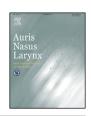
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Case report

A case of extensive pharyngeal vascular malformation successfully treated with Kampo medicine

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ABSTRACT

Objective: To present the efficacy of Japanese-traditional medicine (Kampo) for a case with vascular malformation.

Methods: A case study and literature review.

Patient: A 62-year-old female presented with dysphagia and spitting blood. Esophagogastroduo-denoscopy showed a longitudinal lobulated and septated mass in the posterior pharynx. On MR imaging, the mass showed hyperintensity on T2-weighted images and heterogeneous enhancement on Gadlinium-enhanced T1-weighted images, suggestive of a low-flow vascular malformation. Intervention: According to the Kampo diagnosis, kamisyouyousan and ninjinyoueito were prescribed to this patient. The effect of Kampo medicine was evaluated with improvement of her symptoms and volumetry of MRI findings.

Result: The longitudinal pharyngeal mass was markedly decreased and her symptoms disappeared after 2 years of Kampo administration.

Conclusions: Kampo medicine can be a novel alternative therapy for VM.

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1. Introduction

Vascular malformations remain a therapeutic challenge. They are congenital lesions that never regress and grow with time, trauma, and/or hormonal changes. They are composed of abnormally formed vascular channels lined by endothelium that do not exhibit abnormal cellular turnover or mitosis. They may be of the low-flow variety, including capillary malformations (CM), venous malformations (VM), and lymphatic malforma-

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tions (LM), or of the high-flow variety, including arteriovenous malformations (AVM). In the head and neck, venous malformations (VM) are the third most common vascular mass and may be seen within the muscles of mastication, lips, tongue, or elsewhere within the upper aerodigestive tract. The treatments of VM in the upper aerodigestive tract include laser treatment, resection, and sclerotherapy, but these therapeutic interventions are highly invasive and could cause a serious functional deterioration such as dyspnea or bleeding [1].

The confusion seen in Japan is that both vascular tumors and malformations are often called "hemangioma" in clinical practice. Indeed, VM has been often referred as "cavernous

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hemangioma", "intramuscular hemangioma", "venous hemangioma", and so on. So, if the reviewer meant these types of "hemangioma", VM could be the same as "hemangioma".

The unique role played by traditional Japanese herbal (Kampo) medicine is gradually attracting worldwide attention. It is the most frequently used type of alternative and complementary medicine in Japan [2]. The aim of Kampo therapy is to improve patients' condition regardless of their underlying diseases. Herein, we report on a patient suffering from an extensive pharyngeal low-flow vascular malformation causing chronic dysphagia which was successfully treated with Kampo medicine.

2. Case report

A 62-year-old woman with hemoptysis and dysphagia was referred to our Kampo clinic. She was diagnosed as having pharyngeal "hemangioma" at 50 years of age. She had difficulty in swallowing solid foods, difficulty in singing or talking loudly, and occasionally, was spitting blood. She had a history of endometriosis. Her social history included no drinking and no smoking. She had enjoyed singing karaoke, but she could not sing properly after exacerbation of the disease. Her symptoms exacerbated with mental and physical stress. Laryngoscopic evaluation revealed a broad-based lobulated mass with intact overlying mucosa on the posterior pharyngeal wall. MR imaging revealed a longitudinal lobulated and septated mass measuring 12 cm in diameter at its largest point occupying the posterior to the right portion of the oropharynx. On MR imaging, the mass showed high signal intensity on T2-weighted images, and heterogeneous enhancement on Gadoliniumenhanced T1-weighted images, suggestive of a low-flow vascular malformation (Fig. 1). Although calcified phleboliths, a pathognomonic feature of venous malformations, were not observed, we considered our case as a slow-flow type vascular malformation, most likely venous malformation, based on the clinical and imaging findings including bleeding episodes, high-signal intensity on T2 weighted image and heterogeneous enhancement on post-contrast MR imaging suggesting the blood stagnation. We definitely make the diagnosis of vascular anomalies based on the classification proposed by the International Society for the Study of Vascular Anomalies (ISSVA classification) [1], which is the current standard worldwide. In this way, the most likely diagnosis of pharyngeal venous malformation was made. Radical operation was recommended because of the risk of serious bleeding or asphyxia, but the patient refused to undergo surgery. The previous otorhinolaryngologist had already prescribed Kampo medicine such as kakkonto, hangekobokuto, and syousaikoto according to the symptoms, but any of them did not improve her symptoms. Based on our Kampo diagnosis, in which stagnation of blood flow with heat and deficiency of qi (vital energy) and blood were observed, kamisyoyosan and ninjinyoeito extracts (Kracie pharmaceutical, Japan) were prescribed (Tables 1 and 2). Especially, kamisyoyosan is said to be very effective for upper blood heat, a pathological change in which exuberant heat or fire enters the blood, usually causing hemorrhage. Moreover, she had deficiency of qi and blood after continuous bleeding and anxiety for this disease, ninjinyoeito was also prescribed. In order to cover her symptoms and pathological condition according to the Kampo diagnosis, both of two formulas were indispensable. She continued to take the same Kampo fomulas for two years, and symptoms and MR images were evaluated during the follow-up. The lesion volume was estimated by MR imagery with volumetric software using threshold and region growing method (Aquarius iNtuition Edition ver 4.4.11., TeraRecon. Inc., Tokyo). The extraction of volume was carried out three times for each case, and the estimated lesion volumes were expressed as the mean \pm SD.

One month after her first visit, the frequency of spitting blood decreased and she became able to swallow pieces of bread. She said felt better than before and less depressed or tired. Three months later, she was no longer spitting blood and could swallow almost every type of food and sing karaoke. Eight months later, MR imaging revealed a mild shrinkage and a decrease in signal intensity on T2-weighted images of the

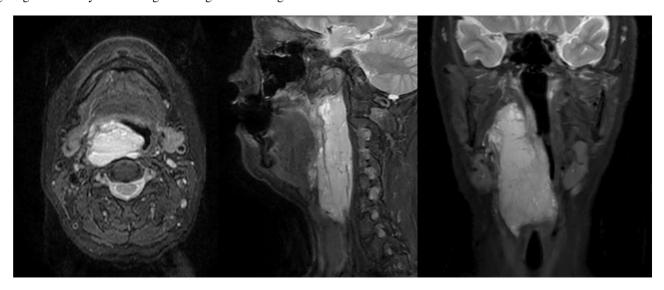


Fig. 1. Fat-suppressed T2-weigted MR images demonstrate a longitudinal lobulated and septated mass with high signal intensity on the posterior pharyngeal wall. The calculated volume of the mass was $85.5 \pm 2.7 \text{ cm}^3$.

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