

Misdiagnosis of Spinal Dural Arteriovenous Fistula: An Analysis of 12 Cases

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Abstract

Background: Spinal dural arteriovenous fistula (SDAVF) is a rare spinal vascular disease. The clinical misdiagnosis rate is very high. The highest misdiagnosis rate is reported in orthopedics. The specific reason for misdiagnosis remains unclear.

Objective: To investigate the clinical and imaging manifestations of SDAVF, analyze the causes of misdiagnosis, propose countermeasures, and improve the orthopedists and other specialists' understanding of this disease.

Methods: The clinical data, diagnosis and treatment of 12 patients who had SDAVF that was misdiagnosed as a different disease were retrospectively analyzed, and the modified Aminoff-Logue Disability Scale (ALS) scores before and during follow-up were compared.

Results: From 2014 to February 2019, 15 patients were diagnosed with SDAVF at our institution. Twelve (80%) were misdiagnosed; of these, 6 (50%) were misdiagnosed more than twice, and 6 patients (50%) were misdiagnosed at least once. The initial diagnoses included lumbar spinal stenosis and lumbar disc herniation (LDH) in 6 patients, cervical spinal stenosis in one patient, benign prostatic hyperplasia (BPH) in one patient, myelitis in 2 patients, and syringomyelia in one patient. After the initial diagnosis, one patient was misdiagnosed with LDH, one was misdiagnosed with subacute combined degeneration of the spinal cord, and 4 were misdiagnosed with myelitis. The clinical manifestations mainly included weakness and numbness of the lower limbs, urinary symptoms, and numbness of the perineal area. In the 12 misdiagnosed patients, magnetic resonance imaging(MRI) showed signs of spinal cord edema and typical or atypical flow-void patterns. One patient had undergone extended cervical decompression and lumbar decompression. All patients eventually underwent microsurgical treatment. The average follow-up duration was 0.9 years. The modified ALS scores showed significant improvement in gait, bladder function and bowel movement, and the differences before and during follow-up were statistically significant ($P<0.05$).

Conclusion: When patients, especially those with bladder and bowel dysfunction, have symptoms such as weakness of the lower limbs and/or numbness, a diagnosis of SDAVF should be considered. Furthermore, if MRI shows signs of spinal cord edema and typical or atypical flow-void patterns, the diagnosis of SDAVF should be strongly considered, and further angiography is needed to confirm the diagnosis.

Background

In the middle-aged and elderly population, patients presenting with lower limb weakness or/and numbness are more likely to seek medical advice first from an orthopedic service, and most orthopedists diagnose diseases based on their own expertise. Since most orthopedists lack experience with spinal dural arteriovenous fistula (SDAVF), they may misdiagnose the disease. According to literature, the misdiagnosis rate for this disease is as high as 80%, and the highest rate is reported in orthopedic

services [1]. Many patients have severely damaged spinal cord function when they are diagnosed, which leads to permanent spinal cord injury [2]. Although digital subtraction angiography (DSA) has become the gold standard for diagnosing SDAVF, spinal-cord magnetic resonance imaging (MRI) is still a preferred imaging method in clinical practice, and its diagnostic accuracy is also closely related to the orthopedist's clinical experience [3]. The purpose of this paper is to retrospectively analyze the clinical data of 12 patients who had SDAVF that was misdiagnosed as another disease, summarize the clinical presentation, and analyze the reasons for the misdiagnosis. This work can enhance orthopedists and other specialists' understanding of this disease to improve the accuracy of the diagnosis.

Methods

General data

From 2014 to February 2019, 15 patients were diagnosed with SDAVF at our institution; 12 (80%) patients were previously misdiagnosed, and the misdiagnosis was corrected later through DSA. The patients included 10 men and 2 women with an average age of 54 years (range: 41-74 years).

Imaging examinations

Among the 12 patients, 2 patients underwent MRI of the lumbar spine at another hospital, and one patient did not undergo MRI before admission (this patient underwent computed tomography (CT) of the lumbar spine). The remaining patients had at least one MRI examination for the entire spine. The diagnosis of all the patients was confirmed by DSA, and single fistulas was presented in all 12 patients. The fistula was located in the thoracic segment in 10 patients, in the thoracolumbar segment in one patient, and in sacrococcygeal region in one patient.

Surgical procedure

The diseased segments were identified before surgery. The patient was placed in the prone position, and fluoroscopy was performed to accurately locate the fistula and the spinous process of the corresponding vertebral segment, which were marked by the injection of a small amount of methylene blue on the skin. A longitudinal posterior midline incision (the marked site was considered the center) was made for laminectomy or semi-laminectomy. Under microscopy, the dural matter was incised and the arachnoid was separated to identify the drainage vein, which was then electrocoagulated and divided.

Follow-up and assessment

The modified Aminoff-Logue Disability Scale (ALS) [4] (Table 1) was used to score the functional status of spinal cord during physical examination before surgery and 6 months after discharge. SPSS 13.0 was

used to perform paired t-tests to compare the modified ALS scores before and after treatment. P<0.05 was considered statistically significant. Spinal cord MRI was performed at follow-up visits from 3 months to 1 year after the surgery.

Table 1
Modified Aminoff-Logue Disability Scale

Gait (G)
0 Normal leg power, stance and gait
1 Leg weakness with no restriction of walking
2 Restricted exercise tolerance
3 Requires a cane or some support for walking
4 Requires crutches or two canes for walking
5 Requires a wheelchair
Urination (U)
0 Normal
1 Urgency, frequency and/or hesitancy
2 Occasional incontinence or retention
3 Persistent incontinence or retention
Defecation (F)
0 Normal
1 Mild constipation, responding well to aperients
2 Occasional incontinence or persistent constipation
3 Persistent incontinence

Results

Clinical manifestations of SDAVF

All 12 patients had subacute onset of SDAVF, which mainly manifested as transverse spinal cord injury (SCI) caused by progressive venous hypertensive myelopathy (VHM). According to the initial manifestations reported in their medical history, 3 patients (25%) had weakness of both lower extremities, 2 patients (17%) had paresthesia in the perineal region or lower limbs, one patient (8.3%) had bladder dysfunction, and 6 patients (50%) had 2 or more of the above symptoms (Table 2). At admission, the course of disease of 12 patients was from 5 to 23 months. Of those 12 patients, 5 had grade III lower

limb muscle strength and increased muscle tone; 2 had grade III left lower limb muscle strength and grade I right lower limb strength with increased muscle tone; 2 had grade III lower right lower limb muscle strength and grade II right lower limb muscle strength with increased muscle tone; one patient had grade I lower limb muscle strength with decreased muscle tone; and 2 had grade IV lower limb muscle strength with increased muscle tone. Twelve patients had numbness of the body and lower limbs below the level of sensory disturbance, and the level of sensory disturbance did not match the location of the SDAVF fistula. Furthermore, 12 patients had difficulty urinating, and 4 patients required an indwelling catheter.

Table 2
Initial clinical and imaging manifestations in 12 patients

Patient number	Age (years)	Male/female	Initial symptoms	Location of fistula	MRI findings
1	55	Male	Lower extremity weakness	T8-9	Hypointense T1 and hyperintense T2 signals, atypical flow void sign in the dorsal spinal cord
2 (Figure 2)	49	Female	Complex symptoms	filum terminale	Hypointense T1 and hyperintense T2 signals, typical flow void sign and tortuous abnormal vascular shadows in the dorsal and ventral spinal cord
3	45	Male	Dysuria	T12-L1	Isointense T1 and hyperintense T2 signals, typical flow void sign in the dorsal and ventral spinal cord
4	62	Male	Lower extremity weakness	T7-8	Hypointense T1 and hyperintense T2 signals, typical flow void sign in the dorsal and ventral spinal cord
5 (Figure 1)	59	Male	Complex symptoms	T9-10	Hypointense T1 and hyperintense T2 signals, atypical flow void sign in the dorsal spinal cord
6	49	Male	Sensory abnormality	T10-11	Isointense T1 and hyperintense T2 signals, typical flow void sign in the dorsal and ventral spinal cord
7	54	Female	Lower extremity weakness	T9-10	Hypointense T1 and hyperintense T2 signals, typical flow void sign in the dorsal and ventral spinal cord
8 (Figure 3)	56	Male	Complex symptoms	T8-9	Hypointense T1 and hyperintense T2 signals, typical flow void sign in the dorsal and ventral spinal cord
9	64	Male	Complex symptoms	T10-11	Isointense T1 and hyperintense T2 signals, typical flow void sign in the dorsal and ventral spinal cord
10 (Figure 4)	73	Male	Complex symptoms	T9-10	Hypointense T1 and hyperintense T2 signals, atypical flow void sign in the dorsal spinal cord
11	60	Male	Sensory abnormality	T6	Hypointense T1 and hyperintense T2 signals, typical flow void sign in the dorsal and ventral spinal cord
12	51	Male	Complex symptoms	T9-10	Hypointense T1 and hyperintense T2 signals, atypical flow void sign in the dorsal spinal cord

Imaging findings

On the MRI images of the spinal cord of the 12 patients, T1-weighted imaging (T1WI) showed abnormal hypointense or isointense signals in the spinal cord; T2-weighted imaging (T2WI) showed hyperintense signals in the enhanced and enlarged spinal cord, such as strip-shaped signals in 8 patients (67.7%), patchy signals in 4 patients (33.3%), and typical bead-like flow voids in the dorsal and ventral spinal cords in 7 patients (58%) (Figure 3) (for visualization purposes, the typical signs of spinal cord edema (hyperintensity) and dorsal and ventral bead-like flow voids are described as a "white radish plus black sesame seeds" sign). Tortuous abnormal vascular shadow was observed in one patient (Figure 2), and weedlike atypical flow voids signs were observed in the dorsal spinal cord in 4 patients (for visualization purposes, hyperintensity associated with spinal edema and dorsal and/or ventral atypical flow voids are described as a "white radish plus weed" sign) (Figure 1, Figure 4) (see Table 2 for complete explanations).

Types of misdiagnosis

Of the 15 patients, 12 (80%) patients were misdiagnosed, and 6 (50%) were misdiagnosed more than twice. The initial misdiagnosis included lumbar spinal stenosis (LSS) and lumbar disc herniation (LDH) in 6 patients, cervical spinal stenosis in one patient, benign prostatic hyperplasia (BPH) in 2 patients, myelitis in 2 patients, and syringomyelia in one patient. After the initial diagnosis, one patient was misdiagnosed with LDH, one was misdiagnosed with subacute combined degeneration of the spinal cord, and 4 were misdiagnosed with myelitis (Table 3)

Table 3
The first and second misdiagnoses of the 12 misdiagnosed patients

Diagnosis	Initial misdiagnosis	Second misdiagnosis
LDH	6	1
Cervical spinal stenosis	1	
Syringomyelia	1	
BPH	2	
Myelitis	2	4
Subacute combined degeneration of the spinal cord		1
Total	12	6

Treatments administered between the first and second misdiagnoses and the confirmed diagnosis

One patient was misdiagnosed with cervical spinal stenosis and underwent expansive laminoplasty, then was misdiagnosed with cauda equina syndrome caused by LDH and underwent removal of the nucleus pulposus and interbody fusion with bone graft and internal fixation (Figure 1). Two patients who were misdiagnosed with BPH received conservative medical treatment (Figure 3). A total of 7 patients were misdiagnosed with myelitis and subacute combined degeneration of the spinal cord at the initial and second diagnoses, respectively, and 4 of them were treated with hormone therapy.

Follow-up and assessment

All patients completed the follow-up. The mean duration of the follow-up was 0.9 years (0.5 to 1 year). The gait score was 3.33 ± 0.89 before surgery and decreased to 1.42 ± 0.91 at the follow-up, and the difference was statistically significant ($t = 5.20$, $p < 0.0001$). The bladder function score was improved from 2.17 ± 0.72 minutes before surgery to 1.17 ± 0.94 at the final follow-up, and the difference was statistically significant ($t = 2.93$, $p = 0.003$). Bowel function was also improved from 2.08 ± 0.67 before surgery to 1.0 ± 0.85 at follow-up, and the improvement was statistically significant (Table 4). Overall improvement was seen in the patients, but the degree of improvement was not identical among the patients. In particular, the patients with severe symptoms and bladder/bowel dysfunction experienced nonideal improvement. Spinal-cord MRI was performed at follow-ups from 3 months to 1 year after the surgery and showed that spinal cord edema was significantly improved or had disappeared, and abnormal flow voids were not noted (Figures 2 and 3).

Table 4
Modified ALS scores of 12 patients before and after surgery

Parameters	Before surgery	Final follow-up	T value	P
Gait	3.33 ± 0.89	1.42 ± 0.91	5.20	< 0.0001
Bladder function	2.17 ± 0.72	1.17 ± 0.94	2.93	0.003
Bowel movement	2.08 ± 0.67	1.0 ± 0.85	3.46	0.0005

Discussion

SDAVF is a type of intraspinal vascular malformation that has been gradually recognized in clinics in the past 20 years. It occurs when the arteries supplying the nerve roots or dura mater communicate with the spinal drainage vein when passing through the dura mater at the intervertebral foramen. Because the incidence of SDAVF is extremely low and the clinical manifestations are not typical [5], early diagnosis is

not easy, and the misdiagnosis rate is high [6, 7]. If diagnosis and treatment are delayed, spinal venous pressure continues to increase and can cause spinal cord ischemia and edema and even irreversible injuries, such as necrosis and demyelination. Therefore, it is necessary to improve the clinical and imaging understanding of this disease to reduce misdiagnosis and missed diagnoses.

Analysis of clinical manifestations

The disease is more common in elderly men, with a male-to-female ratio close to 4:1 [8]. Ten of the 12 patients in this study were men, accounting for 83.3% of all patients. SDAVF usually begins with progressive spinal dysfunction. The most common symptoms include gait abnormality, decreased myodynamia, paresthesia, sphincter dysfunction and sexual dysfunction. Almost half of all patients initially present more than one of these symptoms. In particular, in cases of patients with bladder dysfunction, physicians should be especially vigilant about considering this disease as a possible diagnosis. Within half a year of onset, symptoms of decreased myodynamia, abnormal sensation and bladder/bowel dysfunction can all appear.

Imaging analysis

The typical imaging findings usually include hypointense T1 or isointense signals, hyperintense T2 signals of spinal cord edema, and bead-like flow void signs in the dorsal and ventral spinal cord. However, some atypical images include local “weed” signs locally in the dorsal or ventral spinal cord. When these weed signs are present, the possibility of this disease should be considered to avoid a missed diagnosis.

Analysis of causes of misdiagnosis

1. This is a rare disease with an annual incidence of 5 to 10 per 1 million [9]; therefore, it is easily ignored by some specialists. 2. Additionally, the onset of the disease is not typical, and there are no obvious causes. The clinical manifestations may indicate the involvement of multiple health care specialties, including orthopedics, urology, neurology, pain management, and neurosurgery. Most patients seek medical advice from multiple specialists. However, specialists usually make diagnoses based on their own expertise and thus fail to pay adequate attention to this disease. This is an important reason for the misdiagnosis of this disease [10]. 3. Most patients are middle-aged and elderly men with an onset of sensorimotor dysfunction in the lower limbs; therefore, a diagnosis of degenerative changes of the lumbar spine is often the first choice. BPH is considered when patients present with bladder dysfunction or a decline in sexual function. 4. Physical examinations are not detailed enough to provide a conclusive diagnosis. Although the patient may already have increased muscle tone and positive pathological signs of this disease, the diagnosis of lumbar degenerative diseases, such as LDH, is made only based on MRI findings. 5. MRI may not be available in primary hospitals, so only lumbar CT is used for diagnosis. It is also possible that if MRI is used as a screening method and the initial symptoms are relatively mild,

spinal cord MRI is not considered immediately because of its high cost. 5. Physicians do not fully understand the manifestations of this disease on spinal cord MRI images and only consider inflammatory changes or secondary edema of the spinal cord caused by LDH compression when mild changes in long T2 signals are observed, especially when atypical flow voids are observed near the spinal cord. 6. The level of the spinal cord scanned by MRI is too high or too low, causing a missed diagnosis.

Differential diagnosis

The median time from the onset of the patient's clinical symptoms to the diagnosis of the disease is 12 to 44 months, which is an important reason for the high disability rate associated with the disease. Therefore, early diagnosis is key to reducing the disability rate [11]. In this study, 1. Patients misdiagnosed with LDH accounted for approximately 50% (16/33) of all misdiagnosed patients. Most patients with LDH present with root pain that is obviously related to the LDH. However, the pathological signs are negative, which is a very important indicator of SDAVF. In addition, some patients with SDAVF may also have LDH, and this is also an important reason for missed diagnosis. 2. Myelitis progresses rapidly, and spinal cord swelling can be seen on spinal cord MRI images, but there is no flow void sign. In patients with atypical flow void signs, spinal angiography can be an option for confirming the diagnosis. 3. Patients with BPH usually do not have specific clinical manifestations or signs of lower extremity abnormality, and the diagnosis is generally not difficult to make. For patients who cannot be diagnosed, further spinal cord MRI or spinal angiography should be performed.

How to prevent misdiagnosis and missed diagnosis

Primary physicians should obtain detailed a medical history from patients and perform comprehensive physical and neurological examinations. The relevant specialists should improve their understanding of the clinical features of this disease. MRI can be used as a preliminary screening method to analyze myelitis on longitudinal and axial views. Furthermore, it is necessary to strengthen collaboration among various departments to reduce the occurrence of misdiagnosis and missed diagnosis.

Limitations Of This Study

This study had limitations. Since this was single-center retrospective study, the sample size was small, and the follow-up period was short. Although there were many limitations, the results can deepen different specialists' understanding of this disease and can improve diagnostic accuracy.

Conclusion

When patients, especially those with bladder and bowel dysfunction, have symptoms such as weakness of the lower limbs and/or numbness, the diagnosis of SDAVF should be considered. Furthermore, if MRI shows patterns resembling a white radish plus black sesame seeds or a white radish plus weeds, the

diagnosis of SDAVF should be strongly considered, and further angiography should be performed to confirm the diagnosis.

Abbreviations

SDAVF:Spinal dural arteriovenous fistula; ALS:Aminoff-Logue Disability Scale; LDH:lumbar disc herniation; BPH:benign prostatic hyperplasia; MRI:magnetic resonance imaging; DSA:digital subtraction angiography; CT:computed tomography; SCI:spinal cord injury;VHM:venous hypertensive myelopathy; T1WI:T1-weighted imaging; T2WI: T2-weighted imaging; LSS: lumbar spinal stenosis.

Declarations

Ethics approval and consent to participate

The study was in accordance with the Declaration of Helsinki, and was approved by Ethics Committee of The Second Affiliated Hospital of Xi'an Jiaotong University (Xi'an, China). Written consent to participate in this study was obtained from the participants.

Consent for publication

A written format of informed consent, which includes information on the use, disclosure and publication of patient information on the condition of anonymity, was signed and obtained from all individual participants in the study.

Availability of data and material

The authors declare that the databases, application/tool, etc. described in the manuscript are available for testing.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

Baozhi Yang collected the cases and drafted the manuscript. Shuai Cao analyzed the data. Xijing He designed the study. Haopeng Li performed the operation. All authors read and approved the final manuscript.

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Figures

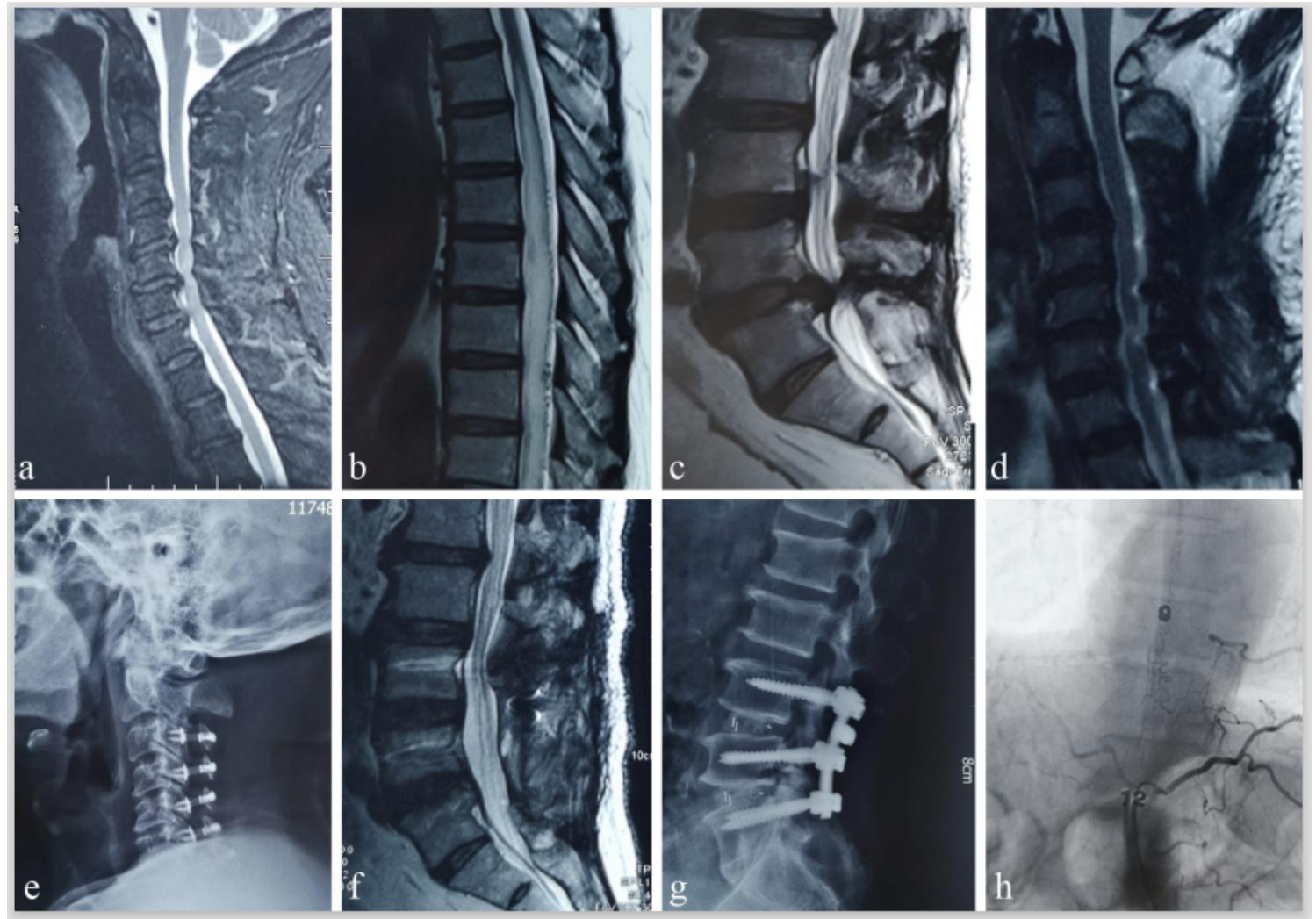


Figure 1

A 59-year-old man first visited another hospital due to "progressive weakness of the lower limbs and difficulty urinating for one month" and underwent cervical, thoracic, and lumbar spine MRIs (Figures 1a, 1b, and 1c). Figure 1b, a thoracic spine MRI image, shows a hyperintense signal consistent with spinal cord edema and weed-like flow voids of the dorsal spinal cord at T9-10 (white radish plus weed pattern). The patient was misdiagnosed with cervical spinal stenosis and was treated with open-door expansive cervical laminoplasty (Figures 1d and 1e). His symptoms were not resolved after surgery. He was misdiagnosed with LDH at his second admission and underwent lumbar decompression (Figures 1f and 1g). His symptoms worsened progressively after surgery. He was admitted to our clinic more than 9 months after the surgery and was diagnosed with thoracic SDAVF, which was confirmed by angiography (Figure 1h).

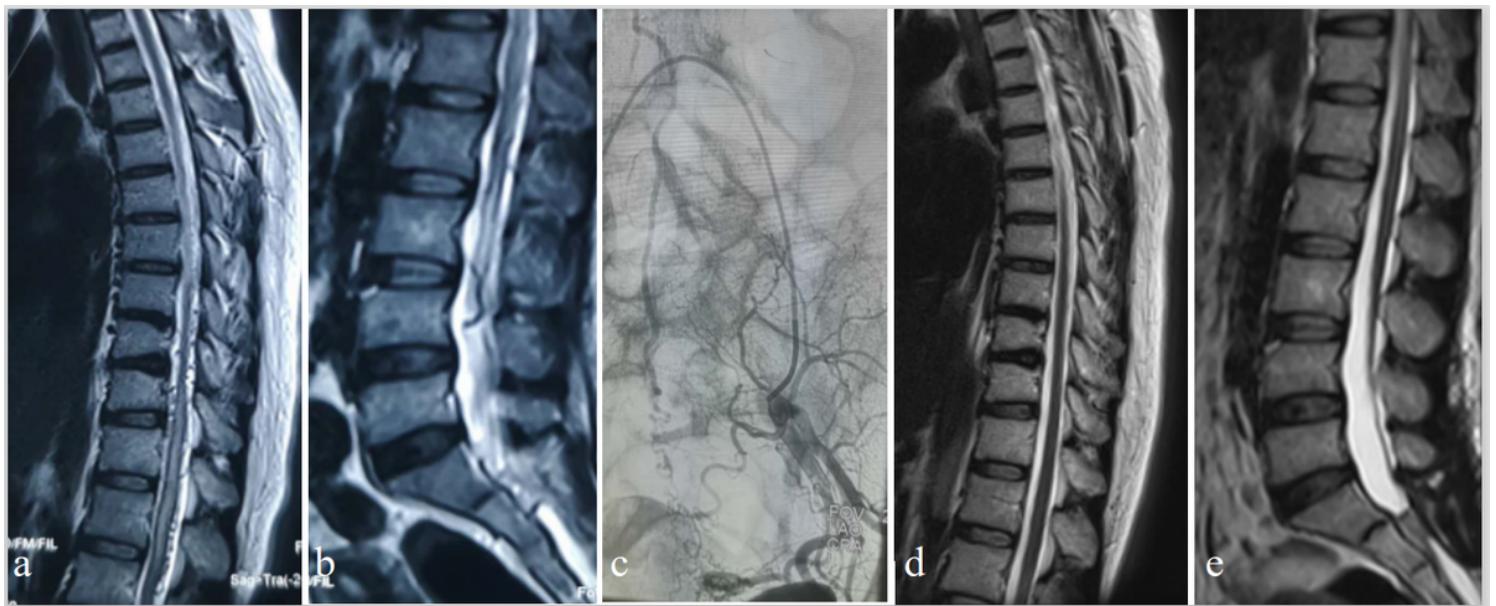


Figure 2

A 49-year-old women was misdiagnosed with primary myelitis and was admitted to our hospital due to "numbness and weakness of both lower limbs and bladder dysfunction for more than 7 months". A: Preoperative thoracic spine MRI shows bead-like flow voids and spinal cord edema (white radish plus black sesame seed pattern). B. Lumbar MRI shows abnormal tortuous vascular shadows around the cauda equina. C. Spinal DSA was performed and confirmed the diagnosis of SDAVF with the fistula in the sacrococcygeal region. D. F. Follow-up MRI at 3 months after surgery shows that flow void signals and spinal cord edema have disappeared.

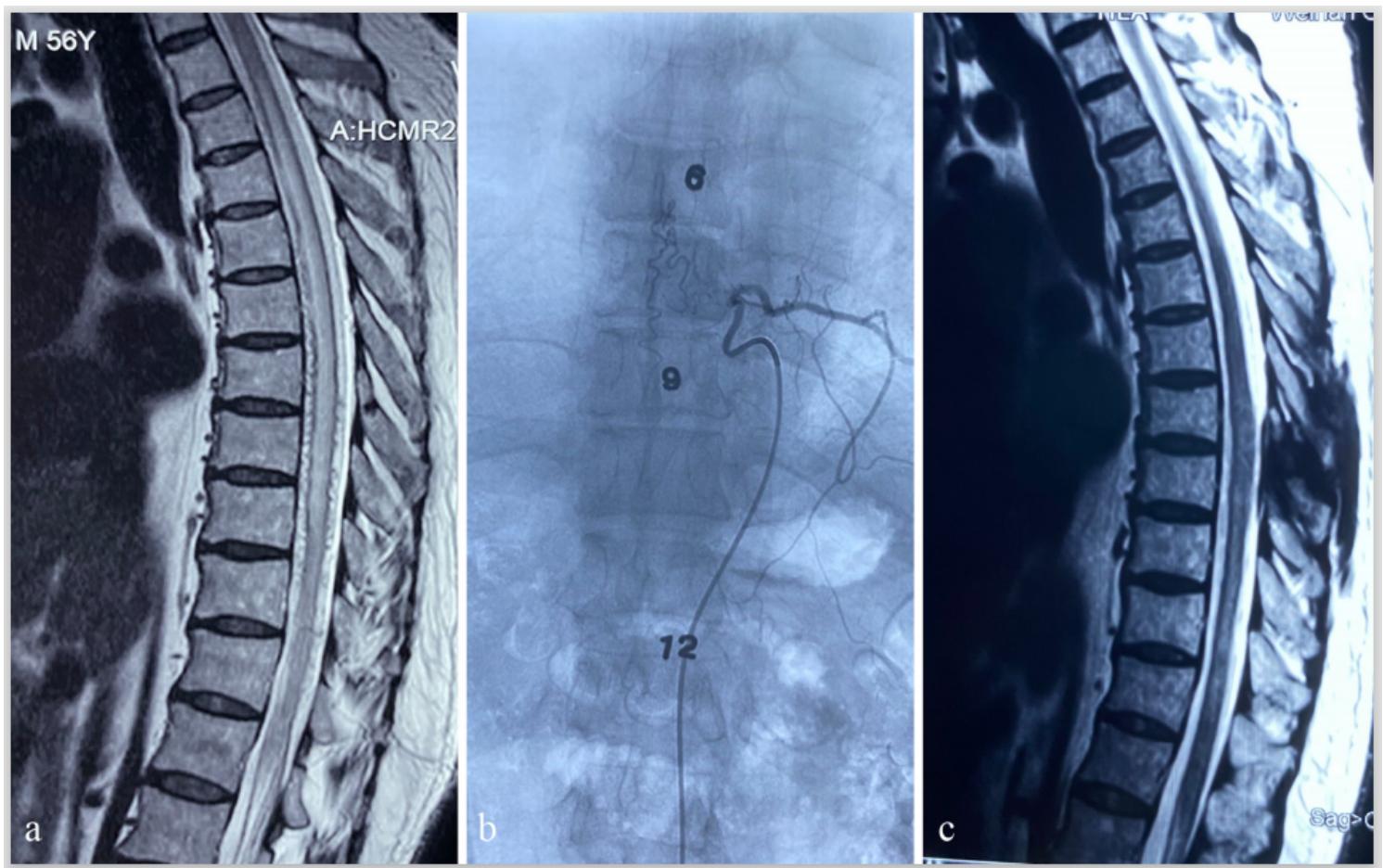


Figure 3

A 56-year-old man was initially misdiagnosed with BPH and then misdiagnosed with myelitis. He was admitted to our hospital due to "lower limb numbness and weakness and bladder dysfunction for more than 9 months". A: Preoperative MRI of the thoracic spine shows signs of flow voids and spinal cord edema (white radish plus black sesame seed pattern). B. Spinal DSA was performed and confirmed the diagnosis of SDAVF with the fistula in the T9 level. C. Follow-up MRI at 3 months after surgery shows the disappearance of flow voids and improvement of spinal cord edema.

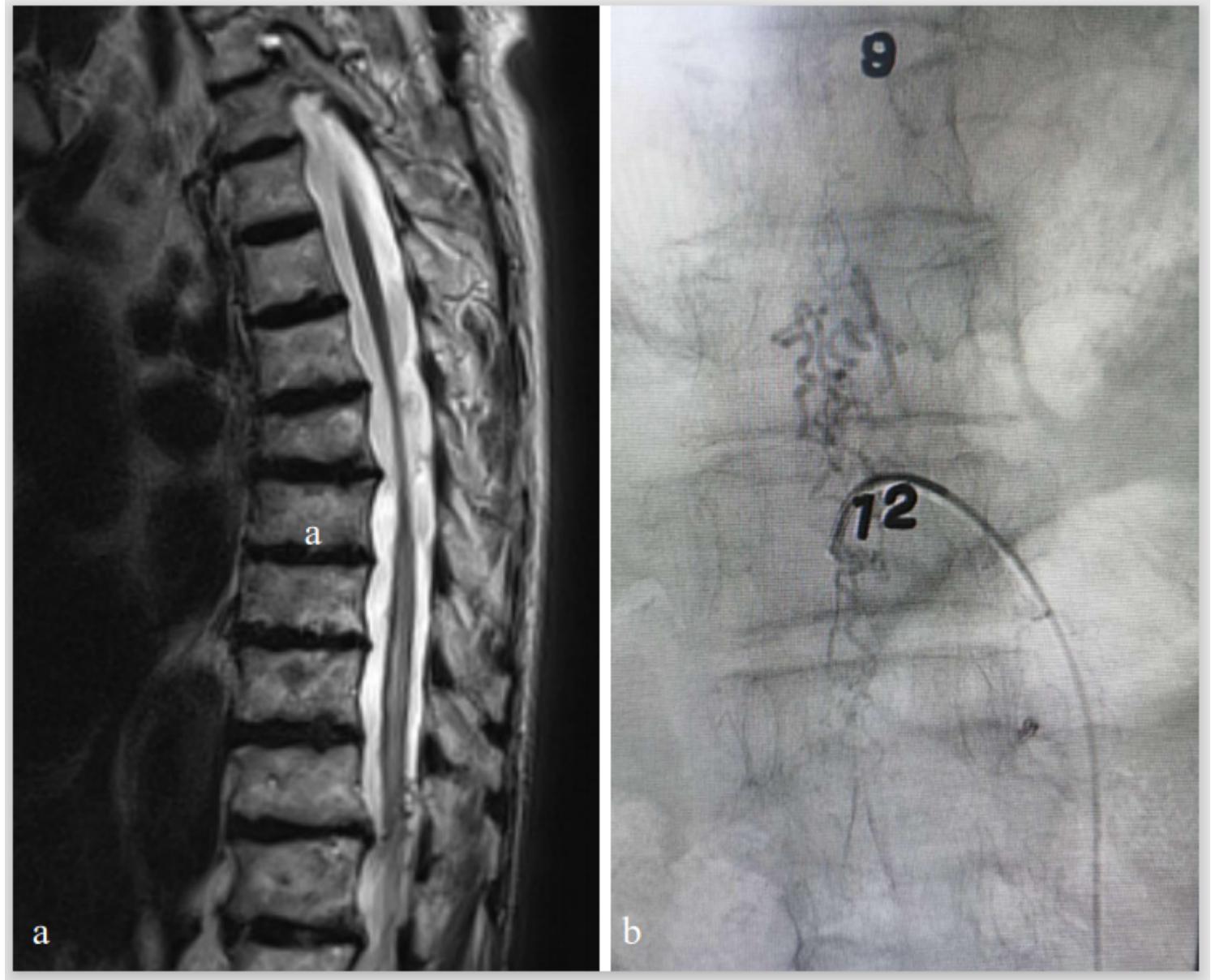


Figure 4

A 73-year-old man was initially misdiagnosed with myelitis and then misdiagnosed with subacute combined degeneration of the spinal cord. a. Thoracic spine MRI shows signals of spinal cord edema and atypical "weed-like" dorsal flow void patterns (white radish plus weed pattern). b. DSA was performed in our hospital and confirmed the diagnosis of SDAVF with the fistula located at the T9-10 level.