Endovascular treatment of intracranial dural arteriovenous fistulas with spinal perimedullary venous drainage

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Intracranial dural arteriovenous (AV) fistulas with spinal perimedullary venous drainage are rare lesions that have distinctive clinical, radiological, and therapeutic aspects. Five patients presented with an ascending myelopathy, which extended to involve the brain stem in three cases. Myelography and magnetic resonance imaging showed slightly dilated spinal perimedullary vessels. Spinal angiograms were normal in the arterial phase. Diagnosis was only possible after cerebral angiography, which demonstrated posterior fossa AV fistulas fed by meningeal arteries and draining into spinal perimedullary veins. Endovascular treatment alone resulted in angiographic obliteration of the lesion in three patients. Two patients required surgery in addition to endovascular therapy. One patient died postoperatively, and in one a transient complication of embolization was observed. Improvement after treatment was good in two cases and fair in two. Transverse sinus thrombosis was observed in three cases and was probably the cause of the aberrant venous drainage of the fistula into the spinal perimedullary veins. The pathophysiology is related to spinal cord venous hypertension. These lesions were classified as Type 5 in the Djindjian and Merland classification of dural intracranial AV fistulas. Endovascular therapy is a safe effective method in the treatment of these fistulas and should be tried first.

Key Words: dural arteriovenous fistula · spinal arteriovenous malformation · vascular myelopathy · embolization · hypertension

Intracranial dural arteriovenous (AV) fistulas account for 15% of all cerebrovascular malformations. Their feeding arteries are of meningeal origin, the shunt is located in the intracranial dura mater, and their venous drainage is intracranial into a sinus or a cortical vein. Intracranial dural AV fistulas with spinal perimedullary venous drainage are rare. Since the first case was published in 1982, occasional cases have been reported. These cases are diagnostically challenging because the presenting symptoms consist of progressive spinal cord deficits related to an ascending myelopathy. Myelography and magnetic resonance (MR) imaging show dilated perimedullary vessels; however, angiograms of the spinal cord are normal (at least in the arterial phase). This is because the fistula is localized on the intracranial dura mater and can be demonstrated only by cerebral angiography. We review our experience with five such patients and present the clinical and radiological findings, as well as the results of endovascular treatment.

Clinical Material and Methods

Case Material

Over the past 10 years, we have treated five patients with spinal cord dysfunction who were found to have intracranial dural AV fistulas draining into the spinal perimedullary veins. Two were men and three were women. The mean age was 52 years (range 35 to 69 years). The first patient in our series corresponds to the case published in 1982 by Womart, et al.

The presenting signs and clinical course are summarized in Table 1. Four patients had suffered progressive bouts of ascending myelopathy; one (Case 3) had exhibited acute deterioration after a cervical laminectomy performed for spinal stenosis. The clinical progression...
Intracranial dural AV fistula with spinal drainage

**TABLE 1**

*Clinical data and treatment of five patients with intracranial dural AV fistulas and spinal perimedullary venous drainage*

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Sex, Age (yrs)</th>
<th>Presenting Symptoms</th>
<th>Clinical Course</th>
<th>Delay in Diagnosis (mos)</th>
<th>Embolization of Artery (material)</th>
<th>Surgery Performed</th>
<th>Angiography Result</th>
<th>Clinical Result</th>
<th>Follow-Up Period</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>F, 35 (1981)</td>
<td>headaches, left ear, dull, then bouts of paraparesis</td>
<td>quadriplegia, bulbar signs</td>
<td>6</td>
<td>OccA &amp; MMA (Gelfoam, clot)</td>
<td>no</td>
<td>cure</td>
<td>good</td>
<td>6 yrs</td>
</tr>
<tr>
<td>2</td>
<td>M, 37 (1981)</td>
<td>bouts of paraparesis</td>
<td>quadriplegia, bulbar signs</td>
<td>9</td>
<td>MMA (dura mater)</td>
<td>yes</td>
<td>died</td>
<td>postop</td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>M, 53 (1982)</td>
<td>progressive paraparesis, deteriorated postcervical laminectomy</td>
<td>quadriplegia</td>
<td>5</td>
<td>meningohypophyseal trunk (IBCA)</td>
<td>no</td>
<td>cure</td>
<td>fair</td>
<td>6 yrs</td>
</tr>
<tr>
<td>4</td>
<td>M, 69 (1988)</td>
<td>bouts of paraparesis</td>
<td>paraplegia</td>
<td>12</td>
<td>ascending PhA &amp; OccA (PVA)</td>
<td>yes, twice</td>
<td>cure</td>
<td>good</td>
<td>1 yr</td>
</tr>
<tr>
<td>5</td>
<td>F, 68 (1989)</td>
<td>headaches, followed by paraparesis</td>
<td>quadriplegia, brain-stem signs</td>
<td>4</td>
<td>OccA, ascending PhA, &amp; MMA (PVA, NBCA)</td>
<td>no</td>
<td>cure</td>
<td>fair</td>
<td>8 mos</td>
</tr>
</tbody>
</table>

* Abbreviations: AV = arteriovenous; OccA = occipital artery; MMA = middle meningeal artery; IBCA = isobutyl cyanoacrylate; PhA = pharyngeal artery; PVA = polyvinyl alcohol; NBCA = N-butyl cyanoacrylate.
† Date of presentation.

**TABLE 2**

*Radiological data of five patients with intracranial dural AV fistula with spinal perimedullary venous drainage*

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Myelography Findings</th>
<th>MR Findings</th>
<th>Site of Fistula (afferent arteries)</th>
<th>Sinus Thrombosis</th>
<th>Visualization of Adam Venous Drainage</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>dilated cervical perimedullary vessels</td>
<td>not done</td>
<td>lateral sinus (MMA, OccA)</td>
<td>thrombosis lt transverse sinus</td>
<td>not studied</td>
</tr>
<tr>
<td>2</td>
<td>dilated cervical dorsal lumbar perimedullary vessels</td>
<td>not done</td>
<td>petrous apex (MMA)</td>
<td>none</td>
<td>not visualized</td>
</tr>
<tr>
<td>3</td>
<td>slightly dilated cervical perimedullary vessels</td>
<td>not done</td>
<td>tentorium cerebelli (meningohypophyseal artery)</td>
<td>none</td>
<td>not studied</td>
</tr>
<tr>
<td>4</td>
<td>dilated cervical dorsal lumbar perimedullary vessels</td>
<td>dilated cervical dorsal-lumbar perimedullary vessels</td>
<td>superior petrous sinus (Ascending PhA, OccA)</td>
<td>thrombosis lt transverse sinus</td>
<td>present on 1st angio during a clinical remission; not seen on 2nd angio</td>
</tr>
<tr>
<td>5</td>
<td>dilated cervical dorsal lumbar perimedullary vessels</td>
<td>dilated cervical dorsal-lumbar perimedullary vessels</td>
<td>superior petrous sinus (Ascending PhA, OccA, MMA)</td>
<td>thrombosis lt transverse sinus</td>
<td>not visualized</td>
</tr>
</tbody>
</table>

* Abbreviations: AV = arteriovenous; MR = magnetic resonance; Adam = artery of Adamkiewicz; MMA = middle meningeal artery; OccA = occipital artery; PhA = pharyngeal artery; angio = angiogram.

was rapid over a few months in a caudocranial direction and resulted in urinary retention, disabling quadriparesis, a sensory level and, finally, brain-stem signs in three cases.

**Radiological Investigation**

Radiological investigations are summarized in Table 2. Magnetic resonance imaging of the spine was performed with a 0.5-tesla unit. Short spin-echo sequences were obtained in the sagittal plane in the two most recently treated patients (Cases 4 and 5). A long spin-echo sequence in the sagittal plane was obtained in Case 4. Spinal cord arteriography was performed under light sedative anesthesia and local anesthesia, with opacification of the sacral, lumbar, thoracic, vertebral, and deep cervical arteries. In three cases, study of the venous phase of the artery of Adamkiewicz was performed by injecting selectively the segmental artery with 10 cc contrast material over a period of 7 seconds. Arteriographic films were obtained at 9, 13, 17, 20, 25, and 30
seconds. The normal venous drainage being at 13 seconds, it was considered "not visualized" when it was not seen by 30 seconds.

**Illustrative Cases**

**Case 3**

This 53-year-old man presented with upper-extremity and low-back pain. Examination revealed a mild hyperreflexia. Cervical myelography was interpreted as showing cervical stenosis with spinal cord compression. A C3–7 laminectomy was performed and, 24 hours later, the patient became quadriplegic with sphincter dysfunction and a C-8 sensory level. Retrospective analysis of the myelogram revealed slightly dilated cervical perimedullary vessels. Total spinal angiography was normal; however, the venous phase of the Adamkiewicz artery was not studied. Cerebral arteriography showed a dural AV fistula of the left tentorium cerebelli, fed by the meningohypophyseal trunk. The fistula drained into the anterior pontomesencephalic vein, then into the anterior and posterior spinal perimedullary veins. A first attempt at embolization resulted in a transient right deficit. A second embolization was carried out using isobutyl cyanoacrylate in the meningohypophyseal trunk with a protection balloon closing the internal carotid artery. Immediately after embolization the arm strength improved. Recovery was fair; 6 years later the patient is still paraplegic and has urgency of micturition.

**Case 5**

This 68-year-old woman presented with acute onset of left-sided headaches and cervical pain. This was accompanied by paraparesis, which resolved over several days, followed by progressive lower-extremity weakness (right > left) and urinary retention over a 3-month period. On examination, the patient had a spastic paraparesis, weakness in the upper extremities, a T-4 sensory level, horizontal nystagmus, and swallowing difficulties. Lumbar and cervical myelograms showed serpiginous perimedullary vessels (Fig. 1), and MR images (short sequences only) revealed dilated perimedullary vessels (Fig. 2). Spinal arteriography did not demonstrate a dural AV fistula, but the venous phase of the artery of Adamkiewicz at the right L-1 level was not visualized. Cerebral angiography (Fig. 3) revealed an intracranial dural AV fistula of the left superior petrosal sinus fed by the petrosal branch of the left middle meningeal artery, the meningeal branch of the ascending pharyngeal artery, and meningeal branches of the occipital artery. The fistula drained into the anterior pontomesencephalic vein and the spinal perimedullary veins. The left transverse sinus was thrombosed. Embolization of the occipital and ascending pharyngeal arteries was performed using 300- to 600-μ particles of polyvinyl alcohol (Ivalon*). The patient's swallowing

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* Ivalon supplied by Nycomed-Ingenor, Paris, France.
difficulties improved over the following few days. Further embolization was then carried out via the remaining petrosal branch of the middle meningeal artery using N-butyl cyanoacrylate glue (Fig. 4), obliterating the superior petrosal sinus and the origin of the draining vein (Fig. 5 left). Angiography with injection of the artery of Adamkiewicz now showed normal venous drainage at 13 seconds (Fig. 5 right). The patient’s condition continued to improve; 4 months later there remained only a mild sphincter disturbance and she became ambulatory with canes.

**Results**

The modalities and results of treatment are summarized in Table 1. In three patients, the dural AV fistula was obliterated by embolization alone. Two patients required surgery because the dural AV fistula reopened after particulate embolization. There was one postoperative death and one embolization-related transient ischemic complication. The clinical follow-up period in the four survivors was 8 months to 6 years (mean 3.4 years). Angiography was performed routinely 3 months after treatment, but no later because no late clinical deterioration occurred. Following treatment, a “good result” (patient independently ambulatory) was obtained in two cases with a mean follow-up period of 3.5 years and a “fair result” (patient with incomplete recovery and not independently ambulatory) in two cases with a mean follow-up period of 3.3 years. Recovery after embolization or surgery (analyzed in four years).

**Fig. 3.** Left external carotid artery angiograms in Case 5. **Left:** Angiogram demonstrating a dural arteriovenous fistula supplied by the petrosal branches of the middle meningeal artery (large arrowhead) and the posterior branch of the middle meningeal artery (small arrowhead). The fistula is located on the superior petrosal sinus (black arrow) and drains into the anterior spinal cervical vein (open arrow) through the perimesencephalic veins. **Center:** Angiogram, lateral cervical view, late phase, demonstrating the cervical anterior (open arrow) and posterior (black arrow) perimedullary venous drainage. **Right:** Angiogram, frontal view, venous phase, showing no filling of the left transverse sinus.

**Fig. 4.** Case 5. Angiogram showing selective catheterization of the petrosal branch of the middle meningeal artery (arrowheads) feeding the superior petrosal sinus (arrow). The dotted line represents the microcatheter.

**Fig. 5.** Case 5. **Left:** Standard x-ray film obtained after embolization. Note the cast of the N-butyl cyanoacrylate glue after embolization of the superior petrosal sinus (arrowhead). A follow-up angiogram performed after embolization (not shown) revealed complete closure of the fistula. **Right:** Spinal cord angiogram, right lumbar artery injection at L-1, showing the venous phase of the artery of Adamkiewicz at 13 seconds. The spinal cord venous drainage is now visualized after a normal delay (arrow). Before embolization, the draining veins were not seen.
of the five cases) occurred in a rostrocaudal direction; brain-stem symptoms and upper-extremity strength recovered quickly in all cases, while sphincter disturbances and lower-extremity deficit resolved in only two cases.

**Discussion**

Four of the five patients presented with partially resolving episodes of an ascending myelopathy. One patient (Case 3) became tetraplegic after undergoing cervical laminectomy for spinal stenosis. A similar postsurgical deterioration was observed in a previously reported case,20 stressing the importance of seeking an associated fistula when spinal surgery is considered. Brain-stem symptoms were the last to appear and were related to the pattern of the venous drainage, which involved the anterior pontomesencephalic vein. Dural fistulas in the cervical or foramen magnum region with drainage into the cervical perimedullary veins only are not accompanied by brain-stem signs.2,6,15,16,18

**Radiological Diagnosis**

Magnetic resonance images were available in only two cases and both showed dilated spinal perimedullary vessels. Reviewing the reported cases with MR imaging, Wrobel, et al.,20 also found dilated vessels in two of three cases. In one case they described enlargement and a bright signal of the cord. It would be interesting to perform long spin-echo sequences in an axial plane in order to disclose intramedullary signal abnormalities; however, those sequences were not used in our cases. As previously discussed,6 total spinal myelography must be performed in cases of myelopathy of unknown origin, even when the MR image is negative. Myelography was carried out in all of our five patients and in each showed dilated spinal perimedullary vessels. In a patient presenting with an ascending myelopathy, MR imaging should be performed first followed by spinal and cerebral angiography if dilated perimedullary vessels are found. If MR imaging is normal, complete spinal myelography must be performed; evidence of dilated spinal perimedullary vessels indicates the need for angiography, which must include cerebral arteriography when spinal angiography is negative.

**Artery of Adamkiewicz Angiography and Spinal Cord Drainage**

Usually the venous drainage of the conus medullaris is visualized 13 seconds after injection of the artery of Adamkiewicz. Stasis in the artery of Adamkiewicz and absence of its venous phase have been well known in cases of dural spinal AV fistula since 1979.10,11 and their diagnostic value has recently been emphasized.18 Before treatment, venous drainage from the artery of Adamkiewicz was studied in Cases 2, 4, and 5. In Cases 2 and 5, there was arterial stagnation and nonvisualization of the venous phase. In Case 4 the first angiography was performed during a clinical remission, and a normal venous phase was found. However, the myelogram obtained at the same time showed dilated cervical perimedullary vessels. Five months later, the patient became paraplegic; angiography in the venous phase showed arterial stagnation of the artery of Adamkiewicz, and the venous drainage was not visualized. This finding suggested spinal cord venous hypertension and spinal cord damage. However, a normal venous phase of the artery of Adamkiewicz does not eliminate the diagnosis of dural intracranial AV fistula with spinal perimedullary venous drainage if angiography is performed during a remission, as has been shown in Case 4. After treatment, the venous phase of the artery of Adamkiewicz was studied in Cases 4 and 5 and drainage was visualized in both. This has not been noted previously and may be correlated to a decrease of spinal cord venous hypertension after closure of the AV fistula.

**Intracranial Dural AV Fistula Recruitment of Spinal Perimedullary Veins**

Transverse sinus thrombosis ipsilateral to the fistula was observed in three of the present cases (Cases 1, 4, and 5). This may be a clue to the etiology of these dural AV fistulas. Anatomical connections between anterior and posterior spinal veins and the veins of the posterior fossa are well known. These can even drain intracranial dural AV fistulas, as discussed by Womant, et al.,19 and Wrobel, et al.20 In Case 1, the ascending myelopathy was preceded 6 months earlier by transient episodes of tinnitus, headaches, and nausea. This may represent a transverse sinus fistula (tinnitus) followed by sinus thrombosis (headaches and nausea), resulting in diversion of flow of the fistula in the spinal perimedullary veins (ascending myelopathy). In Cases 4 and 5, the AV fistula was located on the superior petrosal sinus, which is an anastomotic pathway between the transverse sinus and the cavernous sinus; in both patients the transverse sinus was not seen and no drainage from the AV fistula entered the cavernous sinus, suggesting extensive sinus thrombosis. In Cases 1, 4, and 5 sinus thrombosis was the likely cause of the retrograde venous drainage into

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**TABLE 3**

**Modification of classification of intracranial dural AV fistula by venous drainage**

<table>
<thead>
<tr>
<th>Type</th>
<th>Venous Drainage</th>
<th>Symptoms</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>tinnitus, no neurological risk</td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>free flow drainage in a sinus</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>drainage into a sinus with retrograde flow in sinus or a cortical vein</td>
<td>above symptoms plus seizure, cranial hypertension, risk of hemorrhage from cortical venous reflux</td>
</tr>
<tr>
<td>3</td>
<td>cortical venous drainage</td>
<td>above symptoms, plus cerebral hemorrhage</td>
</tr>
<tr>
<td>4</td>
<td>cortical venous drainage with giant venous ectasia</td>
<td>above symptoms, plus mass effect</td>
</tr>
<tr>
<td>5</td>
<td>perimedullary venous drainage</td>
<td>above symptoms, plus ascending myelopathy</td>
</tr>
</tbody>
</table>

* Original classification by Djindjian and Merland. AV = arteriovenous.
Intracranial dural AV fistula with spinal drainage

the anterior pontomesencephalic and spinal perimedullary veins.

Pathophysiology

The pathophysiology of spinal cord injury in such cases might be due to hemorrhage, arterial steal, spinal cord compression, or venous hypertension. No cases of hemorrhage or spinal cord compression were observed in this series. Arterial steal cannot be incriminated because the feeding arteries were the intracranial meningeal vessels that were not supplying the spinal cord. Therefore, the pathophysiological mechanism of the clinical symptoms in these cases could only be venous hypertension. How such a small, low-flow intracranial dural fistula can cause spinal cord venous hypertension is not clearly understood.

Treatment

Embolization should be performed as the initial procedure since microcatheter technology has improved the feasibility of this approach. The goal is to obtain complete and permanent obliteration; for this purpose, cyanoacrylate remains the best embolic agent, as has been demonstrated in two cases of our series. Failure of distal superselective catheterization occurred in three cases, however, and this was a contraindication to cyanoacrylate embolization. In the three cases, particular embolization was performed, but revascularization occurred in two of them and surgical excision was required.

A New Type of Intracranial Dural AV Fistula

Intracranial dural fistulas have been classified based on venous drainage into four types by Djindjian and Merland.4 As previously stated by several authors,3,5-7 symptoms correlate with the venous drainage. We propose to add a new category to the existing classification as Type 5 (Table 3): intracranial dural AV fistulas with spinal perimedullary venous drainage, which can be responsible for an ascending myelopathy.

References


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