A Patient with Spinal Epidural Arteriovenous Fistula Cured by Balloon-assisted Transarterial Embolization under Flow Control

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Objective: In transarterial embolization (TAE) of spinal epidural arteriovenous fistula (SEDAVF), it is essential to control the blood flow at the shunt point. We report a case of SEDAVF treated with TAE with occluding one of several segmental arteries (SAs) involved in the shunt using a balloon.

Case Presentation: A 68-year-old male presented with gait disturbance and bladder bowel dysfunction. Lumbar spinal MRI showed a dilated and tortuous vein around the spinal conus. Spinal angiography revealed a SEDAVF with intradural venous reflux through the epidural venous plexus fed by the branches of the right 2nd and 3rd lumbar arteries (L2 and L3). We infused 14% n-butyl-2-cyanoacrylate (NBCA) from the feeder of the L2 under the flow control by occluding L3 using a balloon and achieved complete obliteration of the arteriovenous shunt.

Conclusion: In treatment of SEDAVF with feeders from several SAs, TAE with occluding one of the SAs using a balloon is a useful method.

Keywords spinomedullary arteriovenous fistula, transarterial embolization, n-butyl-2-cyanoacrylate, balloon

Introduction

The incidence of spinal dural arteriovenous fistula (SDAVF) is the highest among spinal arteriovenous shunt diseases. The pathology of SDAVF is well known, whereas spinal epidural arteriovenous fistula (SEDAVF) is relatively rare. A After entering the epidural venous plexus via the arteriovenous shunt, SEDAVF causes myelopathy. This is induced by intradural venous reflux or a mass effect of venous plexus dilatation induced by the shunt blood flow. This condition requires treatment, including surgical treatment, endovascular treatment, and a combination of these. In endovascular treatment, when access to the lesion via a vein is difficult, transarterial embolization (TAE) with n-butyl-2-cyanoacrylate (NBCA) or Onyx is selected. To cure SEDAVF complicated by intradural reflux by TAE, sufficient penetration of the shunt point, the epidural venous plexus serving as a venous pouch continuous to the shunt point, and the drainer that refluxes into the intradural region by an embolic material is necessary. Control of the infusion speed is also necessary to prevent unexpectedly early consolidation of the embolic material and its scattering into the drainer. To achieve this, in addition to ensuring wedging a microcatheter into the feeder branching from the segmental artery (SA) selected as a target blood vessel, blockage of blood flow into the shunt from another SA at different levels is important. We report a patient with SEDAVF accompanied by intradural reflux treated with occluding one of two SAs with a balloon followed by TAE using NBCA under flow control and discuss the usefulness of this method.
Case Presentation

68-year-old male
Past medical history: Diabetes, chronic pancreatitis
History of present illness: The patient became aware of weakness in the right lower limb in April 2018. The weakness gradually progressed thereafter and walking became difficult. Backache developed in the middle of May. The patient visited another hospital in the middle of June. Spinal arteriovenous shunt disease was suspected on MRI of the lumbosacral region, and the patient was referred and admitted to our hospital. Urinary tract infection was noted when he was referred and the residual urine volume was large, suggesting complication by dysuria.

Neurological findings on admission: The patients’ consciousness was clear, and no abnormality was found in the cranial nerves or bilateral upper limbs. No muscle weakness of the lower limb was noted on the left side. However, on the right side, muscle strength of the femoral region, such as the iliopsoas muscle and quadriceps muscle, was 3/5 on the manual muscle test (MMT). Crural muscle strength of the tibialis anterior muscle and gastrocnemius was MMT 2/5. There was no sensory impairment in both lower limbs, and numbness was mainly observed in the right lateral thigh over the lateral crus. Urinary retention with overflow incontinence was observed. The Aminoff-Logue grade was G4 M3 and the McCormick classification was grade 3.

Neuroradiological findings: On lumbosacral MRI, swelling of the spinal cord near the medullary cone and a high intramedullary T2 intensity at the Th11-12 level were observed, and a dilated tortuous abnormal vein was present on the ventral side of the spinal cord (Fig. 1A and 1B). On CT angiography, no shunt point could be clearly identified. Spinal angiography: On day 3 after admission, a 4French (Fr) sheath was placed in the right femoral artery and spinal angiography was performed using a 4Fr diagnostic catheter. Two SAs were involved and the radicular arteries (RAs) that bifurcated from the right second and third lumbar arteries (L2 and L3) were the feeders. The shunt point was identified near the intervertebral foramen between the right third and fourth lumbar vertebrae. A dilated epidural venous plexus forming a venous pouch that was continuous to the shunt point was identified. Epidural antegrade venous outflow around the spinal canal and intradural reflux from the emissary radicular vein to the perimedurally vein via the radicular vein was observed (Figs. 2 and 3A). Based on the above findings, the patient was diagnosed with SEDAVF. TAE with NBCA was selected because of predicting difficulty in the transvenous approach to the lesion.

Endovascular treatment: Treatment was performed under general anesthesia on day 4 after admission. Glucagon was intravenously administered to inhibit intestinal peristalsis.
Two SAs, L2 and L3, were involved in the lesion. The vascular diameter of the feeder that bifurcated from L2 was considered appropriate for NBCA infusion while wedging a microcatheter and observing NBCA penetration into the lesion. Occlusion of the feeder from L3 with a balloon during NBCA infusion and flow control in the shunt were considered necessary. In the bilateral femoral arteries, 6Fr sheaths were placed and 3000 units of heparin was administered for systemic heparinization, followed by continuous intravenous administration at 1000 units/hour. Hearttrail II 6Fr JR4 (Terumo Corporation, Tokyo, Japan) and 4Fr Cerulean (Medikit co. ltd., Tokyo, Japan) were placed in L2 with the same axis and used as guiding catheters, and Marathon (Covidien, Irvine, CA, USA) was placed in the RA heading for the shunt point, which was the feeder. Britetip 6Fr JR4 (Cordis, Miami, FL, USA) was placed in L3 and Scepter 4 mm × 10 mm (Terumo) was placed at a site before the origin of the RA (Fig. 3B). The balloon of Scepter was inflated to block the feeder flow from L3. In this state angiography from L2 showed that laminar flow from L3 was lost. This finding suggested that flow could be controlled. NBCA warmed to 37°C and lipiodol were mixed to prepare 14% NBCA. Infusion of this solution through the Marathon placed in the feeder from L2 was initiated (Fig. 4A). After NBCA favorably penetrated the shunt pouch and transferred to the extradural outlet vein (Fig. 4B), infusion was once paused for several seconds. When infusion re-started, reflux into the intradural vein occurred (Fig. 4C). Although intradural transfer of NBCA was incomplete, the venous pouch was smoothly filled (Fig. 4D), and then reflux through the shunt point into the feeder from L3 occurred. Thus, infusion was re-suspended (Fig. 4E). Infusion re-started after several seconds and reflux into the Marathon replacement region occurred, and infusion was completed (Fig. 4F). Disappearance of the lesion was confirmed on angiography of the right L2 and L3 after TAE (Fig. 5), and the procedure was completed.

Course after treatment: On spinal angiography on day 14 after treatment, disappearance of the shunt blood flow was confirmed and no intradural dilated tortuous vein was enhanced. Dysuria did not improve, spontaneous urination did not return, and self-catheterization was introduced. Muscle strength of the right lower limb improved to MMT 4/5 and practice of walking was initiated. On day 19 after treatment, the patient was transferred to a rehabilitation hospital with a modified Rankin Scale (mRS) of 3. At 3 months after treatment, the patient was self-ambulatory without a cane. Dysuria also tended to improve and the frequency of self-catheterization was decreased, and mRS was 2. On lumbosacral MRI, swelling of the spinal cord had improved and the high intramedullary T2 intensity and abnormal intradural vein had disappeared (Fig. 6).
Discussion

SEDAVF is classified into intradural/perimedullary drainage (pMD), paravertebral drainage (pVD), and combined perimedullary and paravertebral drainage (CD). This classification is based on the drainer pattern after flowing into the epidural venous plexus from the arteriovenous shunt as found in a multicenter study performed in Japan.\(^1\) In this previous study, 44, 6, and 9 of 59 cases were pMD, pVD, and CD, respectively.\(^1\) In previous reports on SEDAVF, Huang et al.\(^3\) and Takai et al.\(^2\) closely reviewed the drainer pattern-based clinical characteristics of 101 and 45 cases,
respectively. Huang et al. reported that pVD and CD accounted for 73% and 27% of all cases, respectively. These authors showed that 38% (11 cases) and 62% (18 cases) of cases with pVD and CD, respectively, had lesions in the lumbosacral region, which indicated that CD accounted for the majority. Furthermore, 92% (50 cases) and 7% (4 cases) of cases with pVD and CD, respectively, had lesions in the cervical region, which suggested that pVD accounted for the majority. Takai et al. classified pMD and CD as Type A and pVD as Type B. In Type A, the mean age at the time of diagnosis was 63.5 years, many cases are male, and the location of the lesion was at the lumbosacral level in many cases. In contrast, in Type B, the mean age was 34.3 years, there was no sex difference, and the location of the lesion was at the thoracic cervical level in many cases. All disease types can induce myelopathy. The cause of myelopathy is venous congestion due to intradural reflux in pMD, whereas the cause is a mass effect of marked dilation of the epidural venous plexus by shunt blood flow in pVD, and the pathology differs depending on the drainage pattern. The present case was classified as CD. Our findings are consistent with the above-mentioned characteristics of the age at the time of diagnosis, sex, and level of the lesion. A venous pouch was formed due to dilatation of the epidural venous plexus, but the degree of the dilatation was not so much to show a mass effect, suggesting that the cause of myelopathy was intradural reflux-induced venous congestion. In a review by Huang et al., the disease manifested with hemorrhage in 10% (10 cases), epidural hematoma in 5% (five cases: at cervical, thoracic, and lumbosacral levels in 2, 2, and 1, respectively), and subarachnoid hemorrhage in 5% (cervical level in all five cases), showing that complication by hemorrhagic lesions should be taken into consideration.

For treatment of SEDAVF, surgical treatment and endovascular treatment have been reported. In a review of 45 cases of SEDAVF by Kiyosue et al., the success rate of surgical treatment was 56% (9/16), that of endovascular treatment was 59% (17/29), and disappearance of AVF was achieved. In a review by Huang et al., the success rate of the initial treatment was 74% (23/31) in cases treated with surgical treatment and 55% (36/65) in those who were treated with endovascular treatment. This finding that surgical treatment was superior, although the difference was not significant. Regarding endovascular treatment, cases treated with TAE and those with transvenous embolization (TVE) have been reported. For SEDA VF, the rate of cure by surgical treatment is high compared with the rate by endovascular treatment, but invasiveness of laminectomy and complication by infection are disadvantageous. Opportunities to perform endovascular treatment as the first-line treatment have been increasing, and this tendency may be accelerated with the improvement of treatment techniques and advances of devices. There is no doubt that endovascular treatment will become the main stream of treatment also for SEDAVF. However, the incidence of SEDAVF is low, superiority or inferiority of various treatment modalities has not been sufficiently discussed, and treatment strategy has not been established. Additionally, there are various factors to be investigated, such as the presence or absence of intradural reflux, the quantity of flow into the epidural drainer, and selection of TAE or TVE for endovascular treatment. Treatment strategy corresponding to the individual pathology needs to be determined.

The present case of SEDAVF accompanied by intradural reflux-induced myelopathy and transvenous approach to the lesion was considered difficult. Therefore, TAE was selected and a liquid material was considered for the
embolic material. Onyx has recently been frequently used for the treatment of intracranial arteriovenous shunt due to the advantage that the penetration range is easily controlled owing to slow consolidation compared with NBCA. 11) However, the inferiority of Onyx to NBCA in the long-term outcome of SDAVF treated with TAE in spinal shunt disease has been reported. 8, 12, 13) We selected NBCA for the embolic material in consideration of this issue. To ensure that NBCA reached the distant intradural drainer, it was used at a low concentration (14%) to slow down the consolidation rate.

The key to success of treatment in the present patient was minimizing scattering of infused NBCA to the epidural drainer around the paravertebral region and ensuring penetration into the venous plexus, which dilated from the intradural drainer and formed a venous pouch. Furthermore, we needed to ensure penetration of NBCA to the shunt point, for which blood flow control at the shunt point was essential for favorably controlling the penetration range and speed of NBCA infusion through the target blood vessel. In the present patient, the feeders from two SAs, L2 and L3, were involved. L2 was selected for the target blood vessel because its diameter was suitable for NBCA infusion and it was also considered appropriate for observing penetration of NBCA. In addition to ensuring wedging Marathon into the target blood vessel during NBCA infusion, blood flow from the feeder from L3 was blocked with a balloon to temporarily prepare a vascular architecture close to a single feeder-single drainer structure. Yamakawa et al. 14) simplified the vascular architecture from the feeder to shunt and drainer by blocking blood flow from the feeder other than the target blood vessel with a balloon in TAE to treat SDAVF involving multiple SAs. The same method was used for the present patient. We considered that prevention of early consolidation of NBCA by controlling flow was more important for SDAVF passing through a relatively large venous pouch from the feeder, which may have led to sufficient NBCA traveling to the perimedullary vein located distant from the venous pouch. Yamakawa et al. 14) used a single lumen-type balloon, whereas we used double lumen-type Scepter in our patient considering that additional embolization through the wire lumen of Scepter can be performed in the case that TAE through the target blood vessel was insufficient. Introduction of this method may be useful for TAE of SDAVF accompanied by intradural reflux. The reason for this is that TAE of SDAVF is technically difficult due to the following two conditions compared with TAE of SDAVF: 1) The embolic material infusion range is wide and includes the epidural venous plexus present between the shunt point and drainer and 2) control of scattering of the embolic material to the epidural drainer is necessary.

This method requires caution because of difficulty in guiding a balloon to a small feeder serving as the target blood vessel for infusing the embolic material and the balloon should be placed in a region with a certain size before the feeder. Therefore, sufficient investigation of the placement position is required, and when the anatomy is inappropriate for preparation of the vascular architecture, this method may not be applicable. Moreover, when NBCA is refluxed into the feeder occluded with a balloon through the shunt point during NBCA infusion from the target blood vessel, there is a risk that the balloon catheter is adhered. Therefore, sufficient observation of whether there is reflux toward the balloon placement region is necessary.

I Conclusion

SEDAVF accompanied by intradural reflux involving feeders from two SAs was treated with TAE using NBCA under flow control after occluding one feeder with a balloon, and a favorable outcome was achieved. For TAE of SDAVF, prevention of scattering to the epidural drainer as much as possible and penetration of the embolic material in a wide range due to the presence of the epidural venous plexus between the shunt and drainer are necessary. Our findings suggest the usefulness of this method, which is capable of favorably controlling the shunt blood flow in treatment of SDAVF with feeder of several SAs.

I Disclosure Statement

None of the first and co-authors has conflict of disclosure.

I References

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