Surgical resection of a large hypopharyngeal hemangioma in an adult using neodymium-doped yttrium aluminum garnet laser: A case report

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Abstract

BACKGROUND
Generally, hemangiomas do not require surgical intervention; however, cases of large hemangiomas, potentially involving the throat and trachea, necessitate surgical therapy. Here, we present a case of hypopharyngeal hemangioma in an adult that was successfully treated with neodymium-doped yttrium aluminum garnet (Nd-YAG) laser.

CASE SUMMARY
Laryngoscopic examination of a 61-year-old man demonstrated the presence of a large, submucosal vascular lesion that extended into the epiglottis, left arytenoid cartilage, lateral to the aryepiglottic fold, and pyriform sinus. The lesion was resected and photocoagulated with limited hemorrhage using Nd: YAG laser. The hypopharyngeal hemangioma was completely excised. The patient showed no recurrence of hypopharyngeal hemangioma during the 1.5-year follow-up period.

CONCLUSION
Laser therapy is one of the effective tools for treating hemangiomas with rapid, uncontrolled growth or in functional areas, with few side effects and complications. The present case of a male patient with a large hypopharyngeal hemangioma, treated with YAG laser, demonstrates the efficacy of laser photocoagulation in treating cases of hemangiomas, without the risk of bleeding or airway obstruction. The favorable postoperative outcomes demonstrated by our patient with Nd: YAG laser therapy indicate its consideration in the therapy of similar cases.

Key words: Yttrium aluminum garnet laser; Large; Adult; Surgical resection; Hypopharyngeal; Hemangioma; Case report
INTRODUCTION

Clinically, hemangiomas are divided into pediatric and adult types, and can occur at any age. In adults, hemangiomas usually occur in the larynx, and rarely in the hypopharynx. Laryngeal hemangiomas are slowly progressing vascular tumors, which are commonly diagnosed in children, but rarely, in adults[1].

The terms capillary hemangioma and venous hemangioma were commonly used to describe various types of hemangioma in the past; however, their usage is not precise. Many diagnosed cases of venous aneurysms are actually vascular malformations rather than true hemangioma[1]. Therefore, an important definition is that hemangiomas are true vascular tumors with distinct clinical and biological characteristics, derived from vascular malformations.

Hemangioma can be found all over the body, including the liver[5], gastrointestinal tract[6], central nervous system[7], pancreas[8], gall bladder[9], thymus[10], spleen[11], lymph nodes[12], lung[13], urinary bladder[14] and adrenal glands[15]. In adults, hemangiomas rarely occur in the hypopharynx. Because of the rarity of cases of hypopharyngeal hemangiomas, the exact incidence of the tumor remains unknown. The diagnosis of hemangioma is usually based on clinical history and presentation. In addition, computed tomography and magnetic resonance imaging have both been used in clinical practice. Magnetic resonance imaging and magnetic resonance angiography represent the best diagnostic tools to demonstrate, respectively, soft-tissue components and relationships with nearby structures[14,15]. Time-resolved imaging of contrast kinetics is a recently introduced technique that can obtain dynamic images during the arterial, capillary, and venous phases. It improves temporal resolution with respect to contrast-enhanced magnetic resonance angiography technique[10,11]. It is possible to acquire three dimensional volumes during the passage of gadolinium, to detect the vascularization of the region of interest over time, similar to digital subtraction angiography. Digital subtraction angiography is an invasive, relatively expensive, time-consuming technique, with risk of vascular injury and exposure to ionization[10].

Currently, there is no established consensus for treating hemangiomas because of their rarity. However, large-sized hemangiomas require therapeutic intervention, including surgical resection, corticosteroid injection, ethanol injection, cryosurgery, radiation or gold implants, and interferon treatment[10].

We present the case of a hypopharyngeal hemangioma (12 mm × 15 mm) in an adult that was successfully treated with neodymium-doped yttrium aluminum garnet (Nd-YAG) laser (wavelength, 1064 nm).

CASE PRESENTATION

Chief complaints

On May 2, 2017, a 61-year-old male patient presented to the Jinshan Hospital of Fudan University (Shanghai, China). The patient had no hoarseness of voice, hemoptysis,
expectoration, or dyspnea. Due to upset stomach, he underwent a gastroscopy, which revealed a hypopharyngeal tumor.

**History of past illness**

The patient had no history of cigarette smoking. He has suffered from hypertension for more than 20 years.

**Physical examination**

When the patient presented to our hospital, his temperature was 36.6 °C, heart rate was 80 bpm, respiratory rate was 18 breaths/min, blood pressure was 150/90 mmHg, and oxygen saturation in room air was 98%.

**Imaging examinations**

Laryngoscopic examination performed on April 25, 2017, outside our hospital, demonstrated the presence of a submucosal vascular lesion that extended into the epiglottis, left arytenoid cartilage, lateral to aryepiglottic fold, and pyriform sinus. The blue-black-colored mass showed a grape-cluster-like appearance (Figure 1). Computed tomography was performed to determine the depth and extent of invasion of the lesion (12 mm × 15 mm), and to plan for the therapeutic strategies to protect and preserve important anatomical structures (Figure 2).

**FINAL DIAGNOSIS**

The patient was diagnosed with a hypopharyngeal hemangioma.

**TREATMENT**

Ensuring limited hemorrhage, the lesion was resected and photocoagulated with Nd: YAG laser on May 4, 2017. The Nd: YAG laser (wavelength of 1064 nm, HGL-MY100C, Huaguang, Wuhan, China) was used in the continuous mode at 7 W. Photocoagulation was performed in a sweeping fashion until visible blanching and involution were observed. The hypopharyngeal hemangioma was completely excised. Moreover, the lesion was resected and sent for histopathological examination. Pathological examination confirmed the tumor to be a cavernous hemangioma (Figure 3). CD31 immunostaining was observed on the walls of the blood vessels (Figure 3E and F), whereas, the smooth muscle of some vessels showed positive immunostaining for desmin (Figure 3G and H). Histological examination showed the presence of vascular hyperplasia and hemangiectasis beneath the squamous mucosa.

Postoperatively, the patient showed no signs of airway obstruction or bleeding. The patient resumed regular diet from the subsequent day. A video laryngoscopy performed on May 8, 2017 showed reduction of the mucosal edema and good movement of the arytenoid was observed (Figure 4A). Postoperatively, anti-inflammatory medication and symptomatic treatment were administered. The patient was discharged 6 d after surgery.

**OUTCOME AND FOLLOW-UP**

Forty-seven days after the surgery, an additional video laryngoscopy was performed on June 20, 2017, which showed excellent repair of the arytenoid cartilage mucosal incision (Figure 4B). Furthermore, there was no recurrence of the hypopharyngeal hemangioma at the 1.5-year postoperative follow-up (Figure 5).

The patient did not experience any adverse events from the use of the laser. Excessive bleeding and airway problems were not encountered, and the postoperative pain was limited. The use of Nd: YAG laser provided excellent long-term outcomes and complete symptom resolution without any hemorrhage, and ensured complete healing of the hypopharyngeal areas without any stenosis or scarring.

**DISCUSSION**

For cases of hemangiomas, a pathological examination is usually required to make a definitive diagnosis. Currently, depending on the vessel size and vessel wall thickness, hemangiomas can be classified into three histopathological types: Capillary, cavernous, and mixed. The most common type is cavernous, which shows an
A submucosal vascular lesion extending into the epiglottis, left arytenoid cartilage, lateral to the aryepiglottic fold, and pyriform sinus was visualized. Expanded blood vessel with thin walls\cite{19}. Large hemangiomas require treatment to prevent the development of ulcers, necrosis, or infection of vital organs.

The true pathogenetic mechanism of vascular abnormalities in hemangiomas remains unclear. Various treatment methods have been reported, and the choice of optimal treatment method still remains controversial\cite{20}. The treatment of hemangiomas depends on the primary site and growth phase of the tumor. Barring a few exceptions, surgery should not be the first choice of treatment. It is worth noting that although different laser systems are effective for treating superficial or deep hemangioma, systemic treatment with steroid or β-blockers (still in clinical trials) should assume priority as the first-line treatment. For cases involving the throat or trachea, early treatment is usually required to prevent the occurrence of airway obstruction\cite{20}.

Since our findings were obtained from a single case, additional multicenter, randomized controlled trials are needed to confirm the use of YAG laser resection as an effective treatment for hypopharyngeal hemangiomas.

**CONCLUSION**

Laser therapy is one of the effective tools for treating hemangiomas with rapid, uncontrolled growth or in functional areas, with few side effects and complications. The present case of a male patient with a large hypopharyngeal hemangioma, treated with YAG laser, demonstrates the efficacy of laser photocoagulation in treating cases of hemangiomas, without the risk of bleeding or airway obstruction. The favorable postoperative outcomes demonstrated by our patient with Nd: YAG laser therapy indicate its consideration in the therapy of similar cases. For cases similar to ours, a complete surgical resection with a YAG laser is recommended.
Figure 2  Computed tomography showing a vascular tumor (arrow) in the hypopharynx.

Figure 3  Histopathological examination of the tumor showed a cavernous hemangioma. Vascular hyperplasia and hemangiectasis were observed beneath the squamous mucosal layer. A and C: Hematoxylin and eosin staining (40 ×); B and D: Hematoxylin and eosin staining (100 ×); E: Immunohistochemical staining for CD31 (40 ×); F: Immunohistochemical staining for CD31 (100 ×); G: Immunohistochemical staining for Desmin (40 ×); H: Immunohistochemical staining for Desmin (100 ×).
Figure 4 Postoperative laryngoscopy of the patient. A: Laryngoscopic examination demonstrating reduced mucosal edema and absence of bleeding; B: Laryngoscopic examination showing excellent repair of the arytenoid cartilage mucosal incision.

Figure 5 Laryngoscopy performed on November 14, 2018 showed no recurrence of the hypopharyngeal hemangioma.

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REFERENCES

1. Martins RH, Lima Neto AC, Semenzate G, Lapate R. Laryngeal hemangioma. Braz J Otorhinolaryngol 2006; 72: 574 [PMID: 17143442 DOI: 10.1016/S1808-8694(15)31009-9]
2. Zheng JW, Zhou GY, Wang YA, Zhang ZY. Management of head and neck hemangiomas in China. Chin Med J (Engl) 2008; 121: 1037-1042 [PMID: 18706256 DOI: 10.1097/00029330-200806010-00014]
3. Yoon SS, Charny CK, Fong Y, Jarnagin WR, Schwartz LH, DeMatteo RP. Diagnosis, management, and outcomes of 115 patients with hepatic hemangioma. J Am Coll Surg 2003; 197: 392-402 [PMID: 12946794 DOI: 10.1016/S1072-7515(03)00420-4]
4. Lin J, Bigge J, Ulbright TM, Montgomery E. Anastomosing hemangioma of the liver and gastrointestinal tract: an unusual variant histologically mimicking angiosarcoma. Am J Surg Pathol 2013; 37: 1761-1765 [PMID: 23887160 DOI: 10.1097/PAS.0b013e31829677ec]
5. Abe M, Misago N, Tanaka S, Masuo K, Tabuchi K. Capillary hemangioma of the central nervous system: a comparative study with lobular capillary hemangioma of the skin. Acta Neuropathol 2005; 109: 151-158 [PMID: 15365728 DOI: 10.1007/s00401-004-0921-4]
6. Mundinger GS, Gust S, Micchelli ST, Fishman EK, Hruban RH, Wolfgang CL. Adult pancreatic hemangioma: case report and literature review. Gastroenterol Res Pract 2009; 2009: 839730 [PMID: 19421421 DOI: 10.1155/2009/839730]
7. Crucitti A, La Greca A, Antinori A, Antonacci V, Magistrelli P. Cavernous hemangioma of the gallbladder. Case report and review of the literature. Tumori 2005; 91: 432-435 [PMID: 16459643 DOI: 10.1177/030089160509100511]
8. Papagiannopoulos K, Sheppard MN, Goldstraw P. Thymic hemangioma presenting with recurrent pleural effusion. Ann Thorac Surg 2000; 70: 297-299 [PMID: 10921733 DOI: 10.1016/S0003-4975(00)01273-X]
9. Wilcox TM, Speer RW, Schlinitert RT, Sarr MG. Hemangioma of the spleen: presentation, diagnosis, and management. J Gastrointest Surg 2000; 4: 611-613 [PMID: 11307096 DOI: 10.1016/S1091-255X(00)00110-9]
10. Terada T. Capillary cavernous hemangioma of the lymph node. Int J Clin Exp Pathol 2013; 6: 1200-1201 [PMID: 23686845]
Jin M et al. Hypopharyngeal hemangioma resection using YAG laser

11 Zhao J, Shao J, Zhu L, Yu K, Zhao R, Ding W, Zhang J, Han Y. Solitary pulmonary capillary hemangioma: Clinicopathologic and radiologic characteristics of nine surgically resected cases. Pathol Res Pract 2018; 214: 1885-1891 [PMID: 30268596 DOI: 10.1016/j.prp.2018.09.016]

12 Ikeda T, Shinamoto K, Tanji N, Ohoka H, Nishio S, Yokoyama M, Ikeda M. Cavernous hemangioma of the urinary bladder in an 8-year-old child. Int J Urol 2004; 11: 429-431 [PMID: 15157217 DOI: 10.1111.j.1442-2042.2004.00809.x]

13 Xu HX, Liu GJ. Huge cavernous hemangioma of the adrenal gland: sonographic, computed tomographic, and magnetic resonance imaging findings. J Ultrasound Med 2003; 22: 523-526 [PMID: 12751864 DOI: 10.7863/jum.2003.22.5.523]

14 Josephson CB, White PM, Krishan A, Al-Shahi Salman R. Computed tomography angiography or magnetic resonance angiography for detection of intracranial vascular malformations in patients with intracerebral haemorrhage. Cochrane Database Syst Rev 2014; CD009372 [PMID: 25177839 DOI: 10.1002/14651858.CD009372.pub2]

15 Romano A, Tavanti F, Rossi Espagnet MC, Terenzi V, Cassoni A, Sumi G, Boellis A, Pierallini A, Valentini V, Bozza A. The role of time-resolved imaging of contrast kinetics (TRICKS) magnetic resonance angiography (MRA) in the evaluation of head-neck vascular anomalies: a preliminary experience. Dentomaxillofac Radiol 2015; 44: 20140302 [PMID: 25410709 DOI: 10.1016/j.dmir.20140302]

16 Razek AA, Gaballa G, Megahed AS, Elmogy E. Time resolved imaging of contrast kinetics (TRICKS) MR angiography of arteriovenous malformations of head and neck. Eur J Radiol 2013; 82: 1885-1891 [PMID: 23928233 DOI: 10.1016/j.ejrad.2013.07.007]

17 Kahana A, Lucarelli MJ, Grayev AM, Van Buren JJ, Burkat CN, Gentry LR. Noninvasive dynamic magnetic resonance angiography with Time-Resolved Imaging of Contrast Kinetics (TRICKS) in the evaluation of orbital vascular lesions. Arch Ophthalmol 2007; 125: 1635-1642 [PMID: 18071114 DOI: 10.1001/archopht.125.12.1635]

18 Huang CM, Lee KW, Huang CJ. Radiation therapy for life-threatening huge laryngeal hemangioma involving pharynx and parapharyngeal space. Head Neck 2013; 35: E98-101 [PMID: 22431069 DOI: 10.1002/hed.21919]

19 Abrahamson J, Shandling B. Intestinal hemangiomata in childhood and a syndrome for diagnosis: a collective review. J Pediatr Surg 1973; 8: 487-495 [PMID: 4543245 DOI: 10.1016/0022-3468(73)90211-X]

20 Zheng JW, Zhou Q, Yang XJ, Wang YA, Fan XD, Zhou GY, Zhang ZY, Suen JY. Treatment guideline for hemangiommas and vascular malformations of the head and neck. Head Neck 2010; 32: 1088-1098 [PMID: 19924783 DOI: 10.1002/hed.21274]
