Spinal dural arteriovenous fistulas: early endovascular treatment or surgery?☆

Fístulas arteriovenosas espinales durales: ¿tratamiento precoz endovascular o quirúrgico?

Dear Editor:

It was with great interest that we read the article ''Spinal arteriovenous fistulas in adults: management of a series of patients treated at a neurology department'' by Ortega-Suero et al.[1] In this study, the authors retrospectively analyse the outcomes in a series of 10 patients with spinal arteriovenous fistulas (AVF) treated at their hospital over a 3-year period. Firstly, we wish to congratulate the authors on their series, the second modern series of patients with spinal AVFs to be published in Spain.[1,2] Ortega-Suero et al.[1] include 6 cases of spinal dural AVFs; we will focus on this type of AVF since they represent 75% of all spinal vascular malformations. We agree that spinal dural AVFs are difficult to diagnose despite considerable advances in neuroimaging techniques. Though rare, this type of AVF should be included in the differential diagnosis of patients with symptoms of progressive myelopathy and/or radiculopathy, given the poor neurological outcomes associated with late diagnosis and treatment. Up to 25%-30% of patients present paraplegia by the time spinal dural AVF is diagnosed.[3] Furthermore, a considerable percentage of patients will already have undergone unnecessary spine surgery following incorrect aetiological diagnosis of their neurological symptoms, which are often attributed to spinal canal stenosis, spondylolisthesis, or disc herniation.[4,5] The optimal treatment for spinal dural AVFs is also controversial, with either endovascular therapy or surgery constituting the treatment of choice. The purpose of this letter is to comment on 2 fundamental aspects of treatment for spinal dural AVFs: 1) patients may present rapidly progressive neurological deterioration; in these cases, early occlusion of the fistula should be considered even if the patient already presents paraplegia (neurological improvement has been reported in most cases of early treatment); and 2) analysis of recent case series shows that, despite continuous improvements, endovascular treatment continues to be less effective than surgery; the latter achieves higher rates of complete dural AVF occlusion and is associated with a low morbidity rate.

Patients with spinal dural AVFs may display severe, rapidly progressive neurological deterioration characterised by lower limb paralysis and sphincter dysfunction, known as Foix-Alajouanine syndrome; the pathophysiology is yet to be fully understood.[3] The syndrome has traditionally been thought to be caused by irreversible necrotising myelopathy secondary to venous thrombosis. The high rates of improvement following treatment for spinal dural AVFs, even in patients with complete paraplegia, demonstrate that functional spinal cord alterations underlying severe neurological impairment may be due to decreased nervous tissue perfusion secondary to venous hypertension.[3,5,6] Severe, prolonged reduction of spinal cord perfusion pressure may lead to spinal cord infarction, which may explain why symptom duration, rather than the degree of neurological impairment at diagnosis, is the main prognostic factor in spinal dural AVFs.[3,4,7] Eight years ago, our research group published a study of the prognosis of 107 patients with spinal dural AVFs and paraplegia at the time of treatment. Symptoms improved after treatment in approximately 75% of patients. However, improvements were limited in most cases, with fewer than 6% of patients being able to walk without assistance.[3] Lack of complete recovery in most of these patients was most likely due to diagnostic delays: mean time from symptom onset to definitive diagnosis was 20 months.[3] In our study, paraplegia duration was less than 24 hours in approximately two-thirds of patients with paraplegia secondary to a spinal dural AVF and showing neurological improvement (from an Aminoff-Logue scale score for gait disturbance of 5 [wheelchair-bound] to 1 [leg weakness but no restriction of activity]) after treatment. In the remaining patients displaying such a marked clinical improvement, the mean time of progression of neurological deficits was shorter than 2 months.[3] According to other series, patients with longer symptom progression times are less likely to display significant improvements after surgery.[4,7] Spinal dural AVFs should therefore be considered in the differential diagnosis of patients with rapidly progressive lower limb weakness, paraesthesia, and/or sphincter dysfunction of undetermined origin, particularly in elderly patients displaying unusually fast symptom progression.

Regarding the optimal treatment for spinal dural AVFs, advances in embolisation techniques, with the use of liquid agents that can be introduced into the draining vein, enable treatment of the fistula during diagnostic arteriography, and have led to increases in the rate of endovascular treatment in many centres. Training for young neurosurgeons in cerebrovascular disease and spinal vascular malformations is currently lacking; in Spain, this has led to a predominance of endovascular treatment over surgery, as demonstrated by the series of patients treated between 2012 and 2015 published by Ortega-Suero et al.[1] However, recent case series of spinal dural AVFs published by the most relevant international research groups show that surgery was the initial treatment for occlusion of the fistula in nearly 60% of patients (Table 1).[2,3,5,7–24] Despite the heterogeneity of these series, all show better angiographic and clinical outcomes in patients undergoing surgery than in those undergoing endovascular treatment. The percentage of complete, permanent obliteration of the spinal dural AVF in a single procedure was nearly 100% among patients undergoing surgery, compared to 61% in patients receiving endovascular treatment. Differences in clinical outcomes were more marked: nearly 80% of surgery patients presented clinical improvements, compared to only 63% of those undergoing endovascular treatment. Furthermore, some series report poorer clinical prognosis in patients undergoing surgery following failure of endovascular treatment, whether due to incomplete occlusion of the fistula or to recanalisation.[14,18]

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Table 1 Series of patients with spinal dural arteriovenous fistulas published in the past 20 years.[a]

Author, year	No. patients	No. patients with paraplegia	Mean time to diagnosis (months)	No. acute cases	Initial treatment		Complete obliteration with initial treatment (%)		Overall clinical improvement (%)		Morbidity (%)	
					Embolisation	Surgery	Embolisation	Surgery	Embolisation	Surgery	Embolisation	Surgery
Atkinson et al.[8] 2001	94	30	23	7	0	94	NA	99	NA	98	NA	4
Song et al.[9] 2001	30	8	29	NA	23	7	78	100	57		NA	NA
Van Dijk et al.[10] 2002	49	NA	27.6	5	44	5	25	97.2	92		4.5	2.7
Cenzato et al.[11] 2004	37	7	22	NA	13	24	77	100	78		0	0
Jellema et al.[12] 2004	44	13	13.8	NA	34	10	54	100	70		3	10
Steinmetz et al.[13] 2004	19	5	19.2	1	0	19	NA	100	NA	44	NA	5.2
Andres et al.[14] 2008	21	6	NA	NA	17	4	58	100	65	100	0	0
Cecchi et al.[15] 2008	25	7	18	1	4	21	0	100	40		0	9
Narvid et al.[16] 2008	69	46	19	NA	39	24	69	80	65	50	0	0
Park et al.[17] 2008	18	2	14.4	1	17	1	76.5	100	82.3	100	6	0
Prieto et al.[3] 2009	107	107	20.4	5	25	82	58	97	73		NA	NA
Saladino et al.[5] 2010	154	8	24.7	20	0	154	NA	95	NA	82	NA	3
Ruiz-Juretschke et al.[2] 2011	19	3	12	2	9	10	55.6	100	44	70	16	
Clark et al.[18] 2013	23	0	12	2	15	8	46	90	18	70	NA	NA
Cho et al.[19] 2013	32	21	5	0	27	5	85	100	50		18	20
Gemmete et al.[20] 2013	33	NA	24.6	NA	29	4	82	100	45		3.4	0
Kirsch et al.[21] 2013	78	NA	13.9	NA	61	17	72	100	73.6		1.6	0
Gokhale et al.[7] 2014	27	3	11	NA	10	17	70	100	81		10	11.7
Schuss et al.[22] 2015	29	NA	21	4	0	29	NA	100	NA	76	NA	0
Gross et al.[23] 2017	71	0	NA	NA	28	42	50	100	80	80	11	11
Koch et al.[24] 2017	34	3	NA	16	20	14	65	100	85	93	15	14
Total	1013	269	19	64	406	581	61	98	63	78	6	5

NA: not available.

[a] We only included series with more than 15 patients.

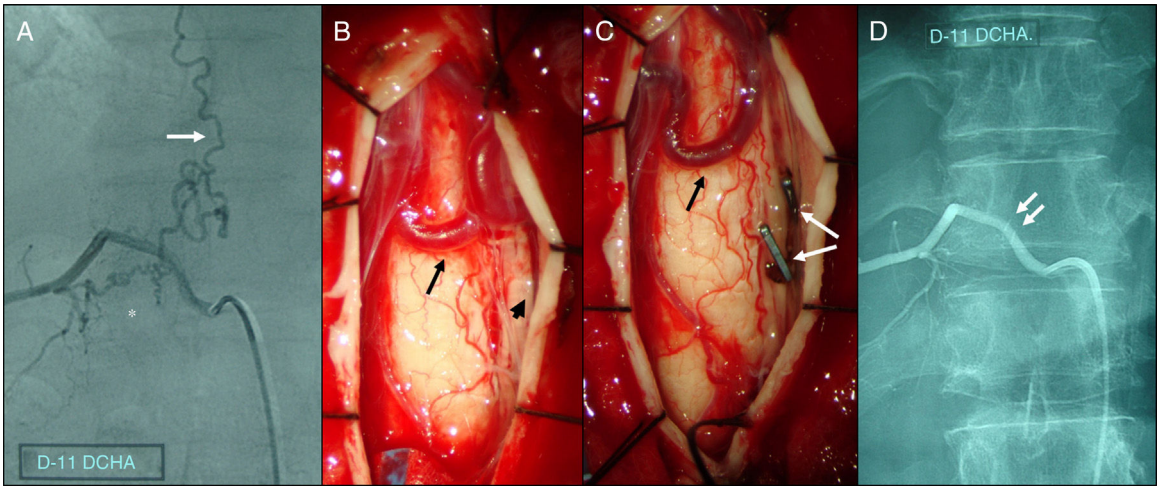

Figure 1 Spinal angiography and intraoperative photographs of a spinal dural arteriovenous fistula affecting the right radicular artery at the level of T11. (A) Selective angiography showing a fistula below the right pedicle of T11 (asterisk), connecting the radiculomeningeal artery and a varicose perimedullary vein (white arrow). (B) Intraoperative photograph following T11 laminectomy and durotomy, showing arterialisation of tortuous perimedullary veins (black arrow), in close contact with the nerve root (black arrowhead). (C) Clipping (white arrows), coagulation, and excision of the draining vein at the level of the nerve root results in immediate collapse and darkening of perimedullary veins (black arrow). (D) Postoperative spinal angiography showing complete occlusion of the fistula; the arrows indicate the vascular clips.

Microsurgical occlusion of spinal dural AVFs is associated with a morbidity rate of approximately 5%, similarly to that observed for endovascular treatment. However, embolisation is associated with more severe complications, which often irreversibly affect neurological function. Introducing embolic material through a catheter should not be considered non-invasive, since it is associated with a considerable risk of migration of embolic material into the venous system and/or radicular artery rupture, potentially leading to such severe outcomes as spinal cord infarction, causing permanent, irreversible motor function loss in the areas supplied by nerves emerging from below the level of the infarction.[7,19,21,23] Furthermore, patients undergoing embolisation are exposed to a radiation dose above 5 Gy.[24] In contrast, the most frequent complications of surgery include cerebrospinal fluid fistulas and pseudomeningocele, which are never associated with permanent neurological deficits. Surgical treatment of spinal dural AVFs consists of disconnection between the dural artery and the intradural draining vein; this technique is relatively easy to perform when the lesion has been correctly diagnosed and located intraoperatively (Fig. 1). The main technical difficulties for less experienced surgeons are determining the precise location of the lesion during the procedure and confirming complete occlusion of the fistula. Both problems have partially been solved with the introduction of indocyanine green, a fluorescent contrast agent enabling the differentiation of arterial from venous blood flow in spinal dural AVFs. Based on the above, we may conclude that microsurgical occlusion of the fistula continues to be the safest and most effective treatment for spinal dural AVFs, regardless of recent advances in endovascular treatment, and should therefore be considered the first-line treatment for these patients. Embolisation should be considered in patients with severe comorbidities contraindicating surgery.

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Reply to the Letter to the Editor ''Spinal dural arteriovenous fistulas: early endovascular treatment or surgery?''☆

Réplica a la carta al editor «Fístulas arteriovenosas espinales durales: ¿tratamiento precoz endovascular o quirúrgico?»

Dear Editor:

We would like to thank the authors of ''Spinal dural arteriovenous fistulas: early endovascular treatment or surgery?'' for their comments on our original article ''Spinal arteriovenous fistulas in adults: management of a series of patients treated at a neurology department.''[1] Most cases of spinal arteriovenous fistulas (AVF) are associated with diagnostic difficulties and treatment delays, given that they frequently present insidiously.[2—4] We agree with the authors that early treatment is critical in any type of spinal AVF.[5] However severe disability may be at the time of diagnosis, symptoms are reversible.[1]

Treatment, whether with surgery or embolisation, aims to eliminate the abnormal connection and re-establish normal spinal cord perfusion and pressure. No treatment guidelines are currently available for spinal AVF. Most studies are case reports or case series including few patients. The consequence of this, as the authors point out, is that selecting the most suitable treatment continues to be controversial, except for those cases of spinal AVFs involving the anterior spinal artery or the artery of Adamkiewicz, where surgery is the treatment of choice due to the high risk of ischaemia associated with embolisation.

Endovascular treatment alone greatly depends on lesion subtype, the angioarchitecture of the fistula, and the type

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