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Case Report

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Reversible Aggravation of Neurological Deficits after Transforaminal Epidural Steroid Injection in a Patient with Undiagnosed Spinal Dural Arteriovenous Fistula

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ABSTRACT

Here we present a case of a 77-year-old man who underwent an epidural steroid injection complicated by delayed monoplegia and urinary incontinence. An MRI showed T2 hyperintensity at the conus along with small serpentine vessels surrounding the spinal cord. An angiogram was performed which showed a spinal dural arteriovenous fistula (SDAVF) with prominent draining vein at the right L3 level. The patient underwent repeat laminectomy and disconnection of spinal dural fistula after failed endovascular repair. His symptoms slowly improved after the lumbar decompression and physical therapy. SDAVF remains a diagnostic challenge. Epidural injection is contraindicated in these patients due to venous hypertension resulting in possible conus ischemia. SDAVF must be considered in the differential diagnosis when unexpected neurological complications occur after epidural steroid injection.

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Abbreviations

SDAVF = spinal dural arteriovenous fistula
MRI = magnetic Resonance Imaging
ESI = epidural steroid injection
TFESI = transforaminal epidural steroid injection
CES = cauda equina syndrome

Introduction

Spinal dural arteriovenous fistula (SDAVF) is a rare malformation resulting in abnormal connections between arterial and venous networks in the spinal column. Although rare, SDAVF comprises 60 to 80% of spinal vascular malformations [1]. The etiology of these lesions are unclear although they appear more likely to form in older male patients. In SDAVF, an abnormal connection is present between radicular artery and medullary vein, resulting in the arterialization of perimedullary veins. Diagnosis remains challenging as MRI findings may be subtle. Congestive myelopathy occurs resulting in progressive weakness which may be acutely exacerbated with injection into the epidural space [2, 3]. Here we present a case of delayed monoplegia after a right sided transforaminal epidural steroid injection (TFESI) in a patient with an undiagnosed SDAVF. A literature review on this diagnostically challenging pathology is also provided.

Case Report

A 77 year-old 79 kg man with a history of degenerative spondylosis status post decompression of the lumbar spine at the L3 and L4 levels one year prior presented to the pain clinic for recurrent radiating right leg pain following the right L4 dermatome. MRI of the lumbar spine with contrast performed 3 months after lumbar decompression showed degenerative disc and hypertrophic osteophyte at L4/5 and L5/S1 level. The patient underwent a right L4-L5 TFESI under fluoroscopic guidance with a mixture containing 80 mg of triamcinolone and 0.25% bupivacaine injected. The patient tolerated the procedure and after one hour, the patient was discharged with no procedural complications.

About 12 hours later, the patient reported weakness in the right lower extremity which progressively worsened. The patient also reported inability to control bowel movements and urinary retention. This progressed to complete paralysis of the right lower extremity a few hours later. An MRI with contrast was obtained showing hyperintensity at the conus along with small serpentine vessels surrounding the spinal cord seen in sagittal but not axial views (Figure 1). The patient was immediately admitted to the Neuro ICU, but about 10 hours upon symptom onset, the patient's paralysis began to improve. A few hours later the patient was able to walk with assistance and able to void. After careful history taking, the patient endorsed history of voiding difficulty over the past month. A spinal angiogram showed a dural arteriovenous fistula with prominent draining vein at the right L3 level and was supplied primarily by the L2 lumbar radicular arteries through collateral flow, since the L3 radicular lumbar arteries were found to be occluded at their aortic origin level (Figure 2). The patient

underwent repeat laminectomy and disconnection of SDAVF under microscope after failed endovascular embolization of the vessels. Multiple dilated vessels were noted on the surface of the nerve roots during the surgical procedure. A large arterialized vessel entering through the right L3 root sleeve and going into the canal heading towards the conus was identified. This was dissected free of its surrounding and transected. Immediately after transection, the dilated vessels on the surface of the nerve rootlets were noted to decrease significantly in caliber. The patient was discharged two days after surgical procedure to rehabilitation. Three months post-procedure, the patient continued to improvement in neurological symptoms.

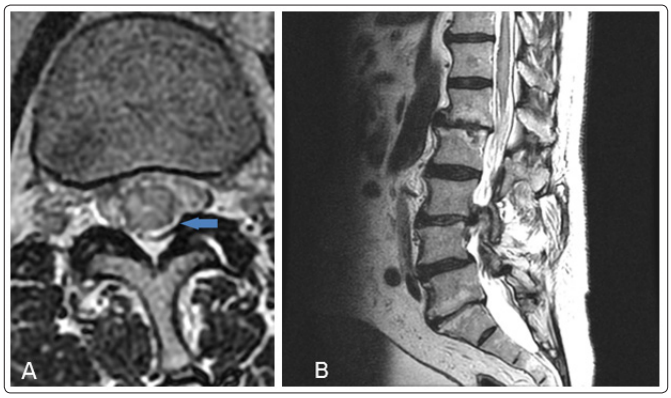

Figure 1: A) T2-weighted MRI axial image of the lumbar spine showing hyperintensity (blue arrow). **B)** Flow void pattern on sagittal image is seen along the L2 vertebral body.

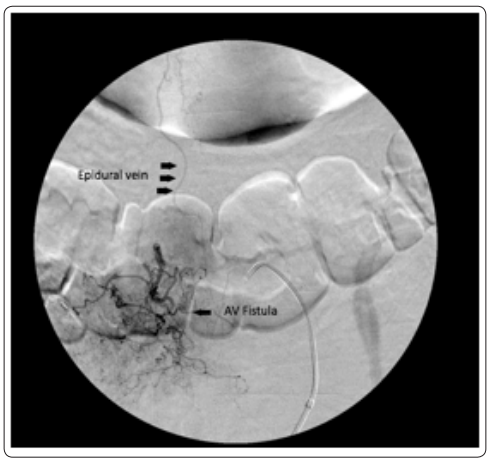

Figure 2: A spinal angiography showing a dural arteriovenous fistula with prominent draining vein at the right L3 level.

Discussion

Epidural steroid injection remains a mainstay treatment for lumbar radiculopathy. Complications from epidural steroid injection are rare, but catastrophic outcomes have been reported when ESI is performed in patients with SDAVF. Fortunately, recovery is possible with appropriate treatment. Epidural steroid injection is contraindicated in patients with SDAVF but diagnosis is often missed. Paralysis from ESI in patients with undiagnosed SDAVF has been described by several case reports (Table 1).

Table 1: Published cases of epidural steroid injection in patients with SDAVF. Ten out of eleven patients with SDAVF experienced neurological complications after ESI

Report	Intervention	Injectate mixture at each site	Total volume injected (mL)	Procedure Outcome	Onset of symptoms post-injection	SDAVF Treatment and outcome
Kim et al Case 1 (2016)	TFESI (bilateral) L5 and S1	Dexamethasone (1.25mg), lidocaine, bupivacaine, normal saline	20	CES, bilateral lower extremity paraplegia	8 hours	Endovascular embolization; incomplete recovery
Kim et al Case 2 (2016)	Caudal ESI	Dexamethasone (unknown dose)	Not described	CES, bilateral lower extremity paraplegia	Not reported	Endovascular embolization; incomplete recovery
Kim et al Case 3 (2016)	Right L5-S1 TFESI	Dexamethasone (10mg), ropivacaine, normal saline	3	Pain relief, no weakness	N/A	N/A
Annaswamy & Worchel (2016)	Left L2-L3 Interlaminar ESI	Betamethasone (12mg), bupivacaine	6	CES, bilateral lower extremity paraplegia	4 hours	Endovascular embolization; incomplete recovery
Sharma & Sharma (2016)	L2-L3 Interlaminar ESI	Betamethasone (12mg), bupivacaine	4	CES	4 hours	Endovascular embolization; incomplete recovery
Oliver et al (2012)	L5-S1 Interlaminar ESI	Not described	Not described	CES, bilateral lower extremity paraplegia	3 hours	Endovascular embolization; near complete recovery
Owen et al Case 1 (2011)	L4-L5 ESI	Triamcinalone (80mg), 8ml lidocaine	Not described	Temporary CES, monoplegia	12 hours	Surgery; recovery not described
Owen et al Case 2 (2011)	L3-L4 ESI	Triamcinalone (80mg), 7ml lidocaine	Not described	bilateral lower extremity paraplegia	24 hours	Endovascular embolization; incomplete recovery
Hetts et al Case 1 (2006)	L2-L3 ESI	12 mL ropivacaine 0.2%, 2 mL methylprednisolone 80 mg/mL	14	CES, Progressive paraparesis,	7 hours	Surgery; incomplete recovery

Hetts et al Case 2 (2006)	Lumbar ESI	Not described	Not described	CES, complete paralysis	~24 hours	Surgery after failed endovascular embolization; partial recovery
Hetts et al Case 3 (2006)	Lumbar ESI	Not described	Not described	CES, Bilateral lower extremity paraparesis	~24 hours	Endovascular embolization; partial recovery

Etiology and Diagnosis

Iatrogenic formation of arteriovenous fistula after lumbar microdiscectomy or other spine surgeries have been described in the literature [4, 5]. Our patient could tolerate epidural steroid injections prior to his initial laminectomy, suggesting that SDAVF may have formed as a complication of the surgery. Review of the initial lumbar laminectomy operative note did not reveal any report of abnormal vasculature, although this does not exclude its pre-existence

Diagnosis of SDAVF is challenging as symptoms are non-specific, MRI findings are often subtle, and spondylopathy is often a comorbidity [6, 7]. Paraparesis has been found to be the most common initial presenting symptom in patients with SDAVF but variability exists in the severity of this symptom [7]. The presence of flow voids in the subarachnoid space is a feature seen on MRI but is not specific to SDAVF. Flow voids may be seen in spinal dural arteriovenous malformations or epidural arteriovenous fistula. Cord edema may also be seen on MRI as a result of venous congestion [8]. In a study of 40 patients with known SDAVF, 34 of the patients showed cord signal changes. Nevertheless, mean time to diagnosis has been found to be one year with about 18% of patients misdiagnosed. Angiography is the gold standard for diagnosis [7].

Epidural Steroid Injection and SDAVF

The normal epidural space can tolerate injection of volume as the pressure is zero or slightly negative, however in the setting of SDAVF, venous hypertension occurs resulting in further compressive ischemia of the spinal cord [9]. Diagnosis is often challenging as symptoms often are similar to lumbar radiculopathy, lumbar stenosis, polyneuropathy, demyelinating disease, and intramedullary tumor. Injection of solution into the epidural space may acutely worsen venous congestion causing spinal cord or nerve root impingement resulting in cauda equina syndrome (CES). Paraplegia after epidural steroid injection have been reported in caudal, interlaminar, and transforaminal approaches to the epidural space [10-15]. Therefore, caution dictates that epidural steroid injection should not be performed in patients with SDAVF due to the significant risk of paraplegia [10]. Injection of a small volume may result in significantly elevated hypertension in the epidural space with as little as 4 mL of volume causing cauda equina syndrome. Reported cases of ESI in patients with SDAVF demonstrates that the onset of symptoms is variable (Table 1). Systemic steroid administration alone has shown to cause aggravation of SDAVF symptoms potentially due to water retention and engorgement of the SDAVF [16, 17]. This may also explain the variable time in the onset of symptoms in patients receiving ESI. Concomitant injection of local anesthetic which have arterial vasodilatory properties may also explain the variability of symptom onset [18].

Treatment

Endovascular transarterial embolization of SDAVF is often technically challenging with success ranging from 25-75% [7, 8]. Although more invasive, surgical repair reaches 100% successful treatment. A meta-analysis performed by Steinmetz et al shows 98% success rate and 1.9% complication rate for surgical repair [19]. A minimally invasive approach using tubular retractor has been suggested but needs further studies [20]. The utility of a lumbar drain in the setting of acute exacerbation of SDAVF induced myelopathy has been demonstrated and should be considered [13]. Effective treatment of the venous congestion caused by SDAVF has been shown to arrest or reverse the changes caused by myelopathy, therefore efficient diagnosis and treatment is recommended.

Conclusion

Spinal dural arteriovenous fistula is a rare condition with potentially devastating consequences if unrecognized. Unfortunately, SDAVF remains a diagnostic challenge due to multiple factors. Epidural injection of steroids is contraindicated with this pathology. Any unexplained neurological complications from epidural injection should alert one to include SDAVF in the differential diagnosis. Interventional pain specialists should be aware of SDAVF and pursue further evaluation including neurological consult as well as angiography should MRI suggest the presence of this pathology.

References

1. Walavan Sivakumar 1, Gabriel Zada, Parham Yashar, Steven L Giannotta, George Teitelbaum, et al. (2009) Endovascular management of spinal dural arteriovenous fistulas. A review. Neurosurg Focus 26: E15.
2. Jellema K, CC Tijssen, J. van Gijn (2006) Spinal dural arteriovenous fistulas: a congestive myelopathy that initially mimics a peripheral nerve disorder. Brain 129: 3150-3164.
3. Koch, M.J (2017) Open and endovascular treatment of spinal dural arteriovenous fistulas: a 10-year experience. J Neurosurg Spine 26: 1-5.
4. Aneesh Mohimen, Santhosh Kumar K, E R Jayadevan, Narendra Kumar Jain, T R Kapilamoorthy (2016) Spinal venous hypertension secondary to pelvic extra-spinal arteriovenous fistula-a previously unreported cause of congestive myelopathy. Spine J 16: e41-42.
5. Huttman D, Mathew Cyriac, Warren Yu, Joseph R O'Brien (2016) The unusual presentation of a vascular injury after lumbar microdiscectomy: case report. J Neurosurg Spine 24: 381-384.
6. H K Yang, J W Lee, S E Jo, C Jung, O-K Kwon, et al. (2016) MRI findings of spinal arteriovenous fistulas: focusing on localization of fistulas and differentiation between spinal dural and perimedullary arteriovenous fistulas. Clin Radiol 71: 381-388.
7. Jookyung Lee, Young-Min Lim, Dae Chul Suh, Seung Chul Rhim, Sang Joon Kim, et al. (2016) Clinical presentation, imaging findings, and prognosis of spinal dural arteriovenous fistula. J Clin Neurosci 26: 105-109.
8. Krings T, S Geibprasert (2014) Spinal dural arteriovenous fistulas. AJNR Am J Neuroradiol 30: 639-648.
9. Pedro L Antibas, Paulo do Nascimento Junior, Leandro G Braz, João Vitor Pereira Doles, Norma S P Módolo, et al. (2014) Air versus saline in the loss of resistance technique

for identification of the epidural space. Cochrane Database Syst Rev 2014: CD008938.

10. Sunwoong Kim, Yuseong Choi, Jinyoung Park, Duk Hyun Sung (2016) Acute Paraplegia after Lumbar Steroid Injection in Patients with Spinal Dural Arteriovenous Fistulas: Case Reports. Ann Rehabil Med 40: 949-954.
11. Annaswamy TM, J Worchel (2016) Paraplegia Following Lumbar Epidural Steroid Injection in a Patient with a Spinal Dural Arteriovenous Fistula. Am J Phys Med Rehabil 96: e147-e150.
12. Sharma K, V.D. Sharma (2016) Delayed onset paraparesis complicating epidural steroid injection with underlying spinal dural arteriovenous fistula. Pain Manag 6: 421-425.
13. Oliver TA, M Sorensen, AS Arthur (2012) Endovascular treatment for acute paraplegia after epidural steroid injection in a patient with spinal dural arteriovenous malformation. J Neurosurg Spine 17: 251-255.
14. NC Owen, L T Smith, L Massey, A J Durnford, C E M Hillier (2011) Decompensation of undiagnosed spinal dural arteriovenous fistulae after lumbar epidural injection and spinal anaesthesia. Br J Anaesth 107: 109-111.
15. Hetts, S.W., J. Narvid, T. Singh S. Meagher K. Corcoran et al. (2007) Association between lumbar epidural injection and development of acute paraparesis in patients with spinal dural arteriovenous fistulas. AJNR Am J Neuroradiol 28: 581-583.
16. Lee CS, Pyun H W, Chae E Y, Kim KK, Rhim S C, et al. (2009) Reversible Aggravation of Neurological Deficits after Steroid Medication in Patients with Venous Congestive Myelopathy Caused by Spinal Arteriovenous Malformation. Interventional Neuroradiology 15: 325-329.
17. Rain S, Udding J, Broere D (2016) Acute Clinical Worsening after Steroid Administration in Cervical Myelitis May Reveal a Subdural Arteriovenous Fistula. Case reports in neurology 8: 234-242.
18. D J Newton 1, G A McLeod, F Khan, J J F Belch (2005) Vasoactive characteristics of bupivacaine and levobupivacaine with and without adjuvant epinephrine in peripheral human skin. Br J Anaesth 94: 662-667.
19. Steinmetz M.P (2004) Outcome after the treatment of spinal dural arteriovenous fistulae: a contemporary single-institution series and meta-analysis. Neurosurgery 55: 77-88.
20. Anderson Chun on Tsang, Philip Yat Hang Tse, Grace Hoi Ting Ng, Gilberto Ka Kit Leung (2015) Minimal access microsurgical ligation of spinal dural arteriovenous fistula with tubular retractor. Surg Neurol Int 6: 99.