Eur Spine J
DOI 10.1007/s00586-017-4946-5

CASE REPORT

Sudden paraplegia after lumbar puncture as a clue in the diagnosis of a patient with spinal dural arteriovenous fistula

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Received: 25 September 2016 / Revised: 4 January 2017 / Accepted: 8 January 2017

Abstract
Purpose Spinal dural arteriovenous fistula (SDAVF) is manifested as congestive myelopathy with progressive motor, sensory and urinary symptoms. Sometimes, clinical picture and magnetic resonance imaging of the spinal cord are not specific and the diagnosis becomes troublesome.
Methods We present a 68-year-old male with a progressive paraparesis of unknown etiology over the course of 12 months. Immediately after LP, the patient remarked a sudden worsening in muscular balance of his inferior limbs and a worsening of urinary retention. This fact was the clue to the SDAVF diagnosis.
Results SDAVF was totally resolved after surgical treatment. During next months, paraplegia slightly improved and he is currently receiving rehabilitation treatment.
Conclusions Acute paraplegia or sudden worsening of previous symptoms secondary to decreasing in cerebrospinal fluid pressure after lumbar puncture has been described, so physicians should be aware of this dramatic and avoidable complication.

Keywords Spinal fistula · Lumbar puncture · Paraplegia

Introduction

Spinal dural arteriovenous fistulas (SDAVF) are the most common vascular malformations of the spinal cord [1]. They consist of a single shunt between an epidural artery, most often a radiculomeningeal artery, and an epidural and/or perimedullary vein that drains in the reverse direction.

SDAVF are relatively rare and their diagnosis is often made late, resulting in morbidity due to the lack of appropriate treatment. While the clinical presentation of this disease is non-specific, they are almost unambiguously revealed by spinal magnetic resonance imaging (MRI). Worsening after lumbar puncture (LP) [2], pleocytosis in cerebrospinal fluid (CSF) [3], and other anomalies have been described in patients with SDAVF, and they may be confusing factors in making the final diagnosis. Therefore, physicians should be aware of these atypical manifestations to avoid delayed and incorrect diagnoses.

Clinical case

We present a 68-year-old male with a progressive paraparesis over the course of 12 months. His past medical history was notable for Lyme disease having been treated correctly with antibiotics 20 years ago. He was on no medication, and he denied drinking alcohol or smoking. His family history was not relevant.

One year earlier, he started with lumbar pain irradiated to the lower limbs. At the same time, he reported perineal sensory loss and difficulty in urination and defecation. At the beginning of the symptoms, spinal MRI with gadolinium contrast was performed without abnormal results. Two months later, the patient began with weakness in the lower limbs and the sphincter disorder worsened. General

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Published online: 01 February 2017

medical examination was normal. Two spinal MRIs and two CSF analyses were performed and both were normal. Laboratory blood examinations performed at different times were normal, including microbiological, autoimmune, and angiotensin converting enzyme analysis.

While complementary studies were carried out, a third LP was performed, CSF analysis showed 31 leucocytes (10^6/L) with lymphocyte predominance, and 108 mg/dL of proteins, there was no glucose consume. Microbiological and oligoclonal bands were normal. Immediately after LP, the patient remarked a sudden worsening in muscular balance of his inferior limbs and a worsening of urinary retention. Neurological examination demonstrated spastic paraplegia and hypesthesia with sensitive level at the tenth dorsal dermatome.

The patient was empirically treated with antibiotics and immunoglobulins with no improvement.

A new gadolinium MRI was performed 10 months after the symptoms started, and this one demonstrated a tumefactive dorsal myelopathy with low enhancement after gadolinium injection. Angiographic MRI sequences were normal (Fig. 1).

At this point, digital substraction arteriography (DSA) was performed showing a dorsal DAVF that involving dural branches of a radicular artery (Fig. 2). Other causes of myelopathy were excluded (normal laboratory examinations and CSF analyses).

After DSA-guided diagnosis, endovascular treatment was planned. Unfortunately, a small spinal branch arose from the SDAVF nidus, so radiologist had to halt the proceedings. Several days later, the patient underwent surgery with positive results. After a week, he developed a surgical bed infection resolved without complications.

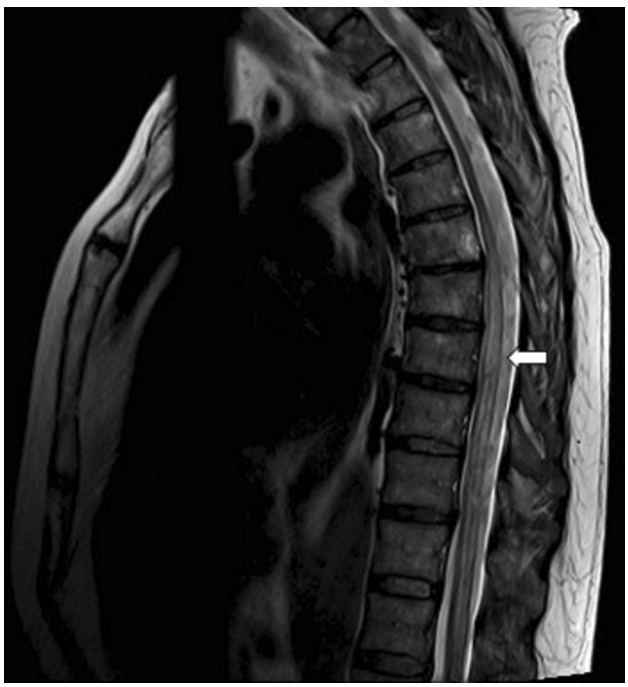

Fig. 1 MRI of the spinal cord. It shows a dorsal myelopathy (*arrow*)

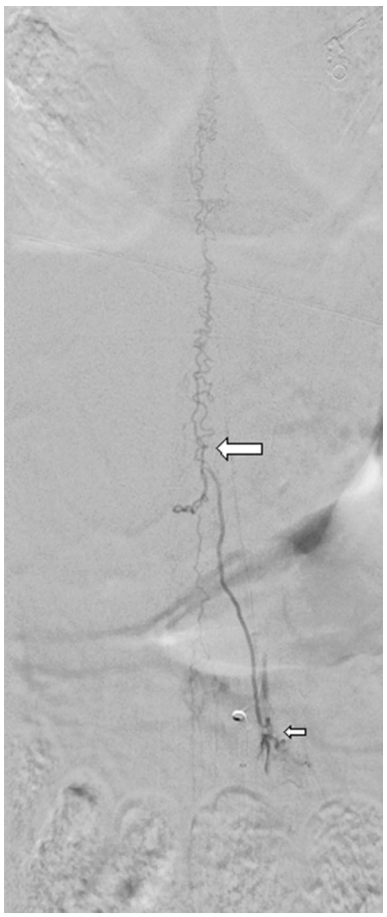

Fig. 2 DSA. It shows SDAVF nidus and venous congestion (see *arrows*)

SDAVF was totally resolved after surgical treatment.

During next months, paraplegia slightly improved and he is currently receiving rehabilitation treatment.

Discussion

In SDAVF, shunting of arterial blood flow causes venous congestion, venous hypertension, and, consequently, progressive myelopathy. High venous pressure results in variations of the arteriovenous pressure gradient in the spinal parenchyma inducing changes in vascular hemodynamics and, eventually, spinal ischemia. This mechanism may explain the clinical worsening described in SDAVF patients after LP due to a sudden decrease of CSF pressure [4].

Four SDAVF patients have been previously described with neurologic deterioration after performing LP or

Table 1 Acute paraplegia after lumbar puncture

Authors	Spinal level	Motor function after LP	Urinary function after LP
Roullet et al. [7]	T6	Paraplegia	Retention
	T7	Paraparesis	Retention
Awad and Barnett [8]	T7–8	Paraplegia	Retention
Aloui-Kasbi et al. [9]	NR	Paraplegia	Incontinency
Koerts et al. [4]	L2–L3	Paraplegia	Retention
Our patient 2015	T8–10	Paraplegia	Retention

LP lumbar puncture, *NR* not referred

myelography (Table 1) [4–9]. In cases of neurologic worsening following LP, the appearance of a compressive hematoma responsible for the clinical aggravation should be ruled out by performing an urgent spinal MRI [6].

In terms of diagnosis, CSF biochemical study usually shows no representative findings; however, an increase in protein accompanied with slight mononuclear pleocytosis may be found in SDAVF patients as the expression of the blood–brain barrier rupture secondary to arteriovenous pressures abnormalities. In addition, spinal MRI and MRI angiograms are highly sensitive and specific; nonetheless, there are some cases with atypical findings or normal spinal MRI at early stages. Therefore, DSA should be performed to confirm the diagnosis. Surgical or endovascular obliteration should be done promptly, because the majority of SDAVF patients experience clinical stabilization or improvement after treatment, reported even in paraplegic patients [5].

The normal results of spinal MRI and CSF analysis at the early stages in our patient demonstrate the progressive character of the myelopathy in relation with SDAVF, and so clinical suspicion is crucial in maximizing the yield of the studies.

The risk of neurologic worsening after LP leads us to recommend avoiding LP in patients with clinical suspicion of SDAVF, especially in patients with long-standing SDAVF.

The main aim of this case report is to highlight the risk of LP in patients with SDAVF, as clinicians cannot depend on an invasive risky procedure that could provoke more deterioration of patients' symptoms.

Compliance with ethical standards

Conflict of interest The first and last authors report no disclosures.

Funding sources of study None.

This paper gets ethical requirements of the hospital.

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