Check for updates

Spinal Dural Arteriovenous Fistulas: Clinical Results and Quality of Life Assessment with Surgical Treatment as a Crucial Therapy. The Joint Experience of Two Centers

Pietro Fiaschi[1], Alessandro Prior[1], Pier Filippo Sbaffi[1], Fabiola Bizzi[2], Alessandro D'Andrea[1], Bernarda Cagetti[3], Filippo Badaloni[3], Luca Allegretti[5], Nicola Mavilio[5], Riccardo Padolecchia[4], Valtero Valsania[3], Gianluigi Zona[1]

■ OBJECTIVE: **Dorsal intradural arteriovenous fistulas (AVFs) consist of a direct connection between a radicular feeding artery and the coronal venous plexus; this direct connection leads to arterialization of the venous plexus, venous congestion, and myelopathy. Controversy still exists regarding the best treatment modality of spinal dural AVFs. Surgical disconnection of spinal dural AVFs is a straightforward procedure with a high success rate and virtually no risk of recurrence or incomplete treatment.**

To identify factors associated with the clinical progression of dorsal intradural AVFs and quantify the range of surgical outcomes in terms of neurologic improvement as well as patients' perception of quality of life (QOL).

■ METHODS: **A retrospective observational study of 19 consecutive patients treated with surgery over a 10-year period was carried out. We analyzed surgical results and clinical outcomes. We also evaluated the impact of this disease and its sequelae on the patients' postoperative health-related QOL.**

■ RESULTS: **The surgical procedure showed good results in terms of neurologic improvement as well as patients' perception of QOL.**

■ CONCLUSIONS: **Our series confirmed that surgical obliteration of dorsal intradural AVFs is an effective and safe procedure. The results of this retrospective analysis make us believe that surgery, given its low morbidity and high success rate, represents a safe and effective first therapeutic option for these spinal vascular malformations. It could be considered to avoid unsuccessful endovascular attempts that could delay the definitive treatment of this disease. The surgical procedure showed good results in terms of neurologic improvement as well as patients' perception of QOL.**

INTRODUCTION

Spinal dural arteriovenous fistulas (SDAVFs),[1] previously referred to as type I dural arteriovenous fistulas (AVFs), are acquired spinal vascular malformations, representing 80% of all spinal vascular malformations. They consist of a direct connection between a radicular feeding artery and the coronal venous plexus. The formation of the fistula leads to arterialization of the venous plexus and venous congestion, which can result in progressive myelopathy.

Disconnection or obliteration of the fistula is necessary to interrupt this pathologic process, giving a chance for recovery to the spinal cord. This process can be accomplished either surgically or endovascularly. It is still debated which is the better treatment; some investigators[2,3] favor endovascular treatment because of the minor invasiveness, claiming a good rate of successful treatment of AVFs, whereas other investigators[4-6] recommend surgery because surgical disconnection is a straightforward procedure with a high success rate and virtually no risk of recurrence or incomplete treatment.

Key words
- Arteriovenous fistulas
- Fistula
- SDAVF
- SF-36
- Spinal vascular malformation
- Surgery quality of life

Abbreviations and Acronyms
ALS: Aminoff-Logue Scale
AVF: Arteriovenous fistula
MRI: Magnetic resonance imaging
mRS: Modified Rankin Scale
HRQOL: Health-related quality of life
SD: Standard deviation
SDAVF: Spinal dural arteriovenous fistula
SF-36: Short-Form 36

From the [1]Section of Neurosurgery, Department of Neurosciences (DINOGMI), San Martino University Hospital, and [2]Department of Educational Sciences (DISFOR), University of Genoa, Genoa; [3]Departments of Neurosurgery and [4]Neuroradiology, S. Corona Hospital, Pietra Ligure; and [5]Department of Diagnostic and Interventional Neuroradiology, San Martino University Hospital, Genoa, Italy

To whom correspondence should be addressed: Pietro Fiaschi, M.D.
[E-mail: pietro.fiaschi@alice.it]

Citation: World Neurosurg. (2019) 122:e270-e278.
https://doi.org/10.1016/j.wneu.2018.10.019

Journal homepage: www.journals.elsevier.com/world-neurosurgery

Available online: www.sciencedirect.com

Table 1. Patients' Data, Symptoms, and Anatomy

Patient Initials	Sex	Age (Years)	Time to Diagnosis (Months)	Initial Signs and Symptoms	Signs and Symptoms at the Time of Diagnosis	Level of Spinal Dural Arteriovenous Fistula	Anatomy
A.D.	M	60	2	Bilateral leg pain	Bilateral leg pain and weakness, gait disturbances, hypertonia, sensory disturbances (L1 level of sensory loss), perineal dysesthesia, acute urinary retention, fecal incontinence	T7	Feeder: left D7 intercostal artery Drainage: perimedullary venous system D4-D9
B.U.	M	51	4	Mild right leg weakness	Right leg weakness and hypoesthesia, urinary retention, fecal incontinence	T5-T6	Feeder: left D5 and D6 intercostal artery Drainage: perimedullary venous system
B.F.	M	30	7	Bilateral leg weakness and gait disturbances	Severe bilateral leg weakness, paresthesias, and hypoesthesia	T6	Feeder: right D6 and D7 intercostal artery Drainage: perimedullary venous system
C.G.	F	79	12	Gait disturbances	Bilateral leg weakness and hypoesthesia, urinary retention, and incontinence	T3	Feeder: left supreme intercostal artery Drainage: perimedullary venous system
F.L.	M	73	12	Right leg pain	Bilateral leg weakness, paresthesias, and hypoesthesia; urinary incontinence	T9	Feeder: left T9 intercostal artery Drainage: perimedullary venous system caudally and cranially
G.G.	M	60	2	Bilateral leg weakness	Bilateral leg weakness and hypoesthesia; urinary retention	T6	Feeder: left T6 intercostal artery Drainage: perimedullary venous system caudally and cranially
R.M.	F	78	2	Moderate right leg weakness	Right leg weakness and hypoesthesia; bilateral leg paresthesias; perineal hypoesthesia, urinary retention, and fecal incontinence	L3	Feeder: right L3 lumbar artery Drainage: perimedullary venous system cranially
S.M.	M	69	1	Bilateral leg pain	Bilateral leg weakness; sensory disturbances (L1 level of sensory loss), fecal incontinence	T11	Feeder: right D11 intercostal artery Drainage: perimedullary venous system
T.A.	M	80	14	Right leg paresthesias	Bilateral leg weakness and paresthesias; gait disturbances	T6	Feeder: right T6 intercostal artery Drainage: perimedullary venous D4-D8
U.M.	F	61	5	Bilateral leg pain	Bilateral leg pain and weakness; sensory disturbances (T11 level of sensory loss); perineal dysesthesia; urinary retention	T6	Feeder: right D6 intercostal artery Drainage: perimedullary venous system cranially and caudally
P.I.	M	66	0,1	Acute low back and left leg pain, gait disturbances	Low back pain and left leg pain, severe bilateral leg weakness, sensory disturbances (D5 level of sensory loss) increased tendon jerks, clonus, Babinski	T4	Feeder: rightT4 intercostal artery Drainage: perimedullary venous system with 1 dilated spinal cord vein extending T4-T11
S.C.	F	75	2	Bilateral leg weakness with gait disturbances	Bilateral leg weakness and dysesthesias; gait disturbances; sensory disturbances T12 level of sensory loss)	T6	Feeder: right D6 intercostal artery Drainage: perimedullary venous system cranially and caudally
M.T.	M	68	6	Bilateral leg weakness	Bilateral leg weakness and urinary retention	T5	Feeder: right D5 intercostal artery Drainage: perimedullary venous system caudally
R.B.	M	63	1	Mild bilateral leg weakness	Mild bilateral leg weakness, perineal dysesthesia, occasional urinary retention	T11	Feeder: left D11 intercostal artery Drainage: perimedullary venous system caudally

M, male; F, female.

Continues

Table 1. Continued

Patient Initials	Sex	Age (Years)	Time to Diagnosis (Months)	Initial Signs and Symptoms	Signs and Symptoms at the Time of Diagnosis	Level of Spinal Dural Arteriovenous Fistula	Anatomy
S.I.	F	63	6	Bilateral leg pain	Bilateral leg pain and paresthesias, urinary incontinence	T10	Feeder: left D10 intercostal artery Drainage: perimedullary venous system cranially
E.A.	M	56	24	Gait disturbances	Bilateral leg weakness and paresthesias, gait disturbances, incontinence urinary	L1	Feeder: right D12 intercostal artery and L1 lumbar artery Drainage: perimedullary venous system cranially
M.A.	M	76	7	Bilateral leg weakness	Severe bilateral leg weakness and paresthesias, sensory disturbances (T10 level of sensory loss); urinary retention and fecal incontinence	T5	Feeder: right T5 intercostal artery Drainage: perimedullary venous system cranially and caudally
C.R.	M	61	2	Bilateral leg pain	Bilateral leg pain, weakness, and paresthesias	T8	Feeder: left T8 intercostal artery Drainage: perimedullary venous system cranially and caudally
P.F.	M	55	6	Occasional urinary retention and constipation	Low back and bilateral leg pain, gait disturbances, bilateral leg weakness, urinary retention, and constipation	T11	Feeder: left D11 intercostal artery Drainage: perimedullary venous system caudally

M, male; F, female.

In this study, we retrospectively analyzed clinical and surgical results in a series of 19 consecutive patients treated with surgery over a 10-year period. Moreover, we assessed the health-related quality of life (HRQOL) in our patients using the Short-Form 36 (SF-36) questionnaire to gain an insight into the impact of this rare but potentially life-changing disease.

METHODS

This retrospective observational cohort study was completed after approval from the research ethics board of our hospitals (Neurosurgical Department of Policlinico San Martino Hospital, Genoa, Italy and Neurosurgical Department of Santa Corona Hospital, Savona, Italy) and after obtaining informed patient consent.

Study Design

A retrospective chart review was performed to identify all cases of patients with SDAVFs at our institutions from April 2007 to March 2017. Patients with intracranial fistulas and retrograde venous drainage causing myelopathy were excluded. Similarly, patients with fistulas located in the region of the foramen magnum and the craniovertebral junction were excluded from the analysis because these lesions have clinical and angiographic characteristics different from those of classic SDAVFs. The medical records of these patients were retrospectively reviewed and clinical history, neurologic examination, and diagnostic workup were obtained. Information collected included patient demographics, clinical presentation, location of the fistula, months of symptoms experienced before diagnosis, treatment, results, retreatment, duration of follow-up, and functional status (preoperative and postoperative). Functional status was measured by use of the Aminoff-Logue Scale (ALS) and the degree of disability or dependence by use of the modified Rankin Scale (mRS), calculated retrospectively from the preoperative and postoperative clinical notes at recorded historic and physical examination. The time before diagnosis was calculated in months from the onset of initial symptoms to diagnosis obtained by means of angiography.

We retrospectively reviewed the records of 19 consecutive patients with SDAVF who were treated with open surgical modality. Recanalization, or recurrence, was defined as the presence of persistent arteriovenous shunting or AVF at the time of the first and/or last angiographic follow-up compared with the initial posttreatment angiogram.

Surgical Technique

All patients in the series underwent preoperative angiography. Each patient was collegially evaluated by an experienced neurosurgeon and neuroradiologist; the day before surgery, a radiopaque marker was placed percutaneously in the pedicle at the level of the fistula, using biplanar fluoroscopy.[7] Alternatively, a radiopaque microcoil was placed in the major feeding artery at the time of the preoperative spinal angiography; the microcoil was easily visualized performing intraoperative fluoroscopy. Preoperative placement of radiopaque markers has been proved to be a safe and effective technique in thoracic spine surgery, making intraoperative localization more reliable and thus avoiding time wasting.[8] In the operating room, after the confirmation of the correct level of interest, a standard 1-level or

Table 2. Diagnostic Features, Treatment, and Outcome

Patient Initials	Level of SDAVF	Anatomy	Spinal Cord MRI Features	Diagnostic Examinations	First Treatment	Resolution After First Treatment	Outcome	Postoperative Examinations and Follow-Up Length
A.D.	T7	Feeder: left D7 intercostal artery Drainage: perimedullary venous system D4-D9	T2 hyperintensity T6-T9	MRI and angiography	Surgery	Yes	Improved	Angiography at 4 months: SDAVF occlusion Follow-up length, 38 months
B.U.	T5-T6	Feeder: left D5 and D6 intercostal artery Drainage: perimedullary venous system	T2 hyperintensity T9-T11	MRI and angiography	Surgery	Yes	Improved	Angiography at 4 months: SDAVF occlusion Follow-up length, 6 months
B.F.	T6	Feeder: right D6 and D7 intercostal artery Drainage: perimedullary venous system	T2 hyperintensity and enlargement T9-conus	MRI and angiography	Surgery	Yes	Improved	Angiography at 6 months: SDAVF occlusion Follow-up length, 26 months
C.G.	T3	Feeder: left supreme intercostal artery Drainage: perimedullary venous system	T2 hyperintensity C6-T9	MRI and angiography	Surgery	Yes	Improved	Angiography at 4 months: SDAVF occlusion Follow-up length, 31 months
F.L.	T9	Feeder: left T9 intercostal artery Drainage: perimedullary venous system caudally and cranially	T2 hyperintensity with enlargement T2-T10	MRI and angiography	Surgery	Yes	Improved	Angiography at 3 months: SDAVF occlusion Follow-up length, 54 months
G.G.	T6	Feeder: left T6 intercostal artery Drainage: perimedullary venous system caudally and cranially	T2 hyperintensity T2-conus	MRI and angiography	Surgery	Yes	Improved	Angiography at 4 months: SDAVF occlusion Follow-up length, 7 months
R.M.	L3	Feeder: right L3 lumbar artery Drainage: perimedullary venous system cranially	T2 hyperintensity T10-conus	MRI and angiography	Surgery	Yes	Stable	Angiography at 4 months: SDAVF occlusion Follow-up length, 24 months
S.M.	T11	Feeder: right D11 intercostal artery Drainage: perimedullary venous system	T2 hyperintensity T10-conus	MRI and angiography	Surgery	Yes	Improved	Angiography at 3 months: SDAVF occlusion Follow-up length, 24 months
T.A.	T6	Feeder: right T6 intercostal artery Drainage: perimedullary venous D4-D8	T2 hyperintensity T7-T12	MRI and angiography	Surgery	Yes	Improved	Angiography at 2 weeks: SDAVF occlusion Follow-up length, 84 months
U.M.	T6	Feeder: right D6 intercostal artery Drainage: perimedullary venous system cranially and caudally	T2 hyperintensity at conus	2 MRI (the first low-field MRI showed only multilevel discopathy) and angiography	Surgery	Yes	Improved	Angiography at 3 months: SDAVF occlusion Follow-up length, 62 months
P.I.	T4	Feeder: rightT4 intercostal artery Drainage: perimedullary venous system with one dilated spinal cord vein extending T4-T11	T2 hyperintensity T2-T7	MRI and angiography	Surgery	Yes	Improved	Angiography at 10 months: SDAVF occlusion Follow-up length, 60 months
S.C.	T6	Feeder: right D6 intercostal artery Drainage: perimedullary venous system cranially and caudally	T2 hyperintensity T10 -L1	MRI and angiography	Surgery	Yes	Improved	Angiography at 3 months: SDAVF occlusion Follow-up length, 74 months
M.T.	T5	Feeder: right D5 intercostal artery Drainage: perimedullary venous system caudally	T2 hyperintensity T5-L1	MRI and angiography	2 unsuccessful endovascular treatments before surgery	Yes	Improved	Angiography at 3 weeks: SDAVF occlusion Follow-up length, 19 months

SADVF, spinal dural arteriovenous fistula; MRI, magnetic resonance imaging.

Continues

Table 2. Continued

Patient Initials	Level of SDAVF	Anatomy	Spinal Cord MRI Features	Diagnostic Examinations	First Treatment	Resolution After First Treatment	Outcome	Postoperative Examinations and Follow-Up Length
R.B.	T11	Feeder: left D11 intercostal artery Drainage: perimedullary venous system caudally	T2 hyperintensity T10-L1	MRI and angiography	Surgery	Yes	Improved	Angiography at 3 months: SDAVF occlusion Follow-up length, 55 months
S.I.	T10	Feeder: left D10 intercostal artery Drainage: perimedullary venous system cranially	T2 hyperintensity T9-conus	MRI and angiography	Surgery (performed transforaminal lumbar interbody fusion a few months before because of misdiagnosis)	Yes	Improved	Angiography at 2 months: SDAVF occlusion Follow-up length, 8 months
E.A.	L1	Feeder: right D12 intercostal artery and L1 lumbar artery Drainage: perimedullary venous system cranially	T2 hyperintensity T7-L1	MRI and angiography	Surgery	Yes	Improved	Angiography at 2 weeks: SDAVF occlusion Follow-up length, 52 months
M.A.	T5	Feeder: right T5 intercostal artery Drainage: perimedullary venous system cranially and caudally	T2 hyperintensity T3-T9	MRI and angiography	Surgery	Yes	Stable	Angiography at 2 months: SDAVF occlusion Follow-up length, 102 months
C.R.	T8	Feeder: left T8 intercostal artery Drainage: perimedullary venous system cranially and caudally	T2 hyperintensity C5-L1	MRI (misdiagnosis with spinal cord tumor) and angiography	Surgery	Yes	Improved	Angiography at 2 months: SDAVF occlusion Follow-up length, 91 months
P.F.	T11	Feeder: left D11 intercostal artery Drainage: perimedullary venous system caudally	T2 hyperintensity T6-L1	2 MRI (at first misdiagnosis with viral infection) and angiography	Surgery	Yes	Stable	Angiography at 1 months: SDAVF occlusion Follow-up length, 3 months

SADVF, spinal dural arteriovenous fistula; MRI, magnetic resonance imaging.

Table 3. Preoperative and Postoperative Aminoff-Logue Scale Disability Score (for Gait, Micturition, and Bowel) and Preoperative and Postoperative Modified Rankin Scale Score

Patient Initials	Time Onset-Diagnosis (Months)	Gait		Micturition		Bowel		Modified Rankin Scale Score	
		Preintervention	Postintervention	Preintervention	Postintervention	Preintervention	Postintervention	Preintervention	Postintervention
A.D.	2	3	1	1	0	1	0	3	1
B.U.	4	2	1	3	3	2	2	1	1
B.F.	7	2	1	0	0	1	1	3	1
C.G.	12	2	1	1	1	1	1	1	1
F.L.	12	4	2	3	3	1	1	3	2
G.G.	2	2	1	1	1	1	1	2	1
R.M.	2	2	3	2	2	2	2	2	2
S.M.	1	5	2	0	0	2	1	4	1
T.A.	14	4	2	1	1	2	1	3	2
U.M.	5	2	1	2	2	0	0	1	1
P.I.	0,1	5	2	2	0	1	1	5	3
S.C.	2	2	1	0	0	0	0	2	1
M.T.	6	5	3	2	2	1	1	4	3
R.B.	1	1	0	1	0	0	0	1	0
S.I.	6	2	1	2	0	0	0	3	2
E.A.	24	3	1	2	1	0	0	3	1
M.A.	7	5	5	2	2	2	1	4	4
C.R.	2	4	3	0	0	0	0	4	3
P.F.	6	5	5	3	3	2	2	4	4

2-level hemilaminectomy or complete laminectomy was performed without major disruption of the facet joints so as not to destabilize the bony elements. The dura was opened to create a working intradural surgical window. The fistula was identified, typically by locating a dorsal arterialized vein into a nerve root sleeve, and then the fistula was obliterated at the artery—vein connection either by coagulation and ligation or by using an aneurysm clip. Under direct microscopic vision, indocyanine green dye was administered to ensure disconnection of the fistula. The dura was sutured in a watertight manner and the wound was closed by a standard method.

Postoperative Course

One patient experienced mild worsening of preoperative strength deficit in a lower limb; in the remaining cases, the postoperative course was uneventful. We did not observe wound infection/dehiscence or cerebrospinal fluid leakage. Two patients experienced a urinary tract infection that required antibiotic therapy.

Follow-Up

Occlusion of the fistula was confirmed by spinal angiography in all patients before discharge from the hospital. Second spinal angiography and magnetic resonance imaging (MRI) were performed within 6 months after treatment in all patients to confirm long-term angiographic occlusion. A clinical evaluation was also performed within 4—6 months after treatment and at distance by telephone interview.

Quality of Life

We assessed the HRQOL in all patients treated for SDAVF with a telephone interview 4—6 months after surgical treatment, and we evaluated clinical factors playing a role in their HRQOL, using the SF-36 questionnaire.[9] This survey consists of 36 questions for the quantification of the HRQOL and addresses 8 items for the self-reporting of physical functioning, physical role, bodily pain, general health perception, vitality, social functioning, emotional role, and mental health. Each item was scored from 0 to 100; the higher the score, the better the perceived outcome. Standardization of these domain scores with Italian population norms[9] showed that the mean score was 50 with a standard deviation (SD) of 10.

Statistical Analysis

The results were analyzed using SPSS version 23.0 (IBM Corp., Armonk, New York, USA). Demographic and symptomatic variables were described using descriptive statistics (frequencies and percentages for the categorical variables, mean and SD for the continuous variables). We used a *t* test to compare the means in preoperative and postoperative ALS/mRS and a nonparametric test (Spearman ρ), which is appropriate for statistically testing small samples, to analyze correlations. The level of significance for all analyses was $P \leq 0.05$.

RESULTS

This study comprised 19 patients; 10 patients were treated at the Neurosurgical Department of Policlinico San Martino Hospital, whereas 9 patients were treated at Neurosurgical Department of Santa Corona Hospital. Fourteen patients were men and 5 were women (male/female ratio, 2.8:1) with a mean age of 64.4 years (SD, 11.99; range, 30—80 years). The mean age was lower in men than in women (62 and 71.2 years, respectively). The mean clinical follow-up duration was 43.1 months. The thoracic location was confirmed to be the most frequent, with 17 patients (89%). Patients' data including symptoms, anatomy, diagnostic features, treatment, and outcome are reported in Tables 1—3.

Table 4. Correlation Analysis Between Short-Form 36 and Neurologic Outcome Scores

	Patients with Spinal Dural Arteriovenous Fistula, Mean (Standard Deviation)	Italian Normative Sample, Mean (Standard Deviation)
Physical functioning	60.77 (29.99)	84.46 (23.18)
Physical role	57.69 (49.35)	78.21 (35.93)
Bodily pain	57.00 (28.01)	73.67 (27.65)
General health perception	51.62 (24.12)	65.22 (22.18)
Vitality	63.46 (22.21)	61.89 (20.69)
Social functioning	69.08 (34.87)	77.43 (23.34)
Emotional role	64.00 (46.06)	76.16 (37.25)
Mental health	71.69 (18.72)	66.59 (20.89)

Diagnostic Delay

The mean time to diagnosis was 6.06 months (SD, 5.92 months). Erroneous initial diagnoses in our series of patients included lumbar spinal stenosis (1 patient), acute infective process (1 patient), spinal cord tumor (1 patient), and disc herniation (1 patient).

Initial and Subsequent Symptoms

The most common early symptoms were leg weakness, gait disturbances, and leg pain. Apart from a case of acute onset of back and leg pain with gait disturbances and rapid progression to paraparesis, the delay between first symptoms and diagnosis did not depend on the nature of the initial symptoms. The initial symptom was gait disturbances in 5 patients (26.3%) and leg weakness in 8 patients (42.1%). Leg pain was the initial symptom in 7 patients (36.8%). At the time of diagnosis, the most common symptoms were leg weakness (19 patients; 94.7%), sensory disturbances (17 patients; 89.4%), and micturition problems (13 patients; 68.4%).

Involvement of sacral segments (disturbance of micturition, fecal incontinence, sensory loss in the buttocks, or erectile dysfunction) occurred in 14 patients (73.6%). Perineal sensory loss was found in 2 patients (10.5%). Sensory loss was reported in 12 patients at the time of diagnosis (63.1%), with a presence of a sensory level of ipo-anesthesia in 6 patients. Paresthesias were reported in 8 patients (42.1%). Eighteen patients (94.7%) had some degree of leg weakness at the time of diagnosis. The average pretreatment and posttreatment ALS score of motor disability was 3.16 (SD, 1.38) and 1.89 (SD, 1.37), respectively.

Micturition and bowel problems were present in 13 (68.4%) and 9 patients (47.3%), respectively, with an average ALS preoperative score of 1.47 (SD, 1.02) and 1 (SD, 0.816) respectively. Postoperative average score improved at 1.10 (SD, 1.15) for micturition and 0.79 (SD, 0.71) for bowel.

The mean preoperative mRS score was 2.78 (SD, 1.22) and the mean postoperative mRS score improved to 1.78 (SD, 1.13).

Table 5. Correlation Analysis Between Short-Form 36 Items and Neurologic Outcome Scores

	Physical Functioning	Physical Role	Bodily Pain	General Health Perceptions	Vitality	Social Role Functioning	Emotional Role	Mental Health
Postoperative Aminoff-Logue Scale score	−0.627*	−0.604*		—	—	—	−0.726†	—
Postoperative modified Rankin Scale score	−0.853†	−0.750†	−0.777†	−0.732†	−0.589*	−0.809†	−0.739†	−0.574*
Postoperative gait	−0.778†	—	−0.610*	—	—	−0.639*	−0.589*	—
Postoperative micturition	—	−0.589*	—	—	—	—	−0.702†	—
Postoperative bowel	—	—	—	—	—	—	—	—

*$P \leq 0.05$.
†$P \leq 0.001$.

Statistical analysis showed significant differences between preoperative and postoperative ALS score ($P = 0.000–0.023$), as well as between preoperative and postoperative mRS score ($P = 0.009$), reaching higher scores in the preoperative phases. Moreover, correlation analysis showed a positive correlation (only marginally significant) between diagnostic delay and postoperative micturition ($\rho = 0.454$, $P = 0.051$). No other significant correlation between age or diagnostic delay and ALS/mRS score was found ($P = 0.222–0.958$).

Diagnosis and Treatment

All suspected lesions on MRI were confirmed by digital subtraction angiography. Preoperative MRI was performed in all patients. All patients presented with spinal cord edema; 68.4% (13 patients) were characterized by the edema extending >4 levels. The treatment strategy was identical for all patients: surgical obliteration was the first choice and endovascular treatment was offered whenever surgical treatment failed or was abandoned for technical reasons, and this option did not happen. Only 1 patient initially underwent endovascular treatment at another center. The first procedure did not resolve symptoms and a second endovascular attempt was performed, before definitive surgical treatment.

Postoperative Quality of Life

The scores for the SF-36 obtained at the follow-up are reported in Table 4. Correlations analysis (Spearman ρ) showed negative relations between SF-36 and ALS score (not for bowel), as well as between SF-36 and mRS score (Table 5).

DISCUSSION

Two main treatment options are available for SDAVFs: open microsurgery and endovascular embolization. Controversies still exist regarding the best treatment modality for SDAVFs. Because of the large heterogeneity, with insufficient longitudinal data, an affordable comparison regarding the efficacy of surgery versus embolization is unfeasible and a definitive treatment recommendation has still to be achieved. Some investigators favor endovascular treatment[2,3] because of the minor invasiveness of the approach, the avoidance of postoperative pain, and the association with a shorter hospital stay. On the other hand, endovascular treatment carries the risk of incomplete SDAVF occlusion and a higher rate of SDAVF recurrence. The results obtained from our series of patients indicate that open surgical treatment has been effective in treating SDAVF. Complete disconnection of the fistula was achieved in all patients during 1 single operative session, with no recurrences noted on angiographic evaluation during the follow-up period. Only 1 patient initially underwent endovascular treatment in another center. The first procedure did not resolve symptoms and a second endovascular treatment was performed. Following a second rapid recurrence, the patient came to our attention and underwent surgical treatment with definitive fistula occlusion. The symptoms did not improve much probably because of the long history of medullary pain.

The initial failure and the higher recurrence rate after endovascular treatment are explained by an anatomic study by Takai et al.[10] SDAVF is made up of multiple arteriovenous connections, mainly on the inner dural surface, between >1 meningeal feeding arteries at the same or adjacent spinal level, and a single proximal radiculomedullary vein is located. For this reason, permanent occlusion is not possible even if embolization materials fill the meningeal arteries on the outer dural surface. Complete penetration of the intradural proximal radiculomedullary vein, which is the only way to achieve permanent occlusion, is not a straightforward procedure, even with the aid of new liquid embolic material (OnyX [Ev3 Neurovascular, Irvine, California, USA]).

A recent meta-analysis[11] seemed to confirm the conclusions of Takai et al. The investigators reported a definite advantage of primary surgical treatment of SDAVF over endovascular treatment in initial failure rate and late recurrence.

After successful surgical obliteration, motor symptoms improved in >80% of patients. The average pretreatment and posttreatment ALS score of motor disability was 3.16 and 1.89, respectively. Regarding sphincter dysfunction, micturition problems improved in 33% of symptomatic patients and bowel problems in 30% of symptomatic patients. Data about postoperative clinical improvement in postoperative scores were statistically significant with respect to preoperative state. A favorable impact of surgery on symptoms was also shown by a meta-analysis by Steinmetz et al.[12] of 16 surgical studies. These investigators found rates of improvement and stabilization of clinical function of 55% and 34%, respectively, with a successful SDAVF occlusion rate of 98%, with 2% morbidity and no mortality.

Regarding the diagnostic delay, our series confirms the long time interval between the initial symptoms and diagnosis (on average, 6.06 months). The diagnostic delay did not seem to be statistically correlated to the type or intensity of initial symptoms or to the quality of the outcome (except for a marginal correlation with micturition postoperative score). In our experience, in line with what emerges from other studies,[13-16] the postoperative outcome is strongly associated with a higher degree of disability at the time of treatment and thus a poorer clinical outcome after treatment.

On the other hand, our experience showed that MRI has an important role in making the suspected diagnosis of SDAVF. The presence of an SDAVF was hypothesized after the first high-resolution MRI examination in all but 2 patients (for which an infective process and a spinal cord tumor had been contemplated, respectively). For those reasons, it may be feasible to attempt to obtain a timely diagnosis of these lesions by also applying a low threshold for performing high-quality MRI in patients with otherwise unexplained symptoms of myelopathy.

Regarding the HRQOL measured with SF-36 during the follow-up, patients with SDAVF showed lower average scores than did those reported in the Italian normative sample, except for vitality and mental health, which were closest to the Italian normative sample. This finding is confirmed also by Sasamori et al.,[17] who similarly investigated the HRQOL of 52 Japanese patients with SDAVF, noting that posttreatment neurologic improvement is often partial. Examination of the correlations between persistent symptoms and HRQOL indicated that the decline of persistent symptoms has a positive impact on emotional functioning. Moreover, analysis of correlations showed that the decline in the SF-36 scores was primarily attributable to an improvement in postoperative mRS score, and only partially in gait and micturition disturbances. However, this finding is not comparable to those of Sasamori et al., because it is the first study in which mRS score was used with patients with SDAVF. In addition, bodily pain was negatively associated with gait and postoperative mRS score, whereas, different from Sasamori et al., posttreatment pain was not independent of the severity of other persistent symptoms. Active intervention to ease postoperative symptoms may be needed to improve patients' HRQOL.

The present study has limitations to drawing a conclusion. The first limitation is that statistical analysis and data research were retrospective and lacked randomization. Despite the multidisciplinary approach, the patients analyzed for the present case series underwent surgical treatment only with no control group. The HRQOL has been evaluated only in follow-up and not preoperatively. In addition, the present results represent a 2-center experience with a small number of patients. However, because SDAVF is a rare entity, larger and prospective study cohorts would be difficult to obtain. The inclusion of additional facilities with a possible comparison group of fistulas managed endovascularly with HRQOL outcomes might be a further development for future multi-institutional study.

CONCLUSIONS

Our series confirmed that surgical obliteration of SDAVFs is an effective and safe procedure. The results of this retrospective analysis make us believe that surgery, given its low morbidity and high success rate, represents a safe and efficient first therapeutic option for these spinal vascular malformations. Surgery could be considered to avoid unsuccessful endovascular attempts that may delay the definitive treatment of this disease. The surgical procedure showed good results in terms of neurologic improvement as well as patients' perception of quality of life. An early diagnosis is crucial for a good outcome and high-field MRI should probably be performed early in all those patients in whom SDAVF is suspected.

REFERENCES

1. Kim LJ, Spetzler RF. Classification and surgical management of spinal arteriovenous lesions: arteriovenous fistulae and arteriovenous malformations. *Neurosurgery*. 2006;59(5 suppl):195-201.
2. Eskandar EN, Borges LF, Budzik RF, Putman CM, Ogilvy CS. Spinal dural arteriovenous fistulas: experience with endovascular and surgical therapy. *J Neurosurg Spine*. 2002;96:162-167.
3. Patsalides A, Santillan A, Knopman J, Tsiouris AJ, Riina HA, Gobin YP. Endovascular management of spinal dural arteriovenous fistulas. *J Neurointerv Surg*. 2011;3:80-84.
4. Bertuccio A, Robba C, Spena G, Versari PP. Intracranial and spinal dural arterio-venous fistula (DAVF): a surgical series of 107 patients. *Acta Neurochir Suppl*. 2016;123:177-183.
5. Saladino A, Atkinson JL, Rabinstein AA, Piepgras DG, Marsh WR, Krauss WE, et al. Surgical treatment of spinal dural arteriovenous fistulae: a consecutive series of 154 patients. *Neurosurgery*. 2010;67:1350-1357 [discussion 1357-1358].
6. Watson JC, Oldfield EH. The surgical management of spinal dural vascular malformations. *Neurosurg Clin N Am*. 1999;10:73-87.
7. Binning MJ, Schmidt MH. Percutaneous placement of radiopaque markers at the pedicle of interest for preoperative localization of thoracic spine level. *Spine (Phila Pa 1976)*. 2010;35:1821-1825.
8. Britz GW, Lazar D, Eskridge J, Winn HR. Accurate intraoperative localization of spinal dural arteriovenous fistulae with embolization coil: technical note. *Neurosurgery*. 2004;55:252-254 [discussion 254-255].
9. Apolone G, Mosconi P. The Italian SF-36 Health Survey: translation, validation and norming. *J Clin Epidemiol*. 1998;51:1025-1036.
10. Takai K, Komori T, Taniguchi M. Microvascular anatomy of spinal dural arteriovenous fistulas: arteriovenous connections and their relationships with the dura mater. *J Neurosurg Spine*. 2015;23:526-533.
11. Bakker NA, Uyttenboogaart M, Luijckx GJ, Eshghi OS, Mazuri A, Metzemaekers JD, et al. Recurrence rates after surgical or endovascular treatment of spinal dural arteriovenous fistulas: a meta-analysis. *Neurosurgery*. 2015;77:137-144.
12. Steinmetz MP, Chow MM, Krishnaney AA, Andrews-Hinders D, Benzel EC, Masaryk TJ, et al. Outcome after the treatment of spinal dural arteriovenous fistulae: a contemporary single-institution series and meta-analysis. *Neurosurgery*. 2004;55:77-87 [discussion 87-88].
13. Cenzato M, Debernardi A, Stefini R, D'Aliberti G, Piparo M, Talamonti G, et al. Spinal dural arteriovenous fistulas: outcome and prognostic factors. *Neurosurg Focus*. 2012;32:E11.
14. Iovtchev I, Hiller N, Ofran Y, Schwartz I, Cohen J, Rubin SA, et al. Late diagnosis of spinal dural arteriovenous fistulas resulting in severe lower-extremity weakness: a case series. *Spine J*. 2015;15:e39-e44.
15. Brinjikji W, Nasr DM, Morris JM, Rabinstein AA, Lanzino G. Clinical outcomes of patients with delayed diagnosis of spinal dural arteriovenous fistulas. *Am J Neuroradiol*. 2016;37:380-386.
16. Muralidharan R, Saladino A, Lanzino G, Atkinson JL, Rabinstein AA. The clinical and radiological presentation of spinal dural arteriovenous fistula. *Spine (Phila Pa 1976)*. 2011;36:E1641-E1647.
17. Sasamori T, Hida K, Osanai T, Yano S, Seki T, Houkin K. Health-related quality of life in patients with spinal dural arteriovenous fistulae. *Neurosurg Rev*. 2017;40:83-86.

Conflict of interest statement: The authors declare that the article content was composed in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Received 18 June 2018; accepted 3 October 2018

Citation: World Neurosurg. (2019) 122:e270-e278.
https://doi.org/10.1016/j.wneu.2018.10.019

Journal homepage: ***www.journals.elsevier.com/world-neurosurgery***

Available online: ***www.sciencedirect.com***