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Original Article

Comparison of surgical and endovascular approach in management of spinal dural arteriovenous fistulas: A single center experience of 27 patients

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Abstract

Background: Spinal dural arteriovenous fistula (SDAVF) is a rare spinal vascular malformation with an annual incidence of 5-10 cases per million. The data on efficacy, recurrence rates and complications of endovascular versus surgical treatment of SDAVF is limited.

Methods: We conducted a retrospective chart review of 27 adult patients with a diagnosis of SDAVF and who underwent treatment at Duke University Hospital between January 1, 1993 and December 31, 2012. We compared the outcome measures by Aminoff–Logue score (ALS) in patients who underwent treatment with endovascular embolization versus surgical ligation of fistula. We compared complication rates, recurrence rates as well as data on long-term follow up in these patients.

Results: Out of 27 patients in the study, 10 patients underwent endovascular embolization (Onyx was used in 5 patients and NBCA in 5 patients) as the first line therapy. Seventeen patients underwent surgical ligation as initial therapeutic modality. Patients in both groups showed significant improvement in clinical status (ALS) after treatment. One patient in endovascular group developed spinal infarction due to accidental embolization of medullary artery. Three patients in embolization group had recurrence of fistula during the course of follow up requiring surgical ligation. Two patients in surgical group developed local wound infection. None of the patients in surgical group had recurrence of fistula during the course of follow up.

Conclusions: Endovascular embolization and surgical ligation are effective treatment strategies for SDAVF. Our observations show that surgical ligation may offer permanent cure without any recurrence. Endovascular approach is associated with higher incidence of recurrence, especially with use of onyx.

Key Words: Aminoff–Logue scale, embolization, recurrence, spinal dural AV fistula, surgery

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INTRODUCTION

Vascular malformations of spinal cord are rare clinical entities.[6,8,15] Traditionally, they are classified into three categories depending upon the arterial supply and anatomic characteristics.[7] Spinal dural arteriovenous fistula (SDAVF) constitutes 70–80% of all spinal vascular malformations with an annual incidence of 5-10 cases per million.[1] The vascular nidus is located on the dural sheath of a spinal nerve root and is supplied by a dural artery, usually a branch of radicular or intercostal artery[3] [Figure 1]. Intradural arteriovenous malformations (AVMs) and cavernous angiomas are less common types of vascular malformations of spinal cord.

SDAVF are generally thought to be acquired lesions and usually present in mid to late adulthood.[20] The pathophysiology is thought to be congestion of the spinal cord medullary venous plexus with edema and neuronal ischemia.[3] The clinical features include radicular or axial back pain, gait imbalance, weakness, numbness, and bladder or bowel disturbances. Aminoff and Logue in their seminal observations described the clinical features and postulated a disability score to classify the severity of SDAVF [3,9] (Aminoff and Logue Scale, ALS) [Table 1].

Traditionally, there are two therapeutic options: (1) Surgical approach of laminectomies and ligation of the vascular nidus [Figure 2] and (2) An endovascular approach with focus on embolization of feeding vessel/s. [Figure 3][1-3] There is little data on superiority of one approach over the other.[3-7] We present our single center experience of 27 patients including clinical features, imaging findings, and response to various modalities of treatment, including complication and recurrence rates.

MATERIALS AND METHODS

This was a retrospective study of patients diagnosed with SDAVF who underwent evaluation and treatment at Duke University Medical Center. Duke Institutional Review Board (IRB) approved the research protocol for this study.

Study population

All adult patients (age more than 18 years at the time of presentation) who were diagnosed with SDAVF and who underwent evaluation and treatment between January 1, 1993 to December 31, 2012 were identified using the appropriate current procedural terminology (CPT) codes and inclusion characteristics using special software database program DEDUCE. DEDUCE is a comprehensive search database of all patients who underwent evaluation at the medical center over past 10 years. The list of patients was further checked manually by a team of health professionals using electronic records (eBrowser™). Patients were excluded from the study if the diagnosis was doubtful or if the feeding vessel for SDAVF was branch of one of the extracranial arteries. A total of 27 patients met inclusion criteria. Clinical details prior to intervention and following intervention at discharge and follow up were used to calculate ALS [Table 1]. Demographic details, intervention details, follow up, and complication-recurrence details were obtained from electronic medical records (eBrowser™) and from the Duke Decision Support Repository (DSR). DSR is a quality assured custom-built data warehouse containing integrated clinical and financial data of all patients admitted to the Duke Health Care System.

Table 1: Aminoff and logue scale score of disability

Classification of gait disturbance
Grade 1: Leg weakness or abnormal gait, no restricted activity
Grade 2: Grade 1 with restricted activity
Grade 3: Requires cane or similar support for walking
Grade 4: Requires walker or crutches for walking
Grade 5: Unable to stand, confined to bed or wheelchair
Classification of micturition
Grade 1: Hesitance, urgency, or frequency
Grade 2: Occasional urinary incontinence or retention
Grade 3: Total urinary incontinence or retention

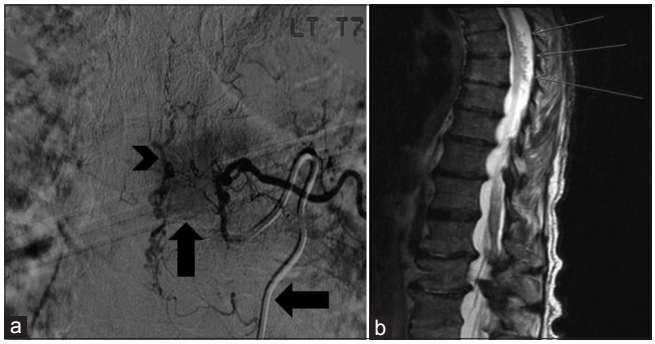

Figure 1: Spinal AV Dural fistula in one of the study subjects. (a) Spinal arteriogram shows a vascular nidus (vertical arrow), and a tortuous draining vein (arrowhead), after contrast injection through a microcatheter (horizontal arrow) at T-7 level on left side. (b) T2-weighted imaging on MRI scan of the same subject shows cord signal change and edema at midthoracic level (Green arrows)

Figure 2: Pre- and post-treatment angiograms in a 44-year-old male with SDAVF at left L1 level. (a) SDAVF at left L1 level, (Marked by arrowhead). He underwent surgical ligation of fistula; (b) post treatment angiogram

Statistical analysis

Mann–Whitney U test or Chi-square tests were used for intergroup comparisons as appropriate. Data was reported as median (interquartile range) for continuous variables and count (percentage, %) for categorical variables. Statistical significance was defined as a $P < 0.05$. All analysis was done using SPSS Inc. software version 20, Chicago, IL, USA.

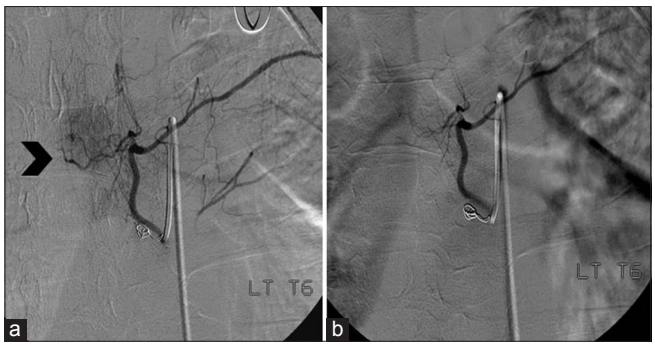

Figure 3: Pre- and posttreatment angiogram in a 37-year-old woman with SDAVF at left T6 level (a); She underwent successful embolization of the fistula (b)

RESULTS

Baseline characteristics

A total of 27 patients were diagnosed with SDAVF; 20 (74%) were male and the majority (22, 88%) patients were older than 50 years at the time of diagnosis. Majority of patients presented with back pain (93%) and lower extremity weakness (85%). Almost half the patients had sensory disturbances. A significant number of patients reported bladder difficulties (59%) while none of the patients in our study reported impotence or sexual dysfunction. All patients underwent conventional digital subtraction spinal angiogram (DSA) for accurate diagnosis and/or intervention. In addition, all patients underwent magnetic resonance imaging (MRI) as part of pretreatment evaluation. MRI scan showed T2 signal hyper intensity involving spinal cord in 24 (89%) patients. In addition, seven (26%) patients underwent

Table 2: Demographic details of study population

Patient number	Age (years)	Gender	Back Pain	Parasthesia	Lower Extremity Weakness	Bladder Symptoms	Duration from symptom onset to diagnosis (months)	Aminoff-Logue score, pre intervention	Aminoff-Logue score, post intervention	Level of Fistula
1	73	Male	Yes	No	No	No	53	5	4	T6, T7
2	75	Female	Yes	No	Yes	No	5	3	1	T5, T6
3	72	Male	Yes	Yes	Yes	Yes	23	5	4	T12
4	59	Male	Yes	Yes	Yes	Yes	8	6	4	T9-T11
5	63	Male	Yes	No	Yes	No	12	3	2	L1
6	37	Female	Yes	Yes	Yes	Yes	2	8	5	T6
7	44	Male	Yes	No	Yes	Yes	7	7	6	T8-L1
8	74	Male	Yes	Yes	Yes	Yes	8	6	5	T7, T8
9	66	Male	Yes	No	Yes	Yes	4	4	3	T7
10	35	Male	Yes	Yes	Yes	Yes	2	4	5	T6
11	50	Male	Yes	Yes	Yes	Yes	4	4	3	T8-TL1
12	45	Male	Yes	No	Yes	Yes	11	4	3	T5
13	57	Female	Yes	No	Yes	Yes	12	4	4	T11, T12
14	60	Male	Yes	Yes	No	No	13	6	6	T10, T11
15	64	Female	Yes	Yes	Yes	Yes	27	5	2	T 11
16	57	Male	Yes	No	No	No	8	4	3	C5, C6
17	20	Male	No	Yes	Yes	No	0.5	1	1	T7
18	59	Male	Yes	No	Yes	No	5	2	1	T12, L1
19	76	Male	Yes	Yes	Yes	Yes	16	4	2	T7
20	72	Male	Yes	No	Yes	Yes	4	6	5	L1
21	71	Male	Yes	No	Yes	No	45	3	6	T9-L1
22	56	Male	Yes	No	Yes	No	2	3	2	T7
23	70	Male	Yes	Yes	Yes	Yes	24	7	6	C7-T1
24	70	Female	Yes	No	No	No	8	0	0	T6
25	54	Female	Yes	Yes	Yes	Yes	25	7	6	T7-T12
26	52	Male	Yes	No	Yes	Yes	11	3	2	T12-L2
27	73	Female	No	Yes	Yes	No	4	3	1	L4
Total (27)			25	13	23	16				

magnetic resonance angiography (MRA), which was diagnostic of SDADF in four patients and was suggestive in three patients. The median ALS was 4 at preintervention and 3 at postintervention. Demographic and clinical details of all patients are summarized in Table 2.

Treatment details and outcome measures

All patients underwent DSA for diagnosis and localization of fistula. After endovascular or surgical correction, a repeat DSA was performed to assess the closure of fistula or any residual feeding vessels. All patients were followed up clinically and a repeat DSA was performed if there was suspicion of recurrence during course of follow up (worsening of residual symptoms or appearance of new symptoms or both). Out of the 27 patients in the study, 10 patients underwent endovascular embolization as the first line therapy, whereas the remaining 17 patients underwent surgical ligation as initial therapeutic modality. The decision to treatment was based on individual fistula characteristics and associated comorbidities. For example, a common segmental artery origin of the artery of Adamkiewicz and the SDAVF feeding vessel was considered a contraindication for endovascular embolization and felt suitable for surgical ligation. Onyx (ethylene vinyl alcohol) was used as an embolization material in five patients and N-butyl 2-cyanoacrylate (NBCA) was used in the remaining five patients. Patients in surgical group underwent removal of vertebral lamina at involved levels in addition to ligation-resection of the fistula. Preoperative steroids were used in one patient in embolization group as compared with four patients in surgical group. Preoperative steroids were used to reduce cord edema.

Patients in endovascular group were slightly older (mean age 65 years) as compared with those in surgical group (mean age 57 years). There was no significant difference in the demographic characteristics and clinical features in both these groups [Table 2]. The average ALS prior to procedure for both these groups were comparable (4.5 in embolization group and 4 in surgical group). The average duration of postprocedure inpatient hospital stay was 3 days in patients who underwent embolization, as compared with 4 days in patients who underwent surgical ligation and resection. The average ALS at discharge improved in both groups by one point (from 4.5 to 3.5 in the embolization group and from 4 to 3 in the surgical group). One patient in embolization group had spinal cord infarction in immediate postprocedure period. None of the patients in embolization group had evidence of local or systemic infection (pneumonia, urine tract infection within 4 weeks of procedure). Two patients in surgical group presented to the hospital within a month of discharge with local wound infection, requiring repeat inpatient admission, intravenous antibiotics, and local wound care. None of the patients in surgical group had postprocedure recurrence on follow up as compared with three patients in embolization group who presented with recurrence. All of the three patients with recurrence required surgical ligation of fistula and none of them had repeat recurrence of fistula. These results are summarized in Table 3.

DISCUSSION

SDAVF is a rare disorder with annual incidence rate of 5-10 per million. It is thought to be an acquired disease, and men are more commonly affected than women, as seen in our patient cohort.[7,8] The presentation is usually insidious and subacute. The underlying pathophysiology is thought to be medullary venous hypertension secondary to a direct abnormal communication between arterial supply and venous drainage.[2,8,9] The increased venous pressure leads to venous congestion, dilation, and tortuous medullary

Table 3: Comparison of patients undergoing embolization and surgical treatment

	Embolization (*n*=10)	Surgery (*n*=17)	*P* value*
Clinical Details			
Age (years, range)	65(57.75-72.25)	57(47.5-71.5)	0.30
Duration of symptoms before diagnosis (months, range)	14(4-24)	8(4-12)	0.20
Pretreatment MRI with T2-hyperintensity	8 (80%)	16 (94.1%)	0.26
Preoperative steroid use	1 (10%)	4 (23.5%)	0.38
Aminoff Logue Scale (Pre intervention, range)	4.5(3.75-6.25)	4(3-6)	0.33
Outcome			
Aminoff Logue Scale (Post intervention, range)	3.5(2-5.25)	3(1.5-5)	0.86
Post procedure inpatient stay (Days, range)	3(1-8)	4(3.5-5.5)	0.53
Recurrence of fistula	3 (30%)	0	0.01
Complications			
Spinal infarction	1 (10%)	0	0.18
Local wound infection	0	2 (11.8%)	0.26
Systemic infection within 4 weeks of procedure	0	0	NA
Follow up			
Duration of follow up (Months, range)	24(7.5-60)	15 (3-34)	0.32
Aminoff Logue Scale(At follow up, range)	4(1.5-6)	3 (1-5)	0.38

*Mann-Whitney U test was used to compare continuous variables and Chi-square test for categorical variables, Data reported as Median (interquartile range) for continuous variables and count (%) for categorical variables, MRI : Magnetic resonance imaging

veins. This results in compression and edema of spinal cord, which is often visualized as T2 signal hyper intensity on MRI scan[10] [Figure 1]. It is thought that increased venous pressure could contribute to decreased tissue perfusion and hypoxia. Frank infarction or hemorrhage in the substance of spinal cord is extremely rare in cases of SDAVF.[4] Almost all the patients present with varying degrees of radicular and back pain. In addition, compressive myelopathy due to edema and tissue hypoxia leads to lower extremity weakness, sensory disturbances, as well as bladder symptoms.[7,10] ALS is widely used for assessing clinical severity of SDAVF.

Advances in neuroimaging have led to better diagnostic modalities for SDAVF.[11-13] As seen from our observations, all the patients underwent MRI scan of spine to evaluate for cause of myelopathy. All of the patients had abnormal cord signal change and a significant proportion of patients (24 out of 27, 88%) had abnormal T2 signal hyper intensity suggestive of cord edema. However, these changes are nonspecific in nature and can be seen in wide variety of spinal cord pathologies such as tumors, demyelinating lesions, or infectious etiologies.[2] One of the patients was wrongly diagnosed as having demyelinating pathology before being diagnosed as SDAVF in our study. Interestingly, seven patients underwent MRA for diagnostic evaluation. MRA was suggestive of SDAVF in all the patients but was able to localize the site of fistula in only three patients. Thus, we suggest that a combination of MRI plus MRA could be a first line set of investigations in a patient suspected to have SDAVF.[2] These tests have high sensitivity and are noninvasive, without radiation exposure or theoretical risk of contrast induced kidney damage, as compared with conventional digital subtraction angiogram.[2,5,21]

The primary principle in management of SDAVF is obliteration or removal of abnormal communicating vessel/s and to relieve spinal cord edema. Traditionally, surgical laminectomy and ligation of fistula is used as definitive treatment strategy. It is an open spinal surgical procedure with inherent complications of general anesthesia, cerebrospinal fluid (CSF) leak and risk of postoperative wound infection.[1,23,6] There is an increasing interest in use of minimally invasive endovascular embolization techniques in past few years.[4,6] This treatment modality consists of advancing a micro catheter in the offending radicular artery and embolizing that artery using various materials. The most commonly used embolization particles are NBCA or onyx.[4,7,14] The major complications of endovascular approach are recurrence or incomplete embolization and accidental damage to medullary spinal vessels leading to spinal cord infarction.[7,15] As seen from our data, the endovascular arm has higher incidence of recurrence rates. Recent advances in neuroanesthesia techniques such as awake procedures and close neuromonitering may help to reduce complication rates of both endovascular and surgical procedures.[16] There is emerging data to support use of neuromonitering and provocative testing to minimize the complication rates.[17,18]

In our observational study, we have compared the functional outcomes and complications following each of the treatment modalities. There is not much data about head to head comparison of these two modalities. The patient population in both groups is comparable in clinical severity (ALS scores) as well as other demographic characteristics [Table 3]. Both of the treatment modalities are effective in immediate post-operative period for symptomatic as well as functional benefit. (ALS score improved by about one point in each group.) There appears to be some correlation between the duration of symptoms before diagnosis and degree of improvement after surgical or endovascular treatment. As noted in Table 2, patients with longer duration of symptoms before being treated were unlikely to show significant change in ALS score after intervention. This underscores the importance of timely diagnosis and urgent intervention to treat the fistula. There was no significant difference in length of postprocedure hospital stay in patients who underwent embolization (3 days) as compared with those who underwent surgical ligation (4 days) [Table 3].

None of the patients in the surgical group had recurrence of fistula during the follow up period. Three patients (30%) in the embolization group had recurrence of fistula during follow up (at 1, 4, and 26 months after initial embolization). All of these patients underwent successful surgical ligation of fistula without any further recurrence. We tried to compare the recurrence rates with different embolization materials. Five patients underwent embolization with onyx and five with NBCA. Two patients in onyx group (40%) had recurrence of fistula, as compared with one patient in NBCA group (20%). While choice of embolization material (NBCA/onyx) may affect recurrence rates, the number of patients in our study is small and hence no definitive conclusions can be made. Further the choice of embolization material depends upon advances in endovascular materials, which may act as a confounder in predicting the complication rates. Regarding postprocedure complications, one patient in the embolization group had accidental embolization of medullar artery resulting in spinal cord infarction. None of the patients in surgical group had demonstrable ischemia of cord. Regarding postprocedure infections, two patients (12%) in surgical group developed local wound infections necessitating repeat inpatient admission. Patients in both groups were followed up for several months after procedure and showed sustained improvement in ALS even at follow up. Many patients, however, remained with neurological deficits and these results are summarized in Table 3.

CONCLUSIONS

SDAVF is a rare acquired disorder of spinal cord. A combination of MRI plus MRA may offer a sensitive screening test in patients suspected to have SDAVF. DSA still remains the gold standard for diagnosis.[1] Endovascular embolization and surgical ligation are effective treatment strategies and offer immediate improvement in clinical status, which is sustained at several months' follow up. Timely diagnosis and treatment is essential to maximize the chances for recovery. Our data shows that surgical ligation may offer permanent cure without any recurrence. Endovascular approach is associated with higher incidence of recurrence and has potential to cause catastrophic injury such as spinal cord infarct. We believe that further advances in micro catheter techniques and embolization materials may reduce the recurrence rates. Our findings underscore the fact that despite modern and surgical and interventional therapy that cured the fistula, many patients were left with residual neurological deficits.

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Commentary

Surgery or embolization for spinal dural fistula?

Surgical ligation of Type I spinal dural arteriovenous fistulas (SDAVFs) remains the gold standard in the treatment of these lesions. However, with advances in endovascular techniques, several authors have proposed embolization as a valid alternative. Embolization has the main advantage of being a less invasive procedure, which is particularly appealing in patients with SDAVFs who often have significant disability by the time they are correctly diagnosed. In this issue of *Surgical Neurology International*, a retrospective observational study of patients presenting with spinal dural arteriovenous fistulas (SDAVF) and seen at Duke University Hospital between 1993 and 2012 is presented. Primary outcome measures included Aminoff–Logue scores (ALS), complications rates, and recurrence rates. Comparison is made between open surgical and compared with endovascular treatment groups, though the authors point out that the small numbers of patients in the

series preclude meaningful statistical comparison. Using a standardized outcome measurement for SDAVF treatment, the ALS, functional outcomes improved in both treatment arms, from 4.5 to 3.5 in the surgical cohort and from 4 to 3 in the endovascular cohort. Significant complications included a spinal cord infarction in the endovascular group and two wound infections requiring readmission in the surgical group. Recurrence was higher in the endovascular group with 3/10 patients requiring subsequent open surgical treatment while none of the open surgical arm required retreatment.

This thoughtful analysis attempts to directly compare surgical and endovascular treatment of SDAVF by experienced microsurgical and endovascular practitioners, although it appears that the authors have used the two techniques in a complementary fashion and attempted embolization as first choice in older patients (mean age 65 years in the embolization group versus 57 years in the surgical group). Despite the obvious appeal, embolization for the treatment of SDAVFs has significant limitations. Akin to intracranial dural arteriovenous fistulas, the *sine qua non* condition to be fulfilled for the successful and permanent obliteration of the fistula is adequate penetration in the proximal portion of the draining vein and its complete obliteration. This is not always easy to achieve given the small size of the feeding pedicles involved, which preclude distal catheterization close to the fistulous connection. Moreover, because of the small size of the vessels involved, apparent immediate angiographic obliteration may not necessarily result in complete obliteration of the fistula as micro fistulous connections may persist while not being evident any longer on catheter angiography. This is a major limitation of embolization for SDAVFs as many of these patients are already neurologically impaired and need immediate complete and permanent obliteration of the fistula. In series presented, Onyx™ (ev3, Irivne CA) was utilized in 5 of the 10 patients treated with embolization first while the remaining ones were treated (probably before Onyx™ introduction in practice) with n-Butyl cyanoacrylate (NBCA). Although some groups have reported good success with Onyx™ embolization for SDAVFs, there are major limitations associated with the use of Onyx for SDAVFs.[2] First, this agent may not achieve good enough penetration to assure complete obliteration of the proximal venous drainage.[1] Second, Onyx™ penetration into very small critical arterial pedicles, which may participate in the vascularization of the spinal cord, may not be immediately evident during the embolization (and this may have played a role in the major complication observed in this series).

Overall, despite the appeal and definitive less invasiveness, embolization for SDAVFs continues to have important limitations, which make surgical ligation a more effective procedure in a significant portion of patients with these lesions.

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