

# BMJ Open Long-term outcomes and prognostic factors in patients with treated spinal dural arteriovenous fistulas: a prospective cohort study

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## ABSTRACT

**Objective** To define the pattern of long-term clinical outcomes and prognostic factors in patients with spinal dural arteriovenous fistulas (SDAVFs).

**Design** Prospective cohort study based on constantly recruiting patients with SDAVFs in two medical centres in China.

**Setting** Patients with SDAVFs were recruited consecutively between March 2013 and December 2014 in two referral centres.

**Participants** A prospective cohort of 94 patients with SDAVFs was included in this study, and 86 patients (mean age 53.0 years, 71 men) completed the study. Patients who had previously undergone endovascular or neurosurgical treatment or had neurological dysfunction caused by other diseases or refused treatment were excluded.

**Interventions** All patients underwent neurosurgery or endovascular embolisation. These patients were evaluated with the modified Aminoff and Logue's Scale (mALS) 1 day before and 3, 6, 12 and 72 months after treatments.

**Results** The duration of symptoms ranged from 0.5 to 66 months (average 12.8 months). The location of SDAVFs was as follows: 33.7% above T7, 50.0% between/include T7 and T12 and 16.3% below T12. 75 patients (87.2%) underwent neurosurgical treatment, and 9 patients (10.5%) underwent endovascular treatment. 58 patients (67.4%) exhibited an improvement in mALS of one point or greater at 72 months. Patients with less disability were more likely to improve at 72 months ( $p<0.05$ ). 48 patients (55.8%) showed deterioration at 72 months compared with 12 months. 61% of the patients suffered numbness, and 22% had pain before treatment. However, 81% of patients had numbness, and 28% had pain after treatment. This deterioration was related to 1-year mALS and age.

**Conclusion** Nearly two-thirds of the patients experienced clinical improvement at 72 months, and preoperative (1 day before treatment) mALS was the strongest predictor of clinical improvement. However, 55.8% of patients showed deterioration after temporary recovery. All patients with SDAVFs should accept treatment as soon as possible.

## BACKGROUND

Spinal dural arteriovenous fistulas (SDAVFs) are a rare disease, although they account for

## Strengths and limitations of this study

- This is a large-scale prospective cohort study of spinal dural arteriovenous fistulas (SDAVFs).
- This study provides the long-term outcomes of patients suffering SDAVFs with a 6-year follow-up.
- This study lacks an analysis of clinical factors such as rehabilitation.
- This study lacks imaging analysis.

70% of spinal vascular malformations, with an annual incidence of 5–10 cases per million persons.<sup>1 2</sup> Digital subtraction angiography (DSA) is the gold standard for the diagnosis of SDAVFs.<sup>3 4</sup> Once diagnosed, either neurosurgery or endovascular embolisation is required for patients to avoid further deterioration.<sup>5</sup> Previous studies suggested that the preoperative severity of disability was the most important prognostic factor. Conflicting results have been obtained regarding the relationship between age, gender and duration of symptoms prior to treatment and treatment outcome. Due to the low incidence of SDAVFs, small sample sizes and unstandardised assessments among retrospective data, previous studies of long-term clinical outcomes and prognostic factors of SDAVFs are not representative.<sup>6–14</sup>

To further understand the natural history of SDAVFs, we continued a prospective cohort study to evaluate the 6-year outcomes of patients with SDAVFs to identify major prognostic factors and long-term outcomes.

## METHODS

### Study design

Under the guidance of Strengthening the Reporting of Observational Studies in Epidemiology,<sup>15</sup> we performed a prospective and

longitudinal cohort study at two referral centres (for SDAVFs that provide neurosurgical and endovascular treatments).

### Participants and settings

We prospectively collected data on patients diagnosed with SDAVFs at the cervical and thoracolumbar regions who received treatment at two referral centres between March 2013 and December 2014. All patients with SDAVFs were recruited to participate. Because lesions located in the cranialcervical region and sacral coccyx area are more complex, they were excluded from this study. Patients who previously had endovascular or neurosurgical treatment or those who had neurological dysfunction caused by other diseases or refused treatment were excluded. The data were analysed by one of three designated investigators who did not participate in the treatment process. Treatment strategies were decided by consensus after review by a team of experienced neurosurgeons and neuroradiologists.

### Treatment and intervention

Spinal angiography was performed in all patients, including angiography of all segmental arteries. Most patients had underwent open surgery in which their fistulas cauterised after haemilaminectomy. Endovascular treatment was considered for patients who were assessed as high risk for general anaesthesia but did not have any arterial feeders from the radicular artery of Adamkiewicz. Onyx (ev3) was injected as close as possible to the fistula until the proximal part of the draining vein was obliterated. If endovascular treatment was unsuccessful to obliterate SDAVFs, neurosurgical treatment was performed. The functional status of the patients was assessed by a standardised scale 1 day before the procedure and 3, 6, 12 and 72 months postoperatively as long-term follow-up.

### Data collection

Clinical data, including age, sex, duration of symptoms, location of fistula, spinal functional status and treatment methods, were collected. The onset of symptoms was considered to be when neurological deficits were first noticed. The preoperative spinal angiogram images were reviewed by one of two senior authors to identify the location of SDAVFs. The functional status of the patients was assessed using the modified Aminoff and Logue's Scale<sup>16</sup> (mALS, online supplemental table 1) 1 day before the procedure and at 3, 6, 12 and 72 months postoperatively. All patients underwent DSA examination after operation and MRI. The modified Denis Scale<sup>17</sup> (mDS, online supplemental table 2) was supplied at the 72-month follow-up to evaluate the patient's sensory impairment.

### Bias

Loss to follow-up might bias the results. Eight patients were lost to follow-up; the follow-up rate was 91.5%. Therefore, recall bias might affect data entry, but this was minimised by ensuring that data were entered in a timely manner.

**Table 1** Baseline demographics and characteristics of patients with spinal dural arteriovenous fistula\*

Characteristics	Number (%)
<b>Age at treatment, average (SD), years</b>	53.0 (10.3)
<b>Men, n (%)</b>	71 (82.6%)
<b>Time interval between symptoms and treatment, average (SD), months</b>	12.8 (12.9)
≤6 months, n (%)	35 (40.7%)
6–12 m, n (%)	23 (26.7%)
>12 m, n (%)	28 (32.6%)
<b>Location of the fistula, n (%)</b>	
Above T7	29 (33.7%)
T7–T12	43 (50.0%)
Below T12	14 (16.3%)
<b>Treatment method, n (%)</b>	
Neurosurgery	75 (87.2%)
Endovascular	9 (10.5%)
Combination	2 (2.3%)
<b>Preoperative mALS, n (%)</b>	
<b>G score</b>	
0	5 (5.8%)
1	8 (9.3%)
2	36 (41.9%)
3	18 (20.9%)
4	6 (7.0%)
5	13 (15.1%)
Average (SD)	2.6 (1.36)
<b>U score</b>	
0	10 (11.6%)
1	38 (44.2%)
2	24 (27.9%)
3	14 (16.3%)
Average (SD)	1.5 (0.9)
<b>F score</b>	
0	7 (8.1%)
1	55 (64.0%)
2	20 (23.3%)
3	4 (4.7%)
Average (SD)	1.2 (0.7)

\*Patient with two fistulas (T8, T10).

F, faeces; G, gait; mALS, modified Aminoff and Logue's Scale; T, thoracic; U, urination.

### Statistical analysis

All data were descriptively presented using the mean±SD for continuous data and frequencies for categorical data (table 1). Paired t-tests (with adjustment for multiple comparisons) were used to assess differences in means for the cohort between baseline and different follow-up

time points. Pearson  $\chi^2$  tests were used to identify factors associated with preoperative status. Pearson  $\chi^2$  tests and unconditional logistic regression were used to identify factors affecting clinical improvement at 72 months (6 years). Clinical improvement was defined as a decrease of at least one point on the mALS compared with baseline assessment at 72 months. In the multivariate model, age ( $\leq 55$  years or  $>55$  years), sex (men/women), time interval between symptom onset and treatment (6 months,  $>6$  months and 12 months or  $>12$  months), location (above T7, T7–T12 or below T12), treatment performed (neurosurgical treatment, endovascular treatment or both) and preoperative mALS were entered. The preoperative mALS was classified as follows: a total score of 0–3 indicated mild disability, a score of 4–7 indicated moderate disability and a score of 8–11 indicated severe disability. Clinical improvement was entered as the dichotomous dependent variable. Interactions were tested in the model. The OR and 95% CI were determined for significant variables in the model. All analyses were performed by an epidemiologist using Python software (V.3.6.0 final, 2016-12-23) and SPSS software (V.23, IBM Corp.).

### Patient and public involvement statement

Patients were not invited to comment on the study design and were not consulted to develop patient-relevant outcomes or interpret the results. Patients were not invited to contribute to the writing or editing of this document for readability or accuracy.

## RESULTS

### Patient population

The baseline demographic characteristics of these 86 patients are presented in [table 2](#). We initially screened 94 patients; 8 patients were excluded due to loss to follow-up ( $n=7$ ) or death from cardiovascular accidents 3 years after treatment ( $n=1$ ). The average follow-up time interval was 74.0 months (range 62–83 months).

In this group of 86 patients (mean age  $53.0 \pm 10.3$  years; 71 men), the average time interval between symptom onset and treatment was 12.8 months (range 0.5–66 months). The most common location for SDAVFs was the lower thoracic region (T7–12, 50%). Before treatment, the patients presented with a median mALS of 5 (range 0–11), a median G score of 2 (range 0–5), a U score of 1 (range 0–3) and an F score of 1 (range 0–3). A total of 75 patients (87.2%) underwent neurosurgical treatment, 9 patients (10.5%) underwent endovascular

treatment, and 2 patients (2.3%) underwent neurosurgical treatment after unsuccessful endovascular treatment. In one patient, the fistula was obliterated after the first embolisation but required neurosurgery for recurrence demonstrated on 8-month follow-up angiography.

### Clinical outcomes

The pattern of changes in mALS scores at different time points postoperation is presented in [figures 1 and 2](#). It is worth noting that in the sixth year after treatment, the spinal cord function of the patients decreased compared with that at 1 year. Most patients improved in the first year after treatment. The mALS score improved the most at the 3-month evaluation after treatment. Except for the change in F score between 3 months and 6 months ( $p=0.09$ ), the difference between two adjacent follow-up time points was significant ( $p<0.05$ ).

At the 1-year follow-up, 74 patients (86.0%) improved their mALS, 63 patients (73.3%) improved gait disability, 51 patients (59.3%) improved urination function and only 38 patients (44.2%) improved their defecation function. For patients with an exercise score of 5, only 38.5% were able to walk independently.

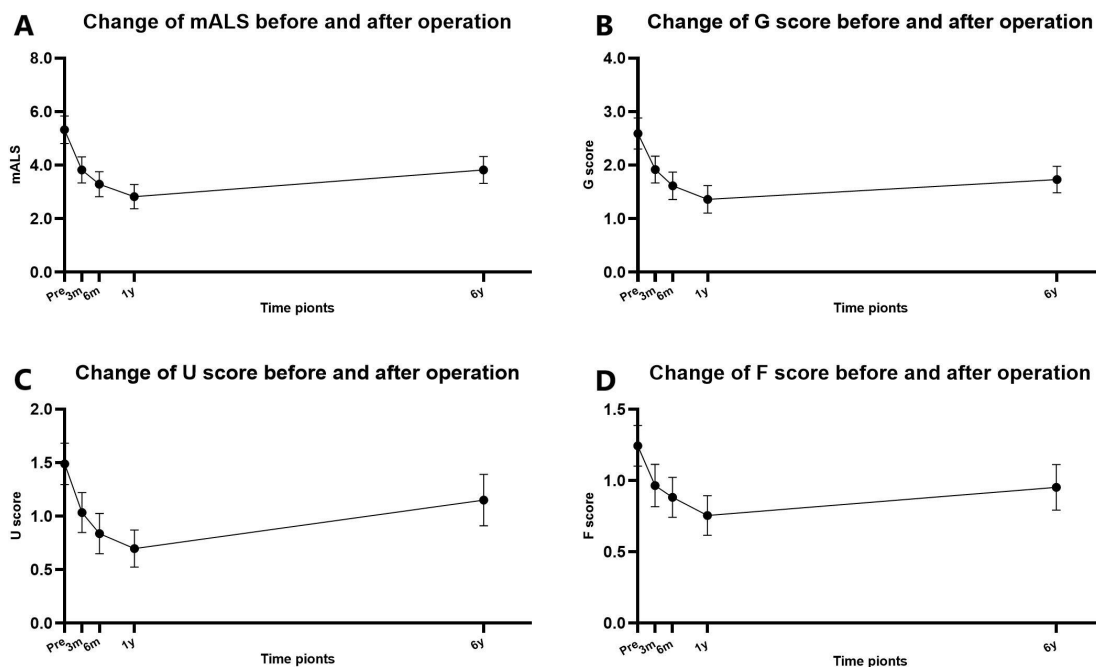
At the 6-year follow-up, only 58 patients (67.4%) showed improvement in their mALS, 50 patients (58.1%) improved gait ability, 38 patients (44.2%) improved urination function and only 30 patients (34.9%) improved their defecation function. Fortunately, for patients with an exercise score of 5, 53.8% of patients were able to walk independently, and 1 patient improved the G score to 0.

We compared the changes between the 6-year and the 1-year periods. Fifteen patients (17.4%) continued to improve spinal cord function, while up to 48 patients (55.8%) were in worse condition than they were a year ago. Fifteen patients (17.4%) had improved gait disorders, 7 patients (8.1%) had improved urination function and 10 patients (11.6%) had improved defecation. Forty patients (46.5%) had worse gait than before, 34 patients (39.5%) had increased urination disorders and 23 patients (26.7%) experienced defecation that was worse than before.

Since the preoperative mDS was not recorded and to ensure the reliability of the data, we only collected whether there was numbness or pain before the operation and the mDS of these patients 6 years after the operation. In this study, 61% of the patients suffered numbness, and 22% had pain before treatment. However, 81% of patients had numbness, and 28% of patients had postoperative pain. Thirty per cent of them developed numbness only after surgical treatments. Thirty-three per cent of patients had local pain before treatment ([figure 3](#)). Few patients could completely relieve their pain or numbness after treatment. At the 6-year time point, the average mDS was 3.7 points (range 2–7), including a pain score of 1.4 points and a numbness score of 2.3 points ([table 2](#)).

**Table 2** Patients' modified Danis Scale (mDS) at 6-year time point

	Numbness	Pain	mDS
Mean	2.3	1.4	3.7
Range	1–4	1–5	2–7
SD	0.87	0.84	1.16



**Figure 1** Compared with the baseline, the modified Aminoff and Logue's Scale (mALS) score improved at all time points. In the sixth year after surgery, the mALS decreased compared with the first year. Except for the change in F score between 3 months and 6 months ( $p=0.09$ ), the difference between two adjacent follow-up time points was significant ( $p<0.05$ ).

### Prognostic factors

The preoperative mALS was found to be related to clinical improvement at 6 years. A higher preoperative mALS, which suggests higher spinal cord disability (score of 4–7, OR 0.305, 95% CI 0.099 to 0.936, and score of 8–11, OR 0.058, 95% CI 0.006 to 0.557), suggests poor clinical improvement at 6 years. The age, duration of symptoms, location of fistula and treatment method used were not related to the outcomes (table 3). There was a trend towards an inverse relationship between age (when entered as a continuous variable) and a higher rate of clinical improvement between 6 years and 1 year ( $p=0.028$ ).

### DISCUSSION

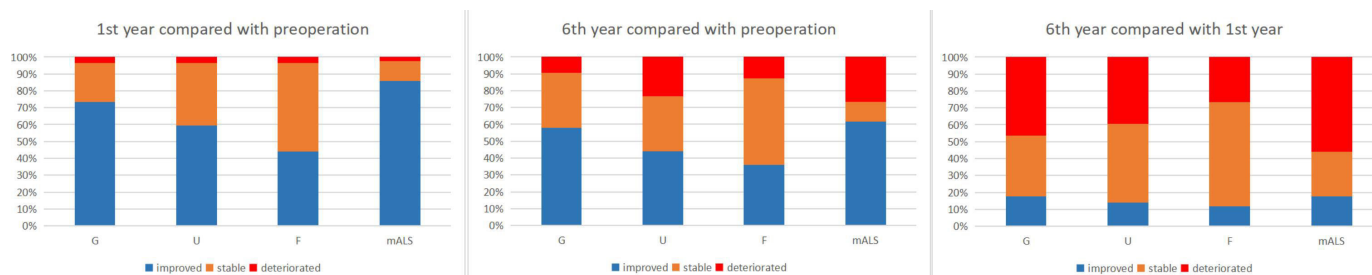
Our study provides outcomes and long-term prognostic factors for a large patient with SDAVF cohort study by using a prospective standardised assessment method.

### Short-term outcome of patients with SDAVFs

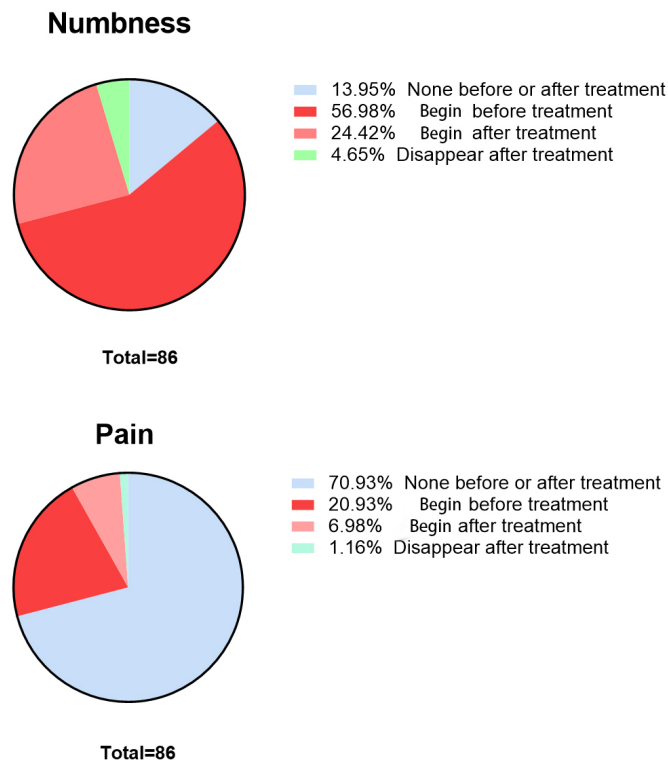
Most patients with SDAVFs can obtain better spinal cord function after neurosurgery treatment.<sup>18</sup> Within 1 year, 74.5% of patients experienced improvement in their motor symptoms. A total of 58.5% of patients recovered urination function, and the percentage of recovered defecation function was 43.6%. The improvement showed a continuing trend and was more obvious in the first 3 months.

### Long-term outcome of patients with SDAVFs

In a study by Jablawi *et al*, clinical outcome was improved in 21/40 (53%), stable in 11/40 (28%) and aggravated in 8/40 patients (20%, follow-up interval 10–231 months).<sup>10</sup> In Tacconi's study, 13/20 (63%) patients showed a deterioration in their follow-up (interval 3–24 months).<sup>13</sup> In our group, although there was an obvious score change between 6 years and 1 year, nearly 67.4% of patients had improved mALS compared with preoperation. A total



**Figure 2** Red indicates deterioration, yellow indicates stability, blue indicates improvement. Compared with first year, 55.8% of patients showed deterioration at sixth year.



**Figure 3** Eighty-one per cent of patients had postoperative limb numbness, 28.0% of patients had postoperative pain and 30% of them developed numb symptoms only after surgical treatments. Thirty-three per cent of patients had local pain symptoms before treatment. Few patients could completely relieve their pain or numbness after surgical treatment.

of 58.1% of patients improved gait disability, 44.2% of patients experienced improvement of urination function and 34.9% of patients improved their defecation function. Moreover, 81% of these patients had body numbness, and 28% of patients had pain after treatment, among which 30% had no numbness previously.

### Prognostic factors

In this study, the strongest factor predicting the long-term outcome was the degree of preoperative spinal cord function, while age, sex, duration of symptoms and location of fistula were not directly correlated. Nagata *et al* suggested that outcome was better in younger patients.<sup>11</sup> However, we found no correlation between age and clinical improvement at 1 or 6 years, consistent with previous studies.<sup>10 13</sup> Although some studies<sup>7 11 14</sup> including ours have shown no correlation between the duration of symptoms and short-term prognosis, Tacconi *et al* believed that a shorter duration of symptoms could predict a better body condition.<sup>10 12 13</sup> SDAVFs are a type of spinal venous hypertensive disease. If not treated in time, the patient's spinal cord function will gradually decline, and even irreversible neuronal damage may occur, so the duration of symptoms may be another predictor of patient outcome. This requires the subsequent inclusion of more cases to confirm their relevance. Cenzato *et al* found that the location of the fistula could predict outcome; patients with a

fistula between T9 and T12 improved more than those with a fistula elsewhere.<sup>8 14</sup> Shinoyama *et al* found that in craniocervical SDAVFs, surgical treatment provides favourable long-term outcomes without a risk of recurrence. In thoracolumbar SDAVFs, irreversible structural changes, such as spinal cord atrophy, may lead to poor recovery.<sup>12</sup> In our results, no association was found between clinical improvement and the level of fistula. In addition, multiple fistula SDAVFs are extremely rare.<sup>10 19 20</sup>

We included 3 (3.5%) patients with dual SDAVFs. In one patient, one fistula was on the right T8 and another on the left T10, and the draining veins were also separated. This patient showed mild disability during the first year but developed severe disability 6 years later. However, the other two patients (one fistula was located on both sides of L1 and the other fistula was located on contralateral L4) were more stable. This phenomenon may be related to the range of the segment covered by the draining vein.

The treatment (microsurgery or endovascular) that benefits patients with SDAVFs has been a controversial topic. However, the primary goal of SDAVF treatment must be the interruption of fistulas. From the perspective of avoiding fistula recurrence, the surgical treatment of SDAVFs is superior to intravascular treatment.<sup>12 20</sup> In terms of therapeutic effects, intravascular treatment is improving. In 2004, Steinmetz *et al* reported a success rate of 46% in the endovascular group. In 2016, the probability of intravascular treatment performed by Sasamori T *et al* achieving complete embolisation was as high as 71.0%. In our study, the success rate of endovascular treatment was 76.9%.<sup>18</sup> To date, it is currently believed that neither microsurgery nor endovascular embolisation shows statistical significance with regard to outcome.<sup>20</sup>

Tacconi *et al* believe that preoperative functional status is related to long-term clinical outcomes.<sup>13</sup> Our results are consistent with this scenario. Jablawi F *et al* speculated that the preoperative neurological condition was not related to long-term prognosis.<sup>10</sup> This might due to their small sample. In our study, there were indeed cases of patients with severe disability preoperatively who achieved extremely high improvement after treatment (among patients with an exercise score of 5, 53.8% of patients were able to walk independently at 6 years, and 1 patient's G score reached 0).

We observed that patients with SDAVFs usually have sensory dysfunction. Muralidharan *et al* believed that acupuncture-like sensation abnormalities in patients before surgery indicate poor clinical improvement.<sup>21</sup> Shinoyama M *et al* found that some patients with SDAVFs have postoperative numbness or pain symptoms and new numbness or pain.<sup>12</sup> Combined with the overall decline in spinal motor function in patients, we suspect that there are often irreversible structural changes in the spinal cord. It is widely recognised that the pathophysiology of SDAVFs is chronic hypoxia and progressive myelopathy induced by venous hypertension.<sup>22</sup> However, venous congestion in SDAVFs does not always cause irreversible tissue destruction. The spinal

**Table 3** Factors associated with clinical improvement at 6 year (compared with preoperation state): univariate and multivariate analysis

Variable	Patients with improvement, n (%)	Patients without improvement, n (%)	Univariate model		Multivariate model	
			$\chi^2$	P value	OR (95% CI)	P value
<b>Age</b>			0.825	0.364	1.527 (0.611 to 3.815)	0.365
≤55 years	37	15				
>55 years	21	13				
<b>Gender</b>			1.305	0.253	2.174 (0.560 to 8.433)	0.262
Men	46	25				
Women	12	3				
<b>Time interval between symptom onset and treatment</b>			1.806	0.405		
≤6 months	21	13			Reference	
6–12 months	14	8			0.923 (0.304 to 2.802)	0.888
>12 months	23	7			0.492 (0.165 to 1.466)	0.203
<b>Location of the fistula</b>			1.171	0.557		
Above T7	18	11			Reference	
T7–T12	29	14			0.790 (0.295 to 2.115)	0.639
Below T12	11	3			0.446 (0.102 to 1.962)	0.286
<b>Treatment method</b>			1.555	0.459		
Neurosurgery	49	26			Reference	
Endovascular	7	2			0.538 (0.104 to 2.781)	0.460
Combination	2	0			0.374 (0.017 to 8.070)	0.530
<b>Preoperative mALS</b>			9.125	0.010		
0–3	7	10			Reference	
4–7	39	17			0.305 (0.099 to 0.936)	0.038
8–11	12	1			0.058 (0.006 to 0.557)	0.014

mALS, modified Aminoff and Logue's Scale; T, thoracic.

cord pathology in some cases is oedema due to congestion or ischaemic penumbra caused by low perfusion, which might be relieved after fistulas are cured.

### Late deterioration

In our study, 48/86 (55.8%) of the patients showed deterioration, which was similar to the findings of Cecchi *et al.*<sup>23</sup> The first cause to be considered was recrudescence. However, this has not been reported in previous surgical series.

The main causes of this reversal might be haemodynamic changes in the local vascularisation induced by the fistulas and the long period of anatomofunctional deficiency of the venous drainage. Patients might obtain significant improvement after surgical treatment, which immediately changes the venous pressure around the lesion and gradually relieves spinal cord oedema. However, late deterioration may result from neuronal loss, which is caused by venous hypertension before treatments. In our rabbit model of spinal venous hypertension, we observed neuronal demyelination and inflammatory changes.<sup>24</sup> We speculate that the temporary recovery of patients with

SDAVF is mainly due to the relief of spinal cord oedema. The inflammatory factors and autoantibodies released by necrotic neurons cause local inflammatory reactions, which persist for a long time, eventually leading to late deterioration of the patient. Durnford *et al.*<sup>25</sup> believed that these patients had a reduced neural reserve, and any future neuronal loss that might have been subclinical in normal individuals may result in clinically overt neurologic deterioration.

We analysed the existing data and found that a poor postoperative mAL grade at 1 year might lead to late deterioration ( $p=0.005$ ,  $<0.01$ ) (table 4) by setting the 1-year postoperative state as baseline. When age was entered as a continuous variable, it was related to deterioration. Furthermore, we examined the patient group characteristics, and the assessment system content included urination function. The average age reached 53, and 4/5 were men. Some diseases, such as prostate diseases in male patients, affect our analysis results. Another manifestation of worsening was that sensory systemic symptoms were aggravated

**Table 4** Factors associated with clinical improvement at 6 year (compared with 1 year after treatment): univariate and multivariate analysis

Variable	Patients with improvement, n (%)	Patients without improvement, n (%)	Univariate model		Multivariate model	
			$\chi^2$	P value	OR (95% CI)	P value
<b>Age</b>			0.387	0.534	0.701 (0.228 to 2.153)	0.535
≤55 years	8	44				
>55 years	7	27				
<b>Gender</b>			1.074	0.300	1.983 (0.534 to 7.370)	0.306
Men	11	60				
Women	4	11				
<b>Time interval between symptom onset and treatment</b>			2.904	0.234		
≤6 months	3	31			Reference	
6–12 months	5	17			0.329 (0.070 to 1.548)	0.160
>12 months	7	23			0.318 (0.074 to 1.364)	0.123
<b>Location of the fistula</b>			3.807	0.149		
Above T7	2	27			Reference	
T7–T12	9	34			0.280 (0.056 to 1.405)	0.122
Below T12	4	10			0.185 (0.029 to 1.173)	0.073
<b>Treatment method</b>			11.924	0.002		
Neurosurgery	10	65			Reference	
Endovascular	3	6			0.308 (0.066 to 1.432)	0.133
Combination	2	0			0.032 (0.001 to 0.716)	0.030
<b>1-year mALS</b>			10.721	0.005		
0–3	6	54			Reference	
4–7	8	17			0.236 (0.072 to 0.777)	0.017
8–11	1	0			0.040 (0.001 to 1.080)	0.056

mALS, modified Aminoff and Logue's Scale; T, thoracic.

or newly occurred. In our study, 81% of patients had body numbness at the 6-year time point, and 30.0% of patients had no previous numbness. The occurrence of numbness was three times more frequent than pain. Shinoyama *et al*<sup>12</sup> found that the existence of postoperative spinal cord atrophy and a residual intramedullary hyperintense lesion in T2WI correlated with poor functional recovery, as well as worsening of neuropathic pain. The specific mechanism was not clear. Furthermore, patient work and economics and attention to the rehabilitation of the disease result in long-term variations in the rehabilitation process, which will cause certain deviations in our research.

Therefore, patients with SDAVFs must undergo surgical treatment as soon as possible; otherwise, their spinal cord function will decline gradually. The irreversible damage to the spinal cord caused by spinal venous hypertension, or any reason for delayed treatment, may also affect subsequent recovery.

## LIMITATIONS

This study has some limitations. First, the investigator analysed spinal function based on mALS, which focuses only on motor and sphincter status. Sensory function development rules cannot be described. Second, there is a lack of analysis of more clinical factors for late deterioration, such as whether the patient has undergone regular rehabilitation, steroid treatment, prostate disease and so on. We continued to collect data to further verify the above factors. Finally, this study lacks imaging analysis. The imaging features on MRI have been studied. Signs of T2WI hyperintensity and flow voids were observed on MRI. Patients with enlarged draining veins (>10 spinal levels) had worse mALS scores, more extensive draining veins were associated with more spinal cord T2 hyperintensity,<sup>26</sup> and the extent of the hyperintensity area was relevant to preoperative neurological deficits.<sup>27</sup> Yamahata *et al* evaluated sagittal T2-weighted MRI scans and assessed the CEOR (the occupation ratio of the cauda equina compared with the sagittal diameter of the corresponding lumbar spinal canal) and found that there was a

significant difference between the preoperative and postoperative values, which could be used as a criterion for evaluating surgery. T2 signal abnormalities of the spinal cord and flow voids were not associated with clinical outcomes.<sup>9 10 28</sup>

## CONCLUSION

This prospective cohort study shows that preoperative mALS is the strongest relevant factor for long-term outcome in patients with SDAVFs. A total of 67.4% of patients recovered after interruption of the fistula. Although the overall spinal cord function of the patients improved in the sixth year after the operation, compared with their preoperative state, a significant decrease was observed compared with the patients' optimal postoperative condition. The postoperative mALS at 1 year and patients' age is inversely related to this decline. Patients' sensory systems may also deteriorate in the long term. Therefore, patients with SDAVFs should undergo treatment (not limited to open surgery or endovascular treatment) as soon as possible, otherwise their spinal cord function will decline gradually. The irreversible damage to the spinal cord caused by spinal venous hypertension, or any reason lead to delayed treatment may also affect subsequent recovery.

## FUTURE EFFORTS

There are 86 patients in our prospective cohort who continued to be followed. By constantly recruiting new participants and supplementing data, we can obtain results that more closely resemble the actual situation, which will be described and analysed in more detail later. Not all patients showed late deterioration. What are the pathological changes in the spinal cords of patients with SDAVFs that can explain this deterioration? How can late deterioration be avoided and treated? The significance of rehabilitation training for long-term outcomes needs further research.

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## REFERENCES

- 1 Thron A. [Spinal dural arteriovenous fistulas]. *Radiologe* 2001;41:955–60.
- 2 Thron AK. Applications in spinal dural AV fistulas. In: *Thron AK. Vascular Anatomy of the Spinal Cord: Radioanatomy as the Key to Diagnosis and Treatment*. 2nd ed. Cham: Springer-Verlag International Publishing, 2016.
- 3 Krings T, Lasjaunias PL, Hans FJ, et al. Imaging in spinal vascular disease. *Neuroimaging Clin N Am* 2007;17:57–72.
- 4 Krings T, Geibprasert S: spinal dural arteriovenous fistulas. *AJNR Am J Neuroradiol* 2009;30:639–48.
- 5 Song JK, Viñuela F, Gobin YP, et al. Surgical and endovascular treatment of spinal dural arteriovenous fistulas: long-term disability assessment and prognostic factors. *J Neurosurg* 2001;94:199–204.
- 6 Steinmetz MP, Chow MM, Krishnaney AA, et al. Outcome after the treatment of spinal dural arteriovenous fistulae: a contemporary single-institution series and meta-analysis. *Neurosurgery* 2004;55:77–88.
- 7 Wakao N, Imagama S, Ito Z. Clinical outcome of treatments for spinal dural arteriovenous fistulas: results of multivariate analysis and review of the literature. *Spine* 2012;37:482–8.
- 8 Cenzato M, Debernardi A, Stefini R, et al. Spinal dural arteriovenous fistulas: outcome and prognostic factors. *Neurosurg Focus* 2012;32:E11.
- 9 Du B, Liang M, Fan C, et al. Clinical and imaging features of spinal dural arteriovenous fistula: clinical experience of 15 years for a major tertiary hospital. *World Neurosurg* 2020;138:e177–82.
- 10 Jablawi F, Schubert GA, Dafotakis M, et al. Long-Term outcome of patients with spinal dural arteriovenous fistula: the dilemma of delayed diagnosis. *AJNR Am J Neuroradiol* 2020;41:357–63.
- 11 Nagata S, Morioka T, Natori Y, et al. Factors that affect the surgical outcomes of spinal dural arteriovenous fistulas. *Surg Neurol* 2006;65:563–8.
- 12 Shinoyama M, Endo T, Takahash T, et al. Long-Term outcome of cervical and thoracolumbar dural arteriovenous fistulas with emphasis on sensory disturbance and neuropathic pain. *World Neurosurg* 2010;73:401–8.
- 13 Tacconi L, Lopez Izquierdo BC, Symon L. Outcome and prognostic factors in the surgical treatment of spinal dural arteriovenous fistulas. A long-term study. *Br J Neurosurg* 1997;11:298–305.
- 14 Cenzato M, Versari P, Righi C, et al. Spinal dural arteriovenous fistulae: analysis of outcome in relation to pretreatment indicators. *Neurosurgery* 2004;55:815–23.
- 15 von Elm E, Altman DG, Egger M, et al. The strengthening the reporting of observational studies in epidemiology (STROBE) statement: guidelines for reporting observational studies. *Ann Intern Med* 2007;147:573–7.
- 16 Aminoff MJ, Logue V. Clinical features of spinal vascular malformations. *Brain* 1974;97:197–210.



- 17 Denis F, Armstrong GW, Searls K, *et al.* Acute thoracolumbar burst fractures in the absence of neurologic deficit. A comparison between operative and nonoperative treatment. *Clin Orthop Relat Res* 1984;189:142–9.
- 18 Ma Y, Chen S, Peng C, *et al.* Clinical outcomes and prognostic factors in patients with spinal dural arteriovenous fistulas : a prospective cohort study in two Chinese centres. *BMJ Open* 2018;8:e019800.
- 19 Rizvi T, Garg A, Mishra NK, *et al.* Metachronous double spinal dural arteriovenous fistulas. Case report and review of the literature. *J Neurosurg Spine* 2006;4:503–5.
- 20 Krings T, Mull M, Reinges MHT, *et al.* Double spinal dural arteriovenous fistulas: case report and review of the literature. *Neuroradiology* 2004;46:238–42.
- 21 Muralidharan R, Mandrekar J, Lanzino G, *et al.* Prognostic value of clinical and radiological signs in the postoperative outcome of spinal dural arteriovenous fistula. *Spine* 2013;38:1188–93.
- 22 Hurst RW, Kenyon LC, Lavi E, *et al.* Spinal dural arteriovenous fistula: the pathology of venous hypertensive myelopathy. *Neurology* 1995;45:1309–13.
- 23 Cecchi PC, Musumeci A, Rizzo P, *et al.* Late deterioration of neurologic function in patients surgically treated for spinal dural arteriovenous fistulas. *Surg Neurol* 2009;72:257–61.
- 24 Zhang HQ, Chen T, S S W. The pathophysiology of venous hypertensive myelopathy—study of an animal model. *Journal of Neurosurgery: Spine* 2013;19:485–91.
- 25 Durnford AJ, Hempenstall J, Sadek AR, *et al.* Degree and duration of functional improvement on long-term follow-up of spinal dural arteriovenous fistulae occluded by endovascular and surgical treatment. *World Neurosurg* 2017;107:488–94.
- 26 Hetts SW, Moftakhar P, English JD, *et al.* Spinal dural arteriovenous fistulas and intrathecal venous drainage: correlation between digital subtraction angiography, magnetic resonance imaging, and clinical findings. *J Neurosurg Spine* 2012;16:433–40.
- 27 Horikoshi T, Hida K, Iwasaki Y, *et al.* Chronological changes in MRI findings of spinal dural arteriovenous fistula. *Surg Neurol* 2000;53:243–9.
- 28 Fugate JE, Lanzino G, Rabinstein AA. Clinical presentation and prognostic factors of spinal dural arteriovenous fistulas: an overview. *Neurosurg Focus* 2012;32:E17.