

척추 해면상 혈관종 출혈로 인해 발생한 하지 강직감을 동반한 보행 장애에 대한 한의학 치료의 효과: 증례보고

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Korean Medicine for Gait Disturbance Accompanying Lower Limb Rigidity Caused by Bleeding of Spinal Cavernous Hemangioma: A Case Report

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해면상 혈관종(cavernous hemangioma)은 인체 내 다양한 장기나 기관에서 발생할 수 있는 혈관 기형으로, 순수 경막외 해면상 혈관종(pure epidural cavernous hemangioma)은 해면상 혈관종의 드문 경우에 속한다. 해면상 혈관종이 원인이 되어 발생한 증상에 한의학 치료를 적용한 연구가 드물게 보고되었으나, 순수 해면상 혈관종의 출혈로 인한 증상에 대한 것은 없었기에 관련 임상증례를 보고하고자 한다. 척추의 순수 해면상 혈관종으로 진단받은 51세 남성이 2022년도에 해면상 혈관종 출혈로 인해 발생한 보행장애에 대해 치료를 받기 위해 한방병원을 방문하였다. 척추의 순수 해면상 혈관종 출혈로 인해 발생한 보행장애 환자는 침 치료, 전기 침 치료, 온침 치료, 한약,推拿, 뜸, 부항 치료를 시행 받았다. 평가지표는 25 foot walk test (25FW), timed up and go test (TUG), comfortable gait speed tests (CGS), 요통과 하지 통증에 대한 numeral rating scale (NRS), Berg balance score (BBS), Tinetti-score, manual muscle test (MMT), EuroQol-five dimension (EQ-5D), EQ-5D visual analog scale (EQ-5D VAS)이었으며, 25FW, TUG, CGS, NRS, BBS, EQ-5D (VAS)에서는 치료 후 호전을 보인 반면, Tinetti Score, MMT, EQ-5D의 지표는 치료 후에도 동일하게 유지되었다. 한의학은 척추 해면상 혈관종의 출혈로 인해 발생한 보행장애나 하지 강직감에 대한 치료의 한 방안으로 고려될 수 있다. (*J Korean Med Rehabil* 2023;33(4):243-249)

Key words Cavernous hemangioma, Gait disturbance, Korean medicine, Case report

Introduction>>>>

Cavernous hemangioma is a vascular malformation that can occur anywhere, including the central nervous system and various organ systems¹. Spinal cavernous hemangioma (SCH) comprises 5~12% of all spinal

vascular malformations². Approximately 50% of SCH are extradural; they originate from within the vertebral bone and extend to the epidural space^{1,2}. Pure epidural cavernous hemangioma (ECH) is extremely rare, accounting for approximately 4% of SCH cases³.

Although the exact mechanism of occurrence has not been determined, cavernous hemangiomas are presumed

to originate from endothelial cells; some theories say it can be generated from dysplastic blood vessel progenitors or telangiectasia, which is gradually increasing¹⁾. SCH causes symptoms such as back pain and insidious or acute paraplegia that imitates the radiculopathy of disc protrusion. Trauma can often lead to radiculopathy, which can be confused with acute disc protrusion⁴⁾.

Several reports exist on the use of Korean medicine (KM) for treating gait disturbance; most of these state that KM is effective in such cases. One study reports on the use of KM for treating gait disturbance caused by SCH⁵⁾. Shin and Kim⁵⁾ report on using KM to treat dysuria caused by neurogenic bladder and gait disturbance. However, to the best of our knowledge, no reports exist on using KM to treat gait disturbances caused by ECH. Therefore, we present a case wherein we used KM to treat gait disturbance caused by ECH.

Case>>>>

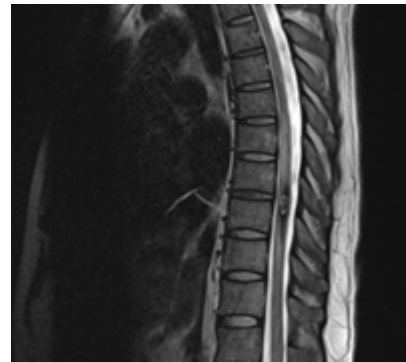
This study has been approved by the Institutional Review Board of Kyung Hee University Korean Medicine Hospital in October, 2023 (KOMCIRB 2023-09-008-001).

1. Chief complaints

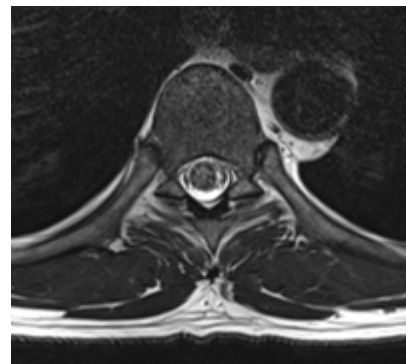
A 51-year-old man visited a KM hospital for treatment for his gait disturbance and lower limb rigidity in July 2022.

2. History of present illness

The patient felt lower limb rigidity on January 2019. On April 2019, he experienced lower limb dysesthesia and gait disturbance. Subsequently, the patient visited Korea University Anam Hospital, was diagnosed with a spinal cord tumor (cavernous angioma), and underwent T8-T9 laminectomy and tumorectomy on April 2019



(A)



(B)

Fig. 1. (A) Sagittal view of thoracic vertebra. We can see the cavernous hemangioma on T8-T9 level. (B) Axial view of T8-T9.

(Fig. 1). Afterwards, the patient was admitted to rehabilitation center of Seoul Soong-in Hospital and recovered well; however, his symptoms worsened without cause on February 2021. He revisited Korea University Anam Hospital, where a T-spine magnetic resonance imaging (MRI) examination showed rebleeding. However, no additional treatment was prescribed. As the symptoms continued, he decided to visit the rehabilitation center of the Kyung Hee University Korean Medicine Hospital for treatment with KM.

3. Past illnesses

The Patient has been taking medicine for hyperlipidemia since 2017, and hepatitis B since 2019. He got Covid-19 at April 2022.

4. Physical examination

The patient's right thigh showed abduction and external rotation tendency while walking. Both legs had motor weakness and numbness: the right and left leg showed 40% and 70% strength, respectively. He had poor scores in the 25 foot walk test (25FW), timed up and go test (TUG), and comfortable gait speed (CGS) tests; the Berg balance score (BBS) was low as well.

5. Imaging examinations

A radiograph of the thoracolumbar spine showed signs suggestive of laminoplasty at the T8-T10 level with posterior instrumentations and disc degeneration at the L3-S1 level with spondylosis. His electrocardiogram was normal.

6. Patient perspective

"I felt gait disturbance and lower limb dysesthesia on April 2019, then I visited Korea University Anam Hospital. I was diagnosed with ECH and underwent T8-T9 tumor-ectomy and laminectomy on April 2019. After surgery, I participated in a rehabilitation program at the local hospital and improved. However, on February 2021, the symptoms worsened again, so I visited the hospital where I underwent the surgery. The MRI showed rehemorrhage, but no additional treatment was provided. Meanwhile, I visited the Kyung Hee University Korean Medicine Hospital. Now I can walk by myself but while walking my right thigh tends to abduct and rotate externally, and the right toe drags. Off and on spasm and rigidity occur in the right lower limb, it gets worse when I sit for long hours and is improved after relaxing. I can barely feel any sensations on the left limb and feel numbness from the waist at the navel to the end of the lower extremity, and the numbness become stronger when it goes lower.

I hope to get well enough to live normal life and get back to work after admission treatment."

7. Final diagnosis

After biopsy, patient was diagnosed with SCH.

8. Treatment

1) Acupuncture

The patient received acupuncture with disposable stainless steel needles (0.2×40 mm, Dongbang Acupuncture Inc.) once a day during admission and ambulatory care. The acupoints were for lung tonification (HT8, LU10, SP3, LU9), Hwatahyeopcheok (Hua-Tuo-Jia-Ji-Xue) points, the first lateral line of the bladder meridian (corresponding to T8-T9), and of the lower limb, such as LR3, ST41, SP6, ST36, GB34, and GB31.

2) Fire needling (FN)

The patient underwent FN therapy once a day during admission, at the point of origin and insertion of the tibialis anterior; the ST36 and SP3 acupoints.

3) Electroacupuncture (EA)

The patient underwent acupuncture with stainless steel needles (0.25×40 mm, Dongbang Acupuncture Inc.) on the part of left limb with paresthesia. The frequency of electrical stimulation was set 120 Hz for 20 minutes, once a day. The acupunctured needles were inserted shallowly at each acupoint.

4) Herbal medicine

The patient was administered Boyanghwanoh-tang (Buyanghuanwu-tang, BHT), which consists of *Astragali Radix*, *Angelicae gigantis Radix*, *Paeoniae Radix Rubra*, *Cnidii Rhizoma*, *Lunbricus*, *Percicae Semen*, *Cartgami Flos*, *Achyranthis Bidentatae Radix*, *Salciae Miltiorrhizae Radix*, *Connamomi Ramulus*, *Polygalae Radix*, *Acori Graminei Rhizoma*. The patient was administered BHT orally thrice a day, in accordance with KM theory.

5) KM physiotherapy

The patient received Chuna, the manual therapy used in KM, once a day.

6) Others

The patient received cupping on the lumbar and hip areas, and moxibustion on the abdominal area.

9. Outcome and follow-up

During 17 days of admission and 20 days of ambula-

tory care, the patient's treatment comprised acupuncture, FN, EA, herbal medicine, moxibustion, cupping, and Chuna. The patient was evaluated with respect to his gait condition and pain, using the following assessment tools: 25FW, TUG, CGS, NRS of low back pain (LBP) and leg, BBS, Tinneti score, manual muscle test (MMT), EuroQol-five dimension (EQ-5D), and EuroQol-five dimension visual analog scale (EQ-5D VAS). The 25FW reduced from 8.39 to 6.9, TUG from 16.47 to 12.51, and CGS from 14.16 to 11.87 at the end of treatment. The NRS for LBP and leg reduced from 3 to 1~2, and finally

Table I. Outcomes and Changes of Gait Evaluation

	7/7	7/14	7/21	8/3	8/17	9/7	9/21	10/5	10/19	11/2
25FW	8.39	8.40	8.81	8.70	7.35	6.47	7.17	7.34	7.48	6.9
TUG	16.47	14.85	14.56	12.30	11.53	10.02	11.41	11.94	11.72	12.51
CGS	14.16	13.99	13.32	13.98	12.65	11.09	11.57	11.88	12.43	11.87
NRS (LBP)	3	1.5	1	2	2	1	1	1	1	1~2
NRS (leg)	3	2	2	2	2	1	1	1	1	0
BBS	46		51							53
Tinneti score	23		23							23
MMT	G/N		G/N							G/N
EQ-5D	22221		22221							22221
EQ-5D VAS	60		66							70

25FW: 25 foot walk test, TUG: timed up and go test, CGS: comfortable gait speed, NRS: numeral rating scale, LBP: low back pain, BBS: Berg balance score. MMT: manual muscle test, EQ-5D: EuroQol-five dimension, EQ-5D VAS: EuroQol-five dimension visual analog scale.

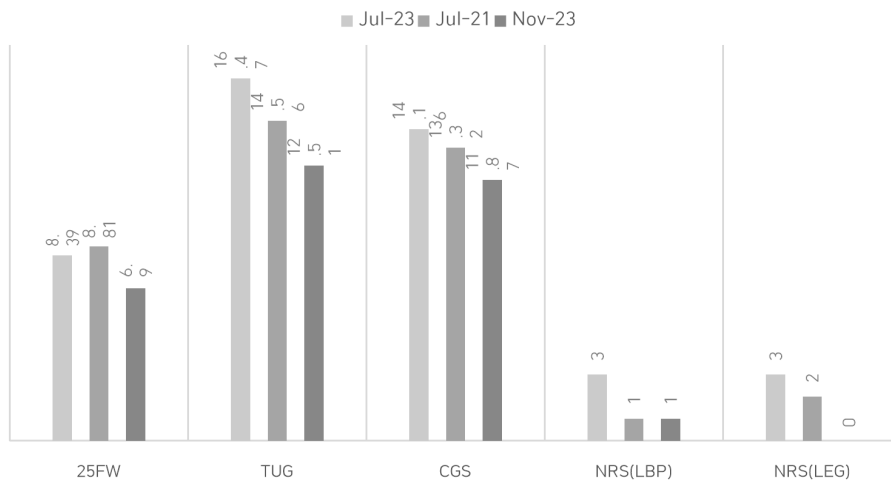


Fig. 2. Outcomes and changes of 25 foot walk test (25FW), timed up and go test (TUG), comfortable gait speed (CGS), numeral rating scale (NRS) of low back pain (LBP), and numeral rating scale (NRS) of leg.

to 0 by the end of treatment. Furthermore, the BBS rose to 53 from 46 and the EQ-5D VAS increased from 60 to 70. However, the Tinetti score, MMT, and EQ-5D were unchanged (Table I, Fig. 2).

The patient's symptoms improved during admission. Therefore, the patient got ambulatory care after.

The patient and professional therapist reported no side effects during the treatment.

Discussion>>>>

Cavernous hemangioma is a rare disease; few reports exist of treating the same with KM. To the best of our knowledge, Choi⁶⁾ and Shin and Kim⁵⁾ are the only two reports. The case by Choi⁶⁾ is of a cavernous hemangioma at the medial side of the right transverse cubital crease in an 8-month-old infant. The case by Shin and Kim⁵⁾ is of an intermedullary SCH that initially presented with dysuria caused by neurogenic bladder and gait disturbance. However, Shin and Kim⁵⁾ did not propose objective evaluation for the gait disturbance, and only used the PULSES profile (physical condition including diseases of viscera, self care activities, mainly upper limb function, mobility activities, mainly lower limb function, sensory components relating to communication & vision, excretory function, bladder & bowel, Intellectual & emotional adaptability, family & financial support), which is used widely for evaluating the ability of self-care. In contrast with these case reports, our study is focused on treating gait disturbance in an adult man, which is caused by spinal ECH. In addition, our study used several objective evaluation tools to rate improvement of gait ability.

After 37 days of KM care, the patient's gait disturbance showed meaningful improvement. The 25FW, TUG, and CGS, which are used for evaluating walking speed and mobility, were elevated. For 25FW, the patient should walk and walk back 25 feet as quickly as possible but

safely within 1 to 5 minutes. In TUG test, patient starts on chair in sitting position, walk 3 m after standing up, and walk back to chair, then sit down on chair again, which should generally performed within 10 seconds. For CGS, patient's gait speed is measured with m/sec during walking 4 m. The gait speed can affect gait pattern in an individual⁷⁾. In the elderly, increased gait speed can provide more stable and superior quality of walking⁸⁾. Based on the improvement in the patient's gait speed and ability, we can state that the patient's gait quality improved. Pain is a significant influence on gait. The NRS, which indicates the degree of pain, decreased in both lumbar and leg⁹⁾. This suggests that the patient's walking ability has improved.

The BBS evaluates patient with each 14 tasks, which is divided by 0 to 4 points. If BBS exceeds 41 points, patient is evaluated to be able to walk independently. The BBS is an effective and appropriate evaluation method used for balance measuring; it increased in the patient^{10,11)}. However, the Tinetti score, which measures both balance and gait, was unchanged^{12,13)}. Tinetti score evaluates patient with nine tasks by scoring from 0 to 28 points. If the score is 25 points or higher, the patient is evaluated as normal. The MMT score and EQ-5D were unchanged as well. The MMT score stays same unless it returns to full normal even if there is an increase in muscular strength. The EQ-5D score comprises 5 questions with 5 choices each; therefore, the patient's selection cannot be changed easily if they have not recovered completely.

The patient underwent various KM treatments based on the KM diagnosis. In acupuncture, the patient received general acupuncture, EA, and FN. The EA was applied at an intensity of 120 Hz by shallow insertion of needles on the lower extremities with dysesthesia. EA suppresses pain by activating sympathetic nerves and consequently blocking cytokine release. A rat model showed improved weight bearing and increased stepping strength in treated legs, along with suppressed spinal nerve activity¹⁴⁾. A study reported that volunteers with paresthesia who re-

ceived EA tended to recover tactile sensitivity faster. When neural transmission fibers are stimulated by EA, it can accelerate repairing and returning to normalized nerve transmission more quickly¹⁵⁾. Based on the several research results, we could apply EA on paresthesia.

The FN treatment was performed at the onset and end of the anterior tibialis to reduce the rigidity of the anterior tibialis and strengthen muscle activity. FN has a better effect on stiffening symptoms than conventional acupuncture. It also improves local blood circulation, removes pathological changes in tissue, and helps restore motor neuronal function¹⁶⁾.

The patient was administered BHT orally three times a day. BHT is a KM herbal medication used to take care of symptoms caused by stroke including speaking disorders, hemiplegia, and facial nerve palsy. Various studies have reported that BHT has neural protection and angiogenesis effects. It has the potential ability to manage typical neurological deficits or cognitive impairments. Thus, by using BHT, we could expect patient's neurological problems would be improved¹⁷⁾. Chuna was used as the KM physiotherapy. When administering Chuna, dorsal relaxation centered around the right lumbar erector was included. Muscle energy therapy for the right iliopsoas, hamstring, and rectus femoris, stretching for the gastrocnemius, guidance for correct ambulation, ankle pumping exercise, and knee flexion exercise were also included. For correct ambulation, the patient was taught to limit right external rotation, and equalize the grounding time of both foot. Additionally, cupping was done on the lumbar and hip area and moxibustion was done on the abdominal area.

The patient showed signs of convulsions while sleeping or lying after he discontinued taking *gabapentin* after discharge. However, the patient continued ambulatory care; on the 8th of August, the convulsions reduced to about 50% compared to right after discontinuing *gabapentin*. Rigidity after exercise decreased after regulating exercise. The patient reported discomfort while climbing upstairs although walking on flat land or downhill became easier.

During ambulatory care, the patient reported that climbing upstairs also became easier from the 23rd of September. By the time the treatment ended, he stated that walking had become convenient compared to when treatment first began. The patient had improved enough to walk 7,000 steps per day, or 12,000~13,000 steps per day if he goes hiking.

This study has a few limitations. First, there was no follow-up on the patient's condition after treatment ended, so we could not know whether improvement had continued after treatment. Second, after the treatment, the patient's walking condition improved but rigidity persisted, leaving questions about the complete recovery of symptoms or rigidity. Third, the degree of improvement was not consistently lowered, there was increase or decrease in symptoms depending on the patient's condition. Finally, the BBS and Tinetti score are both evaluation metrics for measuring balance; the BBS showed improvement but the Tinetti score stayed the same, which is not a good sign when evaluating balance, and it would be better if we could have double-checked the patient.

Summary>>>>

This case study showed improvement both in pain and gait disturbance caused by SCH. KM can be a positive treatment for reducing pain or improving gait disturbance caused by SCH, and it can be considered a treatment option for SCH. Future studies with a larger number of cases and longer period of follow-up in SCH with gait disturbance and rigidity are needed.

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