

Spinal intraosseous epidural arteriovenous fistula; a late complication of vertebral compression fracture: two case reports

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Running title: Long standing VCFs resulted into SEDAVF

Abstract

Introduction: Spinal epidural arteriovenous fistulas (SEDAVFs) are extremely rare and the pathological process still remains unclear. Nevertheless, neurofibromatosis, surgery, as well as trauma have been implicated as causes. Nevertheless, long standing chronic vertebral compression fractures (VCFs) as a cause of SEDAVFs has not been reported in literature.

Case Presentation: Our cases comprised of a male and female with ages 67 and 62 year respectively. They presented with weakness of bilateral lower extremities and back pain. Their symptoms deteriorated into mild recto-bladder dysfunction as well as hypoesthesia of lower limbs. They both had history of road traffic accidents (RTAs) prior to their symptomatology. In both cases, Magnetic resonance imaging (MRI) revealed VCFs prior to the above symptomatology. Spinal digital subtraction angiography (DSA) was used to confirm SEDAVFs in both cases. We utilized the trans-arterial route with Onyx embolization agent to obliterate both fistulae and their symptom resolved after the embolization. Two-years follow-ups revealed no recurrence of their fistulae.

Conclusion: The initial long standing VCFs caused by trauma resulted in perforation of the dorsal somatic branches of the radicular artery leading to the occurrences of SEDAVFs in our cases.

Key words: Embolization, Onyx, SEDAVF, Trans-arterial, VCF, Spine.

Introduction

Spinal epidural arteriovenous fistulas (SEDAVFs) are extremely rare and the pathological process still remains unclear¹⁻³. Only few cases or case series have been reported in literature^{1,2,4-12}. It is postulated that, in SEDAVF, branches of the radicular artery directly communicate with the epidural venous plexus and often manifests

clinically as radiculopathy¹. In most cases, this symptomatology is often progressive as a result of intradural vein reflux as well as venous congestive hypertension⁷. Although the pathophysiological mechanism of SEDAVF remains uncertain, neurofibromatosis, surgery, as well as trauma have been implicated as causes¹³. Song et al categorized SEDAVF into osseous and non-osseous types with relation to bone involvement¹. SEDAVF are often high flow lesions that requires trans-arterial embolization with a balloon or coils to occlude the fistula^{7,8}. Chronic vertebral compression fractures (VCF) as a cause of SEDAVFs has not been reported in literature. We present two cases of SEDAVF with VCF, which we successfully treated via the trans-arterial endovascular approach using Onyx embolization.

Case 1

A 67-year old male was admitted to our department due to weakness of bilateral lower extremities and back pain of 2 years duration. Magnetic resonance imaging (MRI) revealed T12 VCF after a road traffic accident (RTA) three years prior to the above symptomatology. His symptoms deteriorated into mild recto-bladder dysfunction, hypoesthesia of lower limbs on account of which he was referred to our facility. MRI revealed a fracture (**Fig 1A**) as well as hyperintensity and edema at T6 and T7 levels with numerous abnormal dilated vessels as well as flow voids in the T12 vertebral body (**Fig 1B**). Spinal digital subtraction angiography (DSA) confirmed a SEDAVF located in the T12 vertebral body fed by multiple feeding arteries such as bilateral T11, T12 subcostal arteries, and left L1 lumbar artery (**Fig 1C**). The SEDAVF was drained by epidural venous plexus into the paravertebral vein and azygous vein. No obvious perimedullary veins were observed.

Endovascular route was adopted via selective catheterization from the left T11 subcostal artery into the fistula. Subsequently Onyx Liquid Embolic System (Onyx HD-500) (Covidien / ev3 Neurovascular) was utilized to occlude the fistula. The venous sac as well as each intraosseous fistula tract were totally occluded in one session,

and postembolization angiography showed complete obliteration of SEDAVF (**Fig 1D-F**). Post-procedure cast of Onyx was observed in T12 vertebral body (**Fig 1G**). The patient's symptom resolved after the embolization. Six months follow-up MRI showed disappearance of the fistula (**Fig 1H**) and no recurrence of the fistula. The patient symptomology improved remarkably with no further neurological deficits.

Case 2

A 62-year-old female was admitted at our institution with one-year history of unstable walking and lower backache. Her condition deteriorated further into mild fecal and urine incontinence within the last seven months prior to admission. Physical examination revealed paresthesia below the umbilical level, positive right Babinski sign, graded 4 weakness of lower extremities, brisk bilateral ankle reflex. The left knee reflex was also brisk while right knee reflex was weak. Her past medical history revealed VCF at the body of L1 vertebral as a result of RTA three years prior to her current symptomatology. T2-weighted spinal MRI revealed diffused spinal cord edema and flow voids around the cord at T6 and T7 level extending to the L1 vertebral body (**Fig 2A**). Spinal DSA demonstrated a SEDAVF at the L1 level with feeders from bilateral L1 lumbar arteries. The fistula drainage was via the epidural venous plexus with a visible perimedullary vein (**Fig 2 B**).

Endovascular route was adopted to treat the patient. Bilateral 5 french femoral arterial sheath were placed and embolization was performed via selective catheterization into bilateral L1 lumbar arteries. Subsequently Onyx Liquid Embolic System (Onyx HD-500) (Covidien / ev3 Neurovascular) was injected as close as possible to the fistula point via the bilateral lumbar arteries. The venous sac of L1 vertebral body and the connection point between epidural venous plexus and intradural vein were totally occluded in one session. Postembolization angiography confirmed disappearance of the fistula (**Fig 2C**). A casted image showed the connection point clearly (**Fig 2D**). The patient's symptom resolved after the embolization. Six months follow-up MRI showed

disappearance of the fistula (**Fig 2E**) and no recurrence of the fistula. The patient symptomology improved remarkably with no further neurological deficits.

Discussion

SEDAVFs are fistulas originating from the epidural space predominantly between the dorsal somatic branches of the radicular artery and the epidural venous plexus^{1,9,10}. The venous flow often drains only into the epidural as well as the paravertebral plexuses in most reported cases¹⁰. On the other hand, VCFs naturally occur in the mid-thoracic or thoraco-lumbar transition region of the spine¹⁴. The anterior spinal artery as well as paired smaller posterior spinal arteries consists the blood supply to the spinal cord¹⁵. Furthermore, the spinal cord is made up of many anastomoses that allow for excess blood supply to the spinal cord¹⁵.

Trauma is generally believed to be the precipitating event for the development of SEDAVFs although the patho-etiology is inadequately understood because of the rare nature of their diagnosis^{10,16}. In both cases above, the initial finding were VCFs as a result of trauma which aggravated into SEDAVFs years later. Thus, the initial long standing VCFs perforated dorsal somatic branches of the radicular artery resulting in the SEDAVFs in our cases.

In most SEDAVF cases, the feeders typically originated from the bony as well as dural branches of the intersegmental arteries and the fistula nidus is often seen at the site of the bony defect^{7,17}. The fistula nidus usually composes of multiple small arterial feeders from the segmental arteries which join to form a single round venous sac⁷. It is postulated that; the round venous sac often recruits the multiple arterial feeders unlike other draining veins^{10,18}. The distinctive characteristics of the fistula nidus reveals its linkage and drainage into the epidural vein during the venous phase^{7,18}.

In most patients with SEDAVFs, the lesion usually does not show any specific

symptoms until dilation of arterialized veins triggers compressive symptoms or as a result of intradural reflux^{10,19}. The patients usually present with progressive neurologic deficit. The patient usually complains of radicular pain that requires MR imaging to determine any intervertebral disk herniation⁷. All our patients presented with weakness of bilateral lower extremities and back pain. Their conditions deteriorated further into mild fecal and urine incontinence over 3 years period after the initial diagnosis of VCF. The dilated venous sac or trans-medullary venous congestion often results in venous stasis in spinal canal which leads to cord compression¹⁷. Also, retropulsion of fracture pieces often leads to cord or cauda equina compression resulting in weakness and loss of sensation at the lower extremities as well as recto-bladder incontinence¹⁴.

In cases of VCF, Computed tomography (CT) scan although not ideal imaging modality for the spine, is still valuable because, it aids in evaluating the bony anatomy as well as loss of height of the vertebral body, fragment retropulsion, and canal stenosis^{14,20,21}. CT scan is capable detecting chronic fracture through the existence of cortication. MRI is the ideal radiological imaging for patients with VCFs. It is able to detect fracture age due to bony edema during acute fracture. MRI is able to detect nerve roots or spinal cord compression¹⁴. Nevertheless, DSA is the gold-standard radiology modality for the diagnosis of SEDAVFs although magnetic resonance angiograph (MRA) is also very valuable. We did not utilize the MRA because our facility has DSA.

Suh et al classified this distinctive spinal vascular lesion as an osseous SEDAVF because of its distinctive angiographic features as well as the it's efficient treatment via the venous route⁷. They observed disruption or steno-occlusion at the venous outlet which resulted in intradural regurgitation and subsequently spinal cord swelling as well as edema⁷. It is anticipated that, the vasculature of the lesion as well as the surrounding structures be meticulously evaluated to determine suitable treatment modality¹. Therefore, it advocated that, to appropriately evaluated the connections between the feeders, fistulas, as well as the draining veins, routine 3D rotational spinal angiogram should always be obtained^{1,22}.

Endovascular management has proven to be a very effective treatment modality for SEDAVFs although surgery is adapted in some cases^{6,10}. Endovascular route comprises of the trans-artery and the trans-venous routes. The trans-arterial embolization with a balloon or coils to occlude the fistula is often the advocated route because of the high flow nature of the lesions^{7,8,17,19,23}. On the other hand, Suh et al are of the view that, occlusion of the round venous sac facing the numerous arterial feeders via the venous route is very rewarding during endovascular treatment of SEDAVFs^{7,18}. In our cases, we utilized the trans-arterial route with onyx embolization agent to obliterate the venous sacs at the vertebral bodies as well as the connection points between epidural venous plexus and intradural veins in one session.

It is advocated that, the microcatheter tip should be positioned at the point where the blood flows into the intradural vein before embolic agent is injected to achieve occlusion of the fistula¹. Some authors advocate the use of glue via the trans-arterial embolization because it is easy to control, it has a relatively short injection time as well as an enhanced visibility than Onyx^{7,18}. Nevertheless, the usage of Onyx for SEDAVF embolization has been limited because of hitches in attaining venous infiltration¹⁸.

Surgery via posterolateral approach is also another treatment modality for patients with SEDAVFs¹³. Surgery is often complex because the lesions often takes an anterior location which make adequate exposure from a posterolateral approach very challenging. Therefore, surgical resection which involves direct ligation and interruption of the fistula at the site of the shunt and draining vein is often reserved for SEDAVFs without intradural venous drainage^{13,24,25}. Furthermore, surgical resection is often preferred to trans-arterial embolization in SEDAVFs that are around the anterior spinal artery or closer to multiple arterial feeders to a high flow fistula. SEDAVF related mass effect is often lessen after surgical resection because both decompression as well as ligation are often achieved after surgery¹³.

Conclusions

The spinal cord is made up of many anastomoses that allow for excess blood supply to the spinal cord. Thus, the initial long standing VCFs caused by trauma resulted in perforation of the dorsal somatic branches of the radicular artery leading to the occurrences of SEDAVFs in our cases. We utilized the trans-arterial endovascular route with Onyx embolization agent to treat our patients successfully without any further complications.

Abbreviation List: Computed tomography = CT, Digital subtraction angiography = DSA, Magnetic resonance imaging = MRI, Spinal epidural arteriovenous fistulas = SEDAVFs, Vertebral compression fracture = VCF.

Declaration

Ethics approval and consent to participate: This case was reported or written in according to ethical committee of our hospital's criteria for reporting or writing case reports. The patients and their relatives were informed about our intension to involve them in a case study and they agreed to partake in the study.

Consent for Publication: The patients and their relatives were dually informed about our intention to publish their cases and they fully concerted to the use of their documents. In all cases, a written informed consent was obtained. A written concern for publication was signed by the patients. The hospital also concerted to the use of their information for publication.

Author contributions: All authors contributed toward data analysis, drafting and critically revising the paper and agree to be accountable for all aspects of the work. TW and SAR wrote the final paper. All authors approved the final version of this paper.

Competing interests: All the authors have no competing interest to disclose.

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Figure 1 A-H: are diagnostic, procedure as well as post-procedure images of case 1.

A: is MRI showing the fracture. **B:** is MRI showing hyperintensity and edema at T1 and T2 levels with numerous abnormal dilated vessels as well as flow voids in the T12 vertebral body. **C:** is a DSA showing the SEDAVF at T12 vertebral body with feeders from multiple arteries such as bilateral T11, T12 intercostal arteries, and left L1 lumbar artery. **D-F:** are a postembolization angiographies showing complete obliteration of SEDAVF.



Figure 1G &H

G: is a post-procedure cast of Onyx at the T12 vertebral body. **H:** is a follow-up MRI showing no recurrence of the fistula.



Figure 2 A-E: are diagnostic, procedure as well as post-procedure images of case 2. **A:** is an MRI showing diffused spinal cord edema and flow voids around the cord at T2 level extending to the L1 vertebral body. **B:** is a DSA showing SEDAVF at the L1 level with feeders from bilateral L1 lumbar arteries and drainage via the epidural venous plexus with a visible perimedullary vein. **C:** is a post-embolization angiography showing disappearance of the fistula. **D:** is casted DSA image showing the connection point of the fistula. **E:** is a follow-up MRI showing no recurrence of the fistula.

