Spinal dural arteriovenous fistulas: experience with endovascular and surgical therapy

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Object. Although the pathophysiology of spinal dural arteriovenous fistulas (AVFs) has recently been elucidated, the optimal treatment strategy for these lesions has yet to be defined. Current management techniques include endovascular embolization or microsurgical obliteration.

Methods. The authors reviewed the records and angiograms of all patients with spinal dural AVFs treated at Massachusetts General Hospital over a 6-year period (1992–1998). During this period, it was intended initially to treat all patients with embolization and to reserve surgery for those in whom endovascular treatment failed or in cases in which pretreatment evaluation suggested that endovascular therapy would be ineffective or unsafe.

A total of 26 patients with spinal dural AVFs were treated: there were 22 men and 4 women with a mean age of 65 years (range 39–79 years). Lesions were located in the following areas: five in foramen magnum/cervical, 13 in thoracic, five in lumbar, and three in sacral. Twenty-three (88%) of 26 patients underwent embolization and three (12%) of 23 patients underwent surgery as the primary mode of treatment. Of the 23 patients in whom embolization was performed or attempted, nine (39%) ultimately required surgery. All patients were stabilized or improved following definitive treatment, as assessed by the Aminoff–Logue scores. There was one death secondary to a myocardial infarction.

Conclusions. These data demonstrate that endovascular therapy can be successful as an initial treatment for the majority of patients; however, there is a 39% failure rate, which is not observed following surgical therapy. Once a definitive therapy has been achieved using either technique virtually all patients are either stabilized or improved.

Key Words • spine • dural • arteriovenous fistula

S pinal dural AVFs, also called Type I spinal AVMs, are the most common type of spinal vascular malformations, constituting 80% of all spinal AVMs. They are also potentially the most amenable to treatment. The natural history of the disease is typically one of continued symptom progression. In a study by Aminoff and Logue, 19% of patients with spinal AVMs were disabled at 6 months and 50% of patients were disabled at 3 years. Over the last two decades the pathophysiology and clinical manifestations of these lesions have become more clear. These lesions consist of an abnormal communication between a radicular artery and a radicular vein within the dural sleeve of a nerve root. The vein communicates with the coronal venous plexus along the surface of the spinal cord. The clinical manifestations are thought to be related to the resulting venous hypertension, which causes a reduction in the perfusion pressure of the spinal cord.

Despite the improved understanding of the pathophysiology of these lesions, the optimal treatment strategy remains controversial. Endovascular embolization has been advocated as the primary mode of treatment by some, whereas others have advocated microsurgical interruption of the fistula as the preferred mode of primary treatment. Endovascular therapy has the advantages of being less invasive and allowing the possibility of making the diagnosis and providing treatment at the same session. Performing embolization requires a high degree of technical skill, the therapy is not widely available, and, most significantly, it may not provide definitive treatment of all lesions. Microsurgical therapy has the advantages of being relatively straightforward and is believed to provide definitive therapy of the lesions. Surgery has the disadvantage of being more invasive and there are other risks such as wound complications and the potential for longer hospital and rehabilitation stays.

We reviewed our experience in treating patients with spinal dural AVFs over the past 6 years in an effort to determine whether endovascular or surgical therapy should be the primary mode of treatment for spinal dural AVFs.

Clinical Material and Methods

A retrospective study was made of all patients with spinal dural AVFs who were treated at the Massachusetts General Hospital over a 6-year period between 1992 and
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1998. Our strategy during this period was to provide endovascular therapy as the first line of treatment. Liquid acrylic was used for all embolizations. Microsurgery was reserved for patients in whom endovascular therapy failed or when endovascular therapy was thought to be unsafe. Microsurgery consisted of coagulation and excision of the intradural draining vein and coagulation of the dural nidus when possible. Patients were followed clinically and underwent posttreatment MR imaging. Follow-up angiograms were obtained on an as needed basis if the patient exhibited clinical deterioration or if the endovascular team had some concern regarding the possibility of incomplete occlusion or recurrence.

The clinical data were obtained by reviewing both inpatient and outpatient records and by telephone interviews with the patients. Outcome was assessed using the Aminoff–Logue disability scale (1 = onset of leg weakness; abnormal stance or gait, without restriction of locomotor activity; 2 = restricted exercise tolerance; 3 = requires one stick or some support for walking; 4 = requires crutches or two sticks for walking; 5 = unable to stand, confined to bed or wheelchair). Disturbances of bowel and bladder function were rated as mild, moderate, or severe according to the system of Aminoff and Logue.3

Results

A total of 26 patients were identified, 22 of whom were men and four of whom were women. This was consistent with the known 4:1 male/female ratio. The average age was 65 years (range 39–79 years). The clinical presentation was relatively characteristic for these lesions: a gradual onset and progressive deterioration of neurological function including paresis, sensory disturbances, bowel/bladder dysfunction, and pain. The average duration of symptoms prior to the diagnosis was 21 months. Long-term follow-up data were available for 23 of the 26 patients. One patient died and two patients were lost to follow-up review. The mean follow-up duration was 3.5 years (range 3 months–6 years).

Fifteen of the lesions were Anson–Spetzler Type IA with a single feeding vessel. Eleven of the lesions were Type IB lesions with multiple feeding vessels.4 There were 13 lesions in the thoracic area, five in the lumbar area, and three in the sacral area (Fig. 1). Five lesions were in the foramen magnum/cervical region with venous drainage extending inferiorly causing spinal myelopathy. These were fed by the branches of the ascending pharyngeal, vertebral, and/or occipital arteries.

Although our initial treatment strategy was to perform embolization in all patients, 12 of the 26 patients ultimately required surgery (Fig. 1). The factors leading to surgery were as follows: three patients required surgery because the feeding artery was at the same level as a major arterial supply of the spinal cord; six patients underwent surgery because of incomplete occlusion of the fistula, which was detected on control angiography or because recurrence or exacerbation of symptoms prompted a follow-up angiogram; two patients developed collateral supply to the fistula; and one patient developed recanalization of the fistula. All of the foramen magnum and cervical lesions were successfully embolized without need for surgery. The Anson–Spetzler type was not obviously related to the success rate of embolization nor to the decision to proceed with surgery. Of the 23 patients initially treated with embolization 10 (43%) had Type IB lesions, and, of the 12 patients ultimately requiring surgery, five (42%) had Type IB lesions.

The time course of these events is illustrated in a Kaplan–Meier analysis shown in Fig. 2. For the purposes of this analysis recurrence was defined as clinical or radiographic failure. Long-term data were available for 21 of the 23 patients treated with embolization. Of these 21 patients, 12 had a recurrence requiring further treatment. Of these 12 patients, seven underwent surgery and five received a second endovascular treatment. Of the five patients in whom a second endovascular treatment was performed, two ultimately required surgery and one required a third embolization procedure. The recurrence-free survival duration was significantly better in patients treated with surgery than in those treated with embolization alone (Mantel–Cox log-rank test, p < 0.001).

All patients’ conditions were stabilized or improved following definitive treatment (surgery or stable embolization) as assessed by the Aminoff–Logue scores. Of the 23 patients for whom long-term follow-up data were available, 14 (61%) were improved and nine (39%) were stabilized after definitive treatment. The distribution of Aminoff–Logue scores before and after treatment is shown in Fig. 3. Overall, there was a statistically significant improvement in the long-term Aminoff–Logue scores from 3.15 ± 0.26 (mean ± standard error) prior to treatment to 2.22 ± 0.27 following treatment (Wilcoxon signed-rank test, p < 0.05). Only 39% of patients scored 1 to 2 before treatment, whereas 71% scored 1 to 2 after treatment. Four of the 23 patients experienced an improvement in bladder function following treatment but there was no statistically significant difference across the entire group. Seven patients had an improvement in bowel function and in two bowel function deteriorated, although once again there was no statistically significant difference across the entire group.

The patients were subdivided into those treated with embolization alone and those treated with surgery to de-
termine whether either treatment provided a better long-term outcome. Prior to treatment there was no significant difference in Aminoff–Logue scores between the two groups (Mann–Whitney U-test, \( p = 0.81 \)). Following treatment both groups demonstrated a significant improvement in Aminoff–Logue scores (Wilcoxon signed-rank test, \( p < 0.05 \); Fig. 4). There was a trend for patients treated with surgery to have a better long-term outcome than patients treated with embolization alone (mean scores of 1.9 ± 0.23 compared with 2.54 ± 0.49, respectively), although this trend did not reach statistical significance.

Age, duration of symptoms prior to treatment, fistula type (A or B), and pretreatment functional status were all assessed as possible predictors of outcome. The only significant predictor was the pretreatment functional status in that a worse pretreatment score predicted a less favorable functional outcome (Spearman correlation, \( p < 0.005 \)).

Complications following embolization included the following: one death due to development of acute respiratory distress syndrome of an unclear cause and a subsequent myocardial infarction; one toe ulcer secondary to embolic material traveling into the femoral artery; and one axillary hematoma. The complications following surgery included one cerebrospinal fluid leak which was treated by resuturing the wound, and one deep venous thrombosis.

**Discussion**

Our current understanding of spinal dural AVF represents a gradual evolution of ideas regarding venous hypertension and its relationship to spinal cord pathophysiology; however, despite improved understanding, the best management strategy for these lesions remains controversial.

Determining the optimal management for spinal dural AVFs requires consideration of the anatomy and physiology along with review of the empirical data obtained using different treatment strategies. The primary blood supply of the spinal cord is through the single anterior spinal artery, and by the paired posterior spinal arteries. The anterior spinal artery is initially formed by branches of the distal vertebral arteries whereas the posterior spinal arteries are supplied by branches of the posterior inferior cerebella arteries. The spinal dura and nerve roots are supplied by radicular arteries that are present at every segmental level. Most of these vessels do not contribute to the main arterial supply of the cord but rather terminate in branches supplying the dura and the nerve roots. A few of these arteries, called radiculomedullary arteries, also supply the dura and nerve roots at their respective levels and continue on to reach the spinal cord and contribute to the spinal arteries. The most prominent radiculomedullary vessels are the artery of the cervical enlargement (typically at the C-5 or C-6 level) and the artery radicularis magna, also known as the artery of Adamkiewicz (typically at a lower thoracic level on the left). In addition, the paired posterior spinal arteries also have a segmental supply provided by a third class of vessels called radiculopial arteries. The venous drainage of the spinal cord is through longitudinally oriented anterior and posterior veins that in turn drain into 30 to 70 radicular veins. The spinal venous system is valveless.

Kendall and Logue20 were the first to elucidate correctly the nature of spinal dural AVFs and to differentiate them from intramedullary spinal AVMs. The classic spinal dural AVF consists of a single arterial feeding vessel from one of the radicular vessels supplying the dura. The feeding vessel supplies an AVF that is embedded in the dural sleeve around the proximal nerve root and the adjacent spinal dura. The AVF typically drains through a single large radicular vein that communicates with the medullary venous plexus surrounding the spinal cord. For reasons that are unclear these AVFs typically occur at the lower thoracic and upper lumbar levels. Occasionally the spinal dural AVF arises from a dural branch of one of the radiculomedullary arteries: a situation that requires special consideration and will be discussed later. Because the spinal venous system is valveless, the arterial pressure is communicated to the longitudinal venous plexus of the spinal cord causing arterIALIZATION of the vessels and the characteristic dilated and tortuous appearance seen on angiograms and at surgery. Intraoperative measurements of the pressure in these draining veins have shown that the mean intraluminal pressure was 74% of the systemic arterial pressure.15,16 These studies lent considerable support to the idea that the main injury to the spinal cord occurs due to venous hypertension and the associated decrease in perfusion pressure.

Based on our current understanding of spinal dural AVFs, it has become clear that treatment should be aimed...
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at disconnecting the AVF from the spinal venous drainage thereby relieving the spinal venous system of blood at arterial pressure and restoring a normal perfusion pressure to the cord. There are currently two approaches to this end, embolization and surgery. These lesions appear particularly amenable to endovascular techniques and this treatment has been advocated by several groups; however, there are several difficulties associated with endovascular treatment that have prompted some to advocate surgery as the first line of treatment. The theoretical advantages of endovascular therapy include the possibility of diagnosing and treating the lesion in one session, although occasionally more than one session is required. In addition endovascular therapy is potentially less invasive, although a general anesthetic is still required for high-quality spinal angiography. Although arterial angiography remains the “gold standard” for localizing the lesions, authors of recent studies have suggested that MR angiography may provide adequate localization information. With continued advances in MR technology it may be possible in the near future to diagnose and localize the lesions by using MR imaging studies.

An important consideration in the endovascular treatment of spinal dural AVFs is the location of the normal arterial supply of the cord. If the AVF is at the same level as a major contribution to the anterior spinal artery then embolization is contraindicated as it may result in inadvertent occlusion of the anterior spinal artery. Therefore, a small but significant fraction of these lesions, 12% in this series, are not candidates for embolization. For the remaining lesions, adequate endovascular treatment requires a number of criteria to be successful: 1) use of liquid embolic material; 2) penetration of embolic material into the AVF and/or the proximal draining vein; 3) angiographic disappearance of the AVF on control angiography; and 4) no compromise of the venous drainage of the spinal cord. Achieving all of these results is essential but can be difficult. The use of materials other than liquid acrylic results in recanalization. Incomplete occlusion of the AVF can result in early recurrence and penetration of the embolic material distally into the venous system can worsen the venous hypertension and result in further neurological decline. The necessity of using liquid embolic material was demonstrated in a series reported by Morgan and Marsh in which 14 patients with spinal dural AVFs were treated by embolization with polyvinyl alcohol particles or microfibrillary collagen. Of the 14 patients treated in this fashion, angiographic recurrence was demonstrated in 13 (93%) and eight (57%) required surgery. The tendency for lesions treated with polyvinyl alcohol to recanalize has also been reported in other studies. In another series of 70 patients with spinal dural AVFs reported by Mourier et al., embolization was attempted in 63 patients but was successful in only 40 patients (63%). The type of embolic material was not reported. The difficulties in achieving definitive control of these lesion was shown in a more recent study by Niimi et al. In their experience with 49 patients, adequate initial embolization occurred in only 39 (80%) of 49 cases. After adequate embolization was achieved, an additional eight patients suffered a recurrence due to collateralization or to the development of a new AVF. Thus overall, only 63% of patients were definitively treated by the initial embolization. In a recent study by Westphal and Koch, 47 patients with spinal dural AVF were treated using an interdisciplinary approach combining embolization and surgery. The authors concluded that embolization should be attempted as the first mode of treatment, although only 30% of patients were cured by embolization alone and the remaining 70% required surgery. The results of our study are similar to those previous studies in that definitive treatment by embolization occurred in 14 (61%) of 23 patients when embolization was the initial mode of treatment. These results underscore the difficulty inherent in obtaining adequate embolization and the risk of recurrence even after adequate embolization has been performed. Some of the difficulty in achieving adequate embolization is due to the anatomy of the AVF. In a study of the microvascular anatomy of spinal dural AVFs, McCutcheon et al. reported that the feeding artery split into daughter vessels that coalesced one to three times to form a skein of arterial loops in the dura. This anatomical complexity explains the difficulty in achieving adequate embolization of the fistula and the tendency of the AVFs to recur following endovascular treatment.

Microsurgical therapy has the advantages of being relatively straightforward and it is reported to provide definitive therapy of the lesions. Surgery has the disadvantage of being more invasive and has its own risks such as wound complications and the potential for longer hospital and rehabilitation stays. The first successful surgical treatment of a spinal dural AVF was reported by Elsberg in 1916 in a patient with paraparesis and a thoracic sensory level. He ligated a large vein that traversed the dura adja-

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**Fig. 4.** Graphs showing distribution of Aminoff–Logue scores prior to treatment and at long-term follow up for patients treated with surgery (A) and patients treated with embolization alone (B).
cent to the T-8 nerve root, and the patient made a complete recovery. Subsequently, more aggressive surgery with stripping and resection of the entire abnormal venous complex was advocated. This led to equivocal results, which in retrospect, were likely due to worsening of venous drainage by resection of normal spinal veins. Contemporary approaches, which are based on an improved understanding of the anatomy and physiology, limit treatment directed toward interrupting the connection between the dural AVF and the medullary venous drainage. The first modern report made in 1969 on obliteration of the dural AVF alone without stripping of the dorsal veins was by Ommaya, et al. Of nine patients treated by this technique five improved and four stabilized. Since then, a number of other authors relying on surgical interruption of the fistulous connection by ligation of the draining vein or by a combination of interrupting the vein and excising the affected dura have reported similarly good results.

There are relatively few studies in which the long-term outcome of patients with spinal dural AVFs have been evaluated following treatment. The first large surgical series was reported by Logue. A total of 24 patients with spinal dural AVFs were treated by clipping of the feeding artery and excising the affected dura. Of these patients 15 (62.5%) showed some improvement in gait, seven (29%) were stabilized, and two (8%) were worsened by the procedure at a mean 5-year follow up. In a large surgical series of 46 patients reported by Symon, et al. 32 (69%) improved and seven (15%) stabilized, with a follow up ranging from 6 months to 13 years. In a series reported by Rosenblum, et al., 19 of 26 surgically treated patients improved and the remaining seven were stabilized, with a mean follow up of 3.7 years. Anson and Spetzler reported surgically treating 24 patients of whom 17 (71%) improved, six (25%) stabilized, and one (4%) worsened slightly although the length of follow up was not specified. Mouir, et al. reported on a series of 70 patients of whom 30 underwent surgery. Long-term follow up was available in 20 of these patients: 10 (50%) improved, nine (47%) stabilized, and one (3%) was considered a clinical failure. They report that the same results were obtained in patients treated with endovascular therapy (50% improved and 50% stabilized), although the specific details were not reported.

Although excision of the dural AVFs has generally been recommended, there is evidence that simple interruption of the intradural draining vein is curative in most cases. In a study by Afsar, et al., 11 patients underwent excision of the dural AVF whereas simple interruption of the intradural draining vein was performed in eight. In six patients, venous drainage was strictly intradural, and all of these patients were cured by the procedure. Two patients had both intra- and extradural venous drainage. One patient required additional therapy while the other had spontaneous thrombosis of extradural drainage following simple interruption of the intradural draining vein. The only truly long-term study was reported by Tacconi, et al. In this study 25 patients were followed for a mean of 121 months. At midterm follow up (18–36 months) 60% had improved and 24% stabilized. The results at long-term follow up were less encouraging, with 35% of patients having improved or stabilized and 63% of patients showing some evidence of deterioration. The authors suggested that patients who underwent clipping and excision of the AVF had a better long-term outcome than those who underwent clipping alone, although the numbers are too small to make a definitive conclusion.

Spinal dural AVFs most commonly occur in the lower thoracic and lumbar spine; however, authors of a number of studies have reported spinal myelopathy secondary to dural AVFs at either the foramen magnum or the sacrum. These studies illustrate the fact that the lesion may be many levels away from the symptomatic level because of the longitudinal orientation and valveless nature of the spinal veins. Thus, if a lesion at the thoracic or lumbar levels cannot be demonstrated on angiography, and the clinical suspicion is strong, then a thorough search must be conducted in the sacral and cranial vasculature. In this series five patients had dural AVFs at the cranio cervical junction and all were satisfactorily treated with embolization.

Spinal dural AVFs frequently present in an insidious manner and can be mistaken for other lesions, as evidenced by relatively long interval from onset of symptoms to diagnosis reported in this study and previous studies. For optimal management, it is important to diagnose and treat the lesion as early as possible because although some recovery can be expected it is usually not complete; and the best predictor of outcome is the pretreatment functional status.

Given the progressive nature of these lesions it seems clear that definitive treatment should be given at the first opportunity to prevent the possibility of further deterioration. During this 6-year period our intent was initially to treat all patients with embolization although 12 (46%) of 26 ultimately required surgery for a definitive treatment. All patients who were definitively treated experienced an improvement or stabilization of neurological function. Therefore, we would argue that an initial attempt at embolization is appropriate because a majority of patients experience a durable benefit and may therefore avoid having to undergo surgery. The failure rate should be kept in mind, and if there are difficulties with the embolization technique or if there is any question about its adequacy then surgery should be promptly performed to prevent a delay in achieving definitive treatment.

Conclusions

The results of this study demonstrate that endovascular therapy can be successful as an initial treatment in the majority of patients with spinal dural AVFs; however, there is a significant failure rate that is not observed following surgery. Once definitive therapy is achieved with either technique, virtually all patients stabilize or improve. Therefore, although it is reasonable to make an initial attempt at embolization, microsurgical treatment should be promptly performed if the embolization is technically difficult, or if there is any evidence that the embolization is inadequate, to minimize the delay in obtaining definitive treatment.

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