Case Report

Treatment of a Large Pharyngeal Venous Malformation using Transoral Sclerotherapy

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Received 27 May 2017, accepted 17 August 2017
J-STAGE advance publication 27 November 2017

EDITED BY HIROHITO UMENO

Summary: Management of venous malformation (VM) in the pharynx is challenging because of difficulties in needle access and the presence of nerves as well as other critical anatomical structures around the lesion. We describe the successful treatment of a large pharyngeal VM using transoral sclerotherapy. The patient, a 21-year-old male, visited our hospital for sclerotherapy treatment consultation after complaining of difficulty in swallowing because of a pharyngeal VM. Transoral sclerotherapy was successful, and the symptoms showed complete alleviation. Using a flat detector-equipped angiographic C-arm CT (CACT) enabled the precise assessment of sclerosant placement, facilitating safer sclerotherapy in the pharynx.

Key words venous malformation, pharynx, sclerotherapy, C-arm CT, digital subtraction angiography

INTRODUCTION

Venous malformations (VMs) are the most frequent type of vascular malformation [1]. They typically appear as a bluish discoloration of the skin, lips, or inside of the mouth and tend to increase in size with actions such as crying, pushing, or other maneuvers that increase pressure in the venous system. A VM is a collection of abnormally formed veins. Unlike normal veins that have well developed muscle in the wall, a venous malformation has a very poorly formed wall with less muscle than normal [2,3]. The weakness of the wall of the veins is believed to cause them to gradually stretch or enlarge over time. The lesions tend to gradually increase in size, and spontaneous involution is rare; thus, functional or cosmetic problems are indications for therapy [1,3,4]. Pharyngeal VMs often lead to functional or obstructive problems [1,4]. Percutaneous sclerotherapy has become one of the most common less invasive therapeutic options for VMs. However, management of pharyngeal VMs still remains challenging. Here, we describe our experience in successfully treating a large pharyngeal VM with transoral injection sclerotherapy in a patient who presented with severe obstructive symptoms. Sclerotherapy using CACT facilitated safe and precise injection of the sclerosant.

CASE REPORT

A 21-year-old male complaining of difficulty in swallowing and nasal voice visited a hospital and was suspected of having a large pharyngeal VM. At the age of 10 years he had been diagnosed with a pharyngeal tumor after a health checkup. Various tests including MRI were performed, and the patient was diagnosed with a pharyngeal hemangioma; however, no intervention was performed because he had no symptoms...
at that time. Because the lesion gradually increased in size, the symptoms worsened and he developed a dull feeling in the throat and a slight dyspnea especially when lying the right lateral position at the age of 15 years. Potential surgical interventions were planned at several hospitals, but because of the anticipated difficulty in resection, his progress was only monitored for 7 years and no surgery was performed. During this observation period, the lesion continued to grow, causing increasing difficulty in swallowing and eventually in breathing. At this point, i.e., during the year 2012, he visited our hospital for sclerotherapy treatment consultation.

Upon admission, he could not sleep in the supine position owing to his breathing difficulties, and slept in the right lateral decubitus position instead. MRI revealed that the pharyngeal VM was primarily located in the right posterolateral wall, obstructing the pharyngeal airway in the supine and left lateral decubitus positions. Comparison of MR images between 2006 and 2012 revealed enlargement of the pharyngeal VM from $21 \times 10 \times 43$ mm to $33 \times 19 \times 60$ mm (Fig. 1).

**Treatment**

A dark red, elevated lesion was observed on the oropharynx. We planned to perform transoral sclerotherapy owing to the safety of this method. Prior to intralesional sclerosant injection, tracheotomy was performed under local anesthetic by a head and neck surgeon. General anesthetic was administered via ventilation from the tracheal stoma. Immediately prior to injection, 3% sodium tetradecyl sulfate (STS) was mixed with air and ioxaglate (270 mg/ml iodine, Visipaque; GE Health Care) in a 2:1:1 ratio and was agitated by repeated transfer between two syringes to produce foam. Transoral injection of the foam STS mixture was performed under digital subtraction angiography (DSA) guidance using a 23-gauge needle. After confirming the absence of STS mixture spillover by DSA during injection, the dose of foam STS mixture (calculated from the VM volume) was injected. We performed rotational DSA and used the 3D images obtained from this method to confirm sufficient injection of the foam STS mixture (Fig. 2). Finally, we used contrast media to confirm that the sclerosant remained in the venous lake as intended and that no extravasation occurred.

After 3 days of treatment, the patient was safely extubated. Swallowing and breathing difficulties disappeared. Three months after the procedure, MRI showed that the large mass in the pharynx had decreased significantly in size (Fig. 3). To date, no complications have arisen following treatment.

**DISCUSSION**

Although VM management in the pharynx involves several difficulties, percutaneous sclerotherapy is an effective and noninvasive treatment for VM. In particular, superficial musculoskeletal and soft tissue lesions can be easily treated by injection sclerotherapy owing to easy needle access to the lesion. However, it is challenging to treat VM located in deep lesions because of difficulties in needle access as well as the presence of nerves and other critical anatomical structures around the lesion. Hence, because pharyngeal venous malformations are located in deep lesions, their treatment is difficult.

In the present case, the lesion was visible from the

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**Fig. 1.** STIR images show hyperintense and monolocular appearance of the VM because of an abnormal venous lake. Presence of a large mass in the pharynx causes difficulties in swallowing and a nasal voice during initial diagnosis.
mouth; thus, we chose the transoral puncture technique for sclerosant injection. Transoral direct puncture sclerotherapy may be advantageous over an endoscopic approach because bleeding from the puncture site is reportedly the most frequent complication. Recognition and compression of bleeding from the puncture site is easier in the transoral approach than in the endoscopic approach.

Laser therapy is another less invasive therapy for pharyngeal or laryngeal VMs. Kishimoto et al. reported the cases of seven adults with pharyngolaryngeal VMs who were effectively treated using endoscopic potassium titanyl phosphate laser [5]. However, they also described that only surface photocoagulation could be achieved for bulky VMs. Our patient’s VM was considered to be too large to be treated using laser therapy.

Surgical excision was the original treatment of choice and may still offer the best curative option if it can be performed safely [1]. However, surgical intervention to remove large pharyngeal VMs may be difficult and invasive, and resection by a wide neck incision may induce abnormal functioning of the pharynx. Endoscopic piecemeal removal is also dangerous and
is nearly impossible because bleeding from the VM impairs observation of the lesion.

Our patient’s VM was large, and the airway obstruction following sclerotherapy was assumed to be inevitable and likely fatal; hence, a tracheotomy was performed prior to sclerotherapy to avoid such complications. This was beneficial for ventilation during general anesthesia and facilitated an uninterrupted field of vision.

The structures around the pharynx are complicated and comprise multiple components of the nervous and muscular systems. Although pure ethanol is a very effective sclerosant used to treat VM, the potential for nerve injury in ethanol sclerotherapy can be a significant problem [6,7]. We discussed the risks of ethanol sclerotherapy for pharyngeal VM and concluded that this was not the preferred method for initial intervention in our patient. Compared with pure ethanol, polidocanol and STS are less effective in sclerosing VM; however, the effectiveness of this method has increased considerably since the development of the foam method [8]. Thus, we performed sclerotherapy employing the foam method for this patient.

Precise placement is essential for sclerotherapy for pharyngeal VMs because parapharyngeal structures present the risk of sclerosant spillover, even when using STS. Although the use of CACT is unique, this method is preferred because it facilitates more accurate assessment of foam placement and prevents spillover into the systemic venous circulation. Reconstructed images gained by CACT clearly revealed that the sclerosant volume was sufficient for VM; in addition, the images enabled us to determine the point at which no more sclerosant was required. Therefore, this system facilitates easier and more precise assessment of foam placement than that provided by fluoroscopic or endoscopic approaches. We believe that the CACT allows safer and more effective sclerotherapy for VMs in the pharyngeal area or in other high-risk regions. In addition, the C-arm DSA system enables us to determine the endpoint of the sclerotherapy.

In conclusion, we reported here a case of a large pharyngeal VM successfully treated using transoral sclerotherapy. Despite the challenges in managing pharyngeal VMs, we were able to assess the precise placement of the sclerosant and determine the endpoint of the sclerotherapy by using CACT.

**COI/CONSENT/ETHICS STATEMENT:** All authors report no conflict of interest.

**STATEMENT OF INFORMED CONSENT:** Written informed consent was obtained from individual participant included in this study.

**ETHICAL APPROVAL:** All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. For this type of study, formal consent is not required.

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