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Treatment and outcome of spinal dural arteriovenous fistulas

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Abstract Spinal dural arteriovenous fistula (dAVFs) are rare and often misdiagnosed entities. The choice between surgical treatment and embolization remains a matter of debate. We report on the cases of 18 patients (16 men, 2 women) with dAVF, who were treated surgically over an 11-year period. Patient age ranged from 32 to 84 years (mean 60 years). Six patients underwent embolization preoperatively. In three cases, angiography examinations failed to show feeding arteries on first examination at neuroradiological centers. Feeding arteries were at a different level than the fistula point in seven patients, two of them presenting with new anastomoses after embolization. Location of the fistula was midthoracic to lumbar. Seven patients were variously misdiagnosed with tumor, polyneuropathy, Guillain-Barré syndrome, syringomyelia, and knee disease. Clinical history was characterized by slowly progressive and fluctuating deterioration.

Initial symptoms were mainly sensory loss and motor weakness, lasting for between 4 and 45 months before diagnosis (mean 15 months). Recurrent fistula after operation was found in one patient. In another patient, control angiography revealed a fistula at another level, and in a third, a fistula on the contralateral side. All three patients underwent reoperation. Temporary clinical deterioration was found in four patients, seven remained unchanged, and seven improved postoperatively. An attempt at embolization should be made following diagnostic angiography. Otherwise, surgery is our recommended treatment for spinal dural fistulas, as it has a lower failure rate. Because of the progressive natural course with severe deficits, we favor an early definitive treatment.

Keywords Spinal arteriovenous malformation · Spinal dural fistula · Surgical treatment

Introduction

Spinal vascular malformations are rare and often misdiagnosed entities. In principle, we distinguish between three types of spinal vascular malformation: extradural, dural, and intradural [2, 6, 7, 11, 12]. The intradural malformations can be separated into perimedullary fistulas and the real intramedullary arteriovenous malformations (AVMs). Intradural AVMs are supplied by spinal-cord supplying ar-

teries, whereas dural malformations are supplied by meningeal arteries as branches of the radicular artery [6, 12]. Eighty percent of all spinal AVMs are dural fistulas. In contrast to the probably inborn perimedullary fistulas, dAVFs are most likely acquired [11, 12]. The fistula itself is located in the dural layer near the penetration point of the nerve root. Venous drainage runs along the spinal cord veins on the surface of the cord, because the local radicular venous drainage is missing. The high venous pressure is presumed to be the cause of clinical symptoms [2, 6, 9, 11].

The clinical picture presents a slowly progressive myelopathy and/or radiculopathy, with attacks of sudden or fluctuating deterioration up to a transverse lesion [5, 7, 12, 15].

The surgical treatment consists of intradural interruption of the draining vein with coagulation or excision of the dural fistula. Alternatively, endovascular treatment with liquid embolic material is possible [3, 8, 10, 14, 16].

This report summarizes a retrospective analysis of 18 arteriovenous spinal dural fistulas treated at the Neurosurgical Hospital of Duisburg over an 11-year period. The study is designed with special emphasis on surgical approach, extent of surgery, recurrence and clinical features.

Materials and methods

Subjects

The series consisted of 18 patients with spinal dural fistulas who underwent surgery from 1990 to 2001. Our population was composed of 16 men and 2 women. Follow-ups were available for a period of 2–65 months, with a median of 9 months. The mean patient age was 60 ± 12.4 years (range 32–84 years).

Location

The location of the fistulas was midthoracic to lumbar. The fistulas most frequently affected the L1 segment ($n=5$), with a secondary predilection for T6 ($n=3$), T11 ($n=3$), and T8 ($n=2$). Other

affected segments were affected once only (T7, T10, L2, L3, L5).

Two patients were not examined by magnetic resonance imaging (MRI) and were diagnosed by myelography. Nine patients underwent myelography, and pathologically dilated, tortuous, perimedullary vessels were found in eight patients. MRI showed in 14 of 16 cases central cord high-signal intensity on T2-weighted scans. Nine MRI scans revealed pathological vessels.

Definitive diagnosis was made by angiography. However, three angiographies failed to show feeding arteries on first examination at neuroradiological centers. One of those examinations was performed only from L1 to T7, while the feeding artery was coming from T6, as proved by the second angiography.

Information on clinical history, signs, surgical approach, and outcome was obtained retrospectively by review of the patient charts and the radiological reports.

Results

Signs and symptoms

The mean duration of symptoms was 15 months (range 4–45 months). Progressive paraparesis was the earliest and most prominent feature in five patients, and monoparesis in three patients. Sensory deficits were the first sign in six patients with dAVF, and pain was the initial complaint in three patients. One patient initially presented with a bladder disturbance (Table 1).

Acute onset with bladder disturbance or monoparesis was found in one patient each. Fluctuating symptoms with episodes of amelioration were seen in five patients.

Table 1 Clinical symptoms of patients with dural arteriovenous fistula (dAVF), with duration of symptoms in months, initial symptom and symptoms at diagnosis

No.	Duration	Initial symptom	Symptoms at diagnosis
1	28	Hypesthesia	Sensory transverse lesion L1 with spasticity, ataxia, fasciculations, left-sided
2	6	Back pain	Right-accentuated spastic paraparesis, bladder disturbance, acrodistal sensory deficit, ataxia
3	7	Paraparesis	Flaccid paraplegia, bladder disturbance, no sensory deficit, right-accentuated
4	12	Paraparesis	Paraplegia, sensory transverse lesion L1, bladder disturbance, bilateral
5	6	Bladder dysf.	Flaccid mild paraparesis, sensory transverse lesion L1, bladder disturbance, bilateral, ataxia
6	5	Monoparesis	Flaccid severe right-sided monoparesis, acrodistalsensory deficit
7	6	Ataxia	Flaccid mild paraparesis, sensory deficit L3, bladder disturbance, right-accentuated, ataxia
8	8	Ataxia	Flaccid mild atactic paraparesis, sensory deficit L3, bladder disturbance, right-accentuated
9	30	Sensory deficit	Left-accentuated paraparesis, sensory deficit L1, bladder disturbance, fasciculations
10	6	Sensory deficit	Gait ataxia, sensory transverse lesion T10, bladder disturbance, fasciculations, bilateral
11	45	Spasticity	Spastic atactic paraparesis, sensory transverse lesion T12, bladder disturbance, atrophy, left-accentuated
12	13	Monoparesis	Left-sided mild monoparesis, sensory transverse lesion T12
13	40	Monoparesis	Spastic severe left-accentuated paraparesis, sensory transverse lesion T10, bladder disturbance, ataxia
14	17	Sensory deficit	Mild left-accentuated paraparesis, sensory transverse lesion T12, bladder disturbance, ataxia
15	6	Back pain	Spastic atactic mild paraparesis, sensory transverse lesion T10, bladder disturbance, atrophy, right-accentuated
16	18	Back pain	Spastic atactic mild paraparesis, sensory deficit L4, bladder disturbance, right-accentuated
17	20	Sensory deficit	Spastic atactic mild paraparesis, sensory deficit L3, bladder disturbance, right-accentuated
18	4	Sensory deficit	Spastic atactic mild paraparesis, sensory transverse lesion T10, bladder disturbance, right-accentuated

Table 2 Muscle power and walking ability preoperatively, postoperatively and at follow-up. Three patients were lost to follow-up

No.	Muscle power ^a			Gait ^b		
	Preop.	Postop.	Follow-up	Preop.	Postop.	Follow-up
1	5	5	5	II	II	I
2	3	4	5	II	I	I
3	1	1	1	III	III	III
4	1	3		III	II	
5	4	3		III	III	
6	2	3	3	II	II	II
7	4	5	4	I	I	II
8	4	2	4	I	III	II
9	3	3	4	I	II	I
10	5	5	5	I	III	II
11	3	3	4	II	II	I
12	4	4	4	I	I	I
13	2	3	3	III	III	III
14	4	4		I	I	
15	4	4	4	II	I	I
16	4	3	4	II	III	II
17	4	4	4	I	I	I
18	4	4	4	II	I	II

^a Muscle power: 5 normal, 4 active movement against resistance, 3 active movement against gravity, 2 movement without gravity, 1 plegic)

^b Gait: I walking without aids, II walking with aids, III unable to walk

At first presentation, three and eight patients showed upper and lower motor neuron signs, respectively. Seven patients presented with mixed signs.

Sensory impairment was present in 17 patients. Two deficits were exclusively acrodistal, mimicking polyneu-

ropathy, and the others followed a transverse distribution pattern. A sensory deficit above the lesion was found in four patients, all of them presenting with an upwardly directed venous drainage. Clinical sensory level differed by up to three levels from the fistula level in upward drainage and up to seven levels in downward drainage. In two patients the sensory level represented the fistula level.

A bladder disturbance was present in 15 patients. A total of eight could walk only with aids, and four patients became unable to walk altogether (Table 2). Ataxia was present in 14 patients. Marked atrophy could be seen in two patients. Fasciculations were noted in three patients. One patient suffered from local back pain, and six patients with dAVF from radicular or pseudoradicular cramp-like pain. Right-accentuated symptoms were seen in nine patients, left-sided ones in six patients, and bilateral symptoms in three patients (Table 1).

General history

Seven patients were variously misdiagnosed with tumor, polyneuropathy, Guillain-Barré syndrome, syringomyelia, and knee disease. One patient underwent biopsy for suspected tumor, revealing pathological vessels intraoperatively. Histologically, ischemia was diagnosed. Three patients had a prior history of lumbar discectomy. Four patients suffered from diabetes, and eight patients from vascular diseases (coronary heart disease, peripheral arterial occlusive disease, venous thrombosis of the leg).

Embolization

Six dAVFs were embolized preoperatively (Table 3). Embolization was interrupted in two further cases because of

Table 3 Fistula location, level of feeding arteries, level of T2 hyperintensity on magnetic resonance imaging (MRI) scans, direction of venous drainage, angiographically embolized feeders, and postoperative outcome (+better, =unchanged, -worse) in patients with dAVF (l left, r right)

No.	Fistula loc.	Feeders	MRI	Drainage	Embol.	Outcome
1	L1 l	T12+L1 l		Up	No	=
2	L1 r	L1+L2 r		Up	No	+
3	L5 l	L5 l	T4-12	Up	No	=
4	T6 l	T6 l	T3-12	Up	No	+
5	L3 l	L3 l	T4-L1	Up	L3 l	-
6	L1 l	L1 l	T12-L1	Up	L1 l	+
7	L2 r	L2 r	T5-L2	Down	L2 r+l	+
8	T11 r	L1+L3 r	T6-L2	Up	L2 r	-
9	T11 r	T11 r		Up	No	=
10	T7 l	T6+T8 l		Both	T7 l	-
11	T8 l	T9 l	T5-L2	Both	No	=
12	T6 l	T6 l	T5-10	Both	No	=
13	L1 l	L1 l	T9-10	Up	No	+
14	T10 r	T10 r+L1 r+l	T10-L2	Up	T10 r	=
15	T6 r	T6 r	T4-6	Both	Interrupted	+
16	L1 r	L1 r	T5-L2	Up	No	-
17	T8 l	T8+T9 l	T8-9	Up	Interrupted	=
18	T11 r	T11 r	C2-T5	Up	No	+

the appearance of collateral vessels after obliteration of major feeders. These patients were primarily seen by neurologists and treated by neuroradiologists. After treatment failure, they were referred to neurosurgery. One further feeder was only partially embolized and referred for surgery. Four patients underwent definitive embolization in a second treatment. Two patients presented with new anastomoses from the next level, and one from the opposite side after embolization. One of them worsened clinically several weeks after embolization, and showed refilling of the fistula. Two further patients deteriorated clinically immediately after embolization.

Feeders

All dAVFs were located between T6 and L5. Feeding arteries were at a different level than the fistula point in seven cases, varying by up to four levels (Table 3). Two of them presented with new anastomoses from the next level after embolization. Collateral contributions from more than one artery were seen in six fistulas, usually coming from the next level. The side of feeding vessels corresponded to the predominant clinical side in only two-thirds of the patients.

Venous drainage usually ran upwards in dAVF. Only one patient showed a downward directed vein, and four bidirected ones (Table 3).

Surgery

The usual operative procedure of dural fistulas consisted of a limited hemilaminectomy at the side of the draining vein, excision or clipping of the fistula, no stripping of veins, and an intraoperative Doppler control to demonstrate the elimination of the pathological AV communication.

Ten patients were primarily referred for surgery without embolization. Their lesions were unfavorable for interventional approaches with extensive collateral vessels or feeding vessels. One of them was an emergency operation with acute onset of paraplegia, requiring immediate definitive treatment. The most common surgical approach was the hemilaminectomy, used in 15 patients. The single-level laminectomy was used in three patients. The operation time varied from 110 to 210 min (mean 150 min).

Seventeen operations were elective operations. One patient with a thoracic dAVF underwent an emergency operation with acute onset of paraplegia.

Complications were seen in two patients; one patient suffered cerebrospinal fluid leakage, which was sufficiently treated by lumbar drainage, and the other had unclear cerebral subarachnoid hemorrhage 2 weeks postoperatively, which was treated by external drainage.

Outcome and recurrence

Postoperatively, total neurological status improved in seven patients, remained stable in seven patients and worsened in four patients (Table 3).

Recurrent fistulas at the same level after operation were found in one patient with dAVF 2 months postoperatively. This went along with secondary clinical deterioration. In another patient, control angiography revealed a dural fistula at another level, and in a third, on the contralateral side. These latter two patients did not improve after the first operation. All three patients underwent reoperation. One patient showed good benefit from the second operation, two deteriorated.

Follow-up

Follow-ups were available covering a period of 9 months on average (range 2–65 months). Three patients were lost to follow-up. At follow-up, one patient with recurrent fistula showed a worse total neurological status, four remained unchanged and ten showed further improvement. Two patients improved from vegetative disturbances, but nine patients still presented with bladder disturbance. Walking improved in five patients, remained unchanged in seven patients and got worse in three patients (Table 2).

Discussion

The most common type of AVM, with an incidence of 80%, is a fistula of either low or high flow, with the nidus located in the dura [6] (Fig. 1). This nidus is always in the region of an intervertebral foramen in the vicinity of a nerve root supplied by radiculomeningeal branches of the corresponding segmental artery [6, 12, 13] (Fig. 2). Ascending or descending dural branches from neighboring segmental arteries were demonstrated as additional feeders in six of our patients. The cluster of small dural arteries shows a distinct point of communication with usually one arterialized radicular vein on the inner surface of the dura.

Dural AV fistulas first become symptomatic in later adult life. The strong preponderance of male patients is well known [7]. Comparable to other series, in the majority of the dural lesions the AV nidus was in the low-thoracic or lumbar region [12, 16]. The direction of predominant venous drainage of most dAVFs was rostral [12].

The clinical course in general is progressive, with intermissions [7]. Clinical symptoms were a variable combination of lower motor neuron lesion, sphincter disturbance, sensory transverse lesion and partly additional signs of upper motor neuron involvement [13]. Subarachnoid bleeding is only found in AVMs, which corresponds to the findings of Rosenblum et al. [12]. A distinct sensory level

Fig. 1 Angiography demonstrates several meningeal feeders arising from the seventh intercostal artery on the left side, shunting into a dilated and elongated radicular vein, with early upward and then bidirectional drainage into dorsal venous plexus along the spinal cord (⇒ fistula point)

was present in most patients [12]. The frequent discrepancy between the location of the dural fistula and the spinal level responsible for clinical symptoms also supports the theory of inadequacy of the venous drainage system to cope with the blood volume [6, 7].

On average, neurological symptoms stabilized within 1 year in the group of Behrens and Thron [4]. The greatest improvement was found in the category of muscle strength (67%), followed by walking distance (57%). The greatest deteriorations were in male potency (28%) and pain (24%). Tacconi et al. [15] reported on clinical improvement and stabilization following intervention in 84% of patients. However, at last follow-up (mean 147 months), symptoms were improved or stabilized in only 35%, with the remainder deteriorating significantly.

Meisel et al. [8] favor embolization with a permanent liquid substance (NBCA N-butyl 2-cyanoacrylate) as the method of choice in spinal cord AVMs and fistulas. In five of seven patients, the malformation was completely occluded in one therapeutic session. Niimi et al. [10] recommend embolization with an acrylic material as the first choice of treatment for dAVF, unless a spinal cord artery shares the same pedicle as the feeder of the dAVF. The initial success rate of embolization increased from 80 to 87% after the introduction of variable stiffness microcatheters. A total of eight patients (23%) underwent a second embolization for recurrent dAVF after adequate initial embolization. Earlier reports about the use of particulate embolization revealed high long-term failure rates in more than 60% of patients [2, 10, 11]. Recurrence can occur by either recanalization of the embolized vessels, collateralization, or development of the new fistula [10]. In

Fig. 2 Intraoperative aspect of a dural arteriovenous fistula (dAVF) at L1, before (*left*) and after (*right*) clipping of the arterialized vein. The fistula itself is located close to the penetration point of the nerve root (⇒ fistula point)

the series of Song et al. [14], fistulas recurred in 15% of patients despite the use of a liquid embolization material. Westphal and Koch [16] reported on 12 of 47 patients surgically treated after embolization because of the recruitment of collateral vessels. Eight of 47 patients underwent surgery after unsuccessful embolization. Definitive embolization was achieved for 15 patients. Nevertheless, they recommend attempts at embolization at the time of angiography, because no adverse effects were recorded and a 30% chance of a cure was given. Partial embolization with reduction of flow through the fistula resulted in immediate neurological improvement for some of their patients.

Song et al. [14] did not note any significant differences in clinical outcome among treatment groups – surgery alone (7 cases), embolization alone (16 cases), and embolization followed by surgery (7 cases).

Surgical technique has mostly been limited to simply dividing the draining vein, without either stripping the enlarged pial veins or attacking the fistula directly [11, 12]. Tacconi et al. [15] emphasize that excision or coagulation of the nidus, as well as disconnection of the draining vein, offer better results in the long term than clipping of the vein alone. In cerebral dural AV fistulas, ligation of feeding arteries is ineffective because of the recruitment of collateral flow. Successful treatment requires obliteration of the nidus [4]. Afshar et al. [1] found that in most patients with dAVF and only intrathecal medullary venous drainage, surgical interruption of the intradural draining vein provides lasting and curative treatment. Only in patients with both intra- and extradural drainage is complete excision of the fistula or interruption of intra- and extradural venous drainage of the fistula indicated. In patients in whom a common vessel supplies the spinal cord

and the AVF, simple surgical interruption of the draining vein is the treatment of choice. All pathways of venous drainage are interrupted.

The operation is as an easy, safe and definitive treatment of spinal dAVF. We prefer an operative excision, in order to exclude the collateral supply and to preserve the small radicular spinal cord feeders, which may originate close by from a common meningoradicular artery outside the dura. The unfavorable time course of the disease can be stopped and sometimes reversed with the operation. Treatment failure of embolization is still higher than that

of surgery. However, embolization has the advantage of low general morbidity, and may be helpful in patients with reduced physical condition. The chance of complete cure following embolization with a permanent agent is considered to be inferior to surgery. In case of extensive collateral vessels or feeding vessels, the primary option is surgery. We also recommend surgery as the definitive treatment of dAVF in recurrent fistulas with collateralization from the opposite side or other levels. Otherwise, an attempt at embolization following diagnostic angiography should be made.

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