Magnetic resonance imaging (MRI) characteristics of the post-treatment spinal dural arteriovenous fistulas for predicting prognosis

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Directed by Professor Keung Nyun Kim

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Also, I thank to my wife-Dr. Lee- and lovely my son. I could not have enough time to be with them because of a lot of research and hospital work. However, I believe that my family knows my honor and love for them.

So, I want to say, "Dear my wife and son, I am sorry and I love you".

I also appreciate to my parents. They always hope and pray my success, health, and happiness. They have been and will be a great support for me. I don't know all of their love to me, but, I can feel and keep it deeply in my heart.

Finally, I would like to dedicate this accomplishment to God.

Sincerely,

Keun Young Park

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<ABSTRACT>

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Spinal dural arteriovenous fistulas are very rare, but can cause fatal disability. Sometimes they can be misdiagnosed because of their rarity, however, recent developed MRI technique can make them to be diagnosed easily. Although spinal angiography has been still a gold standard of diagnosis of this disease, pre-treatment MRI can help for screening of them. We can expect poor prognosis if there are upward extension of cord signal change above fistula level on pre-treatment MRI. Resolution of cord signal change on post-treatment MRI suggests favorable treatment results. However, remained signal change 1 year or more after treatment means recurrence and this finding requires follow up spinal angiography. Meanwhile, a lot of literatures have reported correlation between MRI findings and clinical outcomes. However, our study cannot show any significant correlation of these two variables.

Key words: spine, dural arteriovenous fistula, Aminoff-Logue's disability scale, MRI

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I. Introduction

Spinal arteriovenous lesions are very rare and about one-tenth of cerebral arteriovenous lesions.[1] Among them, spinal dural arteriovenous fistulas (SDAVFs), also called Type I spinal AVFs or spinal intradural dorsal AVFs,[2,3] are most common and accounts for about 70-80% of all spinal arteriovenous shunts.[4,5] SDAVFs have been considered as acquired lesions and can usually occur at the thoracic spine. They are fed by radicular feeding arteries and drained into perimedullary venous plexus. Fistulous connection is formed at the dural root sleeve. This communication between artery and venous plexus results in venous congestion, ischemia, infarction, and hemorrhage.[6, 7] Although they can be treated sufficiently by endovascular or surgical methods, they can cause severe disability by irreversible damage of spinal cord.[3] However, despite such morbidity, their rarity can make themselves to be missed by clinicians. Recent developed magnetic resonance imaging (MRI) can help clinicians to diagnose the lesions easily and MRI has become replacement of myelography as a first diagnostic tool of the SDAVFs. Their MRI findings are characteristic and T2-weighted MRI shows cord edema and infarction as intramedullary high signal intensity and also shows abnormal perimedullary vessels as subarachnoid flow voids.[8] Additionally, gadolinium (Gd) enhanced T1-weighted MRI can show cord enhancement and abnormal arterialized vessels.[8,9] Although spinal angiography has been still a gold standard of diagnosis of the SDAVFs, MRI can be used for follow up study with comfort. Retrospectively we reviewed data of our institute and analysis was performed to identify ① correlation between pre- and post-treatment MRI findings and functional outcomes, and ② characteristics of MRI findings after treatment and their clinical applications.

II. Materials and Methods

1. Patients

All patients who were treated as spinal dural AVFs in our institute from 1992 to 2007 were reviewed retrospectively and all radiologic examinations of them were reviewed by two radiologists. Among them, the patients who had follow up MRI were fifteen patients, thirteen males and two females. Age was ranged from 19 to 66, average 48.4. We divided the patients into two groups by symptom duration. (Group A<1 year, B≥1 year) Clinical data were obtained from medical records and outpatient charts. Telephone interview were performed to evaluate follow-up functional outcomes.

2. Methods

A. Analysis of radiologic findings and treatment results

Most SDAVFs occurred at the thoracic spine (N: 10) followed by sacral (N: 2), lumbar (N: 1), and double region (thoracic and lumbar) (N: 2). Seven patients were treated by single endovascular embolization and six patients were treated by combined therapy-endovascular and surgical methods. Only two patients needed multiple embolizations. Treatment results (partial obliteration, complete obliteration, recurrence) were decided by follow up spinal angiography. Complete obliteration was done at twelve cases, partial obliteration at two cases, and there was only one recurrent case. Severity grades of MRI findings were measured by 1) number of spinal cord level which showed cord signal change or 2) subarachnoid signal voids severity (+++: severe, ++: present, +: present, but less definite, −: no signal voids). Mean follow-up period was 27.5 months (±31.2, range from 2 months to 8 years).

B. Evaluation of functional status and clinical outcomes

All patients except one had functional disability. Pre-treatment functional status and post-treatment clinical outcomes were measured by Aminoff-Logue's disability scale (ALS) (Table 1). Original scale has three categories- gait (G), micturition (M), and bowel function. However, we used only G and M categories because of insufficient clinical data about bowel function.

Table 1. Aminoff & Logue's disability scale

Gait	**Micturition**	**Bowel function***
G0 Normal	M0 Normal	B0 Normal
G1 Leg weakness, abnormal gait or stance, no restricted activity	M1 Infrequent hesitancy or urgency, altered sensation, but continent	B1 Moderate constipation
G2 Restricted activity but no support required	M2 Occasional urinary incontinence or retention	B2 Severe constipation or occasional incontinence
G3 One stick required for walking	M3 Total incontinence or persistent retention	B3 Total incontinence
G4 Two sticks, crutches, or frame required for walking		
G5 Confined to wheelchair		

*Useless in this study

C. Statistical analysis

Pre-treatment and post-treatment ALS scores were analyzed by Wilcoxon signed rank test. ALS scores between two groups (A and B) were analyzed by Mann-Whitney's U test. Continuous variables were analyzed by Spearman's correlation. A value of $P < 0.05$ was taken to be significant.

III. Results

1. General characteristics of patients

The most common clinical presentation was paraparesis (N: 7) and followed by paraplegia (N: 3), monoparesis (N: 2), bladder retention (N: 1), and sensory disturbance (N: 1). One patient had an incidental lesion after traffic accident. Mean symptom duration was 15.9 months (±24.4, range from 1 day to 8 years). The most common primary pathophysiology was venous ischemia by perimedullary venous congestion. Hemorrhagic accidents have occurred at only two cases and emergent surgical decompression and fistula interruptions proceeded by embolization were performed at these two cases. However, they had poor functional outcome despite successful treatment. Although some literatures insist that symptom duration can affect the functional outcome, there were not significant differences of ALS scores between Group A and B in our study. (① **pre-treatment ALS**; Group A: G3.57M2.14 Group B: G3.57M1.71, ② **post-treatment ALS**; Group A: G3.14M1.57 Group B: G2.86M1.43, **P-value>0.05**)

There were four cases of complications (paresthesia: three, glue migration: one). Among them who complained paresthesia, two patients had paresthesia before treatment, however, they have experienced more aggravating paresthesia after treatment and one patient had newly developed paresthesia after surgery. The patients with paresthesia has obtained long-term analgesics and pain block, however, they still have suffered from the pain. During embolization, glue droplet happened in one case. This was found during embolization and then the patient was treated with two-day heparinization and two week anti-platelet medication. Fortunately, the patient had no embolic event-cord ischemia or infarction, or hemorrhage. There was no wound infection, postoperative hematoma. These clinical characteristics are shown at Table 2.

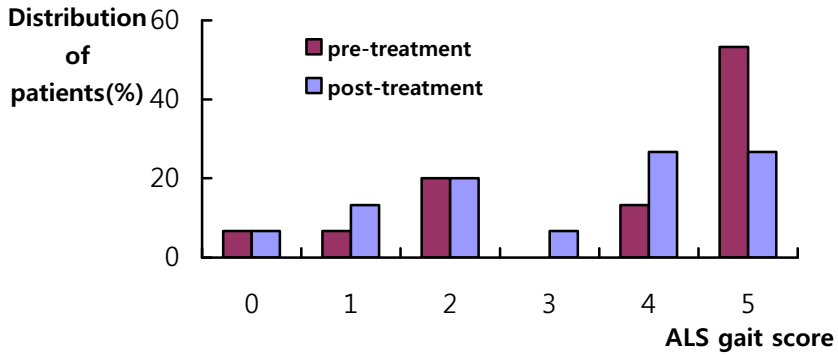
Table 2. Clinical characteristics of the patients

Patient	Sex	Age	Symptom & Sign	Duration	Location *	Treatment **	Treatment results***	Complications
1	M	45	Incidental	1M	Mid T	E	PR	Glue droplet
2	M	57	Paraparesis	2Y	S	E	CR	
3	M	39	Monoparesis	7M	Lower T	E	CR	Paresthesia****
4	M	44	Paraparesis	10M	Mid T	ES	CR	
5	M	49	Sensory change	1.5Y	Upper T	E	CR	
6	M	45	Monoparesis	8Y	T+L	ES	CR	
7	F	60	Paraparesis	1Y	Mid T	ES	CR	
8	M	66	Paraplegia		S	EE	PR	
9+	M	49	Paraplegia	3D	Mid T	ES	CR	
10	M	45	Bladder retention	16M	T+L	E	CR	Paresthesia
11	M	41	Paraparesis	1Y	Mid T	E	CR	Paresthesia
12+	F	19	Paraplegia	1D	T+L	ES	CR	
13	M	46	Paraparesis	3M	L	E	CR	
14	M	64	Paraparesis	3M	Lower T	EE	RR	
15	M	57	Paraparesis	21M	Mid T	ES	CR	

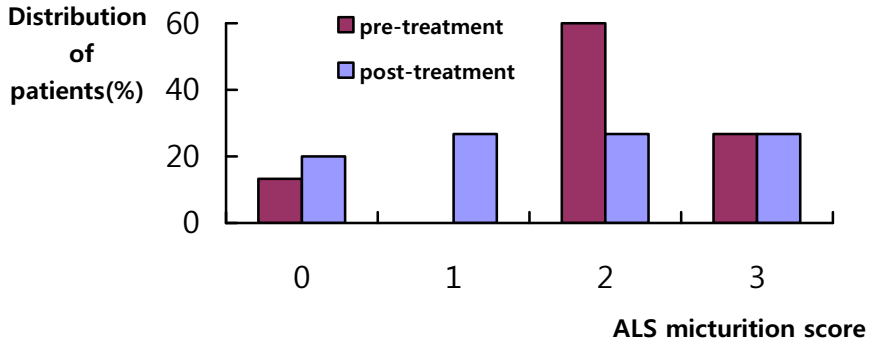
*: **C**: cervical, **T**: thoracic, **L**: lumbar, **S**: sacral
: **E: endovascular treatment, **S**: open surgery
***: **CR**:complete obliteration **PR**:partial obliteration **RR**: recurrence
****: newly occurred after surgery
+: cases with hemorrhagic accident

While pre-treatment ALS was average G3.7M2.0, post-treatment ALS was average G3.1M1.6. This was significant improvement of functional grades after treatment (Gait; P-value 0.033, Micturition; P-value 0.034). However, in our study, there were less improvement in cases who had poor functional grades before treatment (case: 8,9,12,14; pre: G5M3 post: G5M3). We could guess that these patients might have irreversible cord damage. Our study could not reveal that treatment results (CR, PR, and RR) can affect post-treatment ALS scores significantly, although there was much improvement of functional grades in cases of complete obliteration comparing with two partial obliterations and one recurrence.

Gait capacity is less vulnerable than micturition capacity and this discrimination supplies different clinical course. The patients who had poor ALS micturition score (M 3) had a tendency to be less improved after treatment comparing with favorable M 0-2. On the other hand, the patient with poor ALS gait scores (G 4 or 5) had been improved after treatment (Figure 1). According to these findings, we could reach the conclusion that symptomatic SDAVFs patients with poor gait scores also should have any treatment unless contraindication.



(A)



(B)

Figure 1. ALS scores before and after treatment

(A)ALS gait score (B)ALS micturition score; average ALS scores improved after treatment. However, the patients with poor ALS micturition score (M3) had not been improved comparing with favorable ALS score 0-2. On the other hand, the patients with poor ALS gait scores (G4 or 5) could have been improved after treatment.

2. Pre-treatment ALS scores and MRI findings

In most cases, T2-weighted MRI showed ① cord signal change and ② subarachnoid signal voids. Actually, in some cases, subarachnoid signal voids were vague and not evident on T2 image. However, we were able to identify enhanced arterialized perimedullary veins on Gd enhanced T1-weighted MRI in these cases.

MRI findings were measured according to previous mentioned grades and were described at Table 3. We assume that high grade of MRI finding suggest severe venous congestion or significant change of perimedullary venous structure by venous hypertension. However, these MRI findings were not always related with pre-treatment ALS scores in our study and we could not find any significant

correlation between pre-treatment MRI findings and ALS scores (P-value>0.05, Figure 2). In spite of this result, upward extension of cord signal change (above fistula level) could suggest poor ALS scores. For example, pre-treatment MRI of case 2,8,9 showed upward extension of cord signal change and they had a poor functional status before treatment.

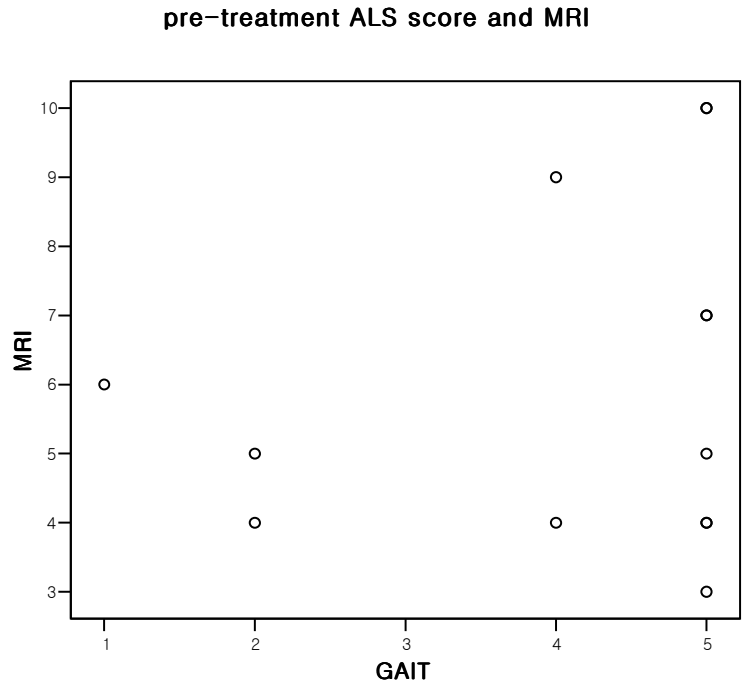


Figure 2. Pre-treatment ALS gait score and MRI
There were no significant correlations between two variables.

3. Post-treatment ALS scores and MRI findings

Pathologic MRI findings had disappeared gradually after any treatment if complete obliteration had been achieved. This result compromised that venous ischemia might be resolved gradually for some months or years after sufficient treatment. On the other hand, in three cases, there were still pathologic findings in spite of sufficient treatment. These cases consisted of two cases of short-term MRI follow up (one month, two months) and one recurrent case. If cord signal change still remains on follow-up MRI after complete obliteration, we can predict disease recurrence. Perimedullary vessel engorgement (shown as subarachnoid flow voids on T2 images or enhanced arterialized vein on Gd enhanced T1 images) was also decreased if any treatment was performed. However, despite complete obliteration of fistula, those pathologic findings still remained in almost of cases. This phenomenon could be

resulted from irreversible structural change of perimedullary vein. Therefore, we cannot use the pathologic vessel findings on follow-up MRI as an indicator of disease recurrence.

We could not identify any statistical correlation between post-treatment MR findings and ALS scores (P-value>0.05). Moreover, some patient who had significant improvement on follow up image study had experienced more aggravated functional deficit (Figure 3). We paid attention that this patient had long symptom duration (18 months) and we guessed that longstanding venous ischemia might cause irreversible cord damage by cord infarction in this case. In fact, follow up MRI showed not cord signal change but cord atrophy.

Table 3. ALS scores and MRI characteristics before and after treatment

Patient	Aminoff-Logue's disability scales				MRI			
	Pre-treatment		Post-treatment		Pre-treatment		Post-treatment	
	Gait	Micturition	Gait	Micturition	Intramedullary signal change	Flow voids♦	Intramedullary signal change	Flow voids♦
**1	0	0	0	0	-	+++	-	+
2♦	5	2	4	2	7	+++	0	++
3	2	2	1	0	5	+++	0	+
4	4	2	4	1	9	+++	0	+
*5	1	2	2	2	6	+++	0	0
6	2	0	2	0	-	+	-	0
7	5	2	4	2	5	+++	4	+
**8+	5	3	5	3	4	-	1	0
9+	5	3	5	3	10	++	-	-
10	2	2	1	1	4	+++	0	+
11	5	2	3	2	10	++	4	+
12	5	3	5	3	7	+++	-	-
13	4	2	2	1	4	++	0	0
***14	5	3	5	3	4	+++	1	++
15	5	2	4	1	3	++	0	0

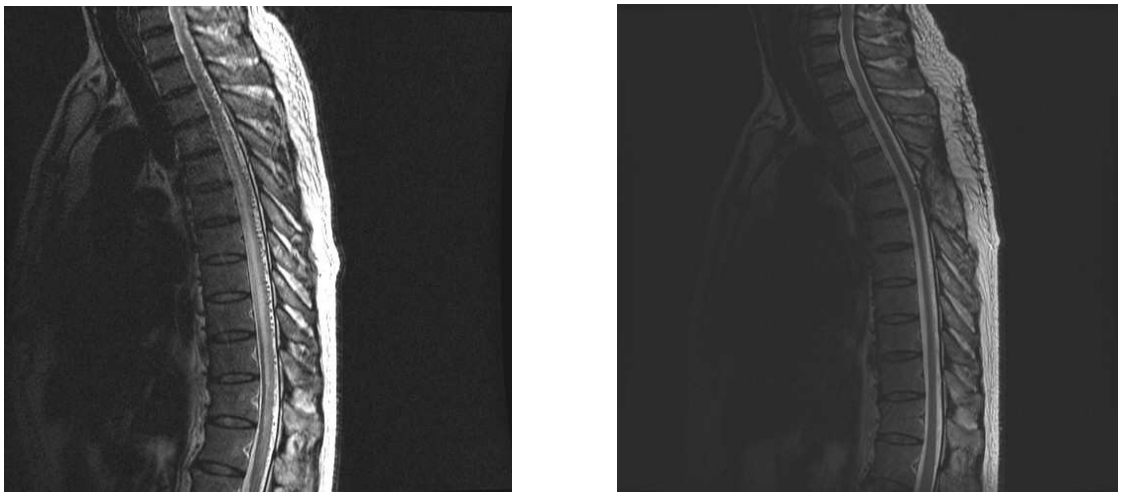
*: possible cord infarction

**: partial obliteration

***: recurrence

+: upward extension of cord signal change above fistula level

♦: +++: severe, ++: present, +: present, but less definite, −: no signal voids



(A) (B)

Figure 3. Case

44 years-old male patient visited our institute for lower extremity weakness for 18 months. Initially, he had mild gait disturbance (G1) and bladder habit change (M2).

(A) Initial T2-weighted MRI showed spinal cord signal change and venous engorgement. Spinal angiography was performed and revealed thoracic dural AVFs. We treated it successfully. (B) About 1.5 years after treatment, follow-up MRI was performed and it showed complete resolution of cord signal change and venous engorgement, but also showed cord atrophy. His functional grades became more aggravated to G2M2 from G1M2.

IV. Discussion

Spinal dural AVFs are very rare and can cause fatal complications (low extremity weakness, paresthesia, back pain, urinary symptoms and so on[10]). However, because of their rarity and undistinguishable clinical symptoms, many clinicians mistake them for other lesions, for example, degenerative disc, spinal cord tumor. Conventional T2-weighted MRI was usually performed to diagnose the AVFs and other spine diseases. On T2-weighted MRI, cord edema resulted from ischemia, infarction, or necrosis is described as intramedullary signal change and high flow velocity of arterialized veins and intravascular pulsations result in subarachnoid flow voids.[8,9,11] However, conventional MRI cannot directly visualize the spinal arteriovenous shunts and sometimes cannot show the flow voids evidently. In cases without flow voids, Gd enhanced T1-weighted MRI can help to visualize the arterialized abnormal veins directly.[9,11] Therefore, we suggested that Gd enhanced T1-MRI plus T2-MRI should be performed to diagnose the SDAVFs evidently and other type of spinal

vascular malformations can be easily differentiated by Gd enhanced T1 MRI. Patrick H. Luetmer and colleagues suggest usefulness of contrast-enhanced MR angiography (MRA) for detection of spinal vascular lesions.[12] Although they insisted that sensitivity of this MRA technique was 91% and specificity 78%, conventional MRI (T2, T1, and Gd enhanced T1) gives clinicians enough clues to predict the spinal vascular lesions and spinal angiography is still a gold standard to diagnose the lesions.

MRI can gives us information about recurrence or recannalization of fistulas.[8] If there still remained cord signal change after sufficient treatment (complete obliteration), we can guess two possibilities, one is short-term follow up and the other is recurrent (Figure 4). Therefore, we suggest follow up angiography in cases with remained cord signal change one year or more after sufficient treatment. As previously mentioned above, some patient whose post-treatment MRI showed no cord signal change has experienced aggravation of clinical symptoms (Figure 3). In this case, cord signal change on T2 image probably expressed not only venous congestion but also cord infarction. Actually, follow up MRI of this patient showed cord atrophy due to cord infarction.

Our study involved three patients with initial poor ALS scores (G 5 M 2.7) and their T2-weighted MRI showed upward extension of cord signal change. Generally, gravity can affect the venous drainage of cord. So, Even if there is not enough venous drain in SDAVFs, cord signal change can usually occur below the fistula level. Therefore, upward extension of signal change suggests severe venous ischemia and poor functional outcomes.

Any treatment is necessary for symptomatic spinal dural AVFs. Our study showed that significant ALS scores' improvement after any type of treatment-endovascular or open surgical methods and pathologic MRI findings can be disappeared after sufficient treatment. However, we could not show any correlation of pre- and post-treatment MRI findings and functional outcomes as previous study.[8,9]

Is there were any relationship between symptom duration and functional outcomes?

Kohno and colleagues showed that short symptom duration groups (<2.5 years) had better functional outcomes (both G and M) than long symptom duration groups (≥2.5 years).[13] However, our study cannot show any outcome differences between group A (<1 year) and B (≥1 year).

There were a few acceptable complications and no mortality in our study.

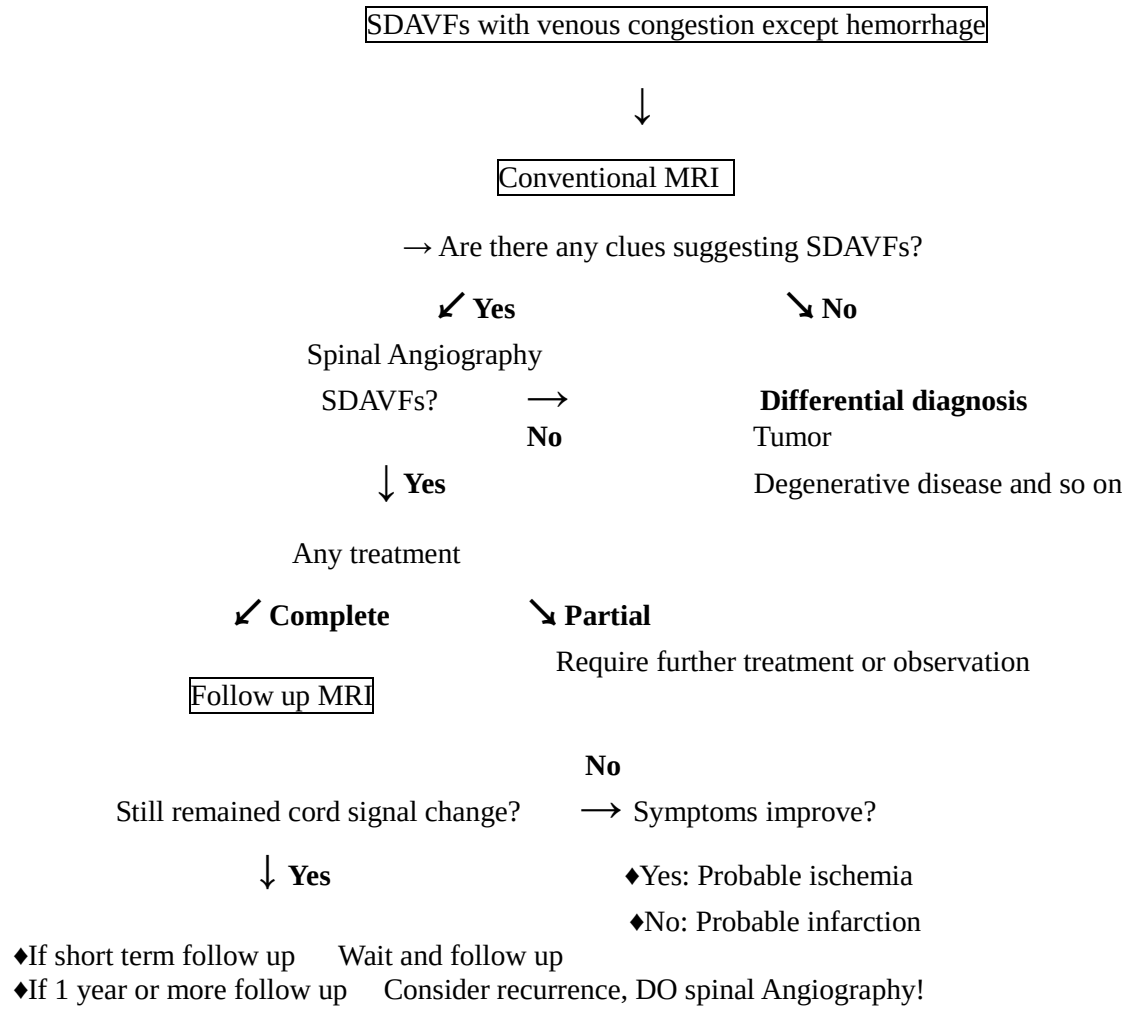


Figure 4. Suggesting algorithms of SDAVFs' MRI finding & treatment

V. Conclusion

Spinal dural AVFs are rare, but devastating diseases. Gd enhanced T1-MRI plus T2-MRI had better give more information than conventional MRI for screening the SDAVFs. Treatment is necessary at symptomatic lesion. Upward extension of cord signal change above fistula level on pre-treatment MRI can suggest poor ALS scores. Still remained cord signal change on follow-up MRI (1 year or more after treatment) can suggest recurrence or partial obliteration. Cord atrophy without signal change on post-treatment MRI can suggest cord infarction or necrosis and this case tend to have poor outcomes. However, there were no significant correlations between pre- and post-treatment MRI findings and clinical outcomes.

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<ABSTRACT IN KOREAN>

척수 경막 동정맥루 치료 후 예후와 관련된 자기 공명 영상 소견

< 지도교수　김 긍 년 >

연세대학교 대학원 의학과

박 근 영

척수 경막 동정맥루는 비록 그 빈도는 적으나 치명적인 결과를 낳을 수 있는 질환이다. 워낙 드물기 때문에 간혹 임상에서 다른 질환으로 오진할 수 있으나 최근 들어서 자기 공명 영상 기기가 발달하면서 보다 손쉽게 진단이 가능해졌다. 비록 아직까지 혈관 조영술이 이 질환의 해부학적 구조 및 병태생리를 가장 잘 설명해주는 중요 진단 기법이지만, 치료 전 척추 자기 공명 영상은 screening test 로서 유용할 뿐 아니라 치료 전에 cord signal change가 동정맥루 level 상방으로까지 퍼진 병변의 경우 나쁜 예후를 의심할 수 있다. 또한 치료 후 자기 공명 영상 상에서 cord signal change의 감소는 적절한 치료가 행해졌음을 의미하며 치료 약 1년 뒤에도 cord signal change가 이미지 상 남아 있는 경우 재발을 의심할 수 있는데 이 경우 추적 혈관 조영술을 시행해야 한다. 한편으로 여러 보고에서 자기 공명 영상에서 보이는 질병 정도와 임상에서의 환자 상태의 상관 관계에 대해 논한바 있으나 본 연구에서는 두 변수 사이의 명확한 상관 관계를 찾을 수는 없었다.

핵심되는 말: 척수, 경막 동정맥루, 아미노프-로그 장애 척도, 자기 공명 영상