

Superselective embolization and transoral ultrasonic surgery of laryngeal hemangioma: a case report

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ABSTRACT

Adult laryngeal hemangioma (LH) is a rare and slowly progressing vascular tumor. The optimal treatment is not yet well-established but can include conservative approaches, especially for asymptomatic cases, or surgical intervention. We report a rare case of an adult LH located in the supraglottis, causing hoarseness, throat foreign body sensation, and mild dyspnea. The large mass was initially embolized by injecting Spongostan and then resected using a transoral approach with an ultrasonic scalpel. After the excision, the patient quickly improved the voice function, and video laryngoscopy revealed reduced arytenoid mucosal swelling with a good movement of the arytenoid cartilages. No recurrence was observed 12 months after resection.

After excision of LH, a close monitoring of the patient is essential because secondary hemorrhage may occur. Regular follow-up controls are needed to monitor the treatment response and to detect any recurrences. Surgical reduction of large LH, achievable by embolization and complete surgical resection with an ultrasonic scalpel, may represent an optimal surgical approach.

Keywords: Embolization, laryngeal hemangioma, transoral ultrasonic surgery, video laryngoscopy

Introduction

Laryngeal hemangiomas (LHs) are relatively rare benign tumors of the larynx, representing about 60% of all hemangiomas that affect the head and neck (1). LH is a slowly progressing vascular tumor that usually occurs in children, although rarer cases have also been identified in adults. Owing to its rarity, it has not been possible to obtain the exact incidence of the disease. Histological subtypes of hemangiomas vary between age groups. Younger age group has more predilections for the capillary type, whereas a cavernous appearance is more common in older age group (2). On the contrary, involution in adults is rare (3). Some of the more commonly reported symptoms include hoarseness, dyspnea, dysphagia, or a pharyngeal foreign body sensation.

There is no consensus on the preferred mode of treatment for hemangiomas owing to insufficient scientific evidence. Observation is usually indicated for small LHs, whereas the larger lesions often require treatment. Several different treatment strategies have been described for the management of LH.

Corticosteroid injections and surgical resection are the most commonly used approaches. Sclerotherapy is an effective and well-tolerated procedure that has been growing in popularity for vascular malformations (4).

In this paper, we present the treatment approach for the management of an adult sizeable LH initially treated by embolization with Spongostan particles (Johnson & Johnson, New Brunswick, NJ, USA) and finally by surgical resection with Ultracision harmonic scalpel (Ethicon, Cincinnati, OH, USA). The applicability of these devices for the treatment of LHs has not been previously advocated in the literature and is the subject of our report.

Case Presentation

A 65-year-old female presented a recurrent pharyngeal foreign body sensation, hoarseness, and mild dyspnea. The patient is a cigarette smoker with a >40 pack-year history. Nasal fiberoendoscopy showed a large mass with a smooth surface and a wide pedicle, with the involvement of the retrocricoid area, ar-

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hyoid complex, and the left pharyngoepiglottic fold. This lesion had a pedunculated configuration in the left aryepiglottic fold that is mobilized during breathing and swallowing, causing a valvular action over the glottic plane resulting in intermittent asphyxiation (Video).

Following hospitalization, an oral prednisone treatment was started at a dose of 20 mg twice a day for 7 days with a slow tapering schedule, and a substantial clinical improvement was achieved. The patient underwent a complete otolaryngological evaluation, gadolinium-enhanced magnetic resonance imaging (MRI), and computed tomography (CT) scan. The MRI revealed a lesion (diameters of 24×23×15 mm), hyperintense on T2, and high restriction of the diffusion-weighted imaging signal, with high and not uniform contrast enhancement (Figure 1). A CT with iodinated contrast (90 mL Iomeron 350) showed a homogenous mass with a sovrafluid-density component on the retrocricoid area and left aryepiglottic fold (Figure 2). The mass caused a 50% reduction in glottic space, without compromising the movement of the left vocal cord during the phonation phase. Both MRI and CT scans were highly suggestive of a hemangioma. Her case was discussed in a multidisciplinary board meeting, and it was decided that she would undergo an embolization with Spongostan followed by a surgical transoral resection of the mass.

The patient was evaluated with selective arteriography through femoral access and was found to have a diffuse vascular lesion. The left superior thyroidal artery was tortuous and dilated. This anomalous venous plexus was located near the site of laryngeal mass, and a limited embolization procedure was undertaken to occlude this vessel (Figure 3). Superselective embolization of the left superior thyroidal artery was performed with microcatheter and Spongostan somministration. The vascular lesion was successfully obliterated without complication.

To prevent airway complications during the surgery, we decided to schedule the patient for preoperative tracheostomy under local anesthesia to secure the patient's airway. The tracheostomy procedure was performed by an experienced ear, nose, and throat surgeon under local anesthesia with 10 mL of lidocaine, and a tracheostomy cannula (8 mm) was placed into the tracheal lumen.

Main Points:

- Laryngeal hemangiomas (LHs) are rare and slowly progressing vascular tumors that are more common in children than in adults.
- LHs are diagnosed primarily on the basis of the patient's history and clinical signs, such as hoarseness, dyspnea, dysphagia, or a pharyngeal foreign body sensation, in conjunction with laryngological examination.
- The management of an adult LH differs from that of pediatric disease. In the adult population, a surgical approach is often required. In children, it usually resolves spontaneously during the first two years of life, and surgery is rarely necessary.
- A surgical approach for a large LH may be feasible by preoperative embolization and transoral ultrasonic resection to provide an almost bloodless surgical field.

Direct laryngoscopy was performed immediately under general anesthesia. The Kleinsasser laryngoscope (KARL STORZ, Tuttlingen, Germany) was introduced transorally, and the larynx was suspended to ensure adequate exposure of the area. The vision was achieved with the use of a surgical microscope (OPMI Sensera, Carl Zeiss, Oberkochen, Germany). The mass was excised using Ultracision harmonic scalpel under microscopy. The powered instrument used was Harmonic ACE shears (Ethicon Endo-Surgery, Cincinnati, OH, USA), with a 23-cm long shaft and 5-mm-diameter shaft, setting the cutting power at 5 W and the hemostasis power at 3 W. The dissection of the lesion was relatively simple, and bleeding was minimal and easy to control by cauterization. A nasogastric tube was applied for feeding.

Postoperative pathological examination of surgical specimen confirmed the diagnosis of cavernous hemangioma. Histolog-

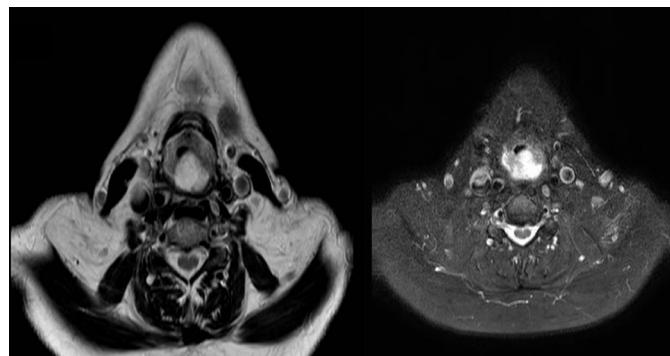


Figure 1. Gadolinium-enhanced magnetic resonance imaging of supraglottic hemangioma.

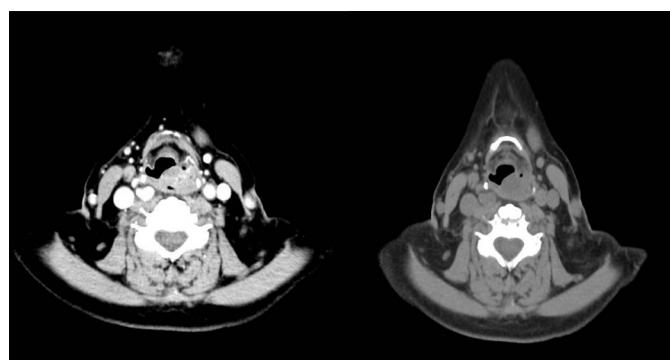


Figure 2. CT imaging of the supraglottic hemangioma. CT: computed tomography.



Figure 3. Selective arteriography of the left superior thyroidal artery.

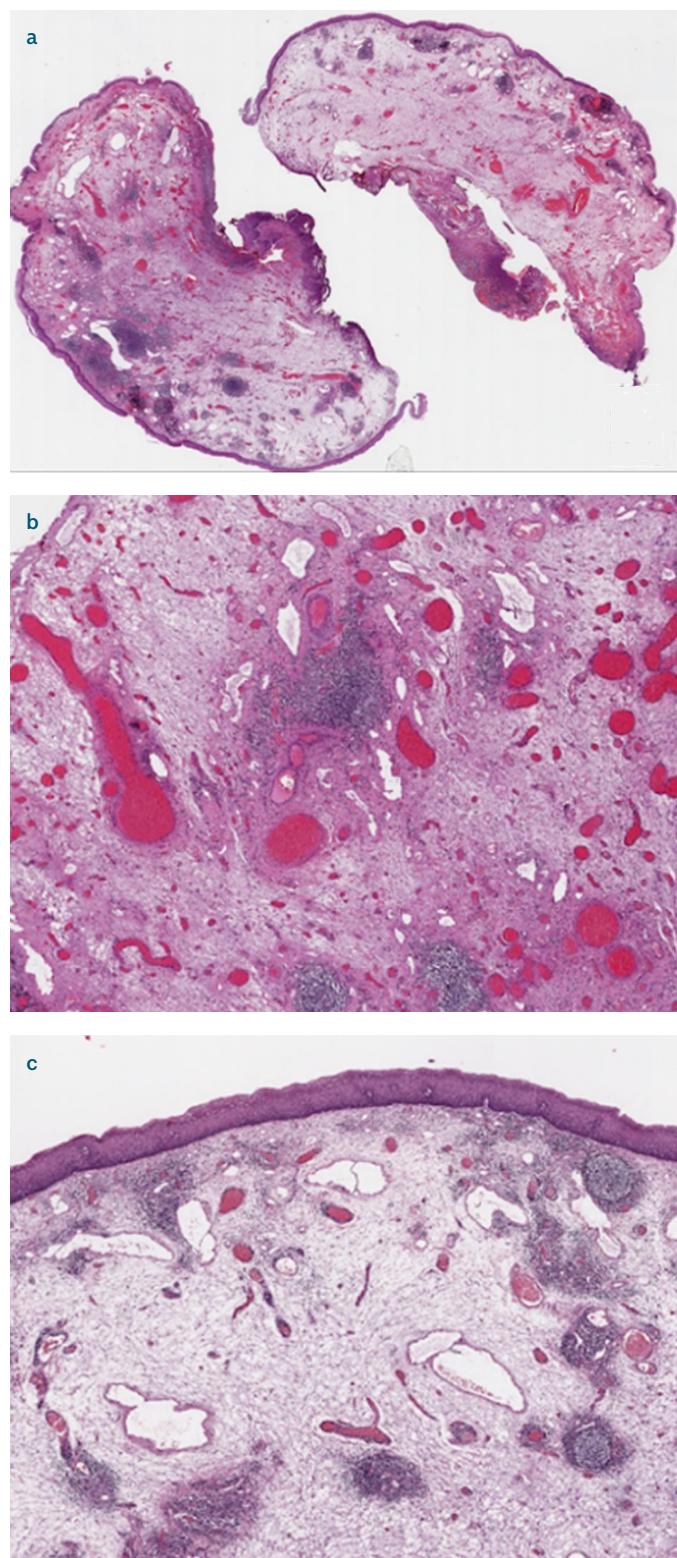


Figure 4. a-c. Histological examination of cavernous hemangioma. (a) At low magnification ($\times 2.5$), the tumor consisted of a proliferation of vascular structures embedded in an edematous stroma. (b, c) At higher magnification, the vessels are variable in size, are lined by endothelial cells, and have walls of variable thickness.

ically, at low magnification, the tumor consisted of a proliferation of vascular structures embedded in an edematous stroma. At higher magnification, the vessels were variable in size, were lined by endothelial cells, and had walls of variable thickness.

Smooth muscle bundles interposed between the edematous matrix were also present (Figure 4). Necrosis, mitoses, and nuclear pleomorphism were absent.

Immunohistochemically, the endothelial cells were positive for ERG, CD31, and CD34. On the basis of both morphological and immunohistochemical features, the diagnosis of cavernous hemangioma of the vocal cord was made.

Videofiberendoscopy on postoperative day 5 showed excellent glottic space patency, decreased mucosal edema, and a good movement of the arytenoid complex. The tracheostomy and nasogastric tubes were removed, and 15 days after admission, the patient was discharged in good overall condition.

One month after surgery, the patient showed complete resolution of the vascular mass with the normal movement of the arytenoid complex. Only mild alteration of voice quality has been reported. A written informed consent was obtained from the patient who participated in this case.

Discussion

In this case report, the treatment of an LH in a 65-year-old female performed by embolization with Spongostan and resection with Ultracision harmonic scalpel is discussed.

MacKenzie was first to describe LH (5). Later, Sweetser differentiated the characteristics between the infantile and adult hemangiomas (6). Adult hemangiomas are rare, and the most common type is the cavernous form (7). It is essential for the disease course and therapeutic procedure to be differentiated between the various types of angiomas. LHs are classified as capillary (plane, tuberonodal, and planotuberous) and cavernous (submucous, intramural, transmural, and extramural) on the basis of their morphology.

The etiologic factors are not well understood, but cigarette smoking, vocal abuse, and laryngeal trauma have been associated with LH (8). The main complaint of our case was dyspnea and hoarseness; moreover, she was a cigarette smoker without any history of trauma or vocal abuse or intubation. LHs are diagnosed primarily on the basis of the patient's history and laryngological examination.

In previous studies, CT and MRI scans have been found to be highly capable of defining the form, size, and surrounding anatomical structures of hemangiomas. Besides, a study by Chetty et al. suggests that contrast CT imaging is a useful method to noninvasively detect subglottic hemangiomas, providing a more accurate determination of airway narrowing (9). Similarly, CT could be used to differentiate between subglottic lesions, vascular rings, and other neck and upper mediastinum extrinsic lesions. This is followed by MRI that can be especially useful for further characterization and to evaluate the extent of and structural involvement. An example of the usefulness of MRI techniques is given in the study by Gelbert et al. where the MRI scans are the best adjunctive indications for capillary-venous hemangiomas (10).

Conditions to consider in the differential diagnosis of LH include the following: branchial cleft cysts, dermoid cysts, and laryngeal webs. Squamous cell carcinoma, usually supraglottic,

may be associated with laryngeal cysts. In this context, a histopathological analysis of the hemangiomas resected could aid in a definitive diagnosis (11).

The treatment of upper airway hemangiomas has included steroid injection, laser ablation (12), cryosurgery, radiotherapy, and surgical resection (13). For large lesions, even a tracheostomy may be required to secure the patient's airway. However, the ideal surgical method of this condition remains controversial because very few reports have been published on this subject. Sometimes, a preoperative selective embolization for bleeding hemangiomas is required, as in our case, to perform an accurate surgical resection.

One of the first reports on the use of the ultrasonic scalpel for the complete resection of a hemangioma is given by Wang et al. who presented a case of LH in a 61-year-old male successfully treated with preoperative artery ligation and surgical resection.

Similarly, in another case report, a 73-year-old man with a large pharyngolaryngeal hemangioma was treated by embolization with an ethylene-vinyl alcohol copolymer (Onyx; ev3, Irvine, California, USA) and resection with an ultrasonic scalpel system, which has proven an effective and feasible therapeutic option in patients with large LH and vital risk (15). These results suggest that the use of an ultrasonic scalpel represents a safe, effective, and less invasive method for the treatment of large LHs.

Our findings in drawing up this report demonstrated that the embolization with Spongostan and a complete resection with the ultrasonic scalpel system are valuable and safe surgical tools for the management of adult LH that has proved resistant or not applicable to conservative treatments.

Video. Visualization of the glottic plane by using fiberoptic endoscope device.

Informed Consent: Written informed consent was obtained from the patients who agreed to take part in the study.

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