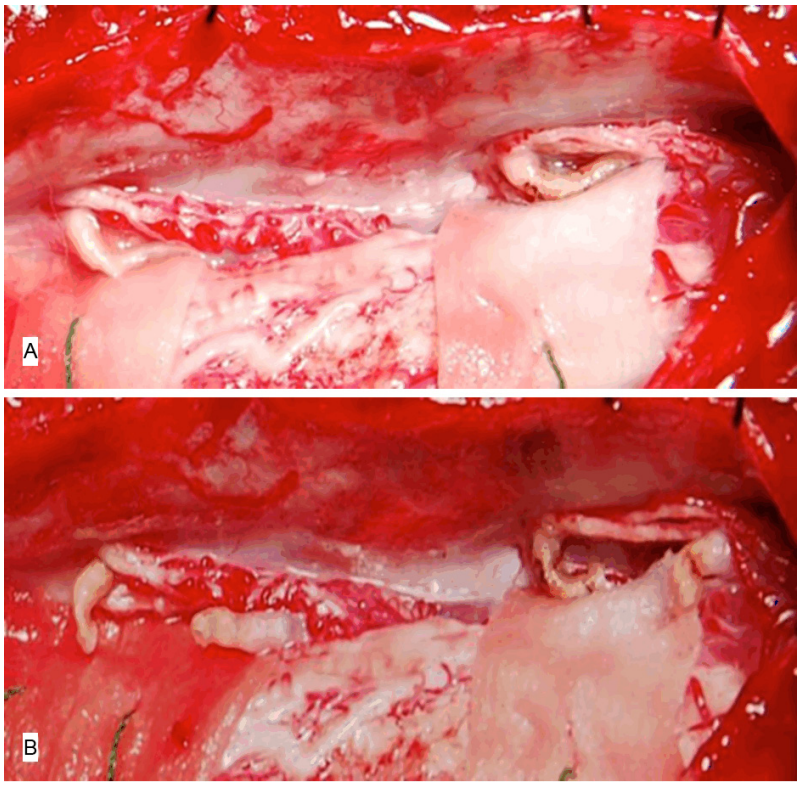

Figure 3. Intraoperative photograph of two arterialized radical veins (A), both veins were coagulated during the surgery (B).

months follow-up angiography confirmed the obliteration of both SDAVFs.

Discussion

Changes in diameter between feeding artery and enlarged vein help us locate the site of fistula. Usually the transition point in the vicinity of the nerve root makes it easy to differentiated SDAVF from PMAVF [4]. In the first case we present, selective injection of the left subcostal artery revealed the tiny branches arose from radicular artery, this obvious narrowing from artery to vein supposed to be the shunting point of the fistula. Yet selective angiography of L1 lumbar artery showed no obvious change in diameter, it was more like a posterior spinal artery of PMAVF than an arterialized drainage vein of another individual SDAVF by angiogram appearance initially. Carefully compared each venous image, two dilated vascular pattern overlapped at T11/T12 intervertebral disc level. Therefore, we assumed that the second fistulous site may locate on the pial surface at the venous overlapping level. But surgical procedure exposed two shunts with two red enlarged radicular veins, which made the final diagnosis as multiple SDAVFs. This angioarchitecture was rarely reported.

Endovascular or microsurgical procedure should aim at obliterating the shunt. PMAVF usually feed by anterior orposterior spinal artery, and the shunt located on the spinal surface [4-6]. Yet in most SDAVF, fistulous point occurred at the level of dural root sleeve rather than along the dilated coronal venous plexus [4]. So once the drainage vein has been identified angiographically, surgeons usually make a small, unilateral interlaminar access to expose the affected radicle, and surgically close the vein through a small dural incision [7]. This straightforward pattern means fistulous point does not need to be identified with precision [8]. If we mistake the dilated drainage veins of SDAVF for the part of PMAVF, endovascular embolization of the distal coronal vein may cause stagnation of venous flow and lead to proximal venous hypertension, which increased risk of postoperative neurological deficits [6, 9]. We carefully reviewed the oblique view of the L1 artery angiography, although the drainage vein was not tortuous, it still could be identified the radicular artery transfer to the spinal vein along the dural sleeve.

Our experience in treating difficult and complicated spinal arteriovenous lesion is that to clearly define the vascular anatomy of feeding artery and draining vein, it is advisable to do the superselective angiography. The detailed understanding of the angioarchitecture of the fistulae is essential for appropriate embolization treatment. Very few literatures reported the result of the obliteration of the two SDAVFs during the same operation, in our case follow-up MRA and neurological examination showed no relapse.

Figure 4. Selective left T6 intercostal artery angiogram (A frontal view, B oblique view), showed a fistula fed by tiny branches arose from the radicular artery and drained into descending coronary venous plexus.

In all three cases of multiple SDAVFs reported before, myelopathy deteriorated after the curative treatment that led to angiographical re-evaluation which detected the second fistula [1, 3, 10, 11]. Such metachronous course, on Sugawara's account, the second fistula was potential or too small to be detected at the first time, closure the first fistula may causing increased pressure of the draining vein then finally enlarged the second fistula [3]. It should be noted that they just performed selective angiography around the abnormal enhancement or hypo-intense areas detected by MRI [3, 8, 11]. Selective angiography was not performed for other sections. However, on occasion, an SDAVF may exist without swelling or edema [7]. As our second case, multi-fistula may also present initially which were distant and independent. These incidental SDAVFs present with no relative symptom, may be easily overlooked on panaortography.

SDAVF cause symptoms through the level of spinal venous hypertension and intramedullary edema rather than the position of fistula. The direct arterial inflow into the venous system

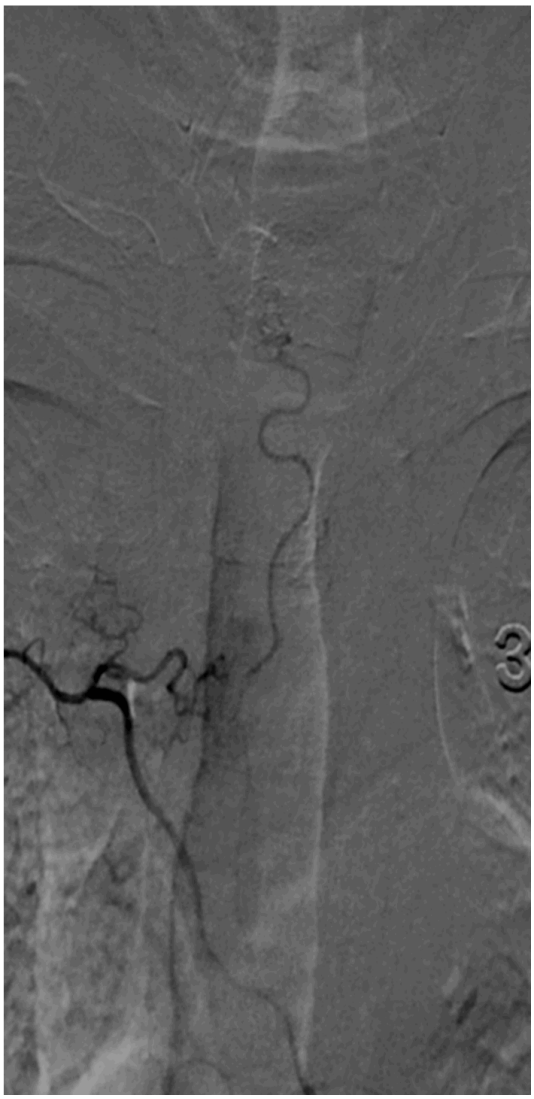

Figure 5. Selective right T3 intercostal artery angiogram, oblique view, revealed another fistula draining into the ascending serpentine venous plexus.

raises the pressure within the coronal venous plexus, thus the site of abnormality on MRI is not a reliable indicator of the location of the fistula, which can be many levels away. Therefore, selective angiography of the entire length of the spinal cord is completely necessary. Every segmental artery supplying the spine and spinal cord—from the skull base to the sacrum—should be selectively catheterized and evaluated.

The authors suggest that both fistulae should be treated at the same operation. Surgical procedure in patients with multiple fistulae, wide exposure was essential, which consists of at least two levels targeted laminectomy and intradural exploration [8]. Current endovascular techniques using various liquid embolic materials have high success and low recanalization rates [10, 12, 13].

As endovascular treatment of the fistula has to be closed on the venous side [8]. If the feeding branch can be superselected, non-adhesive embolic agent should be chosen and delivered to the shunt zone. But in our second case the angioarchitecture was not favorable, microcatheterization seems to be impossible due to the size and tortuous of the pathological vessels. Occlude the initial part of the drainage vein is sufficient to cure the shunt, obliterate the feeding artery may temporarily reduce the shunt flow which improve the clinical system, but have high rate of recanalization [14]. In this situation, high diluted mixture of Glubran and Lipiodol was proved especially useful for easier more effective penetration to reach distal shunts [15]. In spite of that using high diluted embolic agent through the proximal feeding artery requires high levels of operator expertise [16].

Conclusion

The presence of multiple fistulae must be aware in patients being evaluated for spinal dural arteriovenous fistulae. Multidisciplinary approach of multiple SDAVFs is essential, microcatheter angiography significantly improve our ability to identify the angioarchitecture of complicated spinal arteriovenous fistula. Surgical shunt interruption has proven secure and reliable. Improved endovascular technique and imaging equipment made the previously inaccessible lesions manageable by endovascular strategies. For multiple SDAVFs, a therapeutic alternative is closely depends on the anatomic location and angioarchitecture and still need further study.

Disclosure of conflict of interest

None.

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Figure 6. Post embolization angiograms demonstrated no filling of both fistulae.

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