



Case Report

A Case of Syringomyelia with Back and Shoulder Pain

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ABSTRACT

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The purpose of this study was to describe the treatment of the clinical symptoms of syringomyelia using Korean medicine. A patient with syringomyelia complained of back and shoulder pain, numbness of the upper right limb, headaches and dyspnea. He was treated with herbal medicine, acupuncture, moxibustion, and physical therapy. The Visual analog scale (VAS) was used to score sensory symptoms (pain, numbness) and Modified Borg scale (mBorg) was used to score dyspnea. After the treatment, his symptoms were relieved. The patient's chief complaint was back and shoulder pain which was reduced greatly from a VAS score of 6 between 1-2. The results in this study may contribute to the development of the Korean medicine field of syringomyelia in the future.

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Introduction

Syringomyelia is a disease that manifests various neurological symptoms. Cystic cavities are formed in the spinal cord due to various causes such as Chiari deformation, spinal cord tumor, spinal cord injury, and meninges fibrosis [1]. Typical clinical symptoms are ascending palsy of the sensory and motor nerve in the lesion of the spinal cord. Sometimes, this may be accompanied by pain or autonomic nervous system abnormality. Depending on the location or size of the cystic cavities, various symptoms may appear.

It can be easily diagnosed by magnetic resonance imaging (MRI) or diskography. However, the exact cause and pathophysiology of the disease has not yet been elucidated. Various treatment methods including surgical treatment have been tried depending on the cause, location, and size of the onset, but they are a matter of opinion [2]. In Korea, some case reports have been reported such as Kang [3]. In addition, in Korean medicine, only 3 cases have been reported since 2000. The purpose of this study was to investigate the effect of Korean medicine in patients with syringomyelia by herbal medicine, acupuncture, moxibustion and physical therapy (IRB No.: SMJOH-EX-18-01).

Case Report

Patient

Male, 27 years old.

Chief complaint

Back and shoulder pain, headache, numbness of the upper right limb, difficulty in breathing.

Past medical history

Traumatic hemorrhage

He was admitted to Bundang CHA General Hospital and received pharmaceutical treatment in 2015.

Syringomyelia

Syringomyelia was diagnosed at Bundang CHA General Hospital in January 2018.

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Herpes zoster (Rt. arm)

Herpes zoster was diagnosed at Bundang CHA General Hospital in January 2018.

Meniscus cyst (right knee)

The meniscus cyst was operated on at Busan Armed Forces Hospital in 2011.

Present medical history

An intermittent severe headache began in 2015. When the headache began, he treated it with some painkillers in the emergency room. In January 2018, he suffered back and shoulder pain, headaches, and sensory abnormality of the upper right limb. He decided to have an in-depth medical checkup at Bundang CHA General Hospital which resulted in a diagnosis of syringomyelia (medulla oblongata, C5-6/6-7, T8-9/9-10). He was admitted and stayed in hospital for a month receiving Western medication. Even though the number of headaches decreased, the remaining symptoms were unchanged. From April 16th 2018 until June 3rd 2018, he was admitted to and stayed in the Department of Acupuncture and Moxibustion Medicine, Semyung University Jecheon Hospital of Korean Medicine.

Symptoms and physical examination at the first medical examinationBack and shoulder pain

The visual analogue scale (VAS) score for back and shoulder pain was between 4-5. The pain and stiffness had spread across the posterior cervical muscle to rhomboideus and trapezoid muscles. In addition, the pain worsened at night. The Spurling test gave a +++ result. Cervical active range of motion flexure/extension was favorable. For both right and left sides lateroflexion was 25-30°/40°. Valsalva T was +.

Headache

The VAS score for the level of pain experienced during headaches was between 4-5. A headache was not present at the time of admission but onset was sudden during hospitalization. The headaches displayed a tendency to appear as an occipital headache of pulsating pattern which usually appeared when he felt discomfort or heaviness in his chest, this was also accompanied with confusion, nausea, blurred vision, dizziness.

Sensory abnormality

The VAS score for sensory abnormality was between 1-4. A numbness of the upper right limb was not a persistent symptom. When posterior neck pain was severe, he did not feel sensory abnormality. The numbness became worse when he moved his upper right limb, or at nighttime.

Difficulty in breathing

Difficulty in breathing happened twice during hospitalization and onset was sudden and uncomfortable, and its intensity of shortness of breath was a mBorg score of 3. A coughing episode was accompanied with dyspnea. He expressed a feeling of discomfort and heaviness in his chest. Chest pain/tachypnea/hyperpnea -/-/-.

There was no hypotonia, myoatrophy, or neurological abnormalities.

Examination referencesUric-acid

8.5 mg/dL

Chest PA

Nonspecific

EKG

Normal Sinus Rhythm, Rate 68

Normal Axis

Normal ECG

Magnetic resonance imaging

Syringomyelia [medulla oblongata, C5-6/6-7, T8-9/9-10 (Figs. 1 and 2)]

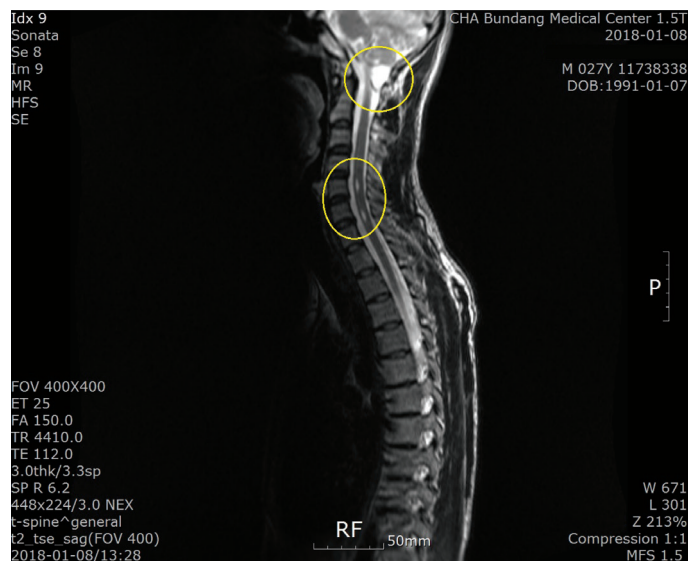


Fig. 1. T2 weighted sagittal plane of magnetic resonance imaging (2018-01-08). The cavities in the spinal cord are shown (medulla oblongata, C5-6 level).

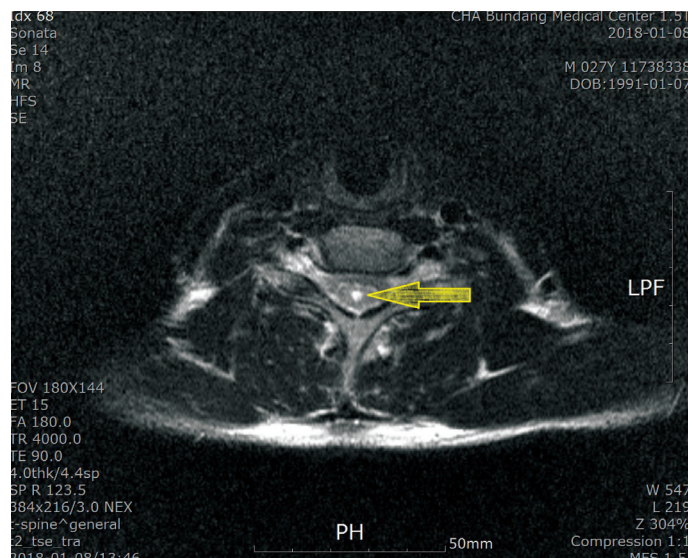


Fig. 2. T2 weighted transverse plane of magnetic resonance imaging at the level C5-6 (2018-01-08). The cavity inside the central canal appears white.

Materials and Methods

Treatment

Herbal medicine

He suffered from severe posterior neck pain. He was underweight and had dark skin. The patient's condition was ascertained by checking the pulse and the coating on the patient's tongue. He was diagnosed as "yin deficiency". He was treated with herbal medicine, *Gami-Seokyong-tang*, *Gami-yugmijihwang-tang*, and *Sipjeondaebo-tang* of Bangyakpyeon was administered to supplement the systemic blood and balance his yin and yang. From 2018-05-11, as the patient's back and shoulder pain reduced, intermittently the headache appeared to worsen VAS score 5-6. *Samul-tang plus Yijin-tang gamibang* was administered with reference to Donguibogam.

Gami-Seokyong-tang (Curcumae longae rhizoma, Angelica gigas Nakai, Kalopanax pictus Nakai, Atractylodis Rhizoma Alba, Paeoniae Radix Rubra, Notopterygii Rhizoma, Glycyrrhiza uralensis Radix, Rehmanniae Radix).

Gami-yugmijihwang-tang (Rehmanniae Radix Preparata, Corni Fructus, Dioscoreae Rhizoma, Moutan Cortex Radicis, Poria cocos Wolf, Alismatis Rhizoma, Paeoniae Radix Rubra, Anemarrhenae Rhizoma, Phellodendron cortex, Zingiberis Rhizoma Recens).

Sipjeondaebo-tang (Ginseng Radix Alba, Glycyrrhiza uralensis Radix, Angelica gigas Nakai, Zizyphi inermis Fructus, Poria cocos Wolf, Paeoniae Radix Rubra, Atractylodis Rhizoma Alba, Zingiberis Rhizoma Recens, Rehmanniae Radix Preparata, Cinnamomum cassia Bark, Cnidii rhizoma, Astragalus membranaceus Radix, Chaenomeles sinensis (Thouin) Koehne, Notopterygii Rhizoma).

Samul-tang plus Yijin-tang gamibang (Angelica gigas Nakai, Cnidii rhizoma, Paeoniae Radix Rubra, Rehmanniae Radix Preparata, Pinelliae Rhizoma, Aurantii nobilis pericarpium, Poria cocos Wolf, Glycyrrhiza uralensis Radix, Cimicifuga heracleifolia, Bupleurum falcatum L., Scutellariae radix, Schizonepeta tenuifolia, Saposhnikovia Radix).

Acupuncture

The patient was treated with acupuncture on the Governor Vessel (GV), Kidney Meridian (KI), painful points, and trigger point mainly while in the sitting position. Acupuncture treatment was performed twice a day and once a day at weekends for 15 minutes. Disposable needles (Dongbang medical, 0.20 x 30 mm stainless steel) were used. The patient complained of severe pain during acupuncture treatment, so additional procedures such as pharmacopuncture or electrical stimulation were not performed.

Moxibustion

Indirect moxibustion treatment (2 ways) was conducted. It was first performed on Gwanwon (CV4) every morning for 20 minutes. Then it was performed on the cervical vertebra and shoulder joint region, for example, Pungbu (GV16), Pungji (GB20), Daechu (GV14), Dodo (GV13), Gyeonjeong (GB21), Cheonryo (TE15), Geogol (LI16). When the upper right limb sensory abnormality worsened at night, moxibustion was performed once again. The procedure took about 15 minutes.

Rehabilitation

For muscle relaxation to cure the posterior neck pain and upper limb sensory abnormality, a hot pack and electrotherapy (STI-500, interference stimulator, manufactured by StraTek, low-frequency current was applied to the affected area to relieve muscle pain) was

used. In addition, aqua massage therapy (Medizen) was performed to promote metabolism and the relaxation of every muscle in his body. These physical therapies were practiced every day for about 20 minutes.

Evaluation

In this study, back and shoulder pain, headache, sensory abnormality on the upper right limb, and difficulty in breathing were the main complaints of the patient. The VAS was used to score the sensory discomfort to evaluate the 3 symptoms (back and shoulder pain, headache, numbness). The dyspnea was measured using the Modified Borg scale (mBorg) (Appendix 1) as a Korean translation, which is widely used.

Results

Back and shoulder pain

The patient complained of severe pain from the time of admission. Initially, he could barely receive acupuncture treatment due to the severe pain he was suffering. In addition, the symptoms worsened at nighttime. As the treatment advanced, the wide range of the pain gradually localized around Pyesu (BL13) and the pain intensity reduced. At discharge, the VAS score was between 1-2 for back and shoulder pain (Fig. 3).

Headache

At the time of admission, the headaches were not severe. However, the severity of pain began to increase from April 30th as back and shoulder pain reduced. It disappeared after treated with herbal medicine *Samul-tang plus Yijin-tang gamibang* curing the headaches from May 11th, except for when the headache was

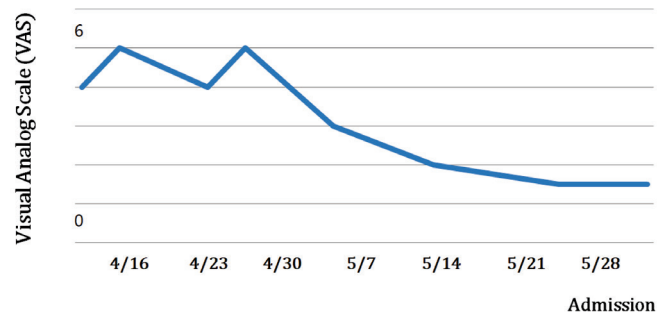


Fig. 3. Back and shoulder pain measured by Visual Analog Scale score. At discharge the pain had gradually decreased to less than half that at discharge.

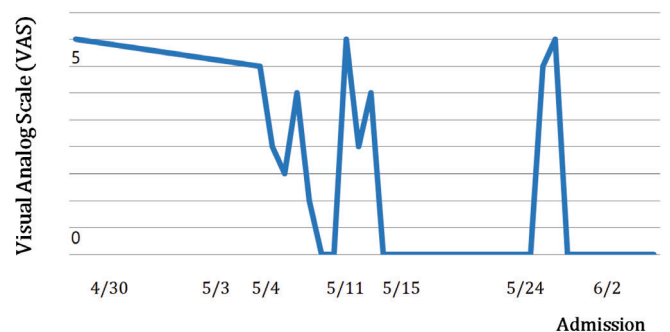


Fig. 4. Headache measured by Visual Analog Scale. Headaches disappeared after May 10th, except when the patient complained of dyspnea on, May 24th and May 25th.

temporarily associated with dyspnea on May 24th and 25th (Fig. 4).

Sensory abnormality

As the back and shoulder pain decreased, the patient began to perceive the sensations of numbness and gradually the sensory abnormality was strongly revealed in maximum intensity VAS score of 4. Symptoms were cured or relieved after moxibustion. The pain was gradually reduced, and had completely disappeared at the time of discharge.

Difficulty in breathing

Difficulty in breathing was not a persistent symptom, it occurred twice in the second half of hospitalization, therefore treatment was not given. Respiratory distress intensity was mBorg 3 and the symptoms disappeared within 4 hours after the onset.

Discussion

In 1546 Estienne first described the cavitation of the spinal cord. In 1827, Ollivier d'Angers was the first to identify the syringomyelia [4]. Syringomyelia is a lesion in which a cystic cavity is formed in the spinal cord. It may form hydromyelia in the central canal or a syrinx of the spinal cord and it may cause gliosis [5]. Epidemiologically, it is not age or gender specific. The incidence area of syringomyelia varies according to the study, but it is reported to be commonly found in the cervical vertebra and thoracic vertebrae [6-8]. Clinical symptoms are characterized by pyramidal signs such as dissociation of sensation, weakening of the upper limb, muscle atrophy, disesthesia, and a tendon reflex. In addition, symptoms such as dyspnea and dysphagia could occur, and various other clinical symptoms may occur depending on the location, extent, and size of the cavity.

Syringomyelia is often accompanied by Chiari 1 malformation and diffuses from the center of the spinal cord to the periphery. Spinothalamic tract at connection is the first to be injured and the lesion progresses to injure the anterior spinal cord and lateral corticospinal tract. As the spinal cord cavity expands and the nerve of the neurotransmission pathway is pressed, most clinical symptoms appear.

The most common etiologic factor is congenital anomalies with hindbrain herniation (Chiari 1 malformation, CM). Syringomyelia is acquired by fibrosis of the meninges caused by meningitis, spinal trauma, or spinal cord tumors. It may also occur idiopathically. The causes of the disease are divided into the communicating, noncommunication by communication with cerebrospinal fluid circulation route, or by hindbrain-related and nonhindbrain-related, depending on the pathophysiology.

The pathogenesis mechanism is not known precisely, but there are many hypotheses. About 75%-80% of syringomyelia is found to be associated with Chiari 1 malformation, CM, and Chiari argued that it originated from congenital hydrocephalus. Ball and Dayan explained that cerebrospinal fluid drains into the spinal cord through the arteries [9]. Williams suggested the mechanism that cerebrospinal fluid inflow from the subarachnoid space into the spinal cord, because the epidural vein expands and pressure is increased [10]. Gardner thought that syringomyelia developed due to the abnormality of outflow route of the fourth ventricle [11].

In general, Chiari 1 malformation, CM is diagnosed when the tonsillar herniation is larger than 5 mm from the foramen magnum in MRI [12]. In this case, the cerebellar tonsil or medulla oblongata did not displace below the foramen magnum in MRI. In addition, this patient had not had any congenital disease, hydrocephalus or surgery history. However, there was a history of Western medication for traumatic cerebral hemorrhage in

2015. Syringomyelia is a post-traumatic complication. It develops spontaneously after a matter of months or several decades, and only 0.3-3.2% individuals show symptoms even if there is a cavity in the spinal cord [13]. The mechanism of spinal cord cavitation after trauma is unclear but there are a few causes. After the occurrence of meningeal fibrosis, spinal cord cavitation develops into cerebrospinal fluid circulatory disorder in the subarachnoid space. Spinal stenosis born of spinal injury may be an important cause [14,15]. In this case study, there were cavities in the medulla oblongata, lower cervical and thoracic vertebrae, but he had no direct trauma to the disease site. However, cerebrospinal fluid leakage due to traumatic cerebral hemorrhage may be associated with the development of lesions.

The treatment of syringomyelia can be through surgical procedure when there is the progression of neurological disorders, and the enlargement of the vertebral column is visible in diagnostic imaging. Myelotomy or syringoperitoneal has been used. Recently, there have been decompressive laminectomy, subarachnoid space scale plastic surgery, and epidural plastic surgery [16]. Drug therapy includes non-steroidal anti-inflammatory drugs, anticonvulsants, narcotics, N-methyl-D-aspartate receptors, antagonists, ketamine and dextromethorphan [17].

Meanwhile, there has been little research into syringomyelia in Korean traditional medicine. Hwang and Kim [8], Jung et al [18], Yoo et al [19], and Park et al [20] have reported cases where patients were treated with Korean medicine for syringomyelia. In previously reported studies, herbal medicine and acupuncture treatment on the Governor Vessel (GV), Kidney Meridian (KI), and painful points were the same treatment in all studies. There were some cases of additional treatment with a pharmacopuncture or cupping therapy. Park et al [20] employed bloodletting-cupping and Yoo et al [19] performed Chuna.

The patient in this case had a post-traumatic intracerebral hemorrhagic syringomyelia. The lesions were medulla oblongata and 4 sites in the spinal cord, the lower cervical and the thoracic vertebral region. He was admitted to a hospital and received Western medication. His headaches only slightly improved, but other symptoms remained after discharge. At the initial time of admission to Jecheon Hospital of Korean Medicine, he had difficulty in sleeping because of severe back and shoulder pain.

During hospitalization, the patient's back and shoulder pain had decreased in its progress. As a result, pain relief at rest was reduced by more than 60% compared with admission. The headaches were intense with a VAS score 4 or higher. Herbal medicine *Ijinhabsamultang* was prescribed for headaches, and the pain disappeared except when accompanied by dyspnea. As the back and shoulder pain reduced, the patient began to experience a greater discomfort in the numbness of the upper right limb. Moxibustion treatment alleviated the abnormal sensation immediately after treatment. The patient experienced intermittent respiratory distress only twice during admission. The herbal medicine *Eogganhwan* (Angelica gigas Nakai, Ramulus et Uncus Uncariae, Cnidii rhizoma, Angelica Dahurica Root, Atractylodis Rhizoma Alba, Poria cocos Wolf, Bupleurum falcatum L., Glycyrrhiza uralensis Radix, Citri Reticulatae Viride Pericarpium, Coptis chinensis, Raphani Semen, Borneolum Syntheticum, Mentha arvensis L.) were effective in the treatment of dyspnea.

The patient was able to perform daily life without outpatient treatment or any medical treatment after discharge. Korean medicine treatment helped to alleviate the symptoms of syringomyelia and improve his quality of life. However, there are limitations in this study, the hospitalization was short (47 days) and the change in symptoms could not be observed after patient discharge.

Although this study has only 1 case of syringomyelia as a refractory disease, the patient had cavities in medulla oblongata as well as in the spinal cord. This is the first case of Korean medicine as an example of cavity in medulla oblongata. Besides, this is the first case report of syringomyelia associated traumatic hemorrhage in Korean medicine. It is considered to be an example showing the possibilities of Korean medicine with remarkable effective treatment. It is thought that Korean medicine should be promoted to widen the scope of treatment.

This is a case report of a patient diagnosed with syringomyelia in January 2018 after traumatic cerebral hemorrhage in 2015. He suffered from syringomyelia symptoms such as back and shoulder pain, numbness of upper right limb, headaches, and dyspnea. We treated him with *Gami-Seokyong-tang* for his severe neck pain and strong stiffness. And we diagnosed him as “yin deficiency” and *Gami-yugmijihwang-tang* was administered. *Sipjeondaebotang* was used to adjust his yin-yang balance. The intermittent headache appeared and *Samul-tang plus Yijin-tang gamibang* of Donguibogam was administered. In this case, electroacupuncture or pharmacopuncture was not practiced. It showed the relief of muscular rigidity only by acupuncture treatment without the need an electroacupuncture or a pharmacopuncture. We diagnosed the numbness of the upper right limb as “deficiency of yin and yang” and the patient was treated with moxibustion on upper right limb and Gwanwon. Korean medicine helped him experience clinical remission and physical therapy made a change for the better.

In conclusion,

1. back and shoulder pain decreased to less than 40% of the initial pain. In particular, numbness and headache were completely eliminated.

2. When the numbness or pain became worse at night, moxibustion was applied again and symptoms were immediately relieved after the procedure.

3. The limitation of range of motion of the cervical vertebra was improved to near the normal level. Consequently, his quality of life improved allowing him to conduct his daily life. High treatment satisfaction was achieved.

Conflicts of interest

The authors have no conflicts of interest to declare.

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Appendix 1.

Modified Borg category scale

10	Maximal
9	Very, very strong (almost max)
8	
7	Very strong
6	
5	Strong (heavy)
4	Somewhat strong
3	Moderate
2	Weak (light)
1	Very weak
0.5	Very, very weak (just noticeable)
0	Nothing at all