Comparison between Spinal Dural Arteriovenous Fistula and Spinal Epidural Arteriovenous Fistula

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Objective: The purpose of this study is to retrospectively assess the differences between spinal dural arteriovenous fistulas (SDAVFs) and spinal epidural arteriovenous fistulas (SEAVFs).

Methods: Subjects consisted of 18 patients with SDAVFs and 7 with SEAVFs admitted to our department between January 2007 and December 2017 exhibiting intradural drainage of shunt flow. Patient background, lesion characteristics, and treatment/follow-up results were compared.

Results: Of the seven patients in the SEAVF group, six patients (86%) had been misdiagnosed with SDAVFs at the time of treatment. The rates of patients with a history of spinal surgery, lumbar vertebral lesions, involvement of a dorsal somatic branch (DSB), involvement of multiple segmental arteries, or involvement of bilateral segmental arteries were significantly higher in the SEAVF group. As for post-treatment course, there were significant difference in the recurrence rate after endovascular treatment (SDAVF group: 6%, SEAVF group: 50%, respectively, \( p < 0.05 \)).

Conclusion: Endovascular treatment may not be effective for SEAVFs if they are misdiagnosed as SDAVFs, and they may recur. For optimal treatment, accurate assessment of the angioarchitecture with the latest diagnostic imaging method may be necessary.

Keywords: spinal epidural arteriovenous fistula, spinal dural arteriovenous fistula, recurrence, transarterial embolization, direct surgery

Introduction

Spinal epidural arteriovenous fistulas (SEAVFs) are classified into types A (with intradural drainage of shunt flow) and B (without intradural drainage).\(^1\,^2\) As demonstrated for spinal dural arteriovenous fistulas (SDAVFs), type A SEAVFs primarily develop in middle-aged or older males, and present with progressive myelopathy due to spinal venous congestion, with similar angioarchitecture. SEAVFs are often misdiagnosed as SDAVFs. Recently, however, 3D-DSA has facilitated the differentiation of SEAVFs from SDAVFs based on the detailed angioarchitectural assessment.\(^3\) In this study, we reviewed the characteristics of SEAVFs and SDAVFs, as well as treatment results in our department to retrospectively assess the differences between the two types of AVFs.

Subjects and Methods

Subjects consisted of 18 patients with SDAVFs and 7 with SEAVFs admitted to our department between January 2007 and December 2017 exhibiting intradural drainage of shunt flow. We excluded type B patients with extradural shunt drainage and those in whom extradural outflow and intradural reflux were simultaneously observed. Prior to undertaking this study, we received approval from the Ethics Review Board of our hospital (1802-027).
Examination items
In the above two groups, we examined the patient background (age, sex, previous spinal surgery), characteristics of lesions (lesion level, sign, diagnosis at the time of treatment), angioarchitecture (feeder, drainer), contents of treatment (endovascular procedure, direct surgery, multiple sessions of treatment), results of treatment, and results of follow-up (recurrence, complete occlusion on the final evaluation, modified Rankin Scale [mRS] score 3 months after the final treatment).

Preoperative examination
In our department, DSA of the segmental arteries was performed under local anesthesia in all patients in which SDAVs or SEAVFs were suspected based on their medical history and MRI findings. When renal function was within normal limits, 3D CTA was conducted before DSA. After predicting a segmental artery branching a feeder, DSA was performed. Imaging of the thoracic and lumbar segmental arteries was conducted to search for the feeder and radiculo-medullary or -pial arteries connecting to the spinal artery. For the segmental artery branching a feeder, 3D-DSA was performed. While instructing patients to stop breathing, rotational imaging for 5 s was consecutively conducted twice to obtain mask and contrast-enhanced images. In our department, an image intensifier (II) was used to acquire DSA until March 2013. After April 2013, a flat panel detector (FPD) was used. After obtaining DSA and digital angiography (DA) images using 3D-DSA with an FPD, a slab maximum intensity projection (MIP) image was prepared from the DA image to confirm the detailed angioarchitecture. As an FPD, an Artis zee biplane (Siemens Healthcare GmbH, Forchheim, Germany) was used. For 3D-DSA, a 4Fr diagnostic catheter was inserted into a target vessel, and 100% contrast medium was infused. We attempted to inject NBCA into an intradural drainer, but stopped when reflux of NBCA into a feeder was noted.

Differential diagnosis of SDAVFs and SEAVFs
For differential diagnosis, 2D-/3D-DSA and slab MIP images were used. Patients in whom the dorsal or ventral somatic branch (VSB) nourishing the vertebral body or prelaminar artery nourishing the vertebral arch was a main feeder, and a venous pouch was present at an epidural site adjacent to the vertebral body or arch were diagnosed with SEAVFs. Those with inflow of a feeder (dural branch from the radiculomeningeal artery), longitudinally aggregating at an area medial to the medial interpedicle line, to an intradural drainer (horizontal T sign) on frontal 2D-DSA images were diagnosed with SDAVFs.3)

Selection of treatment methods
We selected the same therapeutic strategies for both SDAVF and SEAVF patients. When preoperative examination confirmed a pial artery branching from the responsible segmental artery or collateral pathway, direct surgery was selected in principle. When there was no pial artery related to the feeder, an endovascular procedure was selected as a first choice. When shunt occlusion was not achieved, or when recurrence was noted, direct surgery was additionally performed.

For direct surgery, coagulation and disconnection of an intradural drainer were performed in both SDAVF and SEAVF patients. In the latter, epidural shunt-site cautery was not conducted. For endovascular treatment, a 4Fr catheter was inserted into the responsible segmental artery as a guiding catheter under local anesthesia. After inserting a microcatheter into a distal part of the feeder, a provocation test with lidocaine was performed. When there was no neurological deterioration, n-butyl-2-cyanoacrylate (NBCA) was infused. When shunt occlusion was achieved by the endovascular procedure, systemic heparinization was performed for 1 week after the procedure to prevent delayed venous thrombosis. Subsequently, no antithrombotic therapy was conducted. After direct surgery, no systemic heparinization was performed.

Postoperative follow-up
Follow-up was conducted at the outpatient clinic of our department. When neurological examination and MRI suggested recurrence, patients were admitted, and the recurrence was confirmed using DSA to evaluate whether additional treatment should be carried out.

Statistical analysis
For statistical analysis, JMP10 software (SAS Institute Inc., Cary, NC, USA) was used. To compare the values between two groups, the chi squared or median tests were used. A p value of less than 0.05 was regarded as significant.
Results

The mean ages in the SDAVF and SEAVF groups were 66 and 70 years, respectively (range: 51–84 and 59–80 years, respectively). The rates of males were 89% and 100%, respectively. In the latter group, the rate of patients who had undergone spinal surgery was significantly higher. The shunt level ranged from Th5 to L1 (most frequent level: Th6) in the SDAVF group; it ranged from Th11 to L5 (most frequent level: L2/L3) in the SEAVF group, and the incidence of lumbar lesions was significantly higher in this group (Table 1). In the two groups, myelopathy was the primary symptom, and T2-weighted MRI showed high-intensity signals in the spinal cord in all patients. Flow voids were observed in 78% of the patients in the SDAVF group and in 83% in the SEAVF group, showing no difference. Of the seven patients in the SEAVF group, six (86%) had been misdiagnosed with SDAVFs at the time of treatment. In four of these, an II had been used to acquire DSA.

The most common angioarchitecture in the SDAVF group was as follows: the radiculo-meningeal artery functioned as a feeder and the bridging vein as a drainer. In contrast, the most common angioarchitecture in the SEAVF group was as follows: the dorsal somatic branch (DSB) functioned as a feeder and the radiculo-medullary vein (RMV) as a drainer. Patients with feeders from multiple segmental arteries accounted for 17% in the SDAVF group and 71% in the SEAVF group, showing a significant difference. Those in whom feeders branching from the bilateral segmental arteries had flown in the same AVF accounted for 0% in the SDAVF group and 57% in the SEAVF group, respectively, showing a significant difference (Table 1).

Furthermore, epidural venous pouches were noted in all patients in the SEAVF group, and their sites consisted of the ventral space of the spinal canal in six patients, lateral space in one patient, and dorsal space in zero patient. The median duration of disease before initial treatment was 6 months (range: 1–35 months) in the SDAVF group and 9 months (range: 1–51 months) in the SEAVF group, showing no difference. As the initial treatment, endovascular procedure was selected for many patients in the two groups (SDAVFs: 78%, SEAVFs: 100%), but direct surgery was eventually performed in >40% of the patients in each group (44 and 43%, respectively). In the SDAVF group, the reasons why direct surgery was performed included pial artery branching from the feeding artery in four patients, a positive reaction on a provocation test in one, residual AVF after embolization in one, patient’s

Table 1  Patient characteristics and angiographic findings

<table>
<thead>
<tr>
<th></th>
<th>SDAVFs (n = 18)</th>
<th>SEAVFs (n = 7)</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Median age</td>
<td>66</td>
<td>70</td>
<td>n.s.</td>
</tr>
<tr>
<td>Male sex (%)</td>
<td>89</td>
<td>100</td>
<td>n.s.</td>
</tr>
<tr>
<td>Previous spinal surgery (%)</td>
<td>11</td>
<td>57</td>
<td>&lt;0.05</td>
</tr>
<tr>
<td>Lumbar lesion (%)</td>
<td>22</td>
<td>86</td>
<td>&lt;0.01</td>
</tr>
<tr>
<td>Right lesion (%)</td>
<td>44</td>
<td>43</td>
<td>n.s.</td>
</tr>
<tr>
<td>Feeder (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Radiculomeningeal artery</td>
<td>100</td>
<td>14</td>
<td>&lt;0.0001</td>
</tr>
<tr>
<td>Dorsal somatic branch</td>
<td>0</td>
<td>86</td>
<td>&lt;0.0001</td>
</tr>
<tr>
<td>Multiple</td>
<td>17</td>
<td>71</td>
<td>&lt;0.05</td>
</tr>
<tr>
<td>Bilateral</td>
<td>0</td>
<td>57</td>
<td>&lt;0.01</td>
</tr>
</tbody>
</table>

SDAVFs: spinal dural arteriovenous fistulas; SEAVFs: spinal epidural arteriovenous fistulas

Table 2  Treatment results and follow-up results

<table>
<thead>
<tr>
<th></th>
<th>SDAVFs (n = 18)</th>
<th>SEAVFs (n = 7)</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Treatment (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>IVR as initial Tx</td>
<td>78</td>
<td>100</td>
<td>n.s.</td>
</tr>
<tr>
<td>Direct surgery</td>
<td>44</td>
<td>43</td>
<td>n.s.</td>
</tr>
<tr>
<td>Multiple</td>
<td>17</td>
<td>43</td>
<td>n.s.</td>
</tr>
<tr>
<td>Complication</td>
<td>6</td>
<td>0</td>
<td>n.s.</td>
</tr>
<tr>
<td>Follow-up</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Improvement of symptoms (%)</td>
<td>83</td>
<td>100</td>
<td>n.s.</td>
</tr>
<tr>
<td>Recurrence after IVR (%)</td>
<td>6</td>
<td>50</td>
<td>&lt;0.05</td>
</tr>
<tr>
<td>Median mRS (IQR) pre Tx</td>
<td>3 (2-4)</td>
<td>4 (2-4)</td>
<td>n.s.</td>
</tr>
<tr>
<td>Median mRS (IQR) post Tx</td>
<td>2 (1-2)</td>
<td>2 (2-3)</td>
<td>n.s.</td>
</tr>
</tbody>
</table>

IQR: interquartile range; IVR: interventional radiology; mRS: modified Rankin Scale; SDAVFs: spinal dural arteriovenous fistulas; SEAVFs: spinal epidural arteriovenous fistulas; Tx: treatment
wishes in one, and recurrence in one. In the SEAVF group, direct surgery was performed in three patients due to recurrence after embolization (Table 2).

As a treatment-related complication, neurological deterioration related to delayed venous thrombosis was observed in one patient in the SDAVF group. The median follow-up period after the final treatment was 22 months (range: 1–124 months) in the SDAVF group and 23 months (range: 1–86 months) in the SEAVF group. Follow-up for ≥3 months was impossible in three patients in the former group and in one in the latter. Of patients who could be followed up, recurrent AVFs were detected after the endovascular procedure in 6% in the SDAVF group and in 50% in the SEAVF group, showing a significant difference. The median preoperative mRS scores (interquartile range [IQR]) of the SDAVF and the SEAVF groups were 3 (2-4) and 4 (2-4), respectively, showing no difference (p = 0.25). The median mRS scores (IQR) 3 months after the final treatment were 2 (1-2) and 2 (2-3), respectively, showing no difference (p = 0.13) (Table 2).

As a representative case, a patient with a SEAVF in whom direct surgery was finally performed due to repeated recurrence after embolization is presented (Fig. 1).

The patient was a 74-year-old male with a 10 months history of progressive motor and sensory disturbance of the bilateral lower limbs and vesicoarectal disorder. Under a tentative diagnosis of SDAVF, he was referred to our department from a previous clinic. T2-weighted MRI revealed a high intensity intramedullary lesion and flow void around the spinal cord (Fig. 1A). Spinal angiography
showed that two feeders, which were considered to be DSBs, and another one, which was considered to be a VSB from the segmental artery at the right L3 level, had flown into the epidural venous plexus on the ventral space of the spinal canal, draining into the RMV flowing into the intradural vein from its lateral side (perimedullary drainage) (Fig. 1B–1D). Therefore, the final diagnosis was SEAVF. Transarterial embolization (TAE) was performed. A 4Fr catheter was inserted into the right L3 segmental artery, and a microcatheter was inserted into the medial DSB. As a provocation test, 5 mg of lidocaine was injected, causing incomplete paralysis of the left toes/ankle. Its reproducibility was noted, and the reaction was evaluated as positive. Proximal occlusion was conducted by inserting detachable coils. A microcatheter was inserted into the VSB feeder. As a provocation test showed a negative reaction at this site, 15% NBCA was injected, but it did not reach the shunt point and resulted in proximal occlusion of the feeder. At this point, there was no visualization of the shunt from the remaining feeder, and the procedure was completed. After 1 week, DSA showed shunt flow from the bilateral L3 segmental arteries (Fig. 1E). Additional TAE was performed, but the bilateral feeders showed positive reactions on provocation test. Proximal occlusion with coils was conducted and the shunt was confirmed to have disappeared by imaging. As the symptoms reduced, he was referred to a rehabilitation hospital, but they gradually deteriorated again, suggesting recurrence. After 4 months, DSA was performed in our hospital. The DSB branching from the left L2 segmental artery had flown in the contralateral DSB via a retrocorporeal anastomosis, and in the right L3 epidural venous plexus via a vertical collateral pathway (Fig. 1F). Direct surgery was selected. Partial L3 laminectomy was performed, and the RMV ascending along the right L3 nerve root was confirmed by opening the dura mater (Fig. 1G). After confirming no changes in motor-evoked potential (MEP) monitoring through blockage by temporary clipping of the drainer, coagulation and disconnection of the drainer were performed (Fig. 1H and 1I). After surgery, the symptoms reduced, facilitating gait. Angiography confirmed the disappearance of the shunt, epidural venous plexus, and drainer. T2-weighted MRI showed the disappearance of the high-intensity intramedullary lesion.

Discussion

In this study, the characteristics of SDAVFs and SEAVFs, such as patient background and angioarchitecture, in our department were similar to those in the literature.\(^\text{3}\) Furthermore, in our department, most patients with SEAVFs had been misdiagnosed with SDAVF at the time of treatment. The results of treatment showed that the recurrence rate after endovascular treatment was higher in the SEAVF group.

According to previous studies, TAE was more frequently selected for SEAVF patients compared with direct surgery, and favorable results were obtained by sufficiently injecting liquid embolic materials, such as NBCA or Onyx, into the intradural vein via the venous pouch.\(^\text{1,4–6}\) In particular, Kiyosue et al. reviewed TAE of SEAVFs and reported that the occlusion rate of TAE was low when drainage involved epidural outflow, whereas it was 90% in patients with intradural reflux alone.\(^\text{4}\) These studies indicated that a shunt was present on the ventral side of the spinal canal in many patients with SEAVFs, making direct surgery to the shunt difficult and that shunts remained after blockage of a vein with intradural reflux alone, inducing recurrent intradural drainage. The recurrence rate over a long period remains uncertain, however. In contrast, Niizuma et al. and Takai et al. reported that disconnection of an intradural drainer led to shunt occlusion in the presence of simple intradural drainage among their series of direct surgery for SEAVFs, suggesting its long-term efficacy.\(^\text{7,8}\)

In this study, the recurrence rate after TAE was high in the SEAVF group, first because most patients had been misdiagnosed with SDAVF; neither an accurate shunt point nor the multiple shunts have been evaluated, and liquid embolic materials may not have reached the intradural vein. Second, the DSB or VSB with developed collateral pathways may function as multiple feeders in many SEAVF patients, and NBCA injection through a targeted vessel injecting liquid embolic materials, such as NBCA or Onyx, into the intradural vein via the venous pouch.\(^\text{1,4–6}\) In particular, Kiyosue et al. reviewed TAE of SEAVFs and reported that the occlusion rate of TAE was low when drainage involved epidural outflow, whereas it was 90% in patients with intradural reflux alone.\(^\text{4}\) These studies indicated that a shunt was present on the ventral side of the spinal canal in many patients with SEAVFs, making direct surgery to the shunt difficult and that shunts remained after blockage of a vein with intradural reflux alone, inducing recurrent intradural drainage. The recurrence rate over a long period remains uncertain, however. In contrast, Niizuma et al. and Takai et al. reported that disconnection of an intradural drainer led to shunt occlusion in the presence of simple intradural drainage among their series of direct surgery for SEAVFs, suggesting its long-term efficacy.\(^\text{7,8}\)
investigators indicated that a provocation test was unnecessary in SEAVF patients, but we performed this test, considering the risk of NBCA reflux/inflow into the pial artery or vasa nervorum of the nerve root. On the first session of TAE, proximal occlusion of the DSB with a provocation test-positive reaction was performed (occlusion of the somatic branch alone may not have caused neurological symptoms), and NBCA was injected so that it might reach the drainer, regarding the test-negative VSB alone as a feeder. This was, however, completed with only proximal occlusion, which may have contributed to recurrence.

Based on these findings, for radical SEAVF treatment, fine, accurate diagnostic imaging using the latest angiography equipment may be necessary. For accurate diagnostic imaging, patients’ immobilization (respiration/bowel peristalsis in particular) is also important. Examinations and treatment in a state of immobilization under general anesthesia may also be effective although they are not conducted in our hospital.

In this study, direct surgery was finally performed in three patients with SEAVFs. As described above, however, our purpose was to disconnect a vein with intradural reflux, and direct shunt obliteration was not performed. As such, shunt occlusion is not always achieved and, if a shunt remains, the new appearance of intradural reflux may lead to recurrence. As another issue regarding direct surgery, intraoperative hemorrhage control may be difficult when shunt flows in the epidural venous plexus on the dorsal side of the spinal canal.

The limitations of this study include the following. First, this was a retrospective study. Second, the number of patients was small. Third, no long-term outcomes were presented. The number of SDAVF or SEAVF patients is limited in each institution, and a national survey may be useful. A meta-analysis involving SDAVF patients showed that the recurrence rate after endovascular treatment was higher than after direct surgery, with an odds ratio of 3.15. It has, however, been difficult to differentiate SDAVF from SEAVFs until recently, and the greater portion of the patients reported as having SDAVF may actually have had SEAVFs. In the future, this must be retrospectively examined.

**Conclusion**

Endovascular procedures may not be effective for SEAVFs that were not accurately diagnosed, and they may recur. For optimal treatment, it may be necessary to accurately understand the angioarchitecture using the latest diagnostic imaging procedure.

**Disclosure Statement**

Isao Date received a subsidy from Momotaro-Gene Inc. There is no conflict of interest for the first author and other coauthors.

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