Microvascular anatomy of spinal dural arteriovenous fistulas: arteriovenous connections and their relationships with the dura mater

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OBJECT The microvascular anatomy of spinal dural arteriovenous fistulas (AVFs), especially the relationships of the vessels with the dura mater, has yet to be angiographically demonstrated in detail and proven histologically.

METHODS From January 2012 through April 2014, a total of 7 patients with spinal dural AVFs in the thoracic region underwent open microsurgical obliteration at Tokyo Metropolitan Neurological Hospital. The microvascular anatomy of spinal dural AVFs was comprehensively assessed by using advanced microangiography, including 3D computer graphics and intraoperative indocyanine green video angiography, and by histological findings.

RESULTS The 2 microangiography techniques revealed the spatial course and in vivo blood flow of the meningeal vessels and their relationships with the dura mater in sufficient detail. The meningeal branch of the intercostal artery split into multiple meningeal vessels on the outer dural surface adjacent to the root sleeve. After crossing the dura mater to the inner dural surface, these vessels gathered and joined a single intradural draining vessel. On the inner dural surface, the single draining vessel was fed by the surrounding multiple meningeal vessels, which appeared to be caput medusae.

Histological findings revealed that the structure of the meningeal branch of the intercostal artery corresponded to that of a normal artery. The structure of intradural draining vessels corresponded to that of a vein modified by retrograde arterial inflow. On the inner dural surface, more than 1 meningeal artery gathered and joined with the proximal radiculomedullary vein.

CONCLUSIONS Spinal dural AVFs are located on the inner dural surface, where multiple direct AV connections between more than 1 meningeal feeding artery and a single proximal radiculomedullary vein occur at the site where the vein connects to the dura mater.

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KEY WORDS arteriovenous malformation; dural arteriovenous fistula; arteriovenous shunt; angioarchitecture; pathology; endovascular treatment; surgical treatment; vascular disorders
artery, AV connection, and vein, and their relationships with the dura mater by means of comprehensive assessments of advanced microangiography and histological examination. We discuss the pathogenesis and treatment of these lesions.

Methods

This study protocol was approved by the Institutional Review Board at the Tokyo Metropolitan Neurological Hospital. Because this was a retrospective and noninvasive study, written informed consent was not obtained from patients. Instead, a public notice that provided information on this study was posted on the Tokyo Metropolitan Neurological Hospital website.

Patient Population

From January 2012 through April 2014, a total of 9 patients with spinal dural AVF underwent open microsurgical obliteration at the Tokyo Metropolitan Neurological Hospital. Of these, 7 patients (5 men and 2 women, age range 40–85 years) with a dural AVF in the thoracic region were included in this study (Table 1). Two patients with a dural AVF in the lumbar region were excluded because the abnormal AV connection of a thoracic AVF is commonly located dorsolaterally to the thecal sac and that of a lumbar AVF is commonly located ventrolaterally to the thecal sac; therefore, for patients with lumbar lesions, intraoperative microscopic observations were limited.

Angiographic Assessments of Lesion Vessels With High-Resolution 3D Computer Graphics

All diagnoses of spinal dural AVFs were made by spinal angiography. For 6 of the 7 patients, 3D computer graphic analysis was conducted to assess the stereoscopic anatomy of meningeal vessels and their relationships with the dura mater and spinal cord. Briefly, meningeal vessels were visualized as images rendered from rotational angiography data. To visualize meningeal vessels with sufficiently high resolution, the field of view was set to 43–83 mm³ with 512 voxel size: 1 voxel was considered to be 84–162 μm; thus, on a theoretical basis, meningeal vessels with diameters less than 200 μm could be visualized. The dura mater and spinal cord were visualized as a result of surface data rendered from myelographic CT. These high-resolution rendered images from rotational angiography and myelographic CT were fused on a workstation computer (M6700, Dell Inc.) by rendering software (Amira 5.4.3, Visualization Science Group, an FEI company). The dura mater was made half translucent for clear visualization of the spatial course of the meningeal vessels and their relationships with the dura mater.

Intraoperative Assessments of Lesion Vessels With Indocyanine Green Microangiography

During surgery, we investigated microvascular blood flow in the meningeal vessels and their relationships with the thecal sac and root sleeves under the operating microscope. Single-level midline laminectomy at the corresponding level only was performed through a spinous process-splitting approach. We carefully drilled off the spinal lamina and medial facet joint at the corresponding level to expose meningeal vessels on the intact epidural surface. After extensively exposing the epidural vessels, we performed indocyanine green (ICG) video angiography. We injected 12.5 mg of ICG before and after the dural incision. We then attempted to block intradural arterial inflow as much as possible by detaching and removing several meningeal vessels on the outer dural surface. Last, we cut off the proximal subarachnoid portion of the intradural draining vessel to completely block intradural arterial inflow.

Histological Examination of Lesion Vessels

In 4 patients, we additionally removed a small piece of the dura mater suspected to include an abnormal AV connection, which consisted of meningeal vessels and the proximal stump of an intradural dilated vessel. We removed the dura mater en bloc, leaving the corresponding root sleeves intact. The dural defect was covered with

<table>
<thead>
<tr>
<th>Patient No.</th>
<th>Age (yrs), Sex</th>
<th>Level of Feeding Vessels</th>
<th>Meningeal Feeding Vessels on Outer/Inner Dural Surface*</th>
<th>No. of Meningeal Feeding Vessels on Outer/Inner Dural Surface†</th>
<th>No. of Intradural Draining Vessels</th>
<th>Meningeal Feeding Vessels on Outer/Inner Dural Surface (no. examined)</th>
<th>AV Connections w/in Dura Mater</th>
<th>Intradural Draining Vessels</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>85, M</td>
<td>T-11</td>
<td>Yes/ND</td>
<td>3/4</td>
<td>1</td>
<td>Artery (1)/NA</td>
<td>NA</td>
<td>Vein</td>
</tr>
<tr>
<td>2</td>
<td>40, M</td>
<td>T-7</td>
<td>Yes/yes</td>
<td>4/3</td>
<td>1</td>
<td>Artery (2)/artery (1)</td>
<td>ND</td>
<td>Vein</td>
</tr>
<tr>
<td>3</td>
<td>80, M</td>
<td>T-6</td>
<td>Yes/ND</td>
<td>3/2</td>
<td>1</td>
<td>Artery (3)/NA</td>
<td>NA</td>
<td>Vein</td>
</tr>
<tr>
<td>4</td>
<td>48, M</td>
<td>T-6</td>
<td>Yes/yes</td>
<td>4/3</td>
<td>1</td>
<td>Artery (1)/artery (1)</td>
<td>ND</td>
<td>Vein</td>
</tr>
<tr>
<td>5</td>
<td>82, F</td>
<td>T-5</td>
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<td>1</td>
<td>Artery (1)/NA</td>
<td>NA</td>
<td>Vein</td>
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<td>T-8</td>
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<td>4/3</td>
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<td>Artery (2)/artery (2)</td>
<td>Yes (on inner dural surface)</td>
<td>Vein</td>
</tr>
<tr>
<td>7</td>
<td>71, M</td>
<td>T-6</td>
<td>Yes/ND</td>
<td>3/2</td>
<td>1</td>
<td>Artery (3)/artery (1)</td>
<td>Yes (on inner dural surface)</td>
<td>Vein</td>
</tr>
</tbody>
</table>

NA = not available; ND = not detected.
* Detected by 3D computer graphics.
† Detected by ICG angiography.
autologous fat tissue secured to the edges by sutures. The removed specimens (meningeal vessels, the dura mater suspected of including an AVF, and the intradural draining vessel) were fixed in 10% formaldehyde and embedded in paraffin wax. We cut these specimens into step or serial sections with thicknesses of 3 μm and then stained them with H & E and with Elastica van Gieson to detect internal elastic lamina and with Masson trichrome to distinguish between collagen fibrosis and the muscle layer of the vessels. We also immunohistochemically stained the specimens with an antiendothelium antibody to detect endothelial cells in the vessels. We confirmed each lesion vessel as an artery, AV connection, or vein on the basis of objective findings of the vessel wall structures.

**Results**

**3D Computer Graphics**

Spinal dural AVFs were diagnosed in all 7 patients by spinal angiography, which revealed abnormal AV connections between the intercostal artery at the level of T5–11 and the intradural radiculomedullary vein. For 6 of 7 patients, high-resolution 3D computer graphics analysis was successfully performed to assess the stereoscopic anatomy of the microvessels (Fig. 1). The spatial relationships between these small meningeal branches and the dura mater could be visualized in sufficient detail by making the dura half translucent. In 2 of these 6 patients, several meningeal vessels were clearly demonstrated on both the outer and inner dural surfaces at the dorsolateral site of the thecal sac adjacent to the root sleeve. Meningeal vessels had complex anastomoses. A meningeal branch of the intercostal artery split into multiple meningeal vessels on the outer dural surface. These multiple meningeal vessels ran in a longitudinal direction along the thecal sac and crossed the dura to the inner dural surface. After crossing the dura mater, they turned around to gather and join the intradural vessel on the inner dural surface.

**ICG Microangiography**

ICG video angiography revealed the in vivo blood flow in these meningeal vessels and their rich anastomoses between the outer and inner dural surfaces (Fig. 2). For 5 of the 7 patients, before the dura was incised, an intradural draining vessel with a retrograde arterial inflow could be observed through the intact dura mater. Therefore, the site of the abnormal AV connection could be presumed from the epidural space. After the dura was incised, we confirmed that a single dilated vessel was connected to the inner dural surface, where the vessel received arterial inflow from the meningeal vessels. More than 1 meningeal vessel on the inner dural surface gathered to merge into the proximal intradural draining vessel, which appeared to be caput medusae.

We attempted to block arterial inflow by clipping as many of the meningeal vessels on the outer dural surface as possible; however, we could not completely block arterial inflow in all cases. Radical removal of these epidural feeder vessels gave the same results. After the proximal intradural vessel was cut, intradural arterial inflow was completely blocked; however, blood flow in the meningeal vessels on the inner dural surface circumjacent to the proximal stump of the draining vessel persisted on ICG video angiography (Fig. 3).

In the process of removing the dural lesion suspected...
of including an abnormal AV connection, arterial bleeding occurred around the margin of the resected dura although we had already removed as many epidural feeding vessels as possible. Bleeding occurred not only from the feeding vessels on the outer dural surface but also within the dura mater and/or on the inner dural surface. No AVF recurred in any patient during the follow-up period (median 12 months, range 12–41 months).

Histological Findings

Confirmation of each lesion vessel as an artery, AV connection, or vein is shown in Table 1.

Meningeal Vessels

Meningeal vessels on the outer dural surface fed by the intercostal artery had a continuous internal elastic lamina with a regular smooth muscle layer within their vessel wall. The vessel wall structure was compatible with that of a normal artery; no abnormal findings were detected (Fig. 4). In 3 patients, a small epidural vein ran near the epidural arteries.

Intradural Draining Vessels

The vessel wall of the proximal subarachnoid portion of the intradural draining vessels was irregularly thickened by collagen and elastic fibrosis without a continuous internal elastic lamina and a regular smooth muscle layer. The diameter of the vessels was significantly enlarged. The vessel wall structure was compatible with that of a vein modified by retrograde arterial inflow (Fig. 4). These
results were attributed to venous hypertension caused by retrograde arterial inflow.

AV Connections

From 4 patients, a small piece of the dura mater suspected of including an AVF was obtained. The specimen consisted of the dura mater, meningeal arteries, and the proximal stump of an intradural dilated vein. For the first 2 patients, an AV connection was not detected within the specimens, which were cut into step sections. For the last 2 patients, the exact site of the AV connection was eventually identified within the specimens, which were cut into serial sections from the dural part to the proximal stump of the intradural dilated vessel (Fig. 5).

Meningeal arteries crossed the dura mater from the outer to the inner dural surface. After crossing the dura mater, the meningeal arteries gathered and joined to the proximal stump of the intradural dilated vein on the inner dural surface. There was more than 1 direct AV connection around the vein. These results indicated that a spinal dural AVF comprising multiple direct AV connections between more than 1 meningeal feeding artery and a single proximal radiculomedullary vein was located on the inner dural surface (Fig. 5).

The intradural draining vein fed by meningeal arteries terminated at the site at which the vein connected to the dura mater; the vein did not have the epidural drainage route of a normal spinal venous system.

Discussion

This comprehensive assessment, which used advanced microangiography (Fig. 1–3) and histological findings (Figs. 4 and 5), demonstrated the exact location and unique configuration of spinal dural AVFs. The new find-
ing is that a spinal dural AVF exists on the inner dural surface, where multiple AV connections between more than 1 meningeal feeding artery and 1 radiculomedullary vein occur at the site where the vein connects to the dura mater.

Comparisons With Previous Findings

To our knowledge, only 1 previous microangiographic study of spinal dural AVFs, in which blood flow in the meningeal arteries was thoroughly investigated by ex vivo microangiography in resected arteriovenous lesions, has been conducted (Fig. 6). Beyond that study by McCutcheon et al., our findings add new information about the exact location of the AVF: the transition between the arteries and a vein exists on the inner dural surface. As opposed to our study, the previous study did not provide histological proof of whether lesion vessels were arteries, an artery-to-vein transition, or veins; therefore, the transition between the arteries and a vein and their relationships with the dura mater remained unclear.

McCutcheon et al. highlighted the vessel course and its relationship with the dura mater by comparing a microangiogram and an artist’s sketch (Fig. 6). On the outer dural surface, the meningeal artery split into multiple loops adjacent to the nerve root sleeve, and these vessels rejoined on the outer dural surface. After rejoining, the meningeal artery crossed the dura to join the intradural radiculomedullary vein. Complex arterial feeding vessels existed exclusively on the outer dural surface or within the outer dural layer. McCutcheon et al. concluded that an AVF comprised a transdural single channel between the epidural meningeal arteries and the intradural radiculomedullary vein. They did not discuss meningeal arteries on the inner dural surface or their connections.

On the basis of our findings, the microangiogram of McCutcheon et al. could be interpreted as follows (Fig. 6): The meningeal artery split into 2 loops on the outer dural surface. After the 2 meningeal branches crossed the dura mater from the outer to the inner dural surface at each different site, they turned around to gather and join the single intradural radiculomedullary vein on the inner dural surface. The centrally located proximal radiculomedullary vein and 2 meningeal feeding arteries on the inner dural surface may have been misinterpreted as 2 epidural arteries and a single transdural channel to the proximal radiculomedullary vein.

To our knowledge, only 1 histological study of spinal dural AVFs, by Benhaiem et al., has analyzed surgical specimens of the meningeal vessels and AVFs in detail. Our study adds new information about the exact configuration of an AVF: specifically, an AVF comprises multiple AV connections between more than 1 meningeal feeding artery and a single radiculomedullary vein. As opposed to our study, the Benhaiem et al. study did not provide microangiography or operative findings and it remained unclear which findings corresponded to which vessels or which part of the dura mater.

Benhaiem et al. histologically identified an abnormal vessel with its arterial and venous structures within the thickness of the dura mater; however, they did not establish where and how the abnormal vessel within the dura mater was fed by meningeal arteries or where and how the vessel joined the intradural radiculomedullary vein.

FIG. 6. Microangiogram (left) and an artist’s sketch (right) from article by from McCutcheon et al. The microangiogram shows the cannula in the epidural artery (arrowhead), the penetration of the dural artery (thin arrow), and spillage of contrast agent from the cut end of the intradural vein (thick arrow). McCutcheon et al. claimed that when the 2 meningeal arteries rejoin on the outer dural surface, they cross the dura to join the medullary vein (thin arrow). Compared with our results in Fig. 1, their microangiogram can be interpreted as follows: The meningeal artery splits into 2 loops on the outer dural surface. After the 2 meningeal branches cross the dura mater from the outer to the inner dural surface at each site, they turn around to gather and join the single intradural radiculomedullary vein on the inner dural surface. The dural AVF comprises direct AV connections on the inner dural surface between the 2 meningeal feeding arteries and a single proximal radiculomedullary vein. Copyright AANS. Published with permission.

Implications for Treatment

Two treatment options are available for spinal dural AVFs: open microsurgery and endovascular embolization. According to a meta-analysis, the rate of occlusion after initial treatment with endovascular embolization has not yet reached that of open microsurgery; moreover, the lasting effectiveness of open microsurgery is significantly superior to that of endovascular embolization because dural AVF recurrence has been more commonly reported after endovascular treatment. The success rate of permanent AVF occlusion was previously reported to be 98% for open surgery and 10%–75% for endovascular embolization.

On the basis of these findings, we conclude that the cause of the initial failure and the higher recurrence rate after endovascular treatment may have been derived from both the deep location and complicated configuration of dural AVFs and the limited approach route for this treatment. Complex AVFs exist on the inner dural surface and with endovascular embolization can be approached from the extradural side only.

Even if embolization materials fill the meningeal arteries on the outer dural surface, unless they reach and plug the intradural proximal radiculomedullary vein as well as the meningeal arteries on the inner dural surface circumjacent to the proximal vein, permanent occlusion of an AVF cannot be achieved. Permanent occlusion is not possible because the residual meningeal arteries, especially on the inner dural surface, receive arterial blood flow from other
meningeal arteries at the same or adjacent spinal level, leading to AVF recurrence.

Insufficient embolization delays the complete blockage of an abnormal AV connection, resulting in the progression of irreversible neurological deficits in gait, micturition, and the sensory system. \(^2\) With regard to complete penetration of the intradural proximal radiculomedullary vein, even the new liquid embolic material, Onyx (Covidien and ev3 Neurovascular), failed to allow for a more controlled injection than N-butyl cyanoacrylate. \(^1,6,9\)

Therefore, we propose that open microsurgery should be the first-line choice for patients with spinal dural AVFs; the dural AVF on the inner dural surface, a centrally located radiculomedullary vein fed by the surrounding meningeal feeding arteries, which could be easily identified, can be blocked completely on direct microscopic view. In previous studies of the surgical treatment of spinal dural AVFs, the AVF that is consistent with our results could be identified on operative photographs of the inner dural surface. \(^8,11,19\) In addition to blocking the proximal vein, it is ideal to coagulate the AVF itself to block abnormal AV connections completely, including the proximal stump of the radiculomedullary vein and surrounding meningeal arterial feeding vessels, although surgical removal of the dura mater is not necessary. \(^2\)

If endovascular treatment is the first-line choice, it should be planned in combination with preparation for open microsurgery in the same institution for patients in whom embolization fails. Because angiographic evaluations alone cannot predict the risk for recanalization, the embolization cast position must be evaluated by CT immediately after the intervention to determine whether it is in the intradural proximal radiculomedullary vein as well as within the dura mater. \(^10\) If the embolic material does not penetrate the intradural proximal vein, surgical treatment should be performed as soon as possible to try and block an AVF permanently before the progression of incurable neurological deficits. \(^12,25\)

**Limitations**

Because the incidence of spinal dural AVFs is very low, the number of patients in this study was small. The lesion vessels were not compared with those vessels in healthy controls because obtaining normal spinal angiograms and surgical specimens of the dura mater is difficult.

Despite these limitations, our study provides valuable information about the lesion vessels of spinal dural AVFs for stroke specialists (e.g., neurosurgeons, neuroradiologists, and neurologists) to assist with the diagnosis and appropriate treatment for these patients.

**Conclusions**

Our results indicate that a spinal dural AVF is located on the inner dural surface, and a dural AVF comprises multiple direct AV connections between more than 1 meningeal feeding artery and a single proximal radiculomedullary vein at the site where the vein connects to the dura mater adjacent to the root sleeve. The higher rate of recurrence after endovascular treatment may have resulted from the deep location and complicated configuration of dural AVFs.

**References**


Author Contributions
Conception and design: Takai. Acquisition of data: Takai. Analysis and interpretation of data: Takai. Drafting the article: Takai. Critically revising the article: Komori, Taniguchi. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Takai. Study supervision: Takai.

Supplemental Information
Proceedings
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