

## Original Article

# Analysis of the embolization spinal dural arteriovenous fistula and surgical treatments on 52 cases of the patients

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**Abstract:** Background: Spinal dural arteriovenous fistula (SDAVF) highly threatens people's life and health. Effective methods for the diagnosis and treatment of the disease are badly needed in clinical application. Objective: The objective of the present study was to sum up the diagnosis and treatment method of SDAVF to improve the diagnosis and treatment effect of the disease. Methods: The epidemiological data, imaging data, therapeutic methods and postoperative follow-up data of 52 cases of patients with SDAVF received in our hospital in recent 6 years were collected and retrospectively analyzed. Results: There were 43 male patients and 9 female patients with ages of 39-77 years and average age of 59.6 years. The course of disease was 1 to 48 months with an average disease course of 14.4 months. All the patients had syndromes of lower limb numbness, pain, weakness and other sensory and movement disorders mostly accompanied with defecation dysfunction. Magnetic resonance imaging (MRI) results demonstrated that spinal cord abnormalities were found in spinal cord, which could be diagnosed by digital subtraction angiography (DSA) examination. There were 40 cases received surgical treatment and there was no recurrence in the follow-up. There were 12 patients received embolotherapy, of whom 3 patients were operated the second time and 2 patients had embolization again. After 0.5-6 years of follow-up, postoperative symptoms of the 40 patients were improved in different degrees. The modified Aminoff-Logue function scoring was significantly decreased after treatment. Conclusion: SDAVF is the easily diagnosed and delayed spinal cord vascular lesions in clinical applications. The diagnosis relies mainly on MRI and DSA examinations. The surgical treatment effect is good and is not easily relapsed. The trauma of the interventional embolization treatment is small, but the recurrence rate is high.

**Keywords:** Spinal dural arteriovenous fistula (SDAVF), nuclear magnetic resonance, angiography, embolism, operation

## Introduction

Spinal vascular malformation accounts for about 15% of the central nervous system vascular malformation, and its incidence is low. The spinal dural arteriovenous fistula (SDAVF) is the most common disease of spinal vascular malformation, accounting for about half of spinal vascular malformations [1, 2]. Acquired factors play a decisive role in the pathogenesis of SDAVF. The cause of the spinal cord injury patients may be that the slowly increasing coronary venous plexus venous pressure leads to decreasing of the intramedullary arteriovenous pressure gradient [3].

SDAVF is a non-self-limiting disease. Once the disease occurs, the symptoms will be a progressive process, resulting in irreversible neurological damage [4]. After treatment, although the symptoms are reduced or disappeared, they also can increase or recurrence. The duration from onset of symptoms to diagnosis is long with an average of 2.7 years. The disease is a sporadic disease with insidious onset, and longer duration and gradually increasing of progression, so the early diagnosis is difficult [5]. Many patients have a serious loss of active movement ability before diagnosis. The only way to diagnose this disease is the selective



**Figure 1.** Spinal cord was thickened. After T2 weighting, high signal without unclear demarcation line in marrow can be seen. There were tortuous and creeping expanded vascular flow void signal image on the dorsal and/or ventral surfaces. Flow void signal on surface of spinal cord showed significant enhancement in the enhancement scanning. Especially, continuous tortuosity and dilatation of the vessel can be seen in the spinal cord coronal position.

angiography of spinal cord, which can clearly display the abnormal vascular lesion.

SDAVF is often misdiagnosed because of its insidious onset. Spinal cord function is hard to be recovered when SDAVF is confirmed. Therefore, the overall prognosis of the disease is poor and SDAVF is a serious threat to human health and life. The research of SDAVF has made great progress, but there are still many problems to be further elucidated, such as observing the disease in male elderly from the epidemiological point of view. The specific mechanism is still not clear. What are the pathological conditions of the mechanism under normally potential arteriovenous communication open conditions? The experimental animal model of the disease is still failed to be established. Therefore, we should strengthen the basic research of SDAVF to guide the clinical treatment and prevention.

In order to sum up the diagnosis and treatment method of SDAVF to improve the diagnosis and treatment effect of the disease. The epidemiological data, imaging data, therapeutic meth-

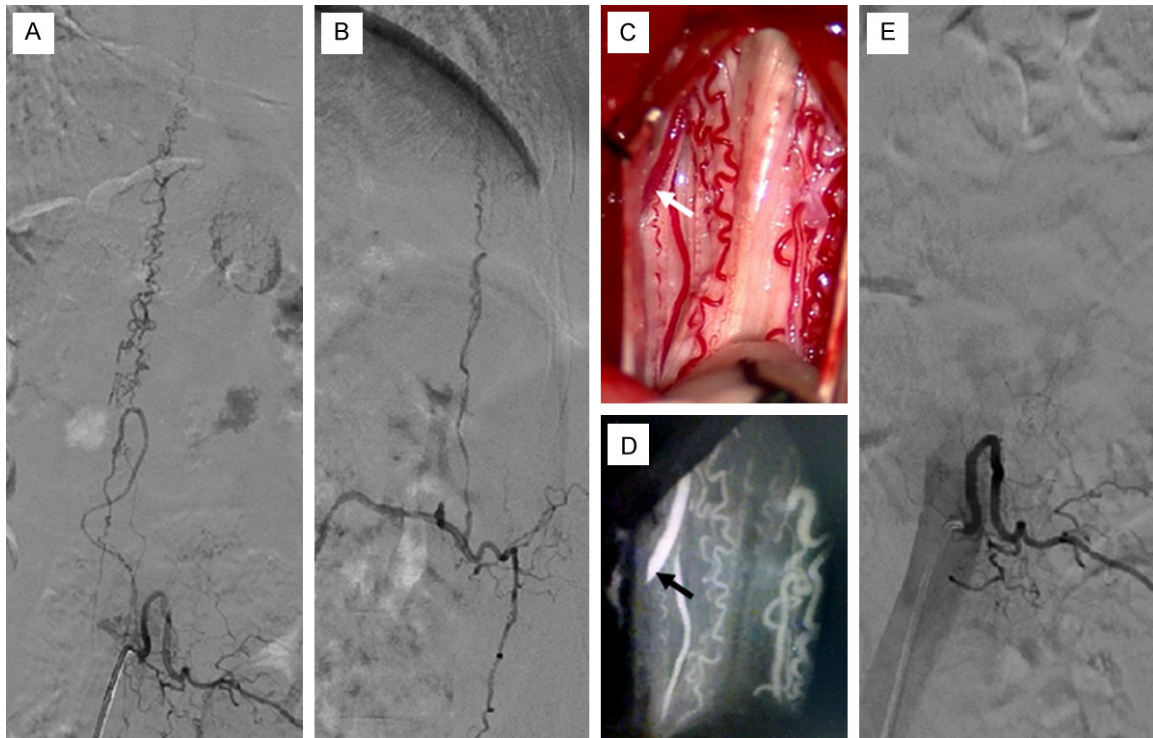
ods and postoperative follow-up data of 52 cases of patients with SDAVF received in our hospital in recent 6 years were collected and retrospectively analyzed.

### Patients and methods

#### General data

For the experiments involving human subjects, approval was obtained from the institutional review board of Changzheng Hospital, The Second Military Medical University. Informed consent was provided according to the Declaration of Helsinki. All the subjects signed the informed consent, and the study protocol was approved by the ethics committee.

There were 52 patients with SDAVF were recruited and treated between January, 2008 and September, 2013. There were 43 male patients and 9 female patients with ages of 39-77 years and average age of 59.6 years. The course of disease was 1 to 48 months with an average disease course of 14.4 months. All the patients had syndromes of lower limb numbness, pain,



**Figure 2.** A and B. SDAVF can be seen in the L1 left anteroposterior and lateral lumbar artery angiography. C. The draining vein drained to the surface of the spinal cord through endorachis. D. Intraoperative fluorescence imaging. E. Fistula was disappeared by the postoperative examinations.

weakness and other sensory and movement disorders mostly accompanied with defecation dysfunction (94.2%).

There were 13 cases had been mistakenly diagnosed as “disc herniation or lumbar spinal stenosis” in all the 52 patients, of whom 8 patients underwent cervical or lumbar operation treatment, but there was no improvement after operation. There were 15 cases were diagnosed as “myelitis, syringomyelia, arachnoiditis or demyelinating disease”, and the conservative treatment was invalid. There was 1 case that was diagnosed as “hyperplasia of prostate” and surgical treatment was conducted. A total of 12 patients were transferred from the Neurology Department to the Department of orthopedic of our hospital.

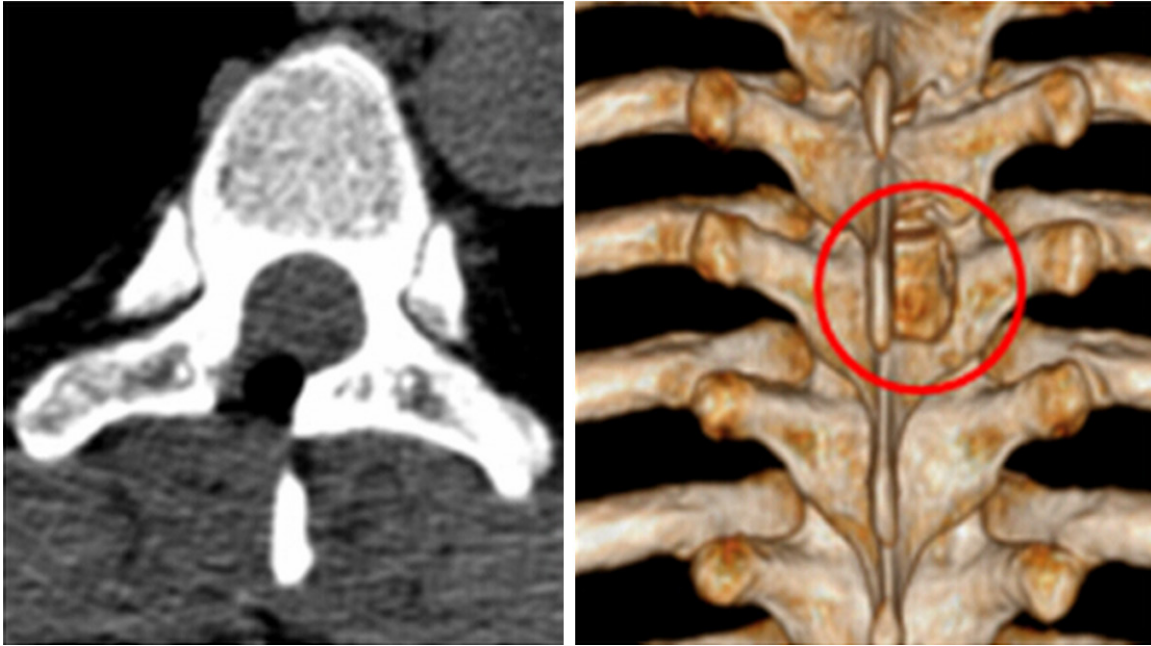
#### *Examination methods*

All patients underwent magnetic resonance imaging (MRI) and digital subtraction angiography (DSA) examinations of spinal cord. Spinal MRI examination was conducted in 52 cases, of whom, 49 patients showed spinal cord thickening, and long segmental diffuse high signal

intensity was observed along central tube distribution in spine in T2 weighting. The data indicate that there was edema of spinal cord in thoracolumbar spine and spinal cord conus.

There were tortuous and creeping expanded vascular flow void signal image on the dorsal and/or ventral surfaces. Flow void signal on surface of spinal cord showed significant enhancement in the enhancement scanning. Especially, continuous tortuosity and dilatation of the vessel can be seen in the spinal cord coronal position (**Figure 1**). There was uneven enhancement in spine of some cases with blurred boundary. There are 2 cases of patients with only spinal cord edema and 1 case with only flow void signal on the surface of spinal cord.

All the cases were confirmed by spinal angiography to be SDAVF. **Figure 2A** and **2B** showed that SDAVF can be seen in the L1 left anteroposterior and lateral lumbar artery angiography. **Figure 2C** showed the draining vein drained to the surface of the spinal cord through endorachis. **Figure 2D** was intraoperative fluorescence imaging. **Figure 2E** showed fistula



**Figure 3.** Postoperative imaging of semi-laminectomy fenestration surgery, which shows little influence on the stability of the spine, and auxiliary fixing is not needed.

**Table 1.** Modified Aminoff-Logue spinal function scoring (Patient score = the added scores of 3 items)

Gait	Urinate	Defecate
0. Normal gait and strength of lower limbs	0. Normal	0. Normal
1. Lower extremity is weakness, but walking is not limited	1. Urgency of urination, frequent micturition, delayed urine	1. Mild constipation, good response to laxatives
2. Exercise endurance is limited	2. Occasional incontinence or retention	2. The occasional incontinence or sustained severe constipation
3. Needing a crutch or some support when walking	3. Continuous incontinence or retention	3. Continuous incontinence
4. Crutches are needed when walking		
5. Cannot standing, bedridden or wheelchair		

was disappeared by the postoperative examinations. There were 51 patients with single fistula, and 1 patient with 2 fistulas.

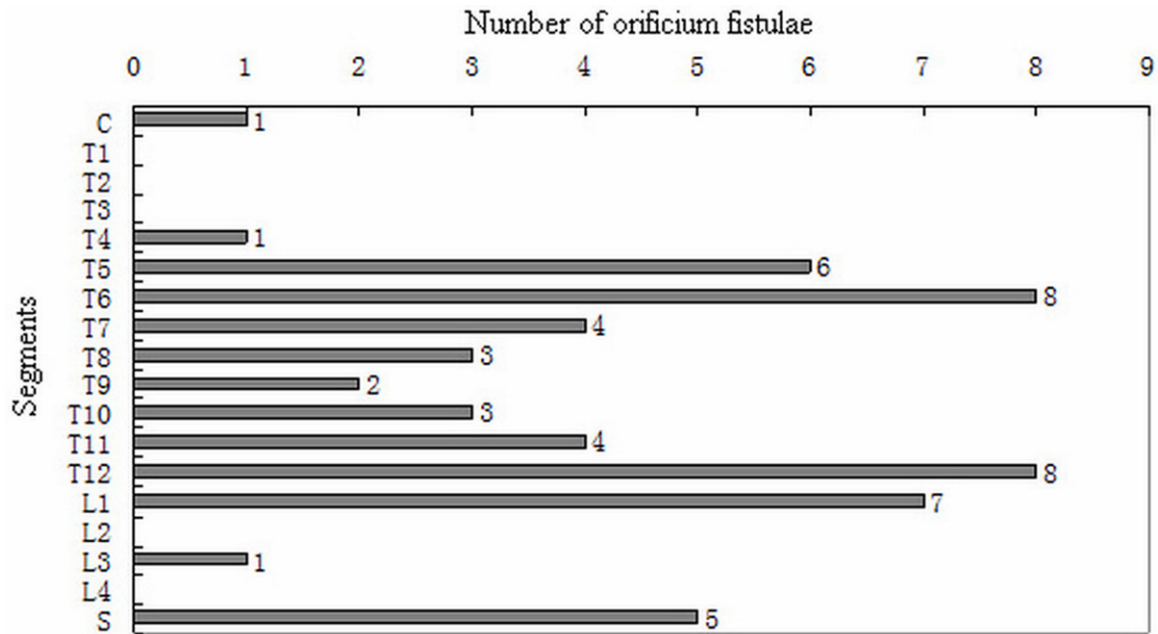
#### *Treating methods*

There were two surgical methods in the present study: operation cut proximal fistula of venous drainage and endovascular liquid glue embolization of fistula.

In the operation treatment, 0.5 ml methylene blue was injected to the corresponding spinous process of orificium fistulae to mark under pre-operative X ray or according to intraoperative fluoroscopy of the twelfth rib positioning on the thoracolumbar vertebra of the patients. After general anesthesia, prone position of the patients was used to make the corresponding

segment of dorsal midline incision. According to the Meilan marker, paraspinous muscle was isolated and spinous process was eliminated to open vertebral plate, or semi laminectomy was conducted at the lesion side of orificium fistulae (**Figure 3**). Endorachis was cut open, and the drainage vein from endorachis to the spinal cord surface was discovered under operation microscope, mostly located in the vicinity of nerve root cuff and close to the nerve root (**Figure 2C**). Intraoperative Doppler ultrasonography is helpful in judging the direction of blood flow and the distinction normal venous drainage. The vein mostly exhibited arterialized changes. Compared with that of the spinal angiography results, it was cut off at the electric coagulation of endorachis sites after confirmation of consensus morphology.

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**Figure 4.** The position distributions of 53 orificium fistula of 52 patients, mainly located in the chest 5 and waist 1, exhibited T6 and T12 double-peak distribution. There were a total of 5 in sacrum tail parts.

**Table 2.** The modified Aminoff-Logue scoring of spinal cord function of the patients with SDAVF before and after surgery

	Preoperative	postoperative	Deviation value
The average value (n=40, $\bar{x} \pm S$ )	7.1 $\pm$ 1.72*	4.65 $\pm$ 2.23*	2.45 $\pm$ 1.16
The maximum value	11	11	5
The minimum value	2	0	-2

\*P < 0.001, Preoperative and postoperative comparison.

### Efficacy evaluation and follow-up

Physical examination and spinal cord function score were conducted on the patients by using the modified Aminoff-Logue score according to the time points before treat-

ment and 6 months after discharge from hospital (**Table 1**). Radiography re-examination was conducted on the patients with occlusion of fistula in operation before discharge or re-admission. Spinal MRI examination was conducted on the patients with operation in 3 to 4 years of outpatient follow-up. A few cases received telephone interviews.

### Statistical analysis

Data were presented as mean  $\pm$  standard deviation (SD). Statistical comparisons were conducted by a one-way analysis of variance (ANOVA), followed by a Student-Newman-Keuls multiple comparison test using SPSS 13.0 software (IBM-SPSS, Inc, Armonk, NY, USA). SPSS 13.0 was used in the paired t-test for statistical analysis on the Aminoff-Logue score before and after treatments. A P < 0.05 was regarded that there was significant difference.

In the embolization treatment, local anesthesia radiography showed that there was no radiculomedullary artery imaging in feeding artery and orificium fistulae after drainage of vein, and other segmental artery also showed no abnormal vascular development. Micro catheter was selected with auxiliary of micro guide wire in operation. Dural vessels were selected to supply vascular fistula as close as possible to the fistula. Micro catheter angiography confirmed again that there was no radiculomedullary artery imaging. NBCA with concentration of 30-50% was injected slowly by micro catheter. When NBCA was first diffused into the draining vein or NBCA had reflux, injection was immediately stopped and tube was drawn. The fistula was no longer developed in the imaging. After embolization, arteriography was conducted on the bilateral upper and low segments at the lesion sites to observe there was fistula developing.



**Figure 5.** The left is T2 weighted MRI of preoperative thoracolumbar spinal cord, in which thoracolumbar spinal cord was thickened and high signal intensity in marrow. The right is the reexamination results 3 years after operation, indicating that high signal intensity in marrow was disappeared.

## Results

### General results

The of 53 fistula positions of the 52 cases were shown in **Figure 4**. There were 45 fistulas between the fifth chest and 1 waist (84.9%), and they exhibited double-peak distribution of T6 and T12. There was 1 orificium fistula in foramen magnum, 1 orificium fistula in T4 and 1 orificium fistula in L3. There were 5 SDAVF in the bilateral internal iliac and the sacrococcygeal region for blood supply at the middle part of sacrum. Orificium fistulae were located at the L5 level and in the sacral canal.

### Surgical results

Surgical treatment was conducted on the 40 patients. There were 3 patients with recurrent embolism and the second time operation was employed on the patients. Reexamination and angiography were conducted on 28 cases, and the results showed that fistula were disappeared. Before intraoperative cut drainage vein, the filling of the vein on the surface of the spinal cord was obvious, the color was bright red and tensile force was high. After cutting off

the venous drainage, intravenous tension was decreased significantly and even collapsed. The color was dark red, or even blood stagnation was obviously seen.

The development time of vein was obviously extended before cutting off of the venous drainage in 5 cases of intraoperative indocyanine green angiography, and the time was up to 20-25 seconds (**Figure 2D**). There were 2 cases of patients with postoperative complications. One case had wound infection and delayed wound healing after surgery. Another case had spinal epidural hematoma, and he was recovered very well after timely operation to remove the hematoma.

### Postoperative results

Orificium fistulae no longer had photographic developing immediately after embolization in 12 cases of patients. A total of 5 patients had palindromia, of whom there were 2 cases had obviously aggregated syndromes 3 hours and 5 hours after embolization with manifestations of Foix-Alajouanine syndrome. Spinal vascular radiography examination was conducted on all the patients, and the results demonstrated

that they all had fistula recurrence. NBCA embolization was conducted again on one patient, and he was improved significantly after treatment. Because his families did not agree to conduct embolism again, emergency operation was used to clamp the fistula of another patient. The postoperative symptom of the patient was slowly recovered, but the state was worse than that of the preoperative state. There was 1 case of recurrence 1 month after embolization outside the hospital, and he was cured in our hospital after another embolization. Aggregated syndromes appeared 1 month and 2.5 months after treatment with embolization the first time in our hospital for another 2 patients. The follow-up angiography showed that fistula had recurrence, and they were cured after clipping operation.

### *Treatment effects*

There were improved lower limb sensory and movement function in all the patients after treatments, exhibiting the recovery of lower limb and lower limb muscle strength recovery, and also with the corresponding recovery of urination and defecation function. However, the recovery of the patients was not consistent, the recovery of the patients with severe preoperative symptoms was not ideal.

### *Aminoff-Logue scoring*

A total of 40 cases were followed-up with a follow up time of 0.5-6 years. The modified Aminoff-Logue scoring was illustrated in **Table 2**. The score before treatment was  $7.1 \pm 1.72$ , and the score after treatment was  $4.65 \pm 2.23$ , indicating that the score after treatment was reduced by  $2.45 \pm 1.16$ . The postoperative score of 1 case of patient was 2 points higher than that of the preoperative score. Spinal cord MRI review was conducted on 32 patients 3 months to 4 years after operation, suggesting that edema of spinal cord was obviously improved or disappeared, and the abnormal vessel flow void was disappeared (**Figure 5**).

### **Discussion**

Although the pathogenesis of SDAVF is still unclear, the patients have similar characteristics: the patients are mostly old male patients. Fistula is commonly located in thoracic segment and lumbar segments. There are slowly

and progressively aggravated lower spinal cord injuries. The data suggest that the disease has a high possibility of acquired disease. SDAVF is believed to be associated with trauma, operation and other factors [1]. SDAVF accounted for about 70% of all the spinal vascular malformations with an annual incidence rate of about 5-10/million people. The incidence rate of the male patients is 5 times that of the female patients with an average diagnosis age of 55-60 years [2]. More than 80% of the patients with fistula located in the T6 to L2, and about 2% of the patients had more than 2 fistula or accompanied with perimedullary arteriovenous shunt [3, 6]. The present study is similar to that is reported at home and abroad.

SDAVF often has insidious onset. The initial symptoms include limb weakness, unsteady gait, limb distal sensory loss and back pain. With time progresses, nerve function damage is slowly increased, and sensory disturbance level rises. In the middle and late stages of SDAVF, there are paraplegia, loss of sensation, constipation, urinary incontinence, urinary retention and sexual dysfunction. Because the disease course progress is slow, dysnesia often is heavy when the disease is confirmed. In addition, about 70% of the patients were combined with sensory, movement and defecation dysfunctions [7-9].

Spinal NMR examination can preliminarily screen the disease, but the confirmation mainly relies on spinal angiography. MR T2 weighting sequences can often detect thoracolumbar spinal cord center edema, spinal cord peripheral vessel expansion, tortuosity and flow, which are the characteristic changes of venous congestions and are also important diagnostic imaging features of the disease [10]. The dilated and tortuous vessel angiography on the surface of spinal cord is dorsal part angiography commonly. With the further development of the disease, spinal cord atrophy can also be seen. The enhanced spinal MRA is helpful in determining the fistula location, so it is necessary to conduct all the segmental artery super selective angiography in the subsequent DSA examination [11]. Spinal CTA can also display the tortuous and dilated spinal cord vein and fistula position. However, because the SDAVF fistula may be located at any level of the foramen magnum to the middle of sacrum, the doses of

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radiation of all the ranges of scanning is too large, and thus it is not practical [12].

Spinal angiography is the gold standard for diagnosis of SDAVF. In the early stage of selective spinal angiography, venous tortuosity and dilatation on the surface of the spinal cord, slow flow and anterior spinal artery velocity delay can be seen. Although arterial blood generally derives from a single root artery, one fistula developing often can be seen when there is adjacent intercostal artery angiography in some cases. Fistula and drainage vein is usually single. The dilated medullary vein is accompanied with obvious stasis and it often drains to the skull and spinal cord partes sacralis at the spinal cord anterior or posterior surface. In a few cases, dural arteriovenous fistula at the skull base can drain to the surface of the spinal cord, which is consistent with that of SDAVF [13, 14]. The same situation may appear in the sacral, so it should be alert to SDAVF and sacral SDAVF when it is the highly suspected SDAVF and the spinal angiography is negative. In the present study, the first time DSA examinations of the 2 cases were negative. After the MRA examination, the result was still suggested to be SDAVF, so DSA was conducted again for confirmation. The data indicate that the complementary role of MRA to DAS is very necessary.

The present investigation had 1 case in pars cervicalis esophagi and 5 cases in partes sacralis, of whom there was 1 case of patient with left internal iliac artery was conducted left common iliac artery angiography in other hospital, but the fistula was undeveloped and so he missed diagnosis. We super elected to the internal iliac artery and found fistula developing and confirmed the disease, suggesting that the super selective angiography of spinal cord has important significance for SDAVF diagnosis.

The exact etiology of SDAVF is not clear, and the prognosis of the patients and disease are closely related to each other, so early diagnosis is depended on by the improvement of the curative effect of the disease. SDAVF has no specific symptoms and signs, so it is easy to be misdiagnosed. The symptom is not typical, and the patients almost have no upper limb disorders, and the sensory disturbance plane is often in the lumbosacral portion. Without treatment, the neural dysfunction is slowly and progressively aggravated. In 2-3 years, complete

paralysis of bilateral lower extremity will be developed in the patients. In the department of neurology, there are many multiple sclerosis and spinal cord inflammation treatment, and there are various protrusions of intervertebral disc operation treatments in Department of orthopedics. The disease is even misdiagnosed as benign prostatic hyperplasia [15]. A few patients may manifest as acute or subacute neurologic deteriorations, and urgent treatment is often required in this case to relieve venous hypertension and prevent spinal cord necrosis [16].

The diagnosis that should be identified with SDAVF mainly includes spinal arthritis and lumbar disc herniation. For the high-risk groups such as the elderly male patients with slowly increased lower limb sensory, movement and defecation dysfunction, spinal MRI examination can be conducted. If there is obvious thoracolumbar spinal cord edema especially tortuous and dilated vascular shadow around the spinal cords, vascular malformation of spinal cord should be highly suspected, and the spinal DSA examination can diagnose [17]. How to improve the rate of early diagnosis and reduce misdiagnosis is the key to improve the prognosis of this disease. Particularly, it is needed to improve the understanding of the disease by the doctors in Department of Neurosurgery, Department of internal medicine and Department of orthopedics.

SDAVF treatments include operation off the fistula draining vein and embolisation of the fistula these two methods [9, 18, 19]. Operation is the long-term and reliable treatment with less complication. Preoperative fistula implanted with spring ring and intraoperative Doppler ultrasonography can increase the safety of operation and reduce the operation wound [20, 21]. However, the trauma of embolization in the treatment is small. For a single blood supply artery, when micro catheter is easy to reach the fistula, endovascular embolization can be considered. The key to embolization treatment is completely and permanently occlude arteriovenous fistula and keep smooth of the drainage vein. The ideal embolization is liquid glue, and at the same time vein is drained at the embolization fistula and fistula proximal part. In the embolization process, it is needed to beware that the glue is prevented into the perimedullary vein so as to prevent aggravating of venous



congestion and spinal cord function barrier [21, 22]. However, the glue is expected to reach fistula and fistula proximal vein when conducting embolism, so it is very difficult to completely prevent entering of glue into perimedullary vein. Although some people think that even a small amount of glue gets into the medullary vein and does not lead to severe symptoms after complete embolization of fistula [23], even a small amount of glue getting into the perimedullary venous will also affect the venous drainage, resulting in spinal cord ischemia when fistula has recurrence or embolism is incomplete. In the present study, aggravated embolization of 2 cases after operation might be related to the above situation. At present, NBCA is still the most usually used embolic agent. The NBCA with antegrade flow toward fistula and injection concentration less than 30% has more opportunities to induce embolism venous drainage [24]. Although the success rate of application of ONYX embolization is high [25], the effect remains to be observed because there is reported high recurrence [26, 27].

In conclusion, the typical clinical manifestations of SDAVF are progressive exacerbation of double lower limbs paralysis and defecation dysfunction. It is a spinal vascular lesion that is easily diagnosed and it often has a delay diagnosis, so it is needed to improve the understanding of this disease. Diagnosis of SDAVF mainly depends on the examination of spinal MRI and DSA. The treatment effect of operation is good with less recurrence. The trauma of the interventional embolization treatment is small, but the recurrence rate is high.

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#### Disclosure of conflict of interest

None.

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