Chronic venous congestion following embolization of spinal dural arteriovenous fistula

Case report

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The authors present a case of spinal dural arteriovenous fistula with fluctuations in symptoms following embolization. Superselective injection of 33% N-butyl cyanoacrylate into the feeding vessel resulted in the complete occlusion of the fistula with traversal of the nidus. The subsequent venous congestion was progressive and treatable with anti-thrombin therapy. Extended medication with dual antiplatelet therapy was required because dose reduction to aspirin monotherapy worsened symptoms. In this case, it took > 2 months for the patient’s symptoms to stabilize. The duration of progressive venous thrombosis after embolization of a spinal dural arteriovenous fistula is not well known, nor is the most adequate treatment. Although it is presumed that prevention of venous thrombosis is best achieved with anticoagulation, dual antiplatelet therapy can be a substitute for patients with poor compliance.

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KEY WORDS • embolization • endovascular therapy • N-butyl cyanoacrylate • progressive venous thrombosis • spinal dural arteriovenous fistula

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PINAL DAVF is an uncommon acquired disease found primarily in men > 50 years of age. Although microsurgical shunt interruption is both secure and reliable, endovascular closure of the spinal DAVF has gained the leading role recently. We report the case of a patient with spinal DAVF in whom embolization was undertaken, after which fluctuations in symptoms emerged in the chronic phase.

Case Report

History and Examination. This 72-year-old man presented with gait disturbance after having fallen 3 years earlier and later developed urinary retention and paresthesia in both lower limbs. After a long period in which various health care professionals were unable to diagnose his disease, MR imaging performed in our department identified a spinal DAVF.

On neurological examination, we noted abnormalities in reflexes (exaggerated bilateral Achilles tendon reflexes and right ankle clonus), as well as in motor (weakness in MMT of 4/5 in the bilateral tibialis anterior muscles and atrophy of the right lower limb), sense (dysesthesia especially in the lateral side of the right lower limb and diminished proprioception in the right knee and ankle), and urinary function (retention of > 1 L). The patient could walk only indoors with crutches.

The findings on MR images showed an increased T2 signal in the spinal cord from T-7 to L-1 and marked flow voids on both T1- and T2-weighted imaging (Fig. 1A and B). A selective injection to the common trunk of the right T6-8 radicular arteries showed a spinal DAVF at the level of T-6, draining into the dilated perimedullary vein (Fig. 2A).
Treatment. Complete closure of the spinal DAVF was accomplished by the superselective injection of 33% NBCA through a coaxial assembly system (Tempo4 and Prowler 14, Cordis). Deposition of the NBCA beyond the nidal-venous junction and into the perimedullary venous system of the spinal cord was suspected (Fig. 2B and C). The patient did not experience any changes in his symptoms immediately following the procedure.

To prevent unexpected thrombosis of the spinal cord venous system, an antithrombin agent (Argatroban) was administered for 2 days prophylactically. On the day following the procedure, the patient showed some improvement in his motor system. After discontinuation of the antithrombin agent, aspirin at a dose of 100 mg per day was given and the patient underwent rehabilitation to improve his activities of daily living. On the 8th day, he was able to walk ~30 m with a cane. His hyperreflexia and urinary retention improved slightly; however, the other disturbances remained constant.

Abasia without local pain began suddenly on the 9th day. Weakness of the right tibialis anterior muscle (MMT 3/5) and sensory disturbance, especially proprioception, deteriorated on neurological examination. Emergency MR imaging showed a marked reduction of the flow voids along the spinal cord, shrinkage of the increased T2 signal area in the cord, and no signs of hematomyelia (Fig. 1C and D). Recurrence of the spinal DAVF or progressive venous thrombosis was suspected. The patient refused our recommendation that he undergo repeated spinal angiography for confirmation, but reinstitution of the antithrombin therapy rapidly restored his symptoms to the level before worsening. Although anticoagulation with warfarin was considered to be the best treatment to prevent this deterioration, the patient was reluctant to accept the necessary dietary restrictions and instead chose to be treated with dual antiplatelets (aspirin 100 mg and cilostazol 200 mg per day).

His symptoms showed gradual improvement until the 27th day, when he stopped taking the cilostazol due to pal-
Recanalization occurs rarely but the inadvertent deposition of embolic material beyond the nidus and into the perimedullary venous system might cause thrombosis of the spinal cord venous system. The most common MR imaging finding of spinal DAVF is an increased T2 signal in the cord. Perimedullary vessels are seen on T2-weighted images as a serpentine or punctate area of signal void in 45–70% of patients. After treatment, the perimedullary vessels are no longer appreciable, but serpentine structures of intermediate signal intensity may be observed. Reopening of the fistula is associated with enlargement of cord hyperintensity and contrast-enhanced perimedullary vessels.

Nii et al. have reported that 2 of their 49 patients treated with embolization suffered progressive venous thrombosis. Their subsequent symptomatic aggravation developed within 1 month of the treatment and improved after heparinization. These authors used heparinization for selected patients who showed significantly compromised venous drainage of the spinal cord for 3 days to maintain the activated partial thromboplastin time at 1.5–2 times normal.

Acute neurological deterioration without evidence of hemorrhage in a patient with a spinal arteriovenous malformation has been referred to as Foix–Alajouanine syndrome. Although it has been suggested that this clinical entity might include spinal cord dysfunction due to venous congestion, which is a potentially reversible process, necrotic myelopathy due to progressive venous thrombosis is thought to be irreversible and untreatable.

The best treatment of progressive venous thrombosis in the chronic phase is not clear. Because the most obvious method of arresting the thrombotic process is anticoagulation, the use of heparin or warfarin may be the best therapeutic option. The optimal duration of oral anticoagulant treatment after the acute phase is also unknown. In the case of cerebral venous thrombosis, vitamin K antagonists are given with a target international normalized ratio of 2.5. Warfarinization may be an adequate treatment for spinal venous thrombosis.

From a pathophysiological point of view, inhibition of platelet aggregation has been associated with impaired thrombus formation both in an experimental model of venous thrombosis and in vivo. It has been reported that antiplatelet therapy significantly reduces the risk of fatal or nonfatal pulmonary embolism by 25%. Given that anticoagulation therapy has shown a superior efficacy and safety profile, the most recent guidelines advise against aspirin monotherapy for thromboprophylaxis in the surgical pa-
A lower risk of major bleeding in patients on long-term low-dose aspirin therapy (annual risk of major bleeding < 1%<sup>14</sup>) compared with vitamin K antagonist therapy (2.7% in 6 months<sup>15</sup>) might support a supplementary role in prolonged use.

With respect to the medication period, vitamin K antagonists for cerebral venous thrombosis are given for 6 months after a first episode of sinus thrombosis unless predisposing factors exist.<sup>16</sup> In the present case, dual antiplatelet therapy was continued for 1 year, because multiple clinical worsening and deposition of NBCA into the venous system were thought to be predisposing factors.

In spinal DAVF, extraspinal injection can reflux into the spinal veins, testifying to an impaired venous protective system.<sup>18</sup> Venous flow for the thoracolumbosacral cord is thought to return with the respiratory cycle.<sup>7</sup> It is unclear in the present case why the patient suffered acute deterioration in a period of 9 days rather than a slow and steady decline. Maneuvers that changed intraabdominal or intrathoracic pressure such as active rehabilitation may have suddenly aggravated his unstable venous outflow.

In the present case, symptomatic aggravation developed on the 9th day and reappeared on the 30th day, and it was determined to be the result of progressive venous thrombosis based on the findings on repeated MR images. We were able to treat the patient successfully with antithrombin/dual antiplatelet therapy even after his symptoms worsened due to the unexpected reduction to aspirin monotherapy; recovery could be achieved only by reinstituting dual antiplatelet therapy. Although it would have been better to perform spinal angiography to confirm the cause of delayed deterioration of symptoms, the present MR imaging findings and the patient’s later course support our diagnosis of progressive venous thrombosis rather than recurrence of spinal DAVF.

**Conclusions**

Progressive venous thrombosis following embolization of spinal DAVF can develop in the chronic phase and can be treated with dual antiplatelet therapy. It should be kept in mind that delayed deterioration is not necessarily caused by the recurrence of spinal DAVF.

**Disclaimer**

The authors do not report any conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

**References**


**TABLE 1**

Functional status determined by the modified Aminoff–Logue grading scheme

<table>
<thead>
<tr>
<th>Grade</th>
<th>Definition</th>
</tr>
</thead>
<tbody>
<tr>
<td>gait</td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>leg weakness, abnormal gait or stance, but no restriction of activity</td>
</tr>
<tr>
<td>2</td>
<td>restricted activity</td>
</tr>
<tr>
<td>3</td>
<td>requiring 1 stick for walking</td>
</tr>
<tr>
<td>4</td>
<td>requiring 2 sticks, crutches, or walker</td>
</tr>
<tr>
<td>5</td>
<td>confined to wheelchair</td>
</tr>
<tr>
<td>micturition</td>
<td></td>
</tr>
<tr>
<td>0</td>
<td>normal</td>
</tr>
<tr>
<td>1</td>
<td>hesitancy, frequency, urgency</td>
</tr>
<tr>
<td>2</td>
<td>occasional urinary incontinence or retention</td>
</tr>
<tr>
<td>3</td>
<td>total incontinence or persistent retention</td>
</tr>
</tbody>
</table>

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