Multidisciplinary Management of Spinal Dural Arteriovenous Fistulas
Clinical Presentation and Long-Term Follow-Up in 49 Patients

J. Marc C. van Dijk, MD; Karel G. TerBrugge, MD; Robert A. Willinsky, MD; Richard I. Farb, MD; M. Christopher Wallace, MD, MSc

Background and Purpose—In the early 1980s, it was demonstrated that surgical intradural division of the shunting vein to the medullary venous plexus cures a spinal dural arteriovenous fistula (DAVF) at low morbidity. There is, however, growing literature to support endovascular therapy.

Methods—The clinical features of 49 consecutive patients with a spinal DAVF treated at a single institution between 1986 and 2001 were studied (mean age, 63 years; range, 28 to 78 years; 80% male). When possible, embolization was offered as the initial treatment. Endovascular treatment was considered adequate only if the proximal shunting vein could be occluded with liquid adhesive embolics. Motor and bladder function was evaluated with Aminoff scores an average of 32.3 months after treatment.

Results—All but 1 patient presented with myelopathy. At a mean of 2.3 years after symptom onset, 48 DAVFs were angiographically demonstrated. Since 1999, gadolinium-enhanced MR angiography was additionally performed in 7 patients to point out the level of the DAVF. Endovascular embolization could be attempted in 44 of the 48 DAVFs and resulted in a cure in 11 (25%). Thirty-five DAVFs were surgically cured; 2 patients refused surgery after failed embolization. Angiographic confirmation of the treatment result was available in 97.7% of the patients. No permanent complications of either embolization or surgery were noted. Motor and bladder function scores were significantly improved in 35 patients who had long-term follow-up (both P<0.005).

Conclusions—Endovascular treatment with liquid adhesive material provided a result equal to surgery in 25% of patients, overall resulting in a significant amelioration in the neurological status of patients with a spinal DAVF. (Stroke. 2002; 33:1578-1583.)

Key Words: arteriovenous fistula ■ central nervous system vascular malformations ■ embolization, therapeutic ■ spinal cord ■ spinal cord diseases ■ surgery

Over the last 2 decades, a definite change has taken place in the way that spinal dural arteriovenous fistulas (DAVFs) are treated. Symon et al1 and Oldfield et al2 made important contributions to the understanding and management of spinal DAVFs. Before these reports, the perimedullary venous plexus was regarded as part of the pathology, and its treatment of choice consisted of extensive intradural surgical stripping of the venous plexus. This procedure often led to postoperative neurological deficits.3 The knowledge that the myelopathy is caused by arterialization of the normal perimedullary venous plexus, resulting in venous congestion of the spinal cord, made a safe cure of spinal DAVFs possible by intradural surgical division of the shunting vein to the venous plexus. This technique has minimal side effects.4 Progressive deterioration is prevented, and improvement to some degree often is possible.

Endovascular treatment has been criticized in the past because of a high recurrence rate with the use of polyvinyl alcohol.5 However, the application of liquid adhesive embolics penetrating through the fistula into the proximal part of the shunting vein without compromising the venous drainage of the spinal cord has resulted in a more durable result.6,7

In this article, the clinical features of a consecutive group of 49 patients with spinal DAVF are presented. Results of either the endovascular or the surgical treatment and its long-term clinical follow-up are outlined.

Methods
The University of Toronto Brain Vascular Malformation Study Group has retrospectively collected data from patients who presented at the Toronto Western Hospital with vascular malformations from 1984 to 1989 and has prospectively kept data since 1989.
A group of 285 consecutive patients diagnosed with a DAVF was selected, consisting of 236 patients with cranial DAVF and 49 patients with spinal DAVF. The spinal cases, assessed between September 1986 and March 2001, are reviewed in this article. Scales were retrospectively assigned by reviewing the charts.

Clinical Features
The patients were clinically evaluated in a multidisciplinary clinic, attended by both neurosurgeons and interventional neuroradiologists. The initial and presenting symptoms were assessed, followed by a full neurological examination.

For comparison purposes, the classification scale for motor and bladder function used by Aminoff and Logue is applied to assess the clinical status before and after treatment. The Aminoff scores were retrospectively applied by a neurosurgeon not directly involved in the treatment on the basis of the full neurological exams that were noted in the patient charts.

Diagnostic Imaging
All spinal DAVFs were diagnosed with conventional digital subtraction angiography (single plane; General Electric). Since 1999, a new technique of gadolinium-enhanced MR angiography (Gd-MRA) was available to point out the level of the fistula before angiography (see the Figure). After validation of this method, the spinal segmental arterial supply was initially screened with Gd-MRA in 7 patients and subsequently confirmed by angiography. Because the need to screen all spinal segmental arteries was obviated, the time of fluoroscopy of the angiograms after Gd-MRA was compared with angiograms without this preceding localizing technique. In 1 case, Gd-MRA was applied for screening of the intracranial vessels to exclude the possibility of a cranial DAVF draining into the perimedullary venous plexus of the spinal cord.

Treatment
Endovascular embolization was initially attempted for all spinal DAVFs except cases harboring a common segmental artery supplying both the anterior spinal artery (artery of Adamkiewicz) and the fistula. Embolization was considered adequate if the proximal part of the fistulous vein could be occluded with liquid adhesive embolics, following the technique by Nimmi et al. However, embolization was considered a failure if the liquid adhesive embolics penetrated the fistula but did not penetrate in the proximal vein. In all cases, n-butyl 2-cyanoacrylate in a mixture with lipiodol was used as a liquid adhesive embolic agent administered through variable-stiffness microcatheters after superselective catheterization of the involved segmental radicular branches.

Surgery was offered after failed embolization or if embolization was considered not feasible (eg, for anatomy-related reasons such as a common trunk with the artery of Adamkiewicz). Surgical treatment consisted of a (hemi)laminctomy at the predetermined level of the fistula, followed by intradural division of the shunting vein to the perimedullary coronal venous plexus.

Follow-Up
The treatment results were confirmed by angiography or, for high-risk patients, by MRI, as indicated by Mascalchi et al, preferably during the same hospital admission as the treatment. Clinical follow-up was obtained in a multidisciplinary clinic attended by both interventional neuroradiologists and neurosurgeons. In case of any delay in recovery or clinical suspicion of recurrent disease, the imaging was repeated.

Statistical significance was calculated with the Wilcoxon rank-sum test for ordinal data and the unpaired t test for interval data.

Results
The mean age at presentation was 63 years (range, 28 to 78 years) with a male predominance. There were 39 men and 10 women. Multiplicity was encountered in 1 patient (2%) with 2 lesions at separate levels that were metachronously discovered after progressive clinical deterioration despite adequate treatment of the first lesion. The other 48 patients (98%) had solitary DAVFs. The highest level involved was the nerve root C7; the most caudal level was the nerve root S1. Ninety-four percent of the fistulas occurred at or below the T5 root level; in 70% of patients, the fistula was located on the left side (Table 2).

Clinical Features
The clinical features of the 49 patients at both symptom onset and presentation to the hospital are summarized in Table 3. At the time of diagnosis, all but 1 patient had signs and symptoms of myelopathy; 17 patients had concomitant lower motor neuron findings. Only 1 patient presented without a myelopathy; he complained of sciatica. The initial signs of the DAVFs were spastic gait in 27 patients (55%), paresthesias in 23 patients (47%), and pain in 16 patients (33%). At presentation, complaints about leg weakness or paraparesis were given by 47 patients (96%). Sensory numbness or paresthesias occurred in 44 patients (90%). Urinary incontinence or retention was a problem in 40 patients (82%), with-related bladder problems in 32 (65%). Pain was a concern in 27 patients (55%); 7 patients had back pain, 8 complained about remote pain or root pain, and 12 patients suffered both. The time interval between the initial symptoms and diagnosis was a median of 10.5 months (range, 1 day to 20.8 years). In most patients, the diagnosis was made within 2 years of presentation (Table 4).

Diagnostic Imaging
The fluoroscopy time at angiography and embolization attempt was measured in patients both with and without a
preceding Gd-MRA that pointed out the level of the DAVF. The mean time of fluoroscopy decreased with the institution of Gd-MRA from 72.3±54.0 to 52.1±22.3 minutes (mean±SD). This is not significant ($P=0.160$).

In 47 patients, the diagnosis of a spinal DAVF was made by angiography. In 2 patients, the angiogram did not reveal a spinal DAVF, although the patients had classic signs and symptoms both clinically and on MRI. In 1 patient, at selective spinal angiography, a slight delay in circulation time was demonstrated. Subsequently, all segmental arterial supply to the spinal cord was visualized, including additional visualization of the cranial circulation to exclude a cranial DAVF draining into the perimedullary plexus. However, a DAVF could not be demonstrated. Three months later, after further progression of the symptoms, the diagnosis of a spinal DAVF was made. In retrospect, it was apparent that initially the supreme intercostal artery had not been catheterized. Another patient severely worsened before an angiographic diagnosis could be made. He was diagnosed with subacute necrotizing myelopathy, the Foix-Alajouanine syndrome, confirmed by MRI. Both patients were excluded from the treatment group of this series.

**TABLE 2. Location of the DAVFs**

<table>
<thead>
<tr>
<th>Level</th>
<th>Left, n</th>
<th>Right, n</th>
<th>Total, n</th>
</tr>
</thead>
<tbody>
<tr>
<td>C7</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>T2</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>T3</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>T5</td>
<td>3</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>T6</td>
<td>5</td>
<td>2</td>
<td>7</td>
</tr>
<tr>
<td>T7</td>
<td>5</td>
<td>4</td>
<td>9</td>
</tr>
<tr>
<td>T8</td>
<td>1</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>T9</td>
<td>1</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>T10</td>
<td>4</td>
<td>1</td>
<td>5</td>
</tr>
<tr>
<td>T11</td>
<td>1</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>T12</td>
<td>3</td>
<td></td>
<td>3</td>
</tr>
<tr>
<td>L1</td>
<td>2</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>L2</td>
<td>4</td>
<td>2</td>
<td>6</td>
</tr>
<tr>
<td>L3</td>
<td>2</td>
<td></td>
<td>2</td>
</tr>
<tr>
<td>L4</td>
<td>1</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>S1</td>
<td>1</td>
<td></td>
<td>1</td>
</tr>
<tr>
<td>Total</td>
<td>35</td>
<td>15</td>
<td>50</td>
</tr>
</tbody>
</table>

**TABLE 3. Symptoms at Onset and at Presentation**

<table>
<thead>
<tr>
<th>Symptoms at presentation (in the hospital)</th>
<th>Patients</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Motor</td>
<td></td>
<td>47</td>
<td>96</td>
</tr>
<tr>
<td>Sensory</td>
<td></td>
<td>44</td>
<td>90</td>
</tr>
<tr>
<td>Bladder</td>
<td></td>
<td>40</td>
<td>82</td>
</tr>
<tr>
<td>Bowel</td>
<td></td>
<td>32</td>
<td>65</td>
</tr>
<tr>
<td>Back pain</td>
<td></td>
<td>19</td>
<td>39</td>
</tr>
<tr>
<td>Remote/root pain</td>
<td></td>
<td>20</td>
<td>41</td>
</tr>
<tr>
<td>Symptoms at onset</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Motor</td>
<td></td>
<td>27</td>
<td>55</td>
</tr>
<tr>
<td>Sensory</td>
<td></td>
<td>23</td>
<td>47</td>
</tr>
<tr>
<td>Bladder</td>
<td></td>
<td>5</td>
<td>10</td>
</tr>
<tr>
<td>Bowel</td>
<td></td>
<td>2</td>
<td>4</td>
</tr>
<tr>
<td>Pain</td>
<td></td>
<td>16</td>
<td>33</td>
</tr>
</tbody>
</table>

n=49.
TABLE 4. Duration of Symptoms Before Diagnosis

<table>
<thead>
<tr>
<th>Duration of Symptoms</th>
<th>Patients</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>n</td>
</tr>
<tr>
<td>&lt;1 mo</td>
<td>5</td>
</tr>
<tr>
<td>1–6 mo</td>
<td>11</td>
</tr>
<tr>
<td>6–12 mo</td>
<td>14</td>
</tr>
<tr>
<td>1–2 y</td>
<td>5</td>
</tr>
<tr>
<td>2–5 y</td>
<td>8</td>
</tr>
<tr>
<td>&gt;5 y</td>
<td>6</td>
</tr>
</tbody>
</table>

Treatment
The 47 patients with an angiographically confirmed diagnosis of 48 DAVFs were offered treatment. According to the management strategy, all fistulas initially had an attempt at treatment by endovascular means except 4 cases with a common segmentary artery (artery of Adamkiewicz) supplying both the anterior spinal artery and the fistula. These 4 DAVFs were treated primarily by surgery. The remaining 44 DAVFs all had an attempt at endovascular embolization, which resulted in a cure in 11 DAVFs (25%). Each embolization result was confirmed by a control angiogram at the end of the treatment. After failed embolization, 31 DAVFs were surgically cured; 2 patients refused subsequent surgery. The preceding embolization attempt did not interfere with the surgical technique in any of the patients.

Endovascular treatment caused 2 dissections of the arterial feeder of the DAVF, which inhibited further embolization therapy but did not have any clinical implication. One surgical case was complicated by an epidural hematoma that required surgical evacuation. No permanent complications of either surgery or endovascular embolization were noted.

Follow-Up
Two patients refused to undergo surgery after failed endovascular treatment, and 2 patients were not diagnosed at our institution with spinal DAVF. Therefore, with these 4 patients excluded, detailed follow-up was available in 45 fully treated patients.

Angiographic confirmation of the embolization result was available in all 11 patients who were considered cured at the time of embolization. In 4 embolization cases, a repeated angiography was done to rule out the clinical suspicion of recurrent disease. However, no recanalization of the embolized fistulas was demonstrated in those 4 delayed angiograms. Including the angiographic control directly after treatment, angiographic confirmation of the result was available in 44 of 45 patients (97.7%). A (second) control angiography was performed in 32 patients (71%) a mean of 198 days after treatment (median, 6 days; range, 2 days to 4 years). A follow-up MRI was performed in 21 patients (47%).

Clinical follow-up in the Toronto Western Hospital multidisciplinary clinic was obtained in 38 of the 45 treated patients (84%); 7 patients were sent back to the referring hospital after treatment. The mean clinical follow-up in these patients was 32.9 months (range, 12 months to 9.9 years). All remained clinically stable or improved. No deterioration was encountered.

In 35 treated patients, the Aminoff score could be applied. The median gait score before treatment was 3; after treatment, it was 2 (Z = −3.997, P < 0.005). The median bladder score before treatment was 3 compared with 2 at the latest follow-up (Z = −3.442, P = 0.001).

Four patients died of nonrelated causes during follow-up: 2 of myocardial infarction, 1 of a ruptured abdominal aneurysm, and 1 of metastatic carcinoma.

Discussion
In 1977, Kendall and Logue12 published their finding that the intradural part of a specific type of spinal arteriovenous malformation, now known as a spinal DAVF, was not to be regarded as an arterial extension of the malformation but as an arterialized venous component. This perception revolutionized the treatment of this type of lesion. Before then, total excision of the vascular lesion, including surgical stripping of the dorsal veins of the spinal cord, was advocated.3 After surgery, this procedure often led to morbidity because of a lack of the normal venous drainage of the spinal cord. Based on the new theory of Kendall and Logue,12 the validity of the surgical method of the intradural division of the arterialized shunting vein to the perimedullary venous plexus was demonstrated.2,14 Nowadays, this is considered the standard treatment, although the same result can also be achieved by endovascular embolization.6,13

Clinical Features
The demographics of the patient group in this series were similar to those of populations reported earlier.1,8,14 Male predominance was common, 80% in this series, and the mean age of diagnosis was in the early sixth decade (63.2 ± 12.1 years).

The clinical presentation of spinal DAVFs nearly always includes signs and symptoms of medullary dysfunction. Hemorrhage, a major presenting sign of cranial DAVFs, virtually never occurs in spinal lesions, probably because of the slow-flow characteristics of these lesions. Because the low thoracic spinal cord and conus are involved in most cases, anterior horn nuclei dysfunction can cause an additional lower motor neuron disease.15 This phenomenon was also reflected in our group of 49 patients.

Many theories have been proposed for the basis of the myelopathy. Venous congestion leading to hypoxia-induced ischemia is accepted as the theory of choice. This is supported by the histopathological findings of an occasional biopsy of the spinal cord16 and fits well with the report of Foix and Alajouanine,17 which described the findings of a subacute necrotizing myelopathy. Venous congestion is also reflected by a delayed venous phase on angiography.18

Diagnostic Imaging
The application of the new Gd-MRA technique published by Farb et al10 proved helpful in pointing out the fistulous level before angiography. Farb et al described that the location of a spinal DAVF could be predicted with Gd-MRA in 6 of 7 cases and in retrospect even determined in 100% of cases. Despite this promising result, Gd-MRA was never intended to replace angiography. However, after localization of the
DAVF by Gd-MRA, the commonly lengthy angiography sessions could be restricted by a mean time of 20 minutes. The reason for the nonsignificant time difference is that, because of the development of this technique in the Toronto Western Hospital, a more extensive angiographic procedure than needed was performed for validation. The expectation is that in the future Gd-MRA will significantly influence the length of angiography, which allows diagnostic procedures to be combined with treatment.

DAVF Location
In this series, 94% of the spinal DAVFs were located at or below the level of nerve root T5 (Table 2). The highest encountered level was at the C7 nerve root. Fistulas at the cervical level are very uncommon. Only 1 case report of a cervical lesion was found in the literature, which remarkably did not present with a medullary dysfunction.19 The same phenomenon was noted by Brunereau et al20 in a study of 12 patients with DAVF in the posterior fossa draining into the perimedullary venous plexus of the spinal cord. In their group, 6 patients had myelopathy and 6 patients did not. On closer look, the latter group had venous outlets at the cervical level; in contrast, no patient in the former group had cervical venous outlets. A possible explanation is that the level of the heart contributes to venous hypertension, with gravity facilitating the venous reflux to the vena cava system at the cervical level.

The fistula was located in the left side in 70% of the patients in this series. This has been reported,21 but the explanation remains unclear.

Treatment
Endovascular embolization with liquid adhesive material is reported to have a very low rate of recanalization.7–22 In experienced hands, it can be applied in a safe way and hence is clearly superior to embolization with polyvinyl alcohol, from which recanalization has been reported in up to 83% of cases.5,23 In this series, recanalization was not encountered after surgery or after endovascular treatment in the repeated angiography of 32 patients. In addition, a recurrence of the DAVF was never suspected on the basis of the repeated MRIs and frequent clinical controls.

Advantages of the endovascular technique are its noninvasiveness and the possibility of an immediate angiographic control of the treatment. Therefore, an initial attempt at endovascular embolization with liquid adhesive material can be justified. If a cure is not obtained or if an endovascular approach is not feasible, surgical therapy can always be done secondarily. In this series, the preceding embolization attempt did not interfere with the surgical technique.

Other large spinal DAVF series in the literature were reported by Nimii et al6 (49 cases), Song et al7 (27 cases), and Westphal et al21 (47 cases). In these studies, the initial success rate of endovascular treatment with liquid adhesive embolics is reported to vary between 30% and 90% of all cases. However, Nimii et al6 (90% initial success) considered penetration of the liquid adhesive embolics into the fistula without penetration into the proximal vein “adequate,” which is reflected in the high recurrence rate of 23%. Song et al7 (74% initial success) reported a failure rate of 25%, despite the fact that they performed a follow-up angiogram in only 65% of the patients. In addition, Song et al reported a 5% permanent complication rate. Following the protocol of Westphal et al,21 a multidisciplinary approach was preferred in this study to offer the patient curative treatment at the lowest morbidity. In our management strategy, endovascular therapy was attempted only if a cure could be expected within a single embolization session, with the additional rule that only penetration into the proximal vein could obviate surgery, because the risk for recurrence is very high. This approach definitely lowered the rate of endovascular success; however, it proved its worth in the absence of permanent complications and clinical recurrence.

Follow-Up
There is definitely a need for close clinical scrutiny of those patients treated both surgically and endovascularly to ensure that clinical or radiological recurrence does not occur. This scrutiny requires regular clinical exams, MRI, and at times spinal angiography. We hope that, with the expected advances in Gd-MRA, the need for invasive catheter spinal angiography will be reduced.

Conclusions
Endovascular embolization with liquid adhesive material provided a stable clinical result equal to that of surgery in 11 of 44 patients (25%) with a mean follow-up of 32.3 months. This result justifies a standard attempt at endovascular treatment with liquid adhesive embolic material before surgery. If embolization does not obtain a cure or is not feasible for anatomic reasons, surgical therapy can easily obtain a cure with low morbidity. The preceding embolization attempt does not interfere with the surgery. Overall, after a mean follow-up of 32.3 months, a significant improvement in motor and bladder function was demonstrated (both P<0.005).

Acknowledgments
We are indebted to Phillip J. Porter, Michael Tymianski, and Walter Montanera for their contribution to the University of Toronto Brain Vascular Malformation Study Group.

References


Multidisciplinary Management of Spinal Dural Arteriovenous Fistulas: Clinical Presentation and Long-Term Follow-Up in 49 Patients
J. Marc C. van Dijk, Karel G. TerBrugge, Robert A. Willinsky, Richard I. Farb and M. Christopher Wallace

*Stroke*. 2002;33:1578-1583
doi: 10.1161/01.STR.000018009.83713.06

*Stroke* is published by the American Heart Association, 7272 Greenville Avenue, Dallas, TX 75231
Copyright © 2002 American Heart Association, Inc. All rights reserved.
Print ISSN: 0039-2499. Online ISSN: 1524-4628

The online version of this article, along with updated information and services, is located on the World Wide Web at:
http://stroke.ahajournals.org/content/33/6/1578

Permissions: Requests for permissions to reproduce figures, tables, or portions of articles originally published in *Stroke* can be obtained via RightsLink, a service of the Copyright Clearance Center, not the Editorial Office. Once the online version of the published article for which permission is being requested is located, click Request Permissions in the middle column of the Web page under Services. Further information about this process is available in the Permissions and Rights Question and Answer document.

Reprints: Information about reprints can be found online at:
http://www.lww.com/reprints

Subscriptions: Information about subscribing to *Stroke* is online at:
http://stroke.ahajournals.org//subscriptions/