

Spontaneous Spinal Osseous Epidural Arteriovenous Fistula with Long Segments of Prominent Epidural Venous Drainage Causing Severe Compressive Thoracic Myelopathy Successfully Treated with Combined Endovascular and Surgical Treatments: A Case Report and Review of the Literature

Abstract

The authors describe an extremely rare case of spinal osseous epidural arteriovenous fistulas (SOEAVFs) with unique characteristic features. A 25-year-old man presented with progressive weakness and paresthesia of the lower extremities for 1 month. Magnetic resonance imaging of the thoracic spine showed an extradural dilated vascular flow void structure extending from T4 to T8 levels with abnormal hyperintense T2 signal from T6 to T8 levels. Magnetic resonance angiography and spinal angiography revealed unique features of SOEAVF supplied by multiple small arterial feeders of intercostal arteries converging into a dilated round venous sac corresponding to a bony defect of T7 lamina and spinous process. The venous drainage directly drained into prominent epidural venous plexus extending from the level of T4 to T8 without intradural venous drainage, causing severe compressive myelopathy. Transarterial embolization was performed using N-butyl cyanoacrylate through the main feeder. Subsequently, he successfully underwent laminectomy and total excision of the fistula and large epidural draining venous plexus. Histopathology confirmed spinal vascular malformations with evidence of previous embolization. He gradually improved until being able to walk independently 3 months later. Follow-up spinal angiography confirmed complete resection of SOEAVF. The patient has remained clinically asymptomatic 5 years after operation.

Keywords: *Compressive myelopathy, round venous sac, spinal osseous epidural arteriovenous fistula, surgical treatment, thoracic spine*

Introduction

Spinal epidural arteriovenous fistulas (SEAVFs) are less common than spinal dural arteriovenous fistulas (SDAVFs).^[1] In comparison between SDAVFs and SEAVFs, patients harboring SEAVFs had often been misdiagnosed with SDAVFs at the time of treatment, and the recurrence rate after endovascular treatment was higher in the SEAVF group. It may be necessary to accurately understand the angioarchitecture of SEAVFs for optimal treatment.^[2] Based on venous drainage, SEAVFs are subtyped into pure extradural drainage, retrograde intradural venous drainage, or combined extradural and retrograde intradural venous drainages.^[3-5] Patients harboring SEAVFs usually presented with congestive

myelopathy (approximately 90%) secondary to intradural venous drainage and may present with compressive symptoms (approximately 10%) resulting from exclusive extradural venous drainage.^[6] Regarding radiologic features, SEAVFs with intradural venous drainage can potentially be distinguished from SDAVFs by the presence of a dilated epidural venous pouch or engorgement of the epidural venous plexus.^[7] It is imperative to differentiate SEAVFs from SDAVFs because the ventral arterialized epidural venous pouches are challenging in hemostasis during surgery.^[8] Primary surgical treatment is less frequently undergone for SEAVFs.^[3] In opposition, surgery remains the best treatment option

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Figure 1: (a-c) Sagittal and (d-f) axial T2-weighted images of the thoracic spine demonstrate multiple dilated epidural flow void structures on the dorsal aspect of the spinal extradural space extending from the level of T4 to T8, causing moderate-to-severe spinal cord compression with long segment of hypersignal intramedullary lesion from T6 to T8 vertebral level, representing spinal cord edema or ischemia

for SDAVFs by obliteration of the intradural arterIALIZED draining vein.^[9]

SEAVFs with intradural venous drainage were diagnosed in patients with a mean age around the sixth decade of life and commonly occurred in the thoracolumbar and lumbar regions, whereas the extradural fistulas with severe mass effect without intradural venous drainage were found in younger patients around the third decade of life and frequently occurred in the cervical and upper thoracic regions.^[10]

Based on the osseous involvement, SEAVFs were subdivided into the osseous and nonosseous types.^[11] Spinal osseous epidural arteriovenous fistulas (SOEAVFs) are extremely rare and characterized typically by recruiting multiple small arterial feeders of segmental arteries converging into a dilated round venous sac corresponding to a bony defect of the vertebral body, the osseous component

of this spinal vascular lesion.^[11-13] The authors described unique characteristic features of SOEAVF at the thoracic spine in a young man who presented with compressive myelopathy caused by long segments of large epidural venous drainage. The authors also reviewed the published case reports and series that had enough clinical description and clearly demonstrated figures of spontaneous SOEAVF with a dilated round venous sac recruiting multiple arterial feeders.

Case Report

A 25-year-old man was admitted to the local hospital due to progressive weakness and paresthesia of the lower extremities for 1 month. He had no history of back pain or trauma. Furthermore, difficulty in urination was noticed 1 week before the hospitalization. Magnetic resonance imaging (MRI) of the thoracic spine showed multiple dilated vascular flow void structures on the dorsal aspect of

the spinal extradural space extending from the level of T4 to T8, causing moderate-to-severe spinal cord compression with intramedullary high intensity on T2-weighted sequence from T6 to T8 vertebral level, representing spinal cord edema or ischemia. There was a partial enhancement of the lesion after gadolinium administration. This lesion involved



Figure 2: Sagittal T1-weighted images (a) before and (b) after gadolinium administration show conglomeration of flow voids within enlarged posterior epidural intraspinal soft tissue, extending from the level of T4 to T8, with partial enhancement of the lesion. This lesion causes severe spinal cord compression from T6 to T8 vertebral level and involves T7 lamina and spinous process

T7 lamina and spinous process [Figures 1-3a]. The initial diagnosis was spinal extradural vascular malformations with compressive myelopathy. The patient was transferred to Prasat Neurological Institute for proper management.

The neurological examination revealed evidence of spastic paraparesis (muscle strength: 4/5), the lack of pinprick sensation below T8 level, impairment of proprioception, hyperreflexia, and presence of Babinski's sign in the lower extremities. Magnetic resonance angiography of the thoracic spine disclosed hypertrophic bilateral T6 to T8 intercostal arteries with evidence of early enhancement of long segment of epidural venous plexus in the posterior spinal canal of the upper thoracic spine and a round venous sac protruding into the T7 spinous process, suggesting of high-flow SOEAVF [Figure 3b and c]. In addition, computed tomography scan of the upper thoracic spine clearly demonstrated osseous involvement in T7 lamina and spinous process [Figure 4].

Spinal angiography showed multiple arterial feeders from bilateral T6, T7, T8, and T9 intercostal arteries converging into the round venous sac at the level of T7 with draining into the large epidural venous plexus extending from the level of T4 to T8 and bilateral dilated intercostal veins from T5 to T8. The main arterial supply of the fistula arose from the right T7 and T8 intercostal arteries. Several small associated feeding vessels arose from left T7, T8, bilateral T6, and T9. Subsequent venous drainage drained cranially into the superior vena cava through the azygos vein and caudally into the bilateral ascending lumbar veins through the azygos and hemiazygos veins [Figures 5 and 6]. No intradural venous drainage into the perimedullary veins was detected. The transvenous approach through the femoral vein was attempted but failed to reach the round venous



Figure 3: (a) Coronal T2-weighted image of the thoracic spine discloses multiple dilated flow voids along the posterior aspect of the upper thoracic spine with a vascular pouch at the midline of the level of T7, (b) Coronal maximum intensity projection, and (c) Sagittal multiplanar reformatted images of contrasted spinal magnetic resonance angiography reveal a long segment of arterialized epidural venous enlargement in the posterior spinal canal of the upper thoracic spine and a round venous sac (black and white arrowheads) protruding into the T7 spinous process

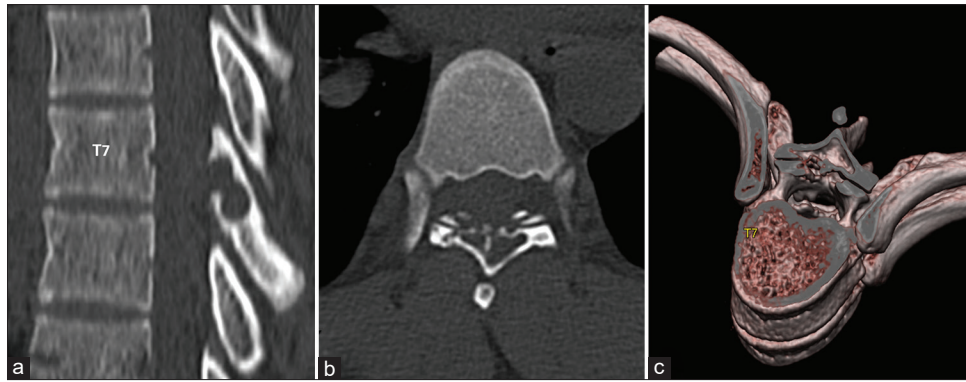


Figure 4: (a) Sagittal, (b) Axial reformatted, and (c) Three-dimensional reconstruction computed tomography images of the level of T7 reveal a bony defect at T7 lamina and spinous process corresponding to the dilated venous sac

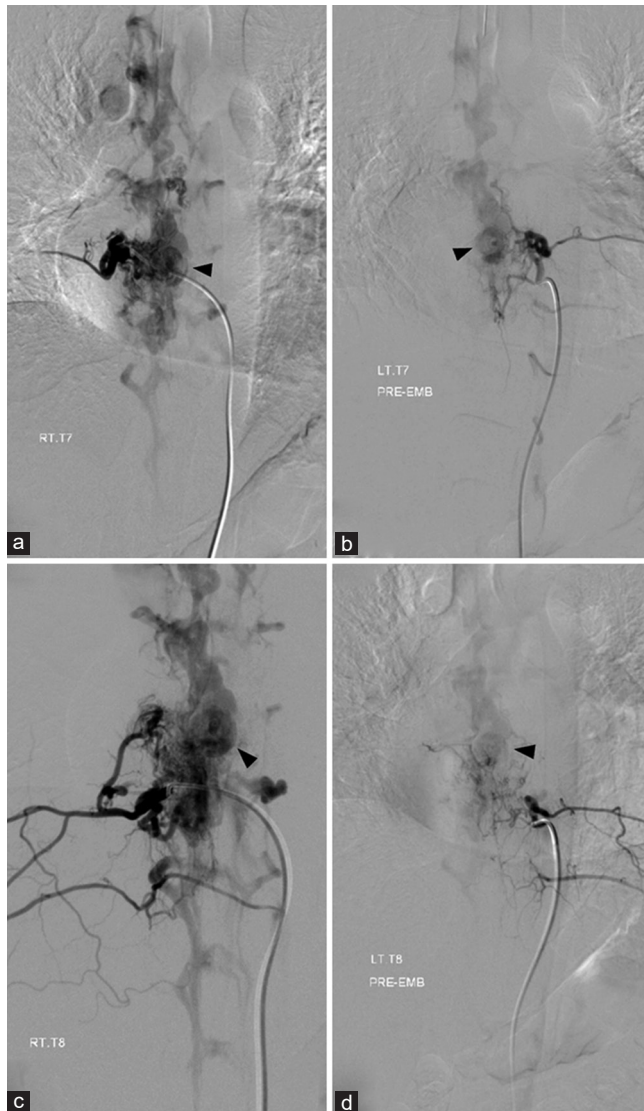


Figure 5: Spinal angiography of the right T7 (a), left T7 (b), right T8 (c), and left T8 (d) intercostal arteries demonstrates high-flow spinal osseous arteriovenous fistula supplied by bilateral T7, T8, and T9 converging to a round venous sac (arrowheads) at the level of T7 with subsequent cranial and caudal venous drainage to dilated epidural venous plexus extending from T4 to T8

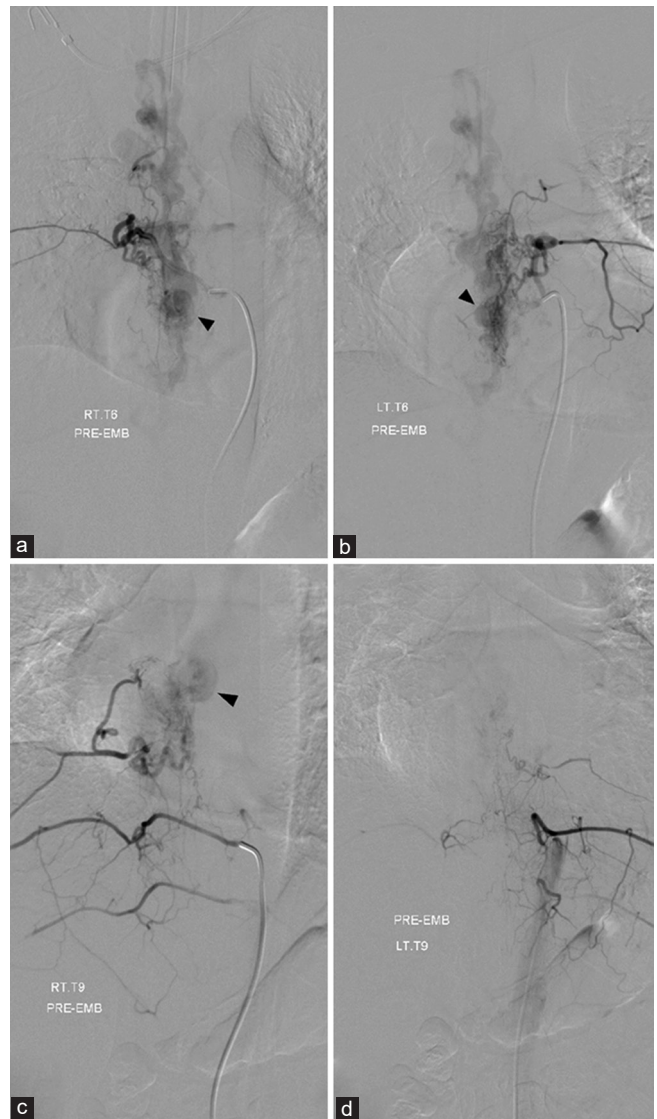


Figure 6: Spinal angiography of the right T6 (a), left T6 (b), right T9 (c), and left T9 (d) intercostal arteries reveals associated arterial feeders from bilateral T6 and T9 converging to a round venous sac (arrowheads) at the level of T7

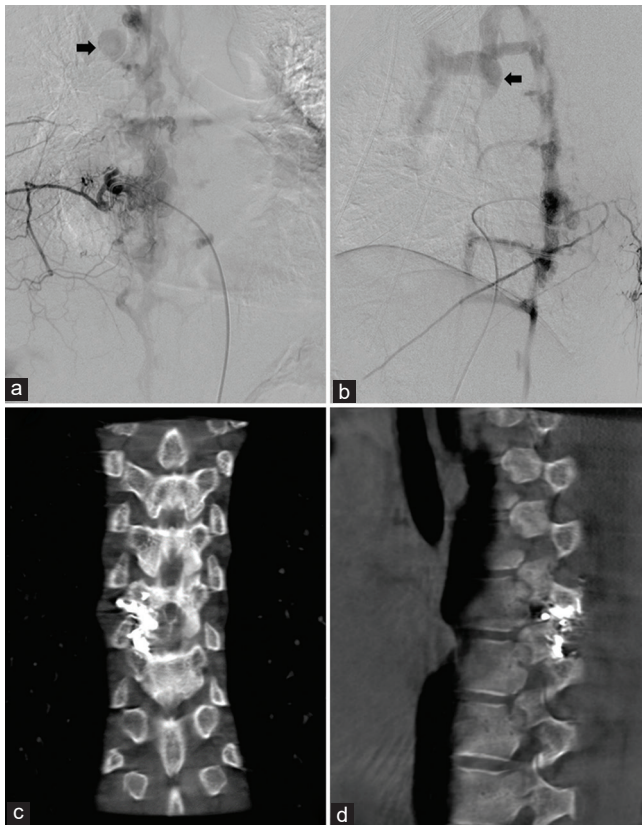


Figure 7: (a) Anteroposterior and (b) Lateral views of selective angiography with the microcatheter into the right T7 intercostal artery before embolization reveal spinal epidural arteriovenous fistula draining into a round venous sac (arrowheads) and prominent long-segment epidural venous plexus with cranial drainage into the superior vena cava (arrows), (c) Coronal and (d) Sagittal views of three-dimensional reconstruction computed tomography obtained after embolization demonstrate high-density embolized glue deposition around the right side of a round venous sac

sac. Therefore, transarterial embolization through the right T7 intercostal artery with N-butyl cyanoacrylate (NBCA) was performed to reduce the flow from the main feeder [Figure 7]. Because the artery of Adamkiewicz or anterior spinal artery (ASA) arose from the right T8 intercostal artery, further endovascular treatment with liquid embolic material was avoided in our patient due to the possibility of reflux of liquid agent into the anastomotic channel connecting with the ASA. The patient and his parents were informed about further surgical treatment and the risks of the surgery, especially uncontrollable bleeding. However, they denied the surgery, and the patient was lost to follow-up.

Two months later, the patient was readmitted to our institute due to worsening symptoms. He had gradually developed weakness of the lower extremities until being inability to walk independently with urinary retention and constipation. The neurological examination revealed evidence of spastic paraparesis (muscle strength: 1–2/5) and the lack of pinprick sensation below T5 level. The patient and his parents accepted the risks of the surgery and signed informed consent. Before the surgery,

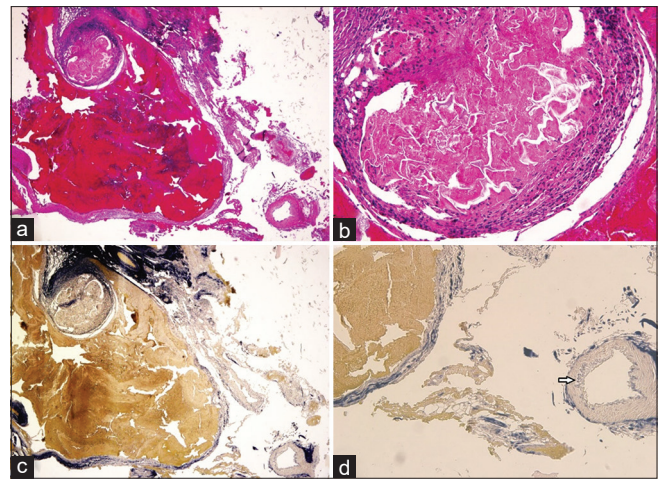


Figure 8: Microscopic examination of the histopathological specimens of extradural mass from the thoracic spine, (a) Vascular wall with marked variation in vascular wall thickness and luminal diameter (H and E, original magnification $\times 40$). Multinucleated giant cells engulfed embolic agents (b) (H and E, original magnification $\times 200$) and (c) (Verhoeff–Van Gieson, original magnification $\times 40$), (d) Presence of internal elastic lamina of artery (arrow) (Verhoeff–Van Gieson, original magnification $\times 100$)

large peripheral and central intravenous accesses with invasive hemodynamic monitoring were prepared. The patient underwent laminectomy of T5–T8, coagulation of all feeding vessels, and total excision of the fistula and large epidural draining venous plexus. Expectedly, sudden profuse bleeding occurred during surgery, leading to hypovolemic shock. Apply pressure over the bleeding site was performed periodically, and massive transfusion with fresh frozen plasma and packed red cells was replaced rapidly. In addition, the patients received intravenous tranexamic acid 1 g bolus in 100 ml of normal saline over 10 min. Fortunately, hemostasis could be achieved in our case. Estimated blood loss was 2000 ml. Histopathology of extradural mass from the thoracic spine revealed fibroadipose tissues with numerous vascular channels with marked variation in vascular wall thickness as well as luminal diameter. Foreign-body materials (i.e., NBCA) surrounding with multinucleated giant cells were noted. Elastic staining confirmed that the vessels were both arteries and veins. These findings were consistent with spinal arteriovenous malformations with evidence of previous embolization [Figure 8].

Spinal angiography of the thoracic spine obtained 2 weeks after the operation confirmed complete resection of SOEAVF [Figure 9]. His postoperative course was uneventful. He gradually improved until being able to walk independently with restoring of bladder and bowel functions in 3 months later. MRI of the thoracic spine obtained 6 months following the surgery demonstrated the disappearance of multiple dilated epidural flow void structures with the resolution of spinal cord edema or ischemia [Figure 10]. The patient has remained clinically asymptomatic 5 years after operation.

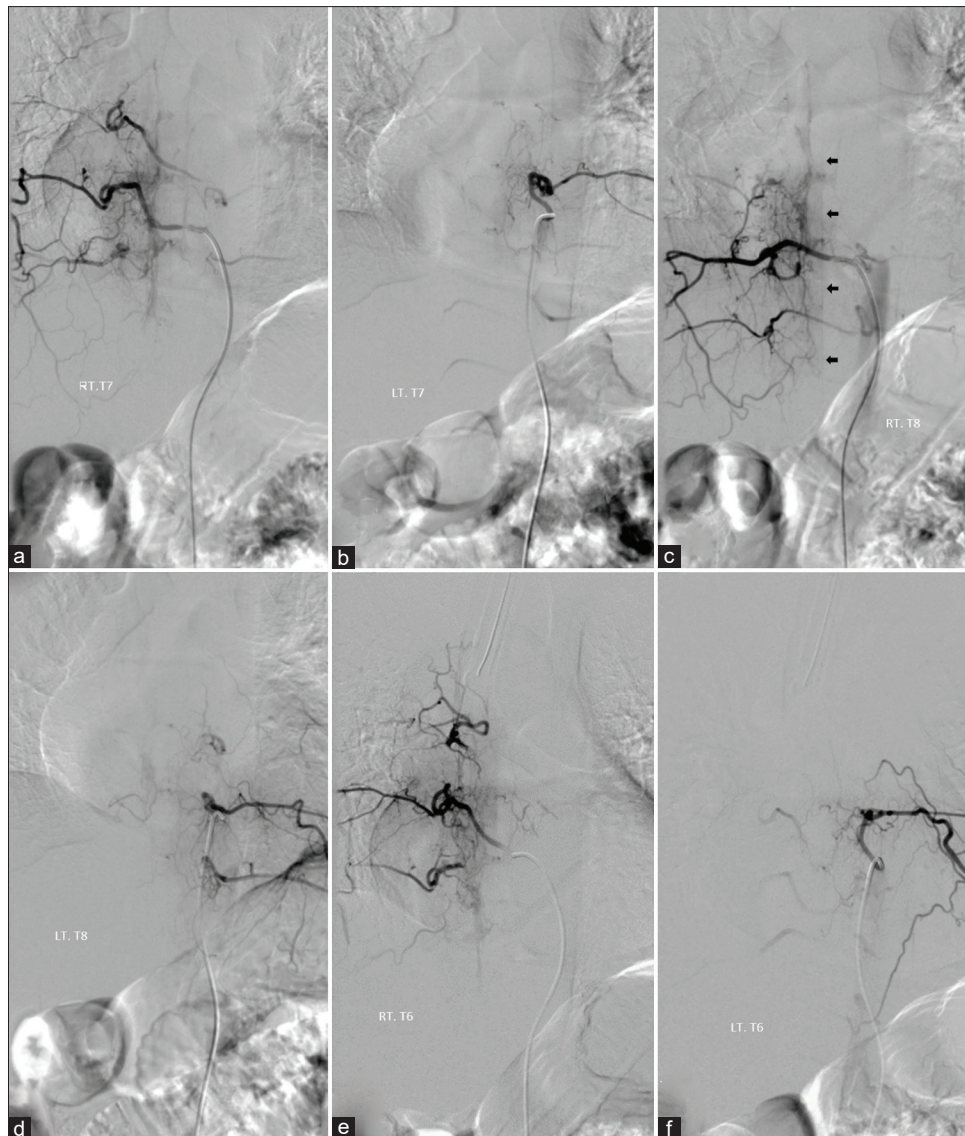


Figure 9: Spinal angiography obtained 2 weeks after the operation. Anteroposterior views of the right T7 (a), left T7 (b), right T8 (c), left T8 (d), right T6 (e), and left T6 (f) intercostal arteries confirm complete resection of spinal osseous epidural arteriovenous fistula

Discussion

We reviewed literature about SOEAVFs with unique feature of a dilated round venous sac recruiting multiple arterial feeders [Table 1].^[11-15] The collected data in this review included demographic data (i.e., gender and age of patient), symptoms and signs, the location of a round venous sac corresponding to the osseous involvement, arterial supply of the fistula, angiographic outcome, and neurological outcome of the patients. From the literature review, there were 8 cases, including our 1 case. Five (62.5%) men and three (37.5%) women with a median age of 33.5 years, range 14–57 years, were included in this review. The symptoms and signs of most patients harboring SOEAVF included compressive radiculopathy and/or myelopathy due to the dilated venous sac. Only one patient presented with progressive myelopathy secondary to perimedullary venous drainage from the fistula.^[12] The fistula was located at the

cervical spine in 4 (50%), thoracic in 3 (37.5%), and lumbar in 1 (12.5%). All fistulas supplied mainly by segmental or intercostal arteries at the level of the bony defect with associated small feeders converging to a round venous sac from the segmental or intercostal arteries arose from the upper or lower segments of the ipsilateral and contralateral arteries. Most patients were treated by endovascular treatment using transvenous and/or transarterial approach. One patient underwent emergency surgery in small fistula presenting with epidural hemorrhage.^[15] Only our case was treated using combined endovascular and surgical therapies. Follow-up spinal angiography confirmed complete obliteration in 3 fistulas. Another 4 fistulas had a small residual shunt. Most patients resulted in good neurological outcome.

In 2004, Chul Suh *et al.*^[12] firstly described unique characteristic features of SOEAVF, i.e., multiple small



Figure 10: Magnetic resonance imaging of the thoracic spine obtained 6 months following the surgery, (a and b) Sagittal and (c-e) Axial T2-weighted images reveal an evidence of previous laminectomy from T5 to T8 with disappearance of multiple dilated epidural flow void structures. The resolution of spinal cord edema or ischemia is noted

arterial feeders of segmental arteries converging into a single round venous sac locating at the site of the bony defect, in 2 patients. First patient, a 50-year-old woman, presented with intractable neck pain, radicular pain, and an audible bruit. There was a bony defect in the right side of the C5 body and pedicle corresponding to the area of the dilated venous sac. The other patient, a 21-year-old woman, presented with progressive myelopathy secondary to perimedullary venous drainage from the fistula. The enlarged venous sac was in the bony defect of the L2 body and pedicle. They suggested that the bony defect in the area of the dilated round venous sac seems to be different from bony erosion causing by a dilated venous ectasia in high-flow pial arteriovenous fistula.

A round venous sac recruiting multiple feeding vessels may be described as a fistula nidus, an osseous nidus, or the fistulous sac.^[11-13] The obliteration of a round venous sac through venous route using coils seems to be the treatment of choice for this unique SOEAVF.^[11,12] In the setting of unsuccessful transvenous approach, transarterial embolization with liquid embolic materials should be considered. In addition, transarterial catheterization into the epidural sac may be another approach.^[14] Transarterial embolization using NBCA by multiple staged sessions may be the first choice of treatment.^[13] However, these high-flow fistulas may need multistage sessions and performing through several feeding arteries to complete closure of the fistulas, probably increasing the risk for radiation exposure.

In addition, the prominent flow into the fistula may obstruct the opacification of the ASA originating at the same level of the feeder during transarterial embolization with liquid embolic materials, resulting in inadvertent complication.

Willinsky *et al.*^[14] reported 2 patients with SEAVF, corresponding to SOEAVF described by Chul Suh *et al.*,^[12] with exclusive epidural venous drainage, causing compressive myelopathy. First, a 57-year-old man was treated with combined arterial and venous approaches using balloons, NBCA, and coils for many years before achievement. Due to unsuccessful closure of the fistula, the cervical SOEAVF recruited new extensive collateral feeding vessels from other segmental arteries. Finally, a 41-year-old man was unable to be treated through venous approach. Therefore, multiple sessions of transarterial embolization were successfully performed using liquid embolic material with a residual small fistula. They suggested that it is important to occlude the venous side of the fistula to cure this unusual condition. However, the tight packing of coils in the large epidural venous pouch, producing compressive myelopathy as in our case, may aggravate worsening symptoms of the patient. Iizuka *et al.*^[13] also concerned the inability to differentiate the intraosseous venous pouch and intracanalicular dilated venous pouch by a venous approach and the mass effect of the dilated venous pouch after coils packing.

Surgical resection of SOEAVF may result in uncontrollable hemorrhage, especially from the osseous component. Rispoli *et al.*^[15] reported a 14-year-old girl presenting with

Table 1: Literature review of spinal osseous epidural arteriovenous fistulas with unique feature of a dilated round venous sac recruiting multiple arterial feeders

Authors	Gender/ age	Symptoms and signs	Location of a round venous sac/bony defect	Arterial feeders	Treatment	Angiographic outcome	Clinical outcome
Willinsky <i>et al.</i> 1993 ^[14]	Male/57	A 4-year history of progressive weakness of his right arm and leg, numbness below the right knee, and urinary hesitancy	Rt. side of C3 and C4 body and pedicle	Bilateral VAs, Rt. ACA, Rt. CCT	TAE (balloons, NBCA) TVE (balloon, coils)	Complete closure	IR
	Male/41	Severe right lower back pain, paresthesia and weakness in both legs, and bladder dysfunction	Rt. side of T9 body and pedicle	Bilateral T7-T11	TAE (NBCA, PVA)	A residual small fistula	GR
Chul Suh <i>et al.</i> 2004 ^[12]	Female/50	A 3-month history of intractable neck pain, audible bruit, and severe right radicular pain	Rt. side of the C5 body and pedicle	CCT, VA	TVE (coils)	A small residual shunt	GR
	Female/21	Both lower leg weakness and void difficulty	Rt. side of L2 body and pedicle	Both L2, Rt. L1, Rt. L3	TVE (coils)	Complete closure	GR
Iizuka <i>et al.</i> 2008 ^[13]	Male/26	A 6-month history of intractable low back pain, progressive paraparesis and paresthesia, and BBD	Lt. side of T11 body and pedicle	Bilateral T10, T11, T12	TAE (NBCA)	A faint residual shunt.	GR
Rispoli <i>et al.</i> 2009 ^[15]	Female/14	Intractable neck pain and acute progressive sensory and motor disturbances of the upper and lower extremities from EDH	Lt. side of the C5 body	VA	Surgery	Complete closure	GR
Song <i>et al.</i> 2019 ^[11]	Male/57	N/A	Rt. lamina and spinous process of the C6 vertebra	Bilateral DCA	TVE (coils)	Nearly complete	N/A
Present study, 2020	Male/25	A 1-month history of progressive paraparesis and paresthesia and BBD	T7 lamina and spinous process	Bilateral T6, T7, T8, T9	TAE (NBCA) Surgery	Complete closure	GR

ACA – Ascending cervical artery; BBD – Bowel and bladder dysfunction; CCT – Costocervical trunk; DCA – Deep cervical artery; EDH – Epidural hematoma; GR – Good recovery; IR – Incomplete recovery; Lt – Left; N/A – Data not available; NBCA – N-butyl-2-cyanoacrylate; PVA – Polyvinyl alcohol particles; Rt – Right; T – Thoracic; TAE – Transarterial embolization; TVE – Transvenous embolization; VA – Vertebral artery

progressive neurological deterioration caused by epidural hematoma from small cervical SOEAVF successfully treated by emergency surgery, whereas the operation in a large thoracic SOEAVF reported by Iizuka *et al.*^[13] was abandoned due to the difficulty in control of hemostasis.

In our case, SOEAVFs tend to be a high-flow shunt with drainage into long segments of large epidural venous plexus. The osseous fistula in our case recruited multilevel of intercostal arteries from the ipsilateral and contralateral arteries converging into a round venous sac. To reduce the flow, transarterial embolization was performed owing to failure of venous approach. Due to the ASA originating at the same level as the feeding vessel in our case, we decided to avoid further transarterial embolization with NBCA. Therefore, the decision-making of surgical treatment was chosen. However, awareness of facing uncontrollable hemorrhage is mandatory. We expected this serious event because the fistula may recruit more blood supply during the surgery was postponed. Before starting the operation, the anesthesiologist was notified for preparation of perioperative blood transfusion during profuse intraoperative hemorrhage.

The pathogenesis of SOEAVF remains unknown. SOEAVF may be associated with traumatic compressive fracture. The traumatic fistula developed in a fractured vertebral body and was termed “spinal intraosseous epidural arteriovenous fistula.”^[16,17] Different to spontaneous SOEAVF characterized by Chul Suh *et al.*,^[12] the trauma-related intraosseous fistula often drains into the epidural venous plexus and connects to the basivertebral vein with reflux into the intradural perimedullary veins. In addition, endovascular treatment through arterial route with liquid embolic material, e.g., Onyx or NBCA, is an effective method for the traumatic type.^[16-18]

According to the comparative analysis of SEAVFs with or without intradural venous drainage by Takai and Taniguchi,^[10] they found that SEAVFs with intradural venous drainage were diagnosed in patients with a mean age around the sixth decade of life and commonly occurred in the thoracolumbar and lumbar regions, whereas the extradural fistulas with severe mass effect without intradural venous drainage were found in younger patients around the third decade of life and frequently occurred in the cervical and upper thoracic

regions. SEAVF with compressive myelopathy from an enlarged epidural venous plexus consists of high-flow, multiple complex anastomoses between arterial feeders and the epidural venous plexus, requiring multisection treatment including endovascular treatment and/or surgical treatment with high rate of incomplete occlusion. Similarly, spontaneous SOEAVF in our review commonly occurred in a young patient at the cervical or thoracic spine. The symptoms of patients frequently resulted from compression of large epidural venous drainage from high-flow fistulas. Treatment of these fistulas required multisection treatment using embolization, surgery, or combined approach.

Conclusion

SOEAVFs tend to be a high-flow shunt presenting with compressive myelopathy. Endovascular treatment through venous and/or arterial routes remains the treatment of choice for SOEAVF with a large epidural vein. Surgical treatment should be used in the setting of unsuccessful endovascular treatment. However, awareness of facing uncontrollable hemorrhage is mandatory. Combined endovascular and direct surgical therapies may be another option for the fistula with symptomatic large epidural venous plexus.

Consent

The patient has given consent to be enrolled and has his data published.

Declaration of patient consent

The authors certify that he has obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published, and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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