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CASE REPORT

Intraoperative direct puncture and embolization (IOPE) using a glue material for spinal cord arteriovenous fistula: a case report

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Abstract
Introduction Spinal arteriovenous fistula (AVF) is treated by embolization or surgery. However, transarterial embolization or surgery is difficult in rare cases when the fistula site is very complicated to access especially as in fistular nidus supplied by posterior and anterior spinal artery. We present the case which was treated with intraoperative direct puncture and embolization (IOPE) using glue material, since the usual transarterial or transvenous neurointerventional approach was difficult to embolize the AVF.
Methods A 36-year-old woman presented with progressive leg weakness and pain after a 20-year history of lower back pain. She had pelvic and spinal AVF combined with arteriovenous malformation (AVM). Despite prior treatment of the pelvic lesion with radiotherapy and coil embolization, the spinal lesion persisted and caused repeated subarachnoid hemorrhages. A spinal angiogram revealed a tortuous and long feeder of the AVF which had growing venous sac, as well as AVM. Two embolization trials failed because of the long tortuosity and associated anterior spinal artery. Four months later, drastic leg weakness and pain occurred, and IOPE was performed using a glue material.
Results The subsequent recovery of the patient was rapid. One month later, the use of a strong opioid could be discontinued, and the patient could walk with aid. A follow-up spinal angiogram revealed that the venous sac of the AVF had disappeared.
Conclusion In spinal AVF which is not feasible to access by usual intervention approach and to dissect surgically, IOPE with glue material can be considered for the treatment.

Keywords Arteriovenous fistula · Glue embolization · Intraoperative direct puncture · Spinal arteriovenous fistula · Direct puncture embolization for spinal AVF

Introduction

The most popular treatment approach for spinal arteriovenous fistula (AVF) is endovascular embolization or surgical ligation of the fistula site. Surgery can be performed when the fistular feeder can be surgically identified or there is a common origin of the normal anterior or posterior spinal artery and the feeder of the fistula [1]. Surgery is also challenging when the feeders from the anterior and posterior spinal arteries are intermingled with other malformed vessels, and thus cannot be dissected owing to the complexity of the angioarchitecture [2]. We here report a case where a glue material was used for an intraoperative direct puncture and embolization (IOPE) of the venous sac of a fistula site: the medical history is summarized in the Fig. 1. This was necessary because access using a neurointerventional approach was complicated by a tortuous and long feeder as well as association with anterior spinal artery (ASA) and surgical ligation was not feasible owing to an intermingled arteriovenous malformation (AVM).

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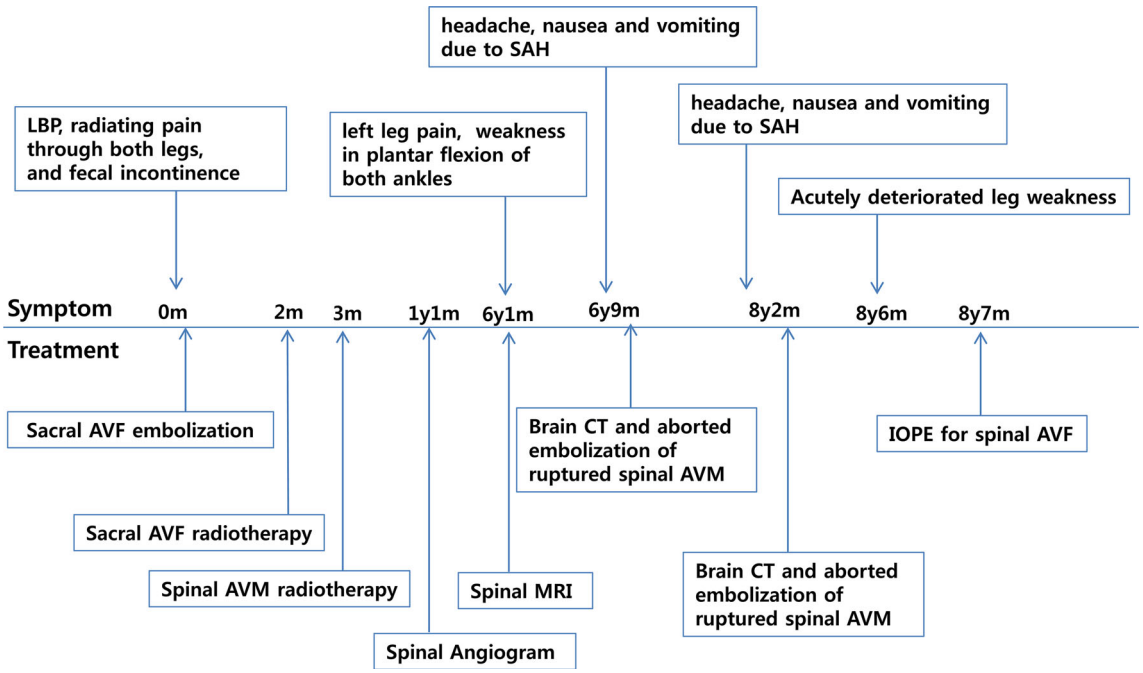

Fig. 1 Schematic diagram demonstrating patient's symptom and treatment procedure. *AVF* arteriovenous fistula, *AVM* arteriovenous malformation, *CT* computed tomography, *IOPE* intraoperative direct puncture and embolization, *LBP* low back pain, *MRI* magnetic resonance image, *SAH* subarachnoid hemorrhage

Case report

History and clinical presentation

A 36-year-old woman presented at our outpatient department with lower back pain (LBP) that she had suffered from for 20 years. Nine years previously, she had been admitted to our hospital for LBP, radiating pain through both legs, and fecal incontinence. A spinal angiogram (first angiogram) revealed a spinal arteriovenous fistula (AVF) at the posterior surface of the conus medullaris supplied by the right T9 posterior spinal artery (PSA) and left T9 ASA and combined with spinal AVM from the left T9 ASA. A metameric multiple AVF at the sacral bony level with high flow shunt supplied by both internal iliac arteries was also identified. For this sacral AVF, she received coil and glue embolization, which relieved both the pain and fecal incontinence. Two months after this sacral embolization, radiotherapy for the sacral AVF and spinal AVM with AVF were also performed 30 Gy and 25 Gy, respectively.

A follow-up spinal angiogram (second angiogram) that was conducted after 11 months indicated an almost complete disappearance of the pelvic AVM, but spinal AVM with AVF at the spinal conus medullaris level was observed with no change. Five years after radiotherapy for spinal AVM with AVF, the patient began to suffer from left leg pain and experienced difficulty in plantar flexion of both ankles. In addition, enlarged draining veins at the conus were noted in magnetic resonance images (MRI). Eight months after the MRI study, she experienced severe headache, nausea and vomiting. Brain computed tomography (CT) analysis revealed subarachnoid hemorrhage (SAH), which was diagnosed as spinal AVM rupture given that the cerebral angiogram did not showed any aneurysm or cerebral vascular abnormality. Although we attempted embolization (third angiogram, first spinal embolization trial) of the spinal AVM with AVF, it was not successful because the left T9 ASA was unstable, and spasm and flow arrest occurred at the right T9 PSA. A venous approach through the draining radicular vein was also unsuccessful. A second spinal embolization trial (Fig. 2) was aborted one and a half years after the first trial, when SAH occurred again.

Four months after the second spinal embolization trial for this patient, the motor power of her lower extremities acutely deteriorated so that she could not walk without a cane, and she had to take oxycodone (30 mg per day) to control her leg pain. We decided to conduct IOPE at the AVF site, which seemed to be a main cause of spinal cord compression, when the patient was confined to a wheelchair (her knee flexion/extension was grade 4 but other joints of lower extremities were grade 1–2 only).

Surgery

The patient was placed in the prone position after endotracheal general anesthesia. The operating area was

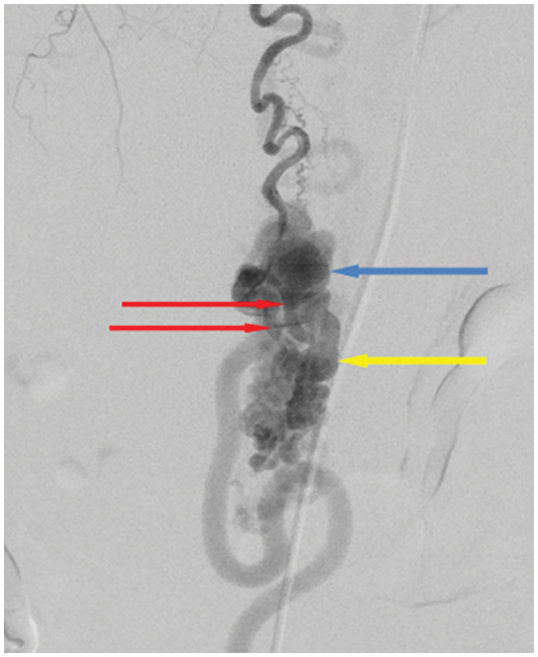

Fig. 2 Preoperative spinal angiogram in the AP view showing the double AVF feeders (*red arrows*) of the PSA from the T9 radiculopial artery and venous sac of the AVF (*yellow arrow*), which is associated with congestive venous myelopathy and aggravation of the patients symptoms through the compression of the spinal cord. The dilated AVM sac is also evident (*blue arrow*). *AP* anteroposterior, *AVF* arteriovenous fistula, *AVM* arteriovenous malformation, *PSA* posterior spinal artery

sterilized and draped, and a monitor to assess intraoperative motor evoked potential (MEP) and somatosensory evoked potential (SSEP) was set up. After a T11 to L2 laminectomy and midline dura incision, the AVM and the venous sac of the AVF on the posterior side of the conus medullaris were exposed (Fig. 3). Indocyanine green angiography (ICG) with a microscopic view (Carl Zeiss, 2009, Germany) (Fig. 4) was first performed to evaluate the anatomy of the lesion. We then dissected a PSA feeder to the venous sac of the AVF to prepare for ligation when direct puncturing failed owing to severe bleeding. During this dissection, however, the feeder was torn but could be controlled by the application of two miniclips. Direct puncture with a 20-gauge needle into the venous sac of the AVF was conducted and the glue (a mixture of 3 cc lipiodol and 1 cc NBCA glue) was injected to embolize the fistula sac (Fig. 5). There was small amount of hemorrhage after the sac was punctured, although this bleeding stopped after the injection of the glue. The successful embolization of the venous sac was confirmed by ICG (Fig. 6). A second puncture and glue injection were carried out at the clipped feeder (Fig. 7) and the miniclips were removed. The wound was closed with duroplasty after no change in MEP and SSEP was ascertained.

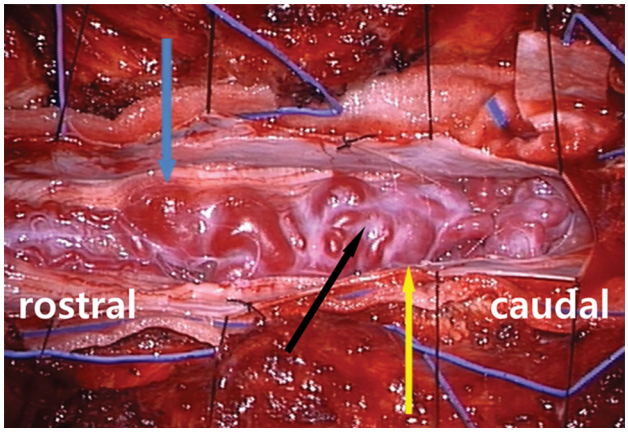

Fig. 3 Microscopic image of the patient before direct puncture. The venous sac of the AVF (*yellow arrow*) on the posterior side of the conus medullaris was exposed via laminectomy. One of two feeders (*black arrow*), which are marked with *red arrows* in Fig. 2, was found to be intermingled with the AVM. The *blue arrow* indicates a dilated venous sac

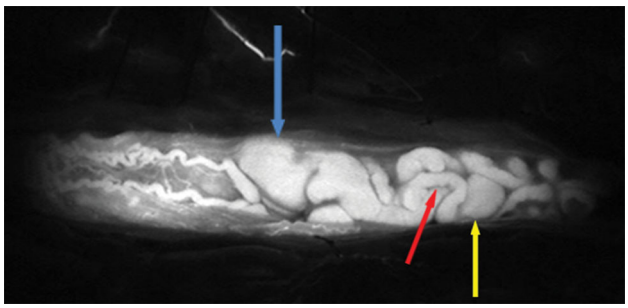

Fig. 4 Microscopic imaging by ICG of the AVM and venous sac of the AVF before direct puncture. The fistular sac of AVF is indicated with the *yellow arrow*. One of the two feeders marked with the *black arrow* in Fig. 3 is indicated here with the *red arrow*. The *blue arrow* indicates a dilated venous sac that corresponds to the same structure shown in Fig. 3. *ICG* indocyanine green angiography

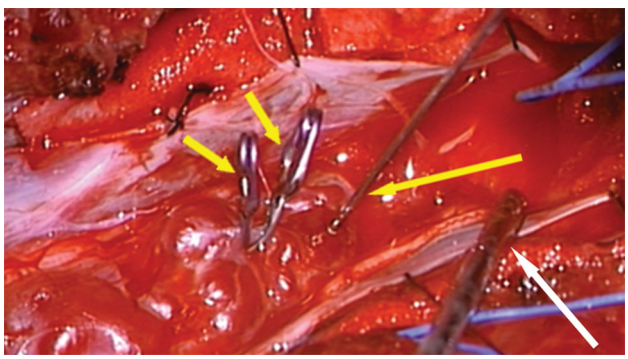

Fig. 5 Microscopic view during direct puncture with a 20-gauge needle (*long yellow arrow*) and glue embolization of the AVF venous sac. Two miniclips were applied for bleeding control (*two short yellow arrows*) to the AVF feeder (*black arrow* in Fig. 3), which was torn during dissection. The suction tip (*white arrow*) is also visible in this view

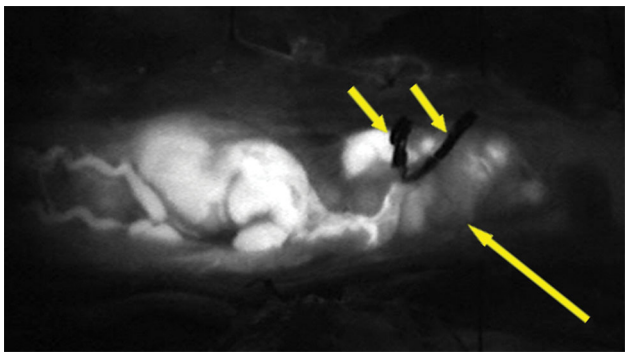

Fig. 6 The ICG under microscopic view after embolization of the AVF venous sac revealed successful embolization of the venous sac (*long yellow arrow*). The two miniclips applied to the feeder appear as dark structures (*two short yellow arrows*)

Follow-up clinical postoperative status

After the operation, the patient recovered rapidly. At the second postoperative day, her leg pain was relieved and the proximal leg motor power improved from grade 2 to 3. At the ninth postoperative day, she could walk with a cane. In a follow-up angiography and MRI 1 week and 2 months later, respectively, the embolized fistula sac was revealed to have disappeared almost completely (Figs. 8, 9). Although the AVM-related lesion and sac was observed, they did not seem to provoke any symptoms.

Discussion

Traditionally, spinal AVM is classified into four groups [3]. Type 1 is the most common form and involves dorsal intradural AVF. It is supplied by a dural artery and drains into the coronal venous plexus. It can cause myelopathic venous

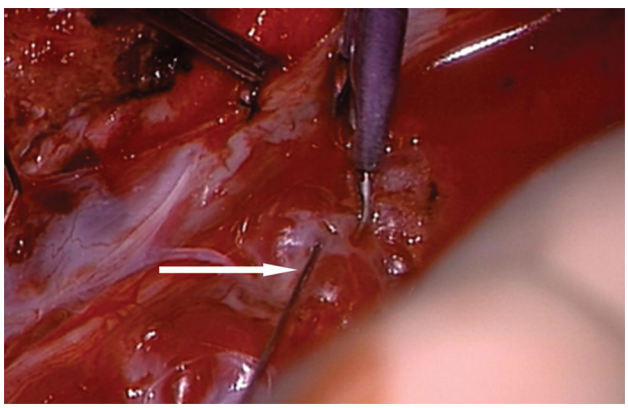

Fig. 7 A second puncture for a further glue injection was performed at the feeder by 20 gauge needle (marked with a *white arrow*) just above the miniclips at the previously torn portion of the feeder. Note that the clip applier is removing the second miniclip after the glue needle was securely punctured

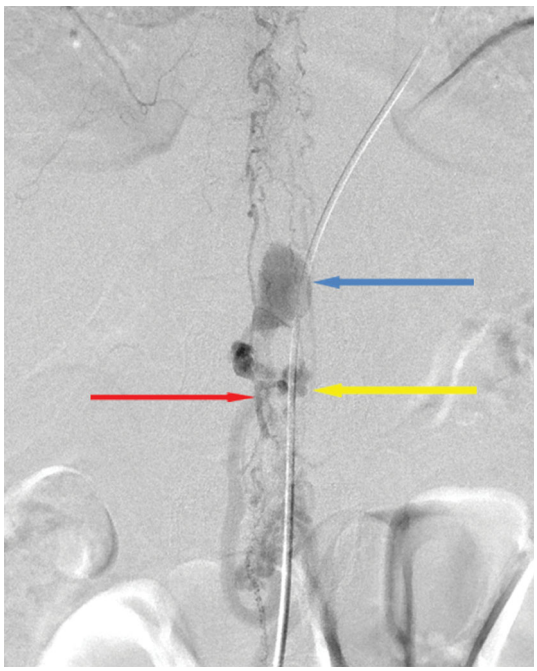

Fig. 8 Postoperative spinal angiogram in the AP view showing a largely disappeared AVM nidus, except for a small reduced embolized fistula sac (*yellow arrow*) and residual AVM-related sac (*blue arrow*). Only one of the two AVF feeder from the PSA (*red arrow*) is observed. Another AVF feeder which was embolized is not visible

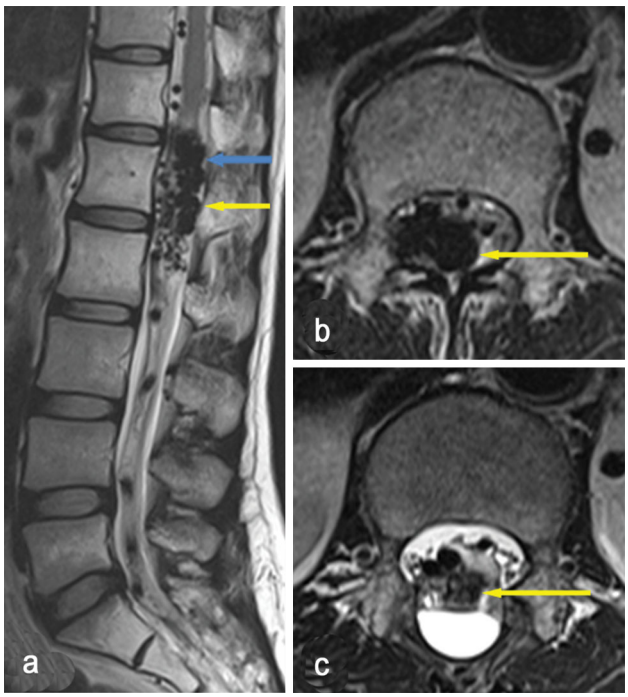

Fig. 9 Preoperative sagittal and axial MRI images are shown in parts **a** and **b**. The fistular component and AVM are indicated by *yellow* and *blue arrows*, respectively. A signal change and decreased size in the glue embolized fistular sac is evident by postoperative MRI 2 months later (**c**)

congestion and myelopathy [4, 5]. Type 2 is an intramedullary AVM or glomus AVM. This involves compact blood vessels that are supplied by single or multiple branches of the ASA and PSA. Type 3 is an extradural–intradural AVM or juvenile AVM. Type 3 AVM has vague boundaries and can involve adjacent soft tissue, such as bone, muscle, and skin. This is fed by enlarged medullary arteries via dilated ASA and PSA. Type 4 is a ventral intradural AVF or perimedullary AVF. This is supplied by medullary arteries through a PSA, and a direct pial fistulous communication lies between an intradural spinal artery and the coronal venous plexus. The case in our present report had a combined lesion that involved a mixture of type-3 juvenile AVM and type-4 perimedullary AVF. Therefore, total excision was difficult, and we designed to treat symptom-related AVF only.

Conventionally, there are three treatment options for spinal cord AVF [6]. These are surgery alone, embolization alone, or surgery after embolization. Surgery involves disconnection of the vessel between the feeding arterial supply and the draining vein [7] or removal of the abnormal vessel nidus. Surgery is indicated when embolization has failed to occlude the fistula site, or if embolization of the main tributaries threatens to occlude the arteries that feed the spinal cord [8]. Embolization is the injection of the embolic material into the vein with selective microcatheterization of the feeding artery [6]. The indications for embolization of spinal cord AVMs are a symptomatic patient with lesions that can be treated [6]. When AVM is complex, surgery after embolization or repeated embolization should be performed [9]. In this case, we performed repeated diagnostic spinal angiography and an embolization trial after the treatment of pelvic AVM. We then decided to use direct puncture and injection of embolic material into the AV fistula site with surgical exposure owing to our failure to access the fistula site due to a long tortuous PSA, the progression of pain and the weakness of the lower extremities in our patient.

There has been no previous report on intraoperative fistula embolization by direct puncture which we abbreviated to IOPE. Theoretically, we can access the fistula site by direct intraoperative puncture [10, 11] but there might be a risk of vessel bleeding when the exposed fistula site, which is not covered by soft tissue, is punctured by the embolization needle. Therefore, we also designed to clip and ligate the fistula site if the bleeding after puncture would be serious and impossible to proceed the embolization procedure. However, the bleeding after puncture in our current case was negligible, and it was feasible to proceed with glue injection. In contrast, the arachnoid membrane around the feeders was very thick in our patient and the vessel complex was friable and intermingled. It was, therefore, difficult to dissect and ligate fistula site selectively. In fact, the feeder was torn during dissection before sac puncture and we could control the bleeding only with vascular clipping. When the needle was withdrawn after glue injection into the fistula sac, the impacted glue material also prevented bleeding.

The residual sac of AVM which was observed in the postoperative angiogram is low pressure status, and we consider the sac does not affect the symptom and the size would not progress, which should be evaluated with follow-up study. The patient's pain was recovered from VAS (visual analogue scale) score [12, 13] 7 to 3 and the weakness recovered from grade 2 (according to ISNCSCI) to walker aided gait (grade 4) [14, 15]. Therefore, we think IOPE using glue material might not be perfect treatment, but accomplished clinically satisfying result for the patient. Long-term follow-up for the residual AVM will be needed.

IOPE procedure should be considered cautiously because there might be several risks in this procedure. Usually percutaneous vessel puncture by needle does not evoke bleeding or rupture because the surrounding soft tissues which compress and protect the vessel prevent from serious bleeding complication. However, needle puncture into operatively exposed vessel might have the risk of bleeding complication. Regarding another risk of IOPE, we have to consider the disparity between radiological image and surgical finding. The discrepancy could mislead into wrong target point. In addition, when the glue is injected excessively in the cases of AVF intermingled with AVM, there is a risk of obliteration of AVM feeder. We can perform fistula site clipping and duroplasty as an alternative procedure when the IOPE fails due to massive bleeding or fistula sac rupture. In the future technique, we could consider coil material to embolize instead of glue material.

Conclusion

We present a case of spinal cord AVF treated by intraoperative direct puncture and embolization (IOPE) using glue material. When both conventional embolization and surgery are not suitable, this procedure could be considered for the treatment of spinal AVF.

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Conflict of interest None of the authors has any potential conflict of interest.

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