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Acute paraplegia in patient with spinal dural arteriovenous fistula after lumbar puncture and steroid administration: A case report

Ibrahim Alhendawy^{a,*}, Bob Homapour^a, Ronil V. Chandra^b, Armin Drnda^a^a Department of Neurosurgery, Monash Medical Centre, Clayton, Victoria, 3168, Australia^b Department of Interventional Neuroradiology, Monash Medical Centre, Clayton, Victoria, 3168, Australia

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ABSTRACT

INTRODUCTION AND IMPORTANCE: Spinal dural arteriovenous fistula (SDAVF) is an uncommon cause of longitudinal extensive transverse myelitis (LETM). It usually presents with vague congestive myelopathy symptoms and diagnosis is usually difficult on initial presentation. Common daily neurological interventions can aggravate the underlying pathophysiology leading to undesirable acute neurological deterioration. Intravenous steroids administration and lumbar (LP) puncture as a diagnostic tool are amongst the most commonly reported aggravating interventions. This rare case presentation highlights this association with its negative impact on the patient outcome in misdiagnosed cases.

CASE PRESENTATION: The authors present a sixty-eight-year-old male with paraplegia following steroid administration and LP for presumed inflammatory/autoimmune LETM in the setting of misdiagnosed SDAVF. The absence of flow voids on the conventional T2-weighted magnetic resonance image (MRI) lead to misdiagnosis. He had satisfactory neurological recovery few hours after surgical disconnection.

CLINICAL DISCUSSION: SDAVF is known to cause congestive myelopathy symptoms. Spinal angiogram is the gold standard for diagnosis. Although the exact mechanism is not fully understood, misdiagnosed cases like our case can develop severe neurological deterioration with steroid administration and lumbar puncture.

CONCLUSION: Although SDAVF is an uncommon cause of LETM, Clinicians should carefully exclude it before proceeding to steroid administration or performing LP as they can lead to devastating neurological deterioration.

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1. Introduction

Spinal dural arteriovenous fistulae SDAVF is the most common spinal arteriovenous malformations encountered in neurosurgical clinical practice and their diagnosis is usually delayed as a cause of myelopathy as majority of patients present with non-specific clinical symptoms secondary to congestive myelopathy [1,2].

Aggravating factors of congestive myelopathy associated with SDAVF have been reported in the literature including steroids administration and lumbar puncture [3,4]. We present a case of acute paraplegia approximately six hours following lumbar

puncture and intravenous steroids administration in patient with congestive myelopathy secondary to undiagnosed SDAVF. He was treated at a tertiary neurosurgery centre.

Twenty-five cases of acute neurological deterioration following intravenous steroid administration and six cases following lumbar puncture have been reported in the literature [3,5]. This case support the evolving evidence of association between certain interventions and neurological deterioration in patients with SDAVF. This case report has been reported in line with the SCARE 2020 Criteria [6].

2. Presentation of case

We present a sixty-eight-year-old male who presented with acute urinary retention on background of recent diagnosis of benign prostatic hyperplasia. His past medical history was significant for diabetes mellitus and hypercholesterolemia managed by oral hypoglycaemic and diet. He has no family history of spinal vascular malformation. According to our institution protocol, urinary

Abbreviations: CISS, constrictive interference in steady state; CSF, cerebrospinal fluid; LETM, longitudinal extensive transverse myelitis; LP, lumbar puncture; MRI, magnetic resonance imaging; SDAVF, spinal dural arteriovenous fistula.

* Corresponding author at: Monash Medical Centre, 246 Clayton Rd, Clayton, Victoria, 3168, Australia.

E-mail addresses: Ibrahim@live.com.au (I. Alhendawy), Bob.Homapour@monashhealth.org (B. Homapour), Ronil.Chandra@monashhealth.org (R.V. Chandra), Armin.Drnda@monashhealth.org (A. Drnda).

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catheter was inserted, and outpatient follow up was arranged at the urology clinic.

Five days later, he presented with progressive bilateral lower limb paraesthesia and feeling unsteady. His neurological examination revealed intact motor power and reflexes in both lower limbs with thoracic 12 sensory level.

Full spine MRI demonstrated diffuse T2 weighted image hyperintensity consistent with cord oedema from T7 to the tip of the conus [Fig. 1a]. Minimal enhancement was noted at the conus on the T1 weighted image, but flow voids were difficult to visualize on the conventional T2 weighted image. Considering these MRI findings and his acute presentation of urinary retention, an underlying SDAVF was thought to be unlikely as bladder and bowel dysfunction is usually a late manifestation of SDAVF.

Subsequently, high volume lumbar puncture was performed, and cerebrospinal fluid (CSF) sent for analysis. One gram of intravenous prednisolone was administered for presumed underlying autoimmune or inflammatory condition while waiting for the CSF results.

Progressive neurological deterioration started after the forementioned interventions and over approximately six hours he became paraplegic with motor power 1/5 in both lower limbs, thoracic 12 sensory level and areflexia.

Urgent MRI spine was done that excluded hemorrhage. On further reviewing of the MRI, Flow voids were detected on the Constrictive interference in steady state (CISS) MRI with suspected DAVF at the level of the second lumbar vertebra [Fig. 1b].

Spinal angiogram confirmed the presence of SDAVF supplied by L2 radicular artery and draining into arterialized tortuous vein [Fig. 2a,b]. CSF was clear and analysis demonstrated normal chemistry with no oligoclonal bands.

As the L2 radicular artery was noticed to be also supplying the posterior spinal artery, microsurgical dissection was considered to be safer.

He underwent laminectomy and the draining vein was disconnected at the point where it entered the dura. The procedure was performed by two senior neurosurgeons.

Interestingly, approximately five hours after the procedure, significant neurological recovery was achieved with near recovery of both sensory and motor functions with motor power 4/5 proximally and 4+/5 distally in both lower limbs.

Post-operative angiogram demonstrated satisfactory disconnection of the fistula. His post-operative course was complicated with pulmonary embolism and embolic bowel infarction that required anticoagulation, laparotomy, bowel resection and colostomy. At four weeks post-operatively, he continued to be catheter dependent for urinary retention without significant recovery of the bladder function and he was discharged to rehabilitation with planned follow up.

Despite suffering from post-operative complications of pulmonary embolism and bowel infarction, he was satisfied with his significant lower limb neurological recovery as he initially thought it would be permanent. He also expressed his willing to publish his uncommon case for learning purpose.

3. Discussion

Spinal vascular malformations are rare pathologies of the spinal cord that may present with a wide variety of non-specific neurological symptoms. They can be simply classified into non-shunting vascular malformations like spinal cord cavernomas or shunting vascular malformations. Shunting vascular malformation can be further subdivided based on the arterial feeders into dural or pial

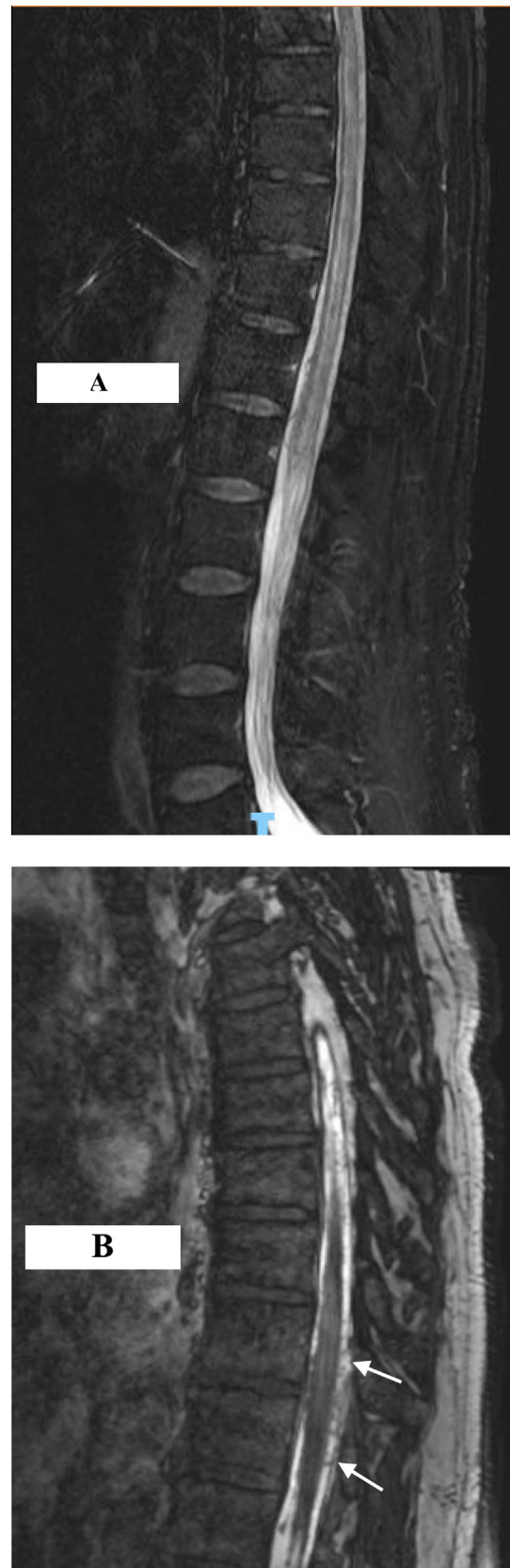


Fig. 1. A: T2 MRI demonstrating thoracic cord oedema involving the conus. B: CISS MRI demonstrating flow voids.



Fig. 2. A,B: 3D reconstruction lateral (A) and anteroposterior (B) spinal angiogram demonstrating arterIALIZED large draining radiculomedullary vein with nidus below L2.

vascular malformations. In pial vascular malformation, the arterial feeders are from the radiculomedullary and radiculopial arteries while in dural vascular malformations the feeding arteries are from the radiculomeningeal arteries. In addition, pial vascular malformation can be subdivided into fistulous type with direct connection between the feeding artery and the vein or nidiform with intervening vascular network between the two [7].

Spinal dural arteriovenous fistulae represent 70% of all spinal vascular malformations and although their exact aetiology is unknown, they are presumed to be acquired lesions. They result from an abnormal intradural connection between multiple small meningeal branches from the meningeal branch of the segmental artery and a single intradural vein resulting in retrograde arterial inflow and giving the appearance of caput medusae Arterialisation of the draining vein results in venous congestion and progressive hypoxic myelopathy [7,8].

They are most commonly located at the thoracolumbar region and their ambiguous presenting symptoms add more to the difficulty of diagnosis on initial presentation with median time of 12–44 months between initial presentation and diagnosis. Majority of patients are middle aged males with gait disturbance, lower limb paraesthesia or sensory loss which may be symmetrical or non-symmetrical. Bowel, bladder and erectile dysfunction is usually a late manifestation of the disease prior to diagnosis [9,10].

Although spinal angiogram is the gold standard for definitive diagnosis, MRI is always the initial imaging technique in almost all undiagnosed cases. The presence of increased T2 weighted image high signal in the cord, enhancement, mass effect, and flow voids should raise the clinical suspicion of underlying SDAVF [11]. Visualization of the flow voids can sometimes be difficult on the conventional T2 weighted MRI increasing the possibility of misdiagnosis, CISS MRI can be superior in that aspect [12].

Acute neurological deterioration following lumbar puncture for CSF analysis and steroid administration for presumed inflammatory or autoimmune pathology has been increasingly reported in the literature [3,5].

Although the exact pathophysiological mechanisms are not fully understood, mechanisms have been suggested. With steroids, transient hydrostatic retention and hypervolemia may aggravate the existing venous hypertension. In addition, as steroids are known to decrease capillary permeability, they may reduce the compensatory retrograde perfusion from the venous side in patients with long standing venous hypertension [13–15].

High venous pressure results in alteration in the spinal parenchymal arteriovenous pressure and sudden drop in the cerebrospinal fluid pressure may increase the venous pressure resulting in decreased spinal cord perfusion [4,16].

In regard to treatment of SDAVF, disconnection of the shunting zone, either by occlusion of the distal part of the feeding artery or the proximal part of the draining vein, is the gold standard. This can be either achieved by surgical disconnection of the draining vein or endovascular occlusion using embolic agents. Distal occlusion of the feeding arteries may result in transient improvement but will eventually have high rate of recurrent due to the rich collateralization of the dura [17].

Considering the less invasive nature of endovascular embolization with rapid recovery, it has evolved as the first treatment option when considered safe despite its lower success rate compared to surgical disconnection. For example, the presence of spinal cord feeder at the same pedicle of the shunt arterial feeder is considered

to be a relative contraindication for endovascular occlusion given the risk of cord infarction. This was the case in our report with the shunt feeder also supplying the posterior spinal artery.

4. Conclusion

Spinal dural arteriovenous fistulae usually present with vague symptoms representing a diagnostic challenge. Although they are uncommon causes of LETM, they should be carefully excluded before certain interventions with aggravating effect. CISS MRI can be helpful in detection of the flow voids where it is difficult to detect them on the conventional T2 weighted MRI.

This case highlights the aggravating effect of steroid and lumbar puncture on congestive myelopathy secondary to SDAVF. With increasing number of reported cases in the literature, clinicians should be aware of the potential outcome of common neurological interventions.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

This case report is exempted from ethical approval as per our institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Ibrahim Alhendawy: Conceptualization, methodology and writing.

Bob Homapour: Editing, supervision.

Ronil V. Chandra: Supervision, final review and approval for submission.

Armin Drnda: Supervision, final review and approval for submission.

Registration of research studies

Not applicable.

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Ibrahim Alhendawy, MBChB.

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The authors verify that this case report has not been published previously and is not under consideration for publication elsewhere. The authors also verify that this publication is approved by all authors and by the responsible authorities at our institution, and if accepted, it will not be published elsewhere in the same form.

References

- [1] M.J. Aminoff, V. Logue, Clinical features of spinal vascular malformations, *Brain* 97 (1) (1974) 197–210.
- [2] M.J. Aminoff, R.O. Barnard, V. Logue, The pathophysiology of spinal vascular malformations, *J. Neurol. Sci.* 23 (2) (1974) 255–263.
- [3] M.A. DiSano, R. Cerejo, M. Mays, Acute paraparesis and sensory loss following intravenous corticosteroid administration in a case of longitudinally extensive transverse myelitis caused by spinal dural arteriovenous fistula: case report and review of literature, *Spinal Cord Ser. Cases* 3 (2017) 17025.
- [4] C. Garcia-Cabo, G. Moris, Sudden paraplegia after lumbar puncture as a clue in the diagnosis of a patient with spinal dural arteriovenous fistula, *Eur. Spine J.* 26 (Suppl 1) (2017) 151–153.
- [5] A. Cakar, H.I. Akcay, T. Gunduz, M. Kurtuncu, Paraplegia following lumbar puncture: a rare complication in spinal dural arteriovenous fistula, *Acta Neurol. Belg.* 118 (4) (2018) 543–545.
- [6] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, Group S, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, *Int. J. Surg.* 84 (2020) 226–230.
- [7] T. Krings, Vascular malformations of the spine and spinal cord*: anatomy, classification, treatment, *Clin. Neuroradiol.* 20 (1) (2010) 5–24.
- [8] K. Takai, T. Komori, M. Taniguchi, Microvascular anatomy of spinal dural arteriovenous fistulas: arteriovenous connections and their relationships with the dura mater, *J. Neurosurg. Spine* 23 (4) (2015) 526–533.
- [9] K. Jellema, C.C. Tijssen, M. Sluzewski, F.W. van Asbeck, P.J. Koudstaal, J. van Gijn, Spinal dural arteriovenous fistulas—an underdiagnosed disease. A review of patients admitted to the spinal unit of a rehabilitation center, *J. Neurol.* 253 (2) (2006) 159–162.
- [10] K. Jellema, C.C. Tijssen, J. van Gijn, Spinal dural arteriovenous fistulas: a congestive myelopathy that initially mimics a peripheral nerve disorder, *Brain* 129 (Pt. 12) (2006) 3150–3164.
- [11] J.R. Gilbertson, G.M. Miller, M.S. Goldman, W.R. Marsh, Spinal dural arteriovenous fistulas: MR and myelographic findings, *AJNR Am. J. Neuroradiol.* 16 (10) (1995) 2049–2057.
- [12] Z. Li, Y.A. Chen, D. Chow, J. Talbott, C. Glastonbury, V. Shah, Practical applications of CISS MRI in spine imaging, *Eur. J. Radiol. Open* 6 (2019) 231–242.
- [13] R.E. Strowd, C. Geer, A. Powers, N. Abou-Zeid, A unique presentation of a spinal dural arteriovenous fistula exacerbated by steroids, *J. Clin. Neurosci.* 19 (3) (2012) 466–468.
- [14] M. Cabrera, C. Paradas, C. Marquez, A. Gonzalez, Acute paraparesis following intravenous steroid therapy in a case of dural spinal arteriovenous fistula, *J. Neurol.* 255 (9) (2008) 1432–1433.
- [15] C.S. Lee, H.W. Pyun, E.Y. Chae, K.K. Kim, S.C. Rhim, D.C. Suh, Reversible aggravation of neurological deficits after steroid medication in patients with venous congestive myelopathy caused by spinal arteriovenous malformation, *Interv. Neuroradiol.* 15 (3) (2009) 325–329.
- [16] G. Koerts, V. Vanthuynne, M. Delavallee, H. Rooijackers, C. Raftopoulos, Spinal dural arteriovenous fistula presenting with paraplegia following lumbar puncture, *J. Neurosurg. Spine* 19 (1) (2013) 57–60.
- [17] T. Krings, S. Geibprasert, Spinal dural arteriovenous fistulas, *AJNR Am. J. Neuroradiol.* 30 (4) (2009) 639–648.

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