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Review

Multiple spinal arteriovenous fistulas: A case-based review

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ABSTRACT

The occurrence of multiple spinal dural arteriovenous fistulas (AVFs) is rare. The majority of cases reported are synchronous and the lesions are mainly found at different spinal levels. Metachronous AVFs have been defined as lesions that manifest in a temporal sequence after treatment of a first AVF. In this report, we present two distinct cases of multiple spinal AVFs. Also, we review the main features of the cases previously reported, with emphasis on the proposed theories for the origin of multiple AVFs. In patients with failure to improve after treatment of a spinal DAVF, a whole-spine angiographic examination is mandatory, not only to ascertain the complete closure of the treated fistula, but also to look for a possible second lesion at a different spinal level.

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1. Introduction

Spinal dural arteriovenous fistulas (AVF) are acquired pathological arteriovenous shunts developed within the layers of the dura. These are complex vascular lesions draining into the perimedullary venous system and producing severe neurologic symptoms due to venous congestion [1,2]. In some cases, these lesions are also associated with significant hemodynamic changes, ranging in severity from spontaneous resolutions to the development of multiple shunts over time [3]. The origin of this dynamic behavior is unclear, but certain related factors such as coagulopathy, trauma, and infection, among others, have been identified.

The majority of multiple spinal AVFs are often described during an initial examination as occurring simultaneously in different locations (synchronous). However, a subset of these multiple malformations is missed in the first examination but later identified during a second examination (metachronous) [4]. True metachronous AVFs have been defined as lesions that manifest at different locations in a temporal sequence after treatment of a first AVF. In the majority of cases, the second AVF is clinically manifested after a short period of improvement [3]. In this work, we review the main features of the cases previously reported [4–19] with emphasis on the proposed theories for the origin of multiple AVFs. We also report two unique cases of multiple spinal AVFs.

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Table 1
Synchronous spinal arteriovenous fistulas reported in the literature.

Author/year	Clinical presentation	Level of the AVFs	Feeders	Drainage	# of therapeutic procedures	Type of treatment
Thiebot/1986	Pain/paraparesis	T4 right, T12 left, L4, L5 right	Radicular arteries	Perimedullary veins	0	No treatment
Pierot/1993	Myelopathy	T6 left, T8 right	Intercostal arteries	Retromedullary vein	2	Embolization/surgery
Pierot/1993	Myelopathy	T8 right, T9 left	Intercostal arteries	Retromedullary vein	N/R	Surgery
Chaloupka/1995	Myelopathy	L1, T9 left	Radiculomeningeal branch	Perimedullary veins	3	Embolization/surgery
Dam-Hieu/2001	Myelopathy	T6 right, L1 right	Intercostal artery, PLSA	Retromedullary vein	2	Surgery
Krings/2004	Myelopathy	L1 right, L2 left	Radicular arteries	Perimedullary veins	2	Surgery
El-Serwi/2006	Myelopathy	T7 right, T5 right	Intercostal arteries, ASA	Perimedullary veins	2	Embolization
Cenzato/2007	Sensory impairment of the legs/sphincter disturbances	T5 right, T6 left	Radicular arteries	Perimedullary veins	1	Surgery
Shankar/2011	Incidental	C1, C2 and C6 left; C3, C6 right	Vertebral artery, ASA	Perimedullary veins	1	Incomplete embolization
Hanakita/2012	Myelopathy	T7 right, T12 left	Intercostal and subcostal arteries	Perimedullary veins	2	Surgery
Hetts/2013	Myelopathy	C5–C6 left, C5–C6 right	Radicular branches of CCT	Posterior spinal vein	1	Surgery

AVFs, arteriovenous fistulas; #, number; N/R, not reported; CCT, costocervical trunk; PLSA, posterolateral spinal artery; ASA, anterior spinal artery.

2. Case reports

2.1. Case 1 (Figs. 1 and 2)

A 51-year-old man presented with a 3-month history of dysesthesias, paresthesias, and "coldness" in both legs, which progressively worsened into an apraxic gait and associated urinary and fecal incontinence. He also complained of decreased coordination in both hands. Neurological examination revealed generalized hyperreflexia, bilateral clonus, and hypoesthesia below the T10 dermatome, with normal motor strength. Cervical magnetic resonance imaging (MRI) revealed extensive myelopathy from the craniocervical junction to the T4 spinal level, with tortuous and dilated vessels on the anterior surface of the spinal cord. A selective angiogram of the right vertebral artery confirmed a right C1 dural AVF fed by a short pedicle arising from the right vertebral artery with anterior venous drainage into perimedullary veins. These features prevented supraselective catheterization for endovascular treatment. The patient underwent surgical intervention consisting of a suboccipital craniotomy and clipping to obliterate the draining vein. Two days after surgery, a spinal angiography showed complete occlusion of the right-sided fistula, and the appearance of a new intradural AV shunt stemming from the left vertebral artery, which was not present in the previous examination. The patient underwent a second surgical intervention with coagulation of the draining vein on the left side. A spinal angiography showed complete occlusion of the AVF on both sides. The patient was discharged to a rehabilitation center to continue gait retraining. An MRI performed 6 months later, showed radiologic resolution of the cervical myelopathy.

2.2. Case 2 (Fig. 3)

A 72-year-old male presented to the emergency department with a 3-month history of gait disturbances and urinary incontinence. Clinical examination revealed bilateral leg weakness, right leg hyperreflexia and left leg hypoesthesia. A spinal cord MRI demonstrated the presence of a dorsal (T3–T8) myelopathy secondary to a spinal dural AVF. Using multilevel spinal cord angiography, the dural AVF was located at the right T7 level, which was embolized with Onyx®. We did not perform a complete angiogram at this time and the patient was discharged without further clinical deterioration. Thereafter, he began a rehabilitation program and partially regained most of his leg strength and ambulatory capacity. However, he did not regain urinary continence.

Five months after the initial procedure, the patient presented once again with worsened gait and decreased leg strength. A complete spinal angiogram confirmed total closure of the previously diagnosed T7 fistula and a new right T12 dural AVF. The new lesion was deemed unsuitable for embolization due to its proximity to the artery of Adamkiewicz. The patient underwent open surgical repair of the T12 dural AVF. A postoperative angiogram showed complete closure of both the T7 and T12 fistulas. Three months later, the patient presented to the emergency department with progressive paraparesis. A spinal angiogram showed recanalization of the previously embolized T7 dural AVF, which was embolized with glue. One month later, the patient was admitted complaining of decreased strength in his lower limbs. A spinal angiogram demonstrated anastomosis from the T6 radiculo-meningeal artery to the previously embolized T7 dural AVF, with abnormal venous drainage to the perimedullary veins. The patient underwent open surgical treatment of this T6 lesion. A postoperative angiogram demonstrated complete closure of all spinal AVFs.

3. Discussion

Multiple spinal AVFs are rare events, with a reported 2% rate of occurrence in the majority of studies [5,6]. However, this event could be underestimated because a complete spinal angiography is rarely performed in all patients [4]. The term "multiple" has been generally used to describe more than one AVF, irrespective of the timeline of the appearance of the lesions [4,6,20]. Synchronous AVFs are lesions occurring simultaneously in different locations and have been extensively reported in both cranial and spinal compartments [3–5,11–17,21] (Table 1). In contrast, metachronous spinal AVFs are lesions occurring at different times. They are extremely rare events with only six cases reported in the English literature