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## Health-related quality of life in patients with spinal dural arteriovenous fistulae

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## **Abstract**

**Background:** Neurological improvement in patients with spinal dural arteriovenous fistulae (SDAVF) is often partial even after adequate treatment. While treatment outcomes have been evaluated primarily on the basis of the postoperative changes in neurological deficits, outcome measures should also reflect the patient-reported outcome (PRO).

**Methods:** We conducted a health-related quality of life (HRQOL) survey in 52 SDAVF patients; 33 (63.5%) completed the short-form 36 Health Survey (SF-36) questionnaire. They were 25 males and 8 females ranging in age from 47 to 85 years (mean age 70.0 years). The mean follow-up period was 95.6 months. We analyzed the completed questionnaires and examined the clinical factors associated with their HRQOL.

**Results:** After treatment, gait- and micturition disturbances persisted in 31 (93.9%) and 31 (93.9%) of our patients; 26 (78.8%) reported chronic leg pain. The SF-36 scores of treated SDAVF patients were significantly lower than the national average of 50 for all 8 sub-items in the questionnaire. The scores for physical functioning (PF) and role-physical (RP) were particularly low. With the exception of bodily pain (BP), there was a significant negative correlation between the Aminoff-Logue scale (ALS) scores for gait and micturition and the sub-item scores. The score for BP showed a significant positive correlation with the scores for the 7 other SF-36 sub-items.

**Conclusions:** The HRQOL of treated SDAVF patients was lower than the national average with respect to both physical and mental aspects. Persistent post-treatment pain and gait- and micturition disturbances were responsible for their lower HRQOL.

## **Key words**

Health-related quality of life (HRQOL), patient-reported outcome (PRO), spinal dural arteriovenous fistulae (SDAVF), short-form 36 Health Survey (SF-36)

## **Introduction**

Spinal dural arteriovenous fistulae (SDAVF) are the most common type of spinal vascular disorders [1]. An abnormal arteriovenous shunt develops between the radicular artery and a radiculomedullary vein, typically within the dorsal surface of the dural root sleeve in the intervertebral foramen. SDAVF is observed most commonly in middle-aged men and most are located in the thoracolumbar region [7]. Congestive myelopathy due to chronic venous hypertension is the most common pathological condition contributing to neurological deficits [4].

SDAVF can be safely treated by surgery and/or embolization [10, 11, 14]. However, neurological improvement even after adequate treatment is often partial because chronic venous congestion can result in irreversible spinal cord changes [4, 6, 8, 9]. Therefore, after the initial treatment, it is important to focus on aftercare. We usually use the modified Aminoff-Logue scale (ALS) to evaluate post-treatment symptoms including gait- and micturition disturbances (Table 1) [1]. However, objective data alone are not enough for a clear understanding of the post-treatment suffering of patients with persistent symptoms. While the patient-reported outcome (PRO) is now widely used as an outcome measure in the healthcare setting [5], PROs in patients treated for SDAVF have not been reported in the literature.

We assessed the health-related quality of life (HRQOL) in patients treated for SDAVF and evaluated clinical factors that play a role in their HRQOL.

## **Patients and methods**

Our study was approved by the institutional review board of our hospital. Prior written consent was obtained from all patients.

Since 1995 we treated 73 patients with SDAVF in a multidisciplinary manner that included first-line embolization. Of these, 52 were followed; among the other 21 patients, 8 died and 13 were lost to follow-up. For a survey of their HRQOL we used the short-form 36 Health Survey (SF-36), Japanese version 2.0 [2, 3]. This survey consists of 36 questions for the quantification of the HRQOL and addresses 8 items for the self-reporting of physical functioning (PF), role-physical (RP), bodily pain (BP), general health perception (GH), vitality (VT), social functioning (SF), role-emotional (RE), and mental health (MH). Each item was scored from 0 to 100; the higher the score, the better the perceived outcome. Standardization of these domain scores using Japanese population norms showed that the mean score was 50 with a standard deviation of 10.

Of the 52 patients, 37 answered the questionnaire; 4 were excluded from further study due to incomplete answers. Consequently, the study population was comprised of 33 patients, 25 males and 8 females, whose mean age was  $70.0 \pm 1.4$  years (range 47-85 years). The mean follow-up period was  $95.6 \pm 8.6$  months (range 12 - 222 months). We analyzed the completed questionnaires and reviewed the medical records of these 33 patients to identify clinical factors that affected their HRQOL.

### **Statistical Analysis**

All data were expressed as the mean  $\pm$  standard error of the mean. A one-sample

mean comparison test versus a fixed value of 50 was used to analyze the SF-36 sub-item scores. The correlation between the SF-36 sub-item scores and the ALS score and between the BP score and the score of the 7 other SF-36 sub-items was analyzed with the nonparametric Spearman test. Differences of  $p < 0.05$  were considered statistically significant.

## **Results**

### **Patient backgrounds**

In all but one patient who presented with subarachnoid hemorrhage, myelopathy progressed gradually. The mean symptom duration from onset to the diagnosis of SDAVF was  $19.2 \pm 4.1$  months (range 1 - 96 months). Of the 33 SDAVF, 22 (66.7%) were located at the thoracic-, 5 (15.2%) at the lumbar-, and 2 (6.1%) at the sacral level; 4 (12.1%) were at the craniovertebral junction.

Treatment consisted of surgery alone (n=14, 42.4%), embolization alone (n=13, 39.4%), and embolization followed by surgery because of incomplete occlusion (n=6, 18.2%). In all cases, spinal angiography revealed the complete occlusion of SDAVF by the end of treatment. At the last follow-up, the ALS scores for gait and micturition had improved in 21 (63.6%) and 8 (24.2%) patients, respectively. As shown in Figure 1, gait- and micturition disturbances were detected in 31 (93.9%) and 31 (93.9%) patients, respectively. Chronic leg pain, i.e. pain that persisted for more than 3 months, was experienced by 26 patients (78.8%).

## SF-36 data

The scores for the 8 SF-36 sub-items obtained at the last follow-up were analyzed by a one-sample mean comparison test versus a fixed national average value of 50. As shown in Figure 2, the patient scores, especially for PF and RP, were significantly lower than the comparison value. The scores for VT and MH were closest to the comparison values.

## Correlation between SF-36 findings and clinical factors

Analysis with the Spearman test showed that the ALS scores for gait and micturition exhibited a significant negative correlation with all sub-item scores except BP (Tables 2 and 3). When we assessed the correlation between BP and the 7 other SF-36 sub-items we found that the BP score showed a significant positive correlation with the scores of the other SF-36 sub-items (Table 4). Our evaluation of the association between PRO and follow-up T2-weighted MRI findings revealed no significant relationship between the scores for the 8 SF-36 sub-items and persistent intramedullary high intensity.

## **Discussion**

The evaluation of the treatment outcome in SDAVF patients has been based primarily on postoperative changes in neurological deficits recorded on the ALS (Table 1) [1]. In a meta-analysis with a mean follow-up of 35 months, gait and micturition were

improved in 55% and 33% of operated patients, respectively [15]. According to others [14], they were improved in 80.7% and 28.6% of patients, respectively, who were followed for a mean of 103.4 months. Elsewhere we reported that gait and micturition were improved in 66% and 32% of 50 SDAVF patients we followed for a mean of 81.2 months [13]. These findings indicate that post-treatment neurological improvement is often partial and does not exclude the persistence of neurological symptoms. In fact, gait- and micturition disturbances persisted in 31 (93.9%) and 31 (93.9%) of our current patients followed for a mean of 95.6 months and 26 (78.8%) reported chronic leg pain.

As outcome measures in treated SDAVF patients should also include the PRO, we surveyed their HRQOL using the SF-36 questionnaire. We found that their SF-36 scores for all sub-items were statistically lower than the national average of 50, indicating that their HRQOL was significantly lower with respect to both physical and mental aspects. Their scores were particularly low for post-treatment PF and RP and reflective of the persistence of gait disturbance in 93.9% of our patients.

Examination of the correlation between persistent symptoms and the HRQOL indicated that, except for BP, the decline in the other 7 SF-36 sub-item scores was primarily attributable to gait- and micturition disturbances. The lack of a correlation between the BP and ALS scores indicated that post-treatment pain was independent of the severity of other persistent symptoms. The BP score exhibited a significant positive correlation with the scores of the other SF-36 sub-items, indicating that postoperative pain exerted a negative effect on the patients' HRQOL. Elsewhere we reported that most operated SDAVF patients experienced moderate to severe chronic leg pain during follow-up [12]. Active intervention



to ease their postoperative pain may be needed to improve their HRQOL.

### **Limitations**

Our study has some limitations. The study population was small and the response rate to our survey was relatively low (33 of 52, 63.5%). Because our study was retrospective, we evaluated the HRQOL of our patients at different intervals after treatment. Studies addressing these limitations are underway.

### **Conclusion**

The HRQOL of treated SDAVF patients was lower than the national average with respect to both physical and mental aspects. Post-treatment pain and gait- and micturition disturbances reduced their HRQOL.

### **Acknowledgements**

None

### **Conflicts of interest**

The authors declare that they have no conflict of interest.

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## **Figure Legends**

### **Figure 1**

Distribution of the ALS gait- and micturition scores of SDAVF patients at the last postoperative follow-up.

**Figure 2**

The scores of 8 SF-36 sub-items were analyzed by a one-sample mean comparison test versus a fixed value of 50. In SDAVF patients the score for each sub-item was significantly lower than the comparison value (\* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$ ). All values are the mean  $\pm$  standard error of the mean.

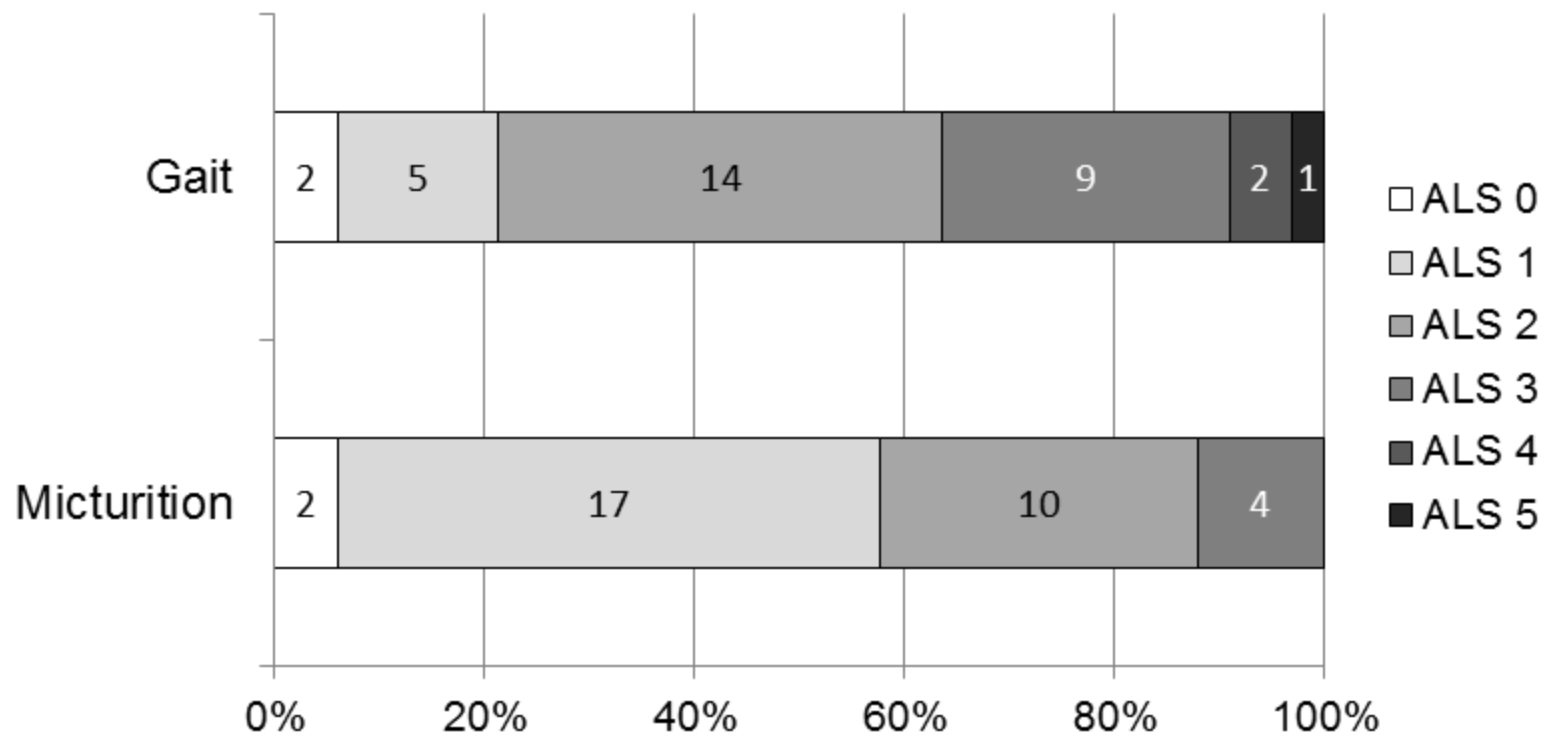


Figure 1

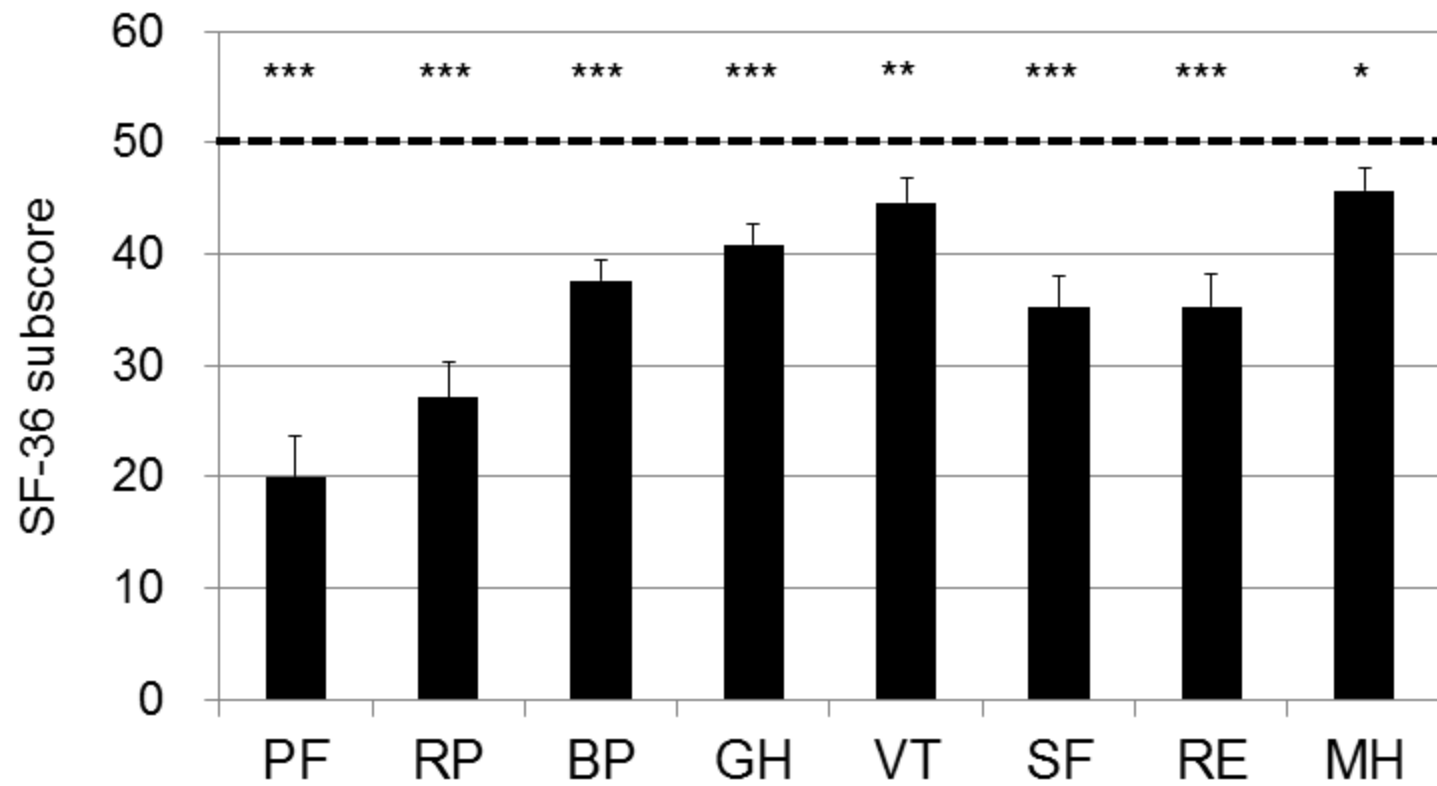


Figure 2

Table 1. Modified Aminoff-Logue scales for gait and micturition

Grade	Definition
gait	
0	normal
1	leg weakness, abnormal gait or stance, but no restriction of activity
2	restricted activity
3	requiring 1 stick for walking
4	requiring 2 sticks, crutches, or walker
5	confined to wheelchair
micturition	
0	normal
1	hesitancy, frequency, urgency
2	occasional urinary incontinence or retention
3	total incontinence or persistent retention

Table 2. Correlation between the ALS gait score and 8 SF-36 sub-items

	Correlation with the ALS gait score	
	Spearman r	p value
PF	-0.7086	<0.0001
RP	-0.7315	<0.0001
BP	-0.3183	0.0711
GH	-0.5515	0.0009
VT	-0.5492	0.0009
SF	-0.6140	0.0001
RE	-0.6216	0.0001
MH	-0.4692	0.0059



Table 3. Correlation between the ALS micturition score and the 8 SF-36 subitems

	Correlation with ALS micturition score	
	Spearman r	p value
PF	-0.7192	<0.0001
RP	-0.6307	<0.0001
BP	-0.3188	0.0705
GH	-0.5853	0.0003
VT	-0.4074	0.0186
SF	-0.5239	0.0018
RE	-0.4555	0.0077
MH	-0.4224	0.0143

Table 4. Correlation between the BP score and the score of the 7 other SF-36 sub-items

	Correlation with the BP score	
	Spearman r	p value
PF	0.5549	0.0008
RP	0.6563	<0.0001
GH	0.5871	0.0003
VT	0.6432	<0.0001
SF	0.6908	<0.0001
RE	0.5898	0.0003
MH	0.5842	0.0004