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Surgical treatment for spinal dural arteriovenous fistulas: Outcome, complications and prognostic factors

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ABSTRACT

Background and purpose: Spinal dural arteriovenous fistulas (SDAVFs) are rare, acquired pathology and they inevitably lead to severe disability if untreated. The aim of this study is to present the outcome and complications, and to find factors that may affect the outcome after surgical treatment.

Methods: Seventeen consecutive patients (men – 14, women – 3, age: 41–79) were retrospectively analyzed. The patients presented with paraparesis (88%), bladder symptoms (71%) and/or sensory disturbances (65%). The fistula was found in the upper thoracic spine in 2 cases, in the lower thoracic (T7–Th12) in 11 cases, and in the lumbar spine in 4 cases. Microsurgical shunt interruption was performed in all, followed by epidural arteries coagulation in 12 cases.

Results: In the long term, improvement or achievement of a good stable condition was observed in 13 patients (76%), and no patient deteriorated. All 5 paraplegic patients improved by at least 1 grade in MCS. Satisfactory results (modified McCormick Scale grades I–II) were found in 10 patients (59%), and 15(88%) were independent. Postoperative complications occurred in 4 patients (24%), two of them (12%) required revision surgery for epidural hematoma. The success rate was 94%; one patient required revision surgery for recurrent SDAVF. Better neurological condition on admission ($p = 0.0098$) and age >60 years ($p = 0.0498$) were the factors associated with satisfactory outcome.

Conclusions: Microsurgical closing of a SDAVF brings good and stable results over time. Aggressive treatment should be attempted even in cases of total loss of spinal cord function. Neurological condition before surgery and age may influence the outcome.

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1. Introduction

Spinal dural arteriovenous fistulas (SDAVFs) classified by Spetzler as intradural dorsal arteriovenous fistulas [1] are the most common vascular malformation of the spinal cord. They are acquired and constitute approximately 80% of vascular malformations in the spinal canal [2]. SDAVFs are rare pathology with an incidence of 5–10 new cases per million per year. Most of them are located in the thoracolumbar region of the spine and they mostly occur in older and middle aged men. SDAVFs are rare in the sacral region (4%) and rarely may present as multi-level fistulas (2%) [1,2].

The essence of the disease is a direct connection between the meningeal arterial branch and the venous coronary plexus of the spinal cord. This causes a direct transmission of arterial blood pressure to the venous system which results in venous stasis in the spinal cord. This is the primary mechanism of spinal cord damage and may lead to irreversible symptoms [2–4]. SDAVFs are characterized by slow progressive symptoms which usually exacerbate with physical effort. Many SDAVFs remain underdiagnosed as the symptoms may be unspecific for along time [2,5].

Currently, microneurosurgical or endovascular methods may be employed for the treatment of SDAVFs [6,7]. Due to the rarity of these spinal vascular lesions, reports of their management and outcomes have been limited to case series and case reports. The purpose of this paper is to present the short-term and long-term outcome and postoperative complications, and to find factors that may affect the outcome on a series of 17 surgically treated SDAVFs.

2. Material and methods

2.1. *Patients, signs and symptoms*

A series of 17 consecutive patients surgically treated for SDAVFs was retrospectively analyzed. The study group consisted of 14 men (82%) and 3 women (18%) aged from 41 to 79 years (average – 61 y.o., median – 60 y.o.).

Progressive paraparesis was the most common initial symptom ($n = 10$; 59%). Other initial signs and symptoms were: pain in the lumbosacral region ($n = 3$; 18%), dysesthesia or paresthesia in the lower limbs ($n = 3$; 18%) and intermittent paraparesis related to physical effort ($n = 1$; 6%). On admission, the patients presented with paraparesis ($n = 15$; 88%), loss of sphincter control ($n = 12$; 71%) and sensory disturbances ($n = 11$; 65%). The symptoms intensified during effort in five cases (29%). According to the modified McCormick Scale (MCS) [8], 7 patients (41%) presented with good condition (MCS grades I–II) and 10 (59%) with significant deficit (MCS grades III–V, Table 2). The duration of symptomatic period ranged from 1 to 60 months (average – 18 months; median – 12 months; <6 months – 3 patients; 7–12 months – 7 patients; >12 months – 7 patients).

2.2. *Work-up*

All patients underwent preoperative magnetic resonance imaging (MRI) and digital subtraction angiography (DSA) with selective segmental artery catheterization. All patients had segmental intramedullary hyperintensive signal on the MRI T2-weighted images: 3 levels in 2 (12%) cases, 4 levels in 1 (6%) case, 5 levels in 3 (18%) cases, 6 levels in 4 (24%) cases, 7 levels in 2 (12%) cases, 8 levels in 1 (6%) case, 9 levels in 3 (18%) cases and the 10 levels in 1 (6%) case (mean – 6 levels).

In DSA, 16 (94%) SDAVFs presented the arterial supply from a single radiculomeningeal arterial branch of the corresponding segmental artery. In 1 case (6%), the fistula was supplied from multiple branches of the segmental artery. The fistula was found in the upper thoracic spine (Th1–T6) in 2 cases, in the lower thoracic (T7–Th12) in 11 cases, and in the lumbar spine in 4 cases (Table 1).

In one patient, after the failed attempt of endovascular treatment in another center, a closed initial section of the right segmental artery at the Th12/Th11 level was demonstrated in DSA. The fistula was supplied by the collateral circulation from numerous small arteries from the Th11 segmental artery on the left side.

2.3. *Method of outcome evaluation*

The modified McCormick Scale was used to assess the neurological status [8], and grades I–II were considered as satisfactory, while grades III–V as unsatisfactory outcome. Relative evaluation of the outcome was performed compared to the preoperative condition, according to three possibilities: 1, "Improved" or "good stable neurological condition"; 2, "neurological deficit unchanged"; 3, "deterioration". "Good stable neurological condition" was defined as the absence of neurological deficit before and after surgery (MCS Grade I).

The patients were invited for a check-up outpatient visit within 2–6 months after surgery. We used data from the last recorded outpatient visit or data obtained in a telephone interview to evaluate the long-term outcome. The follow-up period ranged from 6 to 200 months (average 7 years) and the data were available for all patients. All patients underwent follow-up MRI at least 6 months after surgery. The spinal cord and venous coronary plexus were evaluated on T2-weighted MRI images. Follow-up DSA was performed only in doubtful cases, i.e., in patients with neurological deterioration or with persistent features of SDAVF in an MRI study.

Statistical analysis was conducted to find the factors which may affect long-term outcome and the evolution of a neurological condition. The following factors were taken into account: age (<60 y.o. vs. >60 y.o.), sex, neurological status before surgery (MCS grades I–II vs. MCS grades III–V), symptom

Table 1 – The levels of SDAVFs occurrence.

Spinal level	Number	%
Th2/Th3	1	12
Th5/Th6	1	
Th7/Th8	1	65
Th8/Th9	1	
Th9/Th10	2	
Th11/Th12	3	
Th12/L1	4	
L1/L2	3	24
L4/L5	1	

Table 2 – Neurological status of patients before surgery, at discharge, and in long-term follow-up according to the modified McCormick Scale.

The modified McCormick scale [8]		Neurological condition in different periods		
Grade	Description	At admission Number (%)	At short term follow-up Number (%)	At long-term follow-up Number (%)
I	Intact neurologically, normal ambulation, minimal dysesthesia	2 (12%)	3 (18%)	5 (29%)
II	Mild motor or sensory deficit, functional independence	5 (29%)	6 (35%)	5 (29%)
III	Moderate deficit, limitation of function, independent w/external aid	3 (18%)	3 (18%)	5 (29%)
IV	Severe motor or sensory deficit, limited function, dependent	2 (12%)	5 (29%)	2 (12%)
V	Paraplegia or quadriplegia, even w/flickering movement	5 (29%)	0	0

duration (<1 year vs. >1 year), the fistula level (thoracic vs. lumbar spine), perioperative use of methylprednisolone and coagulation of extradural arteries in the fistula area. Fisher's exact test was used to evaluate significance and a p-value < 0.05 was taken as statistically significant.

3. Results

3.1. *Treatment*

All patients underwent microsurgical treatment. Seven patients (41%) received a perioperative course of methylprednisolone according to the following scheme: an intravenous bolus of 30 mg/kg over at least 30 min was administered preoperatively, then an intravenous infusion at a dose of 5.4 mg/kg for 23 h. The operation was performed under general anesthesia in the prone position. The appropriate spinal level was determined using C-arm. Hemilaminectomy was used in 6 patients (35%): 1 level in 3 (18%), 3 levels in 2 (12%), and 4 levels in 1 (6%). Laminectomy was performed in 11 patients (65%): 1 level in 2 (12%), 2 levels in 7 (41%), and 3 levels in 2 (12%). The fistula was identified after dural incision, tracing the course of the outflow vein to the place where it pierced the dura, usually close to the nerve root sleeve. Then the outflow vein was clipped just behind the fistula location. After a few minutes, when the arterialized veins on the spinal cord surface collapsed and changed their color to dark red, the vein was coagulated and disconnected near the fistula. In 12 cases (71%), epidural small arteries reaching the fistula were also coagulated as an adjunctive. This maneuver was abandoned in 5 patients (29%) because of the fistula location and the higher risk of Adamkiewicz artery occlusion. Intraoperative neuromonitoring (IOM) was used at the beginning of the analyzed series in 4 patients (24%). The shorter latency and higher amplitude of responses was noted after fistula closure in all of these patients. The dura was sutured without additional sealing materials in 7 patients (41%). In 8 cases (47%) a Tachosil® (Takeda) was used additionally, in 1 case (6%) fibrin glue Tissucol® (Baxter) was used, and in 1 case (6%) Tachosil® and Tissucol® were used simultaneously. The wound was closed in standard fashion without drainage. Physical rehabilitation was routinely started from the first postoperative day.

3.2. *Short-term and long-term results of surgery*

Improvement or good stable neurological condition was observed in 11 patients (65%) and no change in 6 patients (35%) at discharge from hospital. No patient deteriorated significantly when evaluated at discharge, however, transient deterioration occurred in 3 patients (see: Postoperative complications). Satisfactory short-term outcome was achieved in 9 patients (53%) and unsatisfactory in 8 patients (47%). Most patients ($n = 14$; 82%) were discharged home with the recommendation of rehabilitation and 3 patients (18%) were transferred to a rehabilitation ward.

In the long term, improvement or good stable condition was observed in 13 patients (76%), stable neurological status in 4 patients (24%) and no patient deteriorated as compared to the preoperative condition. Neurological improvement by at least one MCS grade was observed in 11 patients (73%) of the 15 patients with neurological deficit on admission. Noteworthy is that all paraplegic patients improved by at least 1 grade in MCS. Satisfactory long-term results (MCS grades I–II) were found in 10 patients (59%), and unsatisfactory in 7 patients (41%).

The patient who had had prior failed endovascular treatment improved slightly after surgery in the long term; mild paraparesis still remained (MCS grade II).

Follow-up MRI demonstrated the disappearance of both spinal cord edema and enlarged veins on its surface in all but one patient ($n = 16$; 94%). Four patients (24%) underwent postoperative DSA. The reasons for this examination were: transient postoperative paraparesis in 2 patients, periodic difficulties in walking 20 months after surgery in 1 patient, and persistence of SDAVF features in routine follow-up MRI in 1 patient. The fourth patient presented stable neurological condition postsurgery, however, DSA confirmed the SDAVF recurrence only in this one case (6%, see: Treatment failure).

3.3. *Postoperative complications and treatment failure*

Postoperative complications. Postoperative complications occurred in 4 patients (24%) and two of these patients (12%) required reoperation because of epidural hematoma (EDH). In the first case, the worsening of bladder and motor function from MCS grade II to grade IV appeared on the 4th postoperative day. During revision surgery, EDH was evacuated and the patient quickly improved. Unfortunately, 6 days after the second surgery, an external cerebrospinal fluid (CSF) leak appeared and needed surgical treatment. In long-term follow-up, the patient achieved significant improvement (MCS grade II).

The second patient presented with severe deficit before surgery (MCS grade IV). After surgery, the patient achieved improvement to MCS grade III, but deteriorated again to pre-surgery status (MCS grade IV) on the 4th postoperative day. EDH was urgently evacuated. After several days of temporary improvement, re-aggravation of symptoms occurred. MRI revealed a restriction of the subarachnoid space at the surgery level, while DSA confirmed the fistula closure (Fig. 1). During the third surgery, duroplasty was performed to expand the dural sac. The postsurgery period was complicated with a CSF leak, which was treated by lumbar drainage. In the long term, the patient achieved an improvement; she can walk for short distances (MCS grade III).

The remaining 2 complications were treated conservatively: (1) pneumonia, (2) transient worsening of motor function related to a small EDH. This last patient achieved a good functional outcome (MCS grade I) in the long term.

3.4. *Treatment failure.*

One patient (6%) required revision surgery for recurrent fistula. Preoperative DSA showed SDAVF on the Th11/Th12 level. The fistula was disconnected in a standard fashion, but the epidural arteries were not coagulated. After the surgery, bladder and motor function improved (MCS grade II), and this condition was stable in follow-up. The scheduled MRI, performed 1 year after surgery, found indirect features of SDAVF and fistula recurrence was confirmed by DSA. During the second surgery, two new outflow veins of the fistula located at the same level were found next to the clip previously left. The veins were disconnected with additional coagulation of extradural arteries around the fistula. After the operation, further improvement of the neurological condition was observed (MCS grade I).

3.5. *Factors that may affect the outcome*

An analysis of factors that may affect the long-term outcome and the evolution of the functional status is presented in Tables 3 and 4. Good neurological condition before surgery and age >60 years were the factors significantly associated with better functional outcome. We failed to find factors significantly related with the functional status evolution after surgery. We also found no significant relationship between the functional outcome and longer symptom duration. Satisfactory long-term results were found in 7 (64%) out 11 patients with symptom duration ≤2 years and in 3 (50%) out 6 patients with longer symptomatic period ($p = 1$).

4. Discussion

Spinal vascular malformations constitute about 3–4% of all intradural spinal lesions and 5–9% of all vascular malformations of the central nervous system [9]. The most common of these are spinal dural arteriovenous fistulas which constitute approximately 80% of all vascular malformations within the spinal canal [10]. The etiology is not clearly understood, but it is widely accepted that they are acquired and they are most common in middle-aged and older men [7,9,10]. Untreated SDAVF inevitably leads to severe disability [7,9–11].

4.1. *Microsurgical treatment*

The first attempts of surgical treatment of spinal vascular malformations have been reported from 1910, when Fedor Krause intraoperatively found a vascular malformation of the spinal cord. A few years later, Charles Ellsberg was the first to attempt the surgical excision of a spinal vascular malformation [9,12,13]. Kendall and Logue described the pathophysiology of SDAVF in 1977. They found that the expanded veins of the coronary plexus are normal veins but dilated as a result of diversion of arterial blood from the dural fistula [12,14,15]. Since then, the surgical procedure has not changed significantly. The idea is that the fistula must be disconnected. However, they proposed additional excision of the dura mater to avoid the recurrence of the fistula [16,17]. Simple intradural disconnection of the fistula is now a widely used surgical technique, together with additional coagulation of the arteries on the external surface of the dura. It is only near the Adamkiewicz artery that this should not be performed [3,9,18,19]. In recent years, a trend toward minimizing the surgical approach has appeared, e.g., the microscope-assisted endoscopic interlaminar approach or the hemilaminectomy approach using a tubular retractor [20,21].

Currently, the use of IOM is suggested by some centers as a useful tool for monitoring neurological status after closing the fistula [9,22]. At the beginning of our series, IOM was used in 4 patients (25%). The responses of motor and somatosensory evoked potentials improved quickly in all patients after the closure of the fistula. This group is too small to draw definite conclusions. However, we do not currently use IOM, because in our experience it does not offer any benefit for this procedure.

The use of methylprednisolone in our series was not routine and was dependent on the severity of the neurological deficit before surgery. In addition, the indications for this management have been changing in recent years. In our series, we found no significant association between the use of methylprednisolone and outcomes. In the available literature, there is no data on the rationale for the use of steroids in the perioperative period. However, a few cases of rapid deterioration after steroid administration in patients with SDAVF were reported [23,24].

Endovascular methods are an alternative to surgery. However, they are associated with a higher risk of fistula recurrence and occlusion of the radiculomeningeal arterial branch involved in the vascularization of the spinal cord [3]. The difficulties in reaching the exact site of the fistula by microcatheters and the poor penetration of embolic agents into the fistula are also reported [25–28]. It seems that the microsurgical method currently offers greater efficacy (success rate: 80–98%), as compared to endovascular procedures (success rate: 46–72%) [19,27,28].

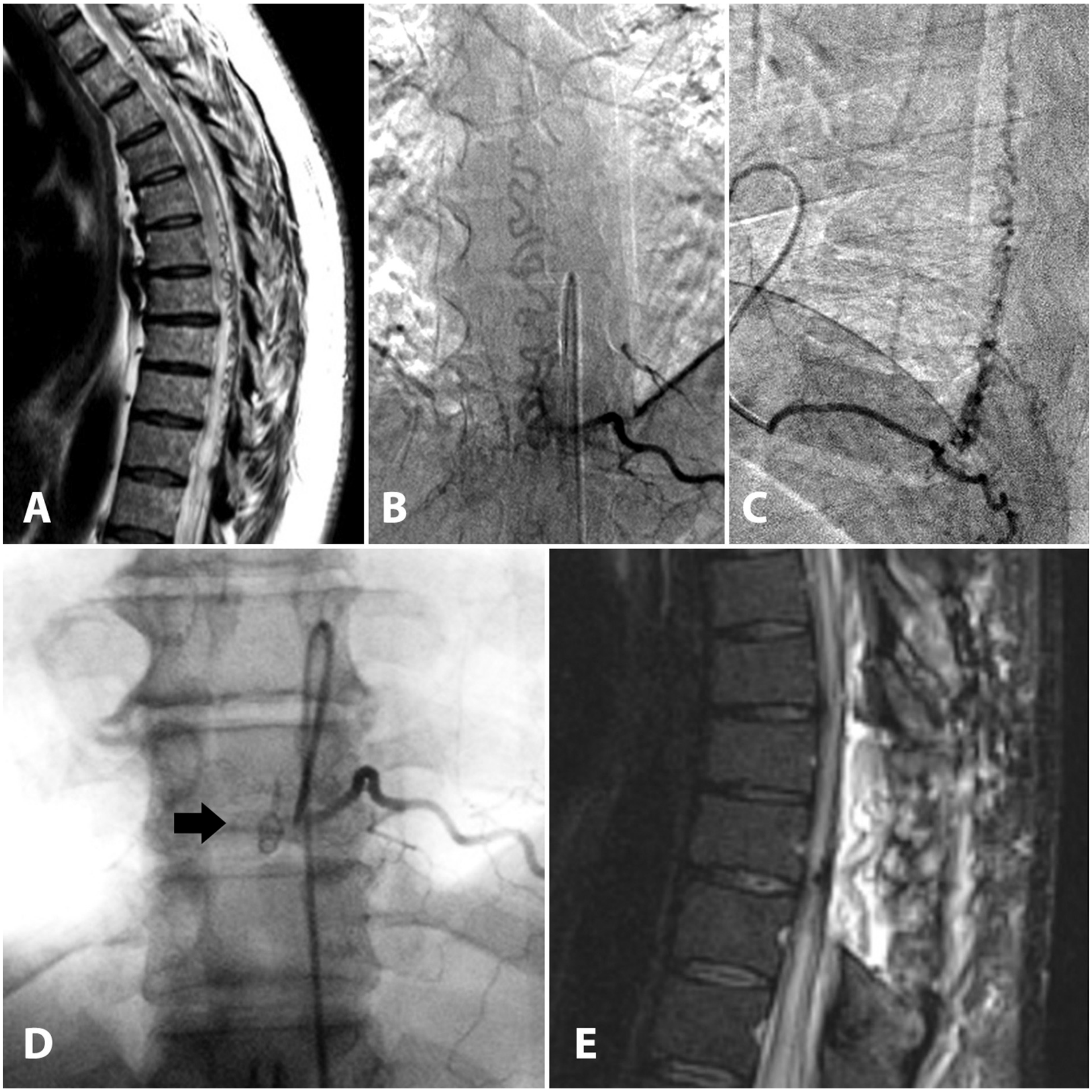

Fig. 1 – Preoperative magnetic resonance imaging (MRI) and digital subtraction angiography (DSA) with selective segmental artery catheterization. (A) T2-weighted sagittal magnetic resonance imaging (MRI) showed the hyperintensive intramedullary signal in the thoracic spinal cord and conus medullaris (myelopathy). Enlarged perimedullary veins are present along the dorsal surface of the spinal cord. (B) DSA in the anterior–posterior projection (a–p) of the left T9 radicular artery showing draining vein and dilated, tortuous, perimedullary veins. (C) DSA the same artery in the lateral projection. (D) Postoperative DSA in a–p projection confirmed the fistula closure. The clip after the fistula closure is visible in the spinal canal (arrow). (E) T2-weighted MRI after the second operation revealed the restriction of cerebrospinal fluid layer around the spinal cord at the surgery level.

4.2. *Short-term and long-term results*

The functional results of surgery for SDAVF are acceptable and the risk of fistula recurrence is low. Saladino et al. presented a series of 154 patients with an average age of 64 years, with a 3.5:1 male to female ratio, with a predilection for the occurrence of fistulas in the thoracic region. Among the 141 patients with available long-term follow-up, [23] improvement in motor function was observed in 82.2%, and stable condition in 14.4% [19]. Cenzato et al. presented data from three centers on 65 patients, of which 10 were treated endovascularly and 55 surgically. In short-term follow-up (6 months), 80% of the patients showed functional improvement (mainly motor function – 63%) and 69% of the patients improved in the long term [2]. In our series, functional improvement or good stable condition was achieved in 65% of the patients at discharge and in 76% in long-term follow-up. The demographic profile and the location of the fistulas were similar to the other series.

Table 3 – Analysis of factors that might influence long-term outcome.

Factor		Long-term outcome (MCS)		p-value (the Fisher Exact test)
		Grades I–II	Grades III–V	
Age	≤60 y.o.	3 (32%)	6 (67%)	$p = 0.0498$
	>60 y.o	7 (88%)	1 (13%)	
Gender	Female	1 (32%)	2 (67%)	$p = 0.5368$
	Male	9 (63%)	5 (36%)	
Neurological condition at admission (MCS)	Grades I–II	7 (100%)	0 (0%)	$p = 0.0098$
	Grades III–IV	3 (30%)	7 (70%)	
Symptomatic period	≤1 year	6 (60%)	4 (40%)	$p = 1.0000$
	>1 year	4 (56%)	3 (43%)	
SDAVF location	Thoracic spine	7 (54%)	6 (45%)	$p = 0.6221$
	Lumbar spine	3 (75%)	1 (25%)	
Perioperative use of methylprednisolonum	/+/	1 (14%)	6 (86%)	$p = 0.133998$
	/−/	6 (60%)	4 (40%)	
Epidural vessels coagulation	/+/	5 (45%)	6 (55%)	$p = 1$
	/−/	2 (33%)	4 (67%)	

MCS = modified McCormick Scale, SDAVF = spinal dural arteriovenous fistula.

Table 4 – Analysis of factors that might influence the evolution of neurological condition from admission to the long term.

Factor		Long-term outcome (MCS)		p value (the Fisher Exact test)
		Improvement or stable good neurological condition	Stable neurological deficit or deterioration	
Age	≤60 y.o.	6 (67%)	3 (33%)	$p = 0.576471$
	>60 y.o	7(88%)	1 (13%)	
Gender	Female	3 (100%)	0 (0%)	$p = 0.541176$
	Male	10 (71%)	4 (29%)	
Neurological condition at admission (MCS)	Grades I–II	6 (86%)	1 (14%)	$p = 0.60294$
	Grades III–IV	7 (70%)	3 (30%)	
Symptomatic period	≤1 year	6 (60%)	4 (40%)	$p = 0.10294$
	>1 year	7 (100%)	0 (0%)	
SDAVF location	Thoracic spine	9 (69%)	4 (31%)	$p = 0.51932$
	Lumbar spine	4 (100%)	0 (0%)	
Perioperative use of methylprednisolonum	/+/	5 (71%)	2 (29%)	$p = 1$
	/−/	8 (80%)	2 (20%)	
Epidural vessels coagulation	/+/	8 (73%)	3 (27%)	$p = 1$
	/−/	5 (83%)	1 (17%)	

MCS = modified McCormick Scale, SDAVF = spinal dural arteriovenous fistula.

4.3. *Complications of surgery*

Postoperative complications occurred in 4 patients (24%) in our series, including 3 patients (18%) with transient worsening of neurologic status. Complications included: EDH ($n = 3$), CSF leak ($n = 2$) and pneumonia ($n = 1$). Saladino et al. reported functional deterioration or a new deficit in 16% (including permanent deterioration in 2.6%), gait deterioration in 0.6%, abnormal urination in 1.9%, surgical wound infection in 1.3% and deep vein thrombosis in 1.9% [19]. Steinmetz et al. observed pseudomeningocele in one case (5%) [28]. Ropper et al. in a series of 15 patients reported transient paresis in 13% and epidural hematoma in 7% [14].

4.4. *Treatment failure*

The success rate in our series was 94%, with only 1 patient requiring revision surgery for recurrent SDAVF. In the series of Saladino et al., two patients (1.3%) with neurological deficit required reoperation because of incomplete disconnection of the fistula [19]. A comparative analysis of MRI before and after treatment was carried out by Kaufman et al. on a group of 34 patients [29]. The authors noted that MRI does not always correlate with clinical status. Follow-up DSA was performed on 10 patients (29%) due to their clinical condition and 3 of them (9%) had recurrent fistula. However, only 2 patients presented subtle changes in the MR imaging of the spinal cord. The authors suggest that DSA should be performed in any clinically doubtful case, even if MRI reveals no pathological changes.

4.5. *Factors that might influence the outcome*

We failed to find factors that would influence the evolution of the neurological condition after surgery (Table 4). However, we found that better neurological condition on admission and older age are factors that are associated with better outcome in the long term. It is not surprising that all patients with good condition and only 30% of the patients with significant deficit

(MCS grades III–IV) before surgery achieved satisfactory long-term functional result (MCS grades I–II, $p = 0.0098$). Apparently, this is due to irreversible damage to the spinal cord before surgery in patients with severe deficits [2]. On the other hand, it must be emphasized that all paraplegic patients improved by at least 1 grade in MCS. Therefore, aggressive treatment should be attempted even in cases of total loss of spinal cord function. Nevertheless, it would be interesting to know why patients over the age of 60 years achieved better results than their younger counterparts (MCS grades I–II, 88% vs. 32%, $p = 0.0498$). The borderline statistical results and the low number of patients may suggest that this is only an accidental finding that needs to be confirmed in a study with a larger series (this implies that this finding might disappear as the size of the series increases). Furthermore, 63% (5/8) of older patients, and only 22% (2/9, p – non significant) of younger patients presented with good preoperative condition (MCS grades I–II) in our series. On the other hand, it is also possible that the aging process in the spinal cord may help to tolerate some pathological processes as occurs in the aging brain, e.g., in case of chronic subdural hematoma [30,31]. However, Cenzato et al. observed faster recovery after surgery in younger patients, but the difference was not statistically significant [2].

We found no significant relations between the functional outcome and the following factors: gender, symptomatic period, level of SDAVF, steroid neuroprotection and epidural vessel coagulation. However, Cenzato et al. observed a relationship between the level of the fistula and the outcomes [2]. The authors obtained better results when the fistula was located in the lower thoracic spine and they explained that this was as a result of better anatomical vascularization of this region of the spinal cord by the Adamkiewicz artery.

5. Conclusions

Intradural, microsurgical closing SDAVF brings good and stable results over time. All paraplegic patients improved by at least 1 grade in MCS after surgery, therefore, aggressive treatment should be attempted even in cases of total loss of spinal cord function. The only case of SDAVF recurrence after disconnection alone indicates that the additional coagulation of the epidural vessels is reasonable.

Disclosure

The authors have no personal financial or institutional interest in any of the drugs, materials, or devices described in this article.

Conflict of interest

None declared.

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