



ADULT LARYNGEAL HEMANGIOMA: A RARE CASE REPORT

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ABSTRACT

Introduction: Benign vascular tumors known as hemangiomas are frequently observed in the head and neck area. Hemangiomas are more common in infantile and can regress spontaneously. In adults, laryngeal hemangioma is extremely rare. They may occur in supraglottic and glottic regions that can cause hoarseness, globus sensation, dysphagia, hemoptysis, and shortness of breath. As a result of the uncommon nature of these conditions, there are no clear treatment guidelines. Surgical excision under micro laryngoscopy with laser is still the first choice for laryngeal hemangiomas in adults. **Aim:** Give information on how to diagnosis adult laryngeal hemangioma and give information about the effectiveness and their efficacy using diode laser for the management. **Case Report:** A 51 year old woman appeared with a one-month history of gradually worsening shortness of breath and a five-month history of dysphonia. On fiberoptic laryngoscopy, examination revealed a purple-red mass in color, covering the vocal folds and almost entire of rim glottic. We performed tracheostomy procedures under local anesthesia before excision the hemangioma by microlaryngeal surgery with diode laser under general anesthesia. **Method:** Literature searching through Google Scholar, Pubmed and Science Direct using keywords of “laryngeal hemangioma”, “management of laryngeal hemangioma” and “microlaryngoscopic surgery”. **Result:** Selection based on inclusion and exclusion criteria, found only 1 literature was relevant with the topic. **Conclusion:** Microlaryngoscope surgery with diode laser is a best choice for treatment of adult laryngeal haemangioma because it is the safest and most effective. In cases accompanied wuth shortness of breath that causes by upper airway obstruction, a tracheostomy could be conducted.

Keywords: Laryngeal hemangioma, Microlaryngeal surgery, Diode laser

INTRODUCTION

Hemangiomas are a type of vascular tumor that can frequently be discovered in the head and neck area.¹ In children, 60-70% of hemangiomas arise from birth with an incidence 2.5% in neonates. It has been observed that approximately 4% of children are affected by laryngeal hemangioma. Meanwhile, the incidence of laryngeal hemangiomas in adults still unclear because of it was extremely rare case.²

Rapid proliferation of infantile hemangioma are caused by local hypoxia that triggers angiogenesis and vasculogenesis.³ In adults, the causes of laryngeal hemangiomas are not fully understood.¹ The etiology and predisposition factors for hemangiomas are not known for certain.⁴

Based on age category, laryngeal hemangiomas is classified into 2 types, infantile type and adult type.⁴ Infantile laryngeal hemangioma is located in the subglottic region and is occurs more frequently in females. Infantile hemangiomas are characterized by the presence of stridor and intermittent respiratory distress as the primary symptoms.⁵ Laryngeal hemangiomas in adults are more common in men than women¹ and are commonly found in the supraglottic and glottic

regions.⁶ The clinical symptoms may vary based on their severity and where they are located, the most common symptom in adult laryngeal hemangiomas includes foreign body sensation in the pharynx or larynx, voice changes, dysphagia, blood-tinged sputum, hemoptysis, and dyspnea.⁷

Hemangioma can be identified through the clinical symptoms, but in some patients, hemangiomas can be found incidentally during a routine physical examination.⁸ Physical examination using an endoscopy is almost sufficient to establish a diagnosis of hemangioma.⁴ Radiologic evaluations including CT (Computed Tomography) Scan and MRI (Magnetic Resonance Imaging) provide informations about the location, size, and extent of the hemangioma, as well as its relationship to other anatomic structures.⁷ Angiography can be performed on large hemangiomas and for surgical patients with respiratory symptoms.⁴

The management of hemangiomas in infants and adults is different.¹ Until now there is no clear agreement about how to treat laryngeal hemangioma in adults due to its very rare case. Observations can be made in cases of small or asymptomatic laryngeal hemangiomas. In large or symptomatic hemangiomas, corticosteroids systemic or intralesional, beta-blockers, microlaryngeal surgery with laser, sclerotherapy,

embolization, cryosurgery, and radiotherapy are recommended.⁷

Management of laryngeal hemangioma is very challenging because of the intricate nature of the anatomical structures that are involved, the limited scope of treatment that is currently being offered, and the possibility of experiencing life-threatening complications following surgical intervention, including life-threatening hemorrhage and airway obstruction and a high recurrence rate.⁷ With the development and use of suspension laryngoscopy for microlaryngeal surgery, patients with hemangiomas have had laser therapy under general anesthesia for the treatment of their condition. Laser has two important functions: tissue resection and cautery. Laser therapy also has the following advantages, such as a rapid tumor ablation or cautery, a speedy recovery time, and an effective treatment for the condition.⁹

This report is aimed to know about how to diagnose adult laryngeal hemangioma and the management with diode laser.

CASE REPORT

A woman aged 51 came forward to ORL-HNS outpatient clinic of Dr. M. Djamil Hospital Padang on June 2nd 2022 with a worsening shortness of breath since 3 days before admission. She had a history of shortness of breath since 1 month ago. Shortness of breath unaffected by activity, exposure to dust or cold air. She had hoarseness since 5 months ago and getting worse since a month ago. There was globus sensation since 3 months ago. There were no complaints of coughing with blood, pain or difficulty in swallowing. There is no history of long cough and taking regular medication for 6 months. There was no history of diabetes mellitus, hypertension and heart disease. There was no history of stroke or limb weakness. There was no history of surgery on head and neck region. There was no history of cancer in family. There is no history of weight loss in the last 6 months. The patient is a food vendor.

According to the findings of the physical examination, the patient's general condition was moderately unwell, their compositis was cooperative, and their blood pressure was 132/78 mmHg, heart rate 92x/minute, respiratory rate 26x/minute, SpO₂ 98% and body temperatures was 36,6°C. Physical examination of the neck did not reveal enlarged lymph nodes. On thorax region there was vesicular based sound, there was inspiration stridor, no wheezing or rhonchi. The Voice Handicap Index (VHI) – 30 and Grade-Roughness-Breathiness-Asthenia-Strain (GRBAS) are 98 and (G₁R₁B₁A₁S₀).

Ears and nose examination were normal limits. On fiberoptic laryngeal examination, the arytenoid and epiglottis were not hyperemic, there was a hypervascular mass purple-red in colour covering both of vocal fold and almost the entire of glottic rim. There was no standing secretion on pyriform sinus. (Figure 1).



Figure 1. Fiber Optic Laryngoscopy pre operation, showed a hypervascular mass purple-red in colour (red arrow)

The result of a contrast CT Scan of the larynx in axial section showed an isodens mass with strong homogeneous pascu contrast enhancement on the anterior side of the glottic with well-defined lobulated edges measure 13,1 x 11,5 x 1,2 mm.



Figure 2. Contrast CT scan Isodens mass on the anterior side of glottic (red arrow)

Patient was diagnosed with impending upper airway obstruction caused by mass suspicious laryngeal hemangioma and was prepared to undergo tracheostomy under local anesthesia then microlaryngoscopic surgical excision biopsy under general anesthesia by diode laser on June 9th 2022. Laboratory finding and PA x-ray examination was performed to this patient and the result was within normal limit.

Tracheostomy was conducted under local anesthesia, tube number 7 inserted into the stoma, filled cuff with 10 cc air, air passage was good then fixated. Bleeding is evaluated and treated. General anesthesia was performed and a kleinsasser laryngoscope was inserted into the patient's mouth to reveal the uvula and the posterior pharyngeal wall was traced until the epiglottis was visible. The epiglottis is slightly hooked and insertion is continued until the vocal folds are visible. The kleinsasser laryngoscope is fixed with chest support on the patient's chest. Followed by evaluation with 0° scope which showed the hypervascular mass, purple-red in color with smooth surface. The mass was mobile and the base of mass attached on left vocal fold. A roll gauze moistened with 0.9%

NaCl was placed in the subglottic area. The mass was excised using a diode laser. An incision was made between the normal mucosa and the margin of hemangioma. Excision was carried out until all the mass was removed. An evaluation was carried out using a 0° and 70° scope, intraoperative bleeding was controlled by applying local pressure with an adrenaline-soaked cotton swab.

There was no active bleeding and residual mass, the roll gauze and laryngoscope kleinsasser were deattached, the operation was finished.

The patient was hospitalized in ORL-HNS Department wards and given therapy infusion of Ringer lactate 20 drops per minute, Ceftriaxone injection 2x1gram, Dexamethasone injection 3x5mg and Paracetamol tablets 3x500mg and deflate the tracheostomy cuff 6 hours postoperative.

One day after surgery, June 10th 2022, there was no shortness of breath, no difficulty swallowing, no bleeding from the mouth. There is minimal pain in the surgical area with VAS 4. In the anterior colli region, the tracheostomy tube was in

good position, air passage was good. The medications were continued.

Four day after surgery, June 12th 2022, the patient had no complaints. In the anterior colli region, the tracheostomy tube was in good position, air passage was good. Patient was discharged and given therapy Cefixime tablets 2x200mg orally, Paracetamol tablets 3x500mg orally, Dexamethasone 3x0.5mg orally and N-Acetylcysteine 3x200mg orally.

One week after surgery, June 16th 2022, patient controled to ORL-HNS outpatient clinic Dr. M Djamil Hospital. Padang The patient has no complaints, there was no shortness of breath, no difficulty in swallowing, no blood coming out from the mouth On the anterior colli region, tracheostomy tube was in a good position, air passage was good. The patient brought the histopatologic examination that indicated a capillary cavernous hemangioma (Figure 3). The patient was diagnosed with post tracheostomy and post excision biopsy of laryngeal capillary cavernous hemangioma. We suggest the patient to voice rest and given therapy N-Acetylcysteine 3x200mg orally.

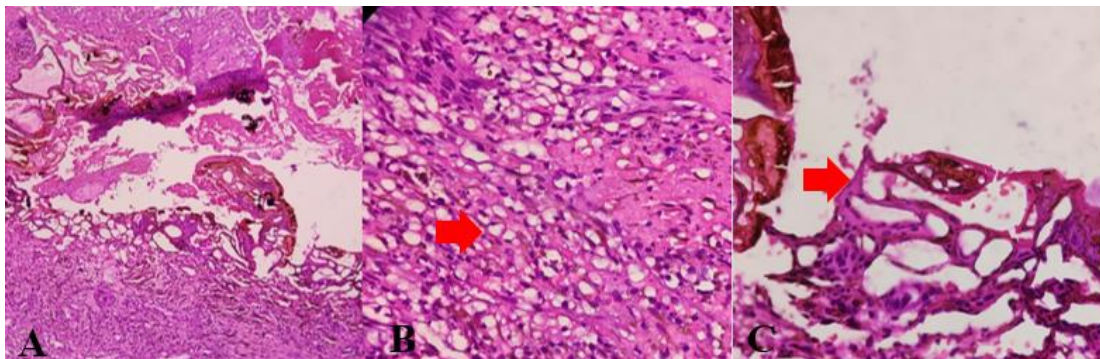


Figure 3. A) Microscopic appearance of cavernous capillary hemangioma, (B) Small spaces of blood vessels covered by endothelium and (C) Cavernous blood vessels covered by endothelium, with a larger cavity size, in which there are erythrocytes

Two weeks after surgery, June 23th 2022. The patient has no complaints. On the anterior colli region, the tracheostomy tube was in a good position, air passage was good. Fiber-optic laryngoscopy showed the vocal fold and ventricular movements were symmetrical, the glottic rim was open, and the surgical wound was healing (Figure 4). We planned for a repeat fibreoptic laryngoscope for the next 2 weeks and given therapy N-Acetylcysteine 3x200mg orally.



Figure 4. Fiberoptic laryngoscope 2 weeks after surgery, the surgical wound was healing

Four weeks after surgery, July 9th 2022, The patient has no complaints. There was improvement of voice, the Voice Handicap Index (VHI) – 30 and Grade-Roughness-

Breathiness-Asthenia-Strain (GRBAS) are 19 and (G₀R₀B₀A₀S₀) and there was no sign of any complication or upper airway symptoms on the anterior colli region, the tracheostomy tube was in good position, air passage was good. A fiberoptic laryngoscopy examination showed the vocal fold and ventricular movements were symmetrical, the glottic rim was open, there were no visible lesions or adhesions from the surgery. No recurrence was found (Figure 5).



Figure 5. Fiberoptic laryngoscope 4 weeks after surgery, there were no visible lesions or adhesions from the surgery

Is the management of adult laryngeal hemangioma with diode laser was satisfied?

- P : Adult patient with laryngeal hemangioma
- I : Microlaryngeal surgery with diode laser
- C : Management adult laryngeal hemangioma with cold steel surgery
- O : Compared to surgical excision, laser surgery is relatively effective and less invasive. Lasers can reduce trauma, improve control of hemostasis, produce superior postoperative functional outcomes, shorten hospital stays, and reduce sociohealth costs.

METHOD

Literature searching was performed through Google Scholar, PubMed and Science Direct using keywords “Laryngeal hemangioma”, “Management of laryngeal hemangioma” and “Microlaryngoscope surgery” (Figure 6).

The search was using inclusion criteria: 1) Adult patient with laryngeal hemangioma, 2) Location on the glottis, 3) Laser; and exclusion criteria: 1) Infantil hemangioma, 2) Supraglottis and subglottis hemangioma, 3) Non-surgical management, 4) More than 10 years

RESULT

The search obtained literatures which were published in the last 10 years, and found only 1 article was relevant with topic.

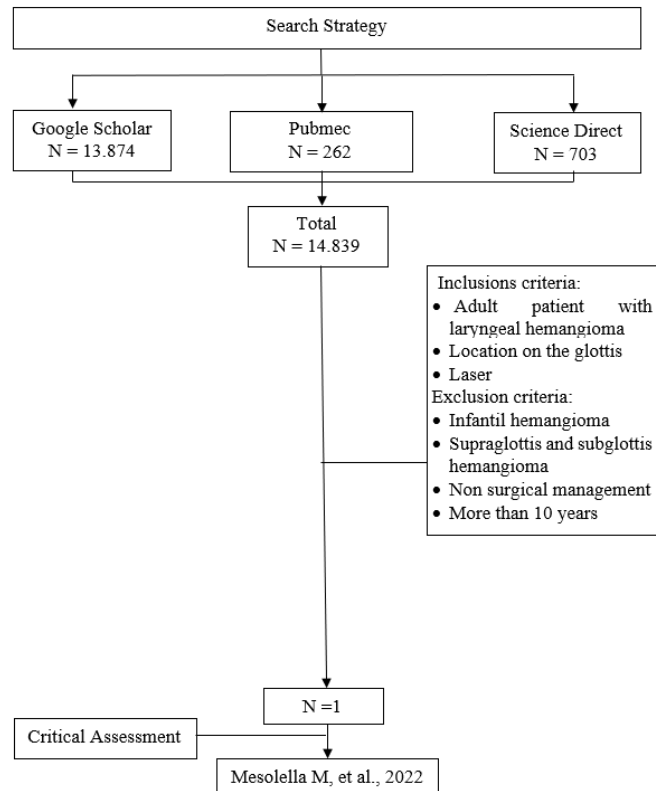


Figure 6. Literature search flow

Table 1. Search strategy

Literature	Result	Keywords
Google Scholar	13.874	'Laryngeal hemangioma'AND 'Management of laryngeal hemangioma AND 'Microlaryngoscopy surgery'
PubMed	262	
Science Direct	703	

Mesolella M et al's reported on their case report that hemangiomas of the vocal cords are extremely uncommon, and they typically cause problems with the patient's voice. Indirect endoscopy is sufficient for diagnosis. The surgical excision of

the larynx using microlaryngoscopic techniques yields satisfactory results ⁴

Table 2. Literature review

No	Writer - journal	Patients/ Problem	Intervention	Comparison	Outcome
1	Mesolella M et al. Ear, Nose Throat J. 2022	Adult patients with glottis hemangioma	Microlaryngoscopy surgery using laser	Cold steel surgery	Compared to surgical excision, CO2 laser surgery is less invasive and more effective. The additional benefits of lser include reduced trauma, improved control of hemostasis, excellent postoperative functionality outcomes, brief hospitalization, and low socio health costs.

DISCUSSION

Reported a case of adult laryngeal hemangioma in a 51 years old female. This was a rare case: Laryngeal hemangioma of the adult extremely rare and is almost always located in the supraglottic region. There have only been a few cases of vocal cord hemangioma that have been described and talked about in the medical literature. Kimmelman was the first person to report a case of hemangioma of the vocal fold in an adult in 1979. Twenty examples of solitary adult vocal fold hemangiomas have been detected after conducting a study of the scientific literature that was published between 1979 to 2020.¹

History and a laryngoscopic examination are used to diagnose adult laryngeal hemangiomas. A histological study is used to confirm the diagnosis and provide a definitive diagnosis.¹ Laryngoscopy is usually always adequate for making a diagnosis of a hemangioma. Other exams, such as CT, MRI Contrast-enhanced imaging and angiography are procedures that are reserved for patients who have respiratory symptoms as well as those who have larger lesions.⁶ It is not known what causes adult laryngeal haemangiomas; however, it is speculated that smoking, vocal abuse, and laryngeal trauma (such as from intubation) may play a role in the development of these tumors. There are additional accounts in the medical literature of cases of adult laryngeal haemangiomas multiplying during pregnancy and later decreasing in size postpartum. This lends credence to the hypothesis that these tumors may react to hormones containing oestrogen and/or progesterone.¹⁰

On anamnesis, it was found that patient had a shortness of breath and dysphonia. Although the incidence of

hemangioma larynx is low, the hemangiomas that occur in these areas have the potential to interfere with the functioning of respiration, speaking, and swallowing. Foreign-body sensation in the pharynx and larynx, hemoptysis, bloody sputum, sore throat, and hoarseness were the main symptoms of our patients; dysphasia and dyspnea, which frequently develop in the late stages of the disease were absent.⁹

The patient was diagnosed by her history and fiberoptic laryngoscope examination. The diagnosis of hemangiomas in adults is predominantly based on the patient's medical history and laryngoscopic examination, while histopathological examination confirms the diagnosis. Direct visualization by fiber-optic laryngoscopy or videostroboscopy for comprehensive evaluation of vocal cord lesion remains the investigation of choice for adult hemangiomas.¹ Prior research has demonstrated that CT and MRI are highly capable of defining the shape, size, and adjacent anatomical structures of hemangiomas. Contrast CT imaging is a useful method to noninvasively detect subglottic hemangiomas, providing a more accurate determination of airway narrowing.¹¹ Contrast CT of this patient was suggested that the mass was obstructing the airway with measure 13,1 x 11,5 x 1,2 mm on the anterior side of glottic.

There is no well-established protocol for the management of adult laryngeal hemangioma. Depending on the extent and location of the lesion, various treatment options for adult type are available. Adult vocal fold hemangiomas that are modest in size and asymptomatic are conservatively managed.¹ Due to their rarity, laryngeal haemangiomas are almost never taken into consideration as a differential diagnosis. Furthermore, the diagnosis will ultimately need to be verified

through histopathology, which is why surgery is the treatment of choice for adults.⁵ After performing a tracheostomy on the patient while they were under local anesthesia, the surgeon then proceeded to resect the hemangioma while they were under general anesthesia. This was done because of the potential for complications with bleeding from the mass during intubation. Temporary tracheotomy can be performed in larger hemangiomas.¹² The patient's airway was secured by performing a tracheostomy prior to the surgery, which was done under local anesthesia to avoid any airway complications.¹¹

In otorhinolaryngology, The laser has historically been employed in a manner similar to a scalpel or cautery, allowing for precise incision, cauterization, and coagulation of tissue. This is especially beneficial in cases where a collimated beam can offer non-contact and accurate tissue ablation, addressing tissue targets that would pose challenges when treated with conventional instruments. Over the course of time, additional uses of lasers have emerged in the regions of the head, neck, and upper airway. These applications take advantage of the laser's precise nature, as well as its capacity to achieve selective photothermolysis, spatial selectivity, confined thermal damage, and the production of photoacoustic waves.¹³

Diode lasers (DLs), which were initially introduced in 1962, continue to be the most energy-efficient and economically viable laser technology. Deep learning (DL) has demonstrated its efficacy in the treatment of vascular lesions through a therapeutic approach known as photosclerosis or thermocoagulation. This technique involves the utilization of DL technology, which emits radiation at a wavelength of 810 nm. Notably, DL has proven to be highly effective in achieving desirable outcomes without causing any scarring. After conducting a series of 22 endoscopic DL (810 nm) interventions for congenital subglottic hemangiomas, it can be concluded that this treatment modality is both highly safe and effective, with a reported efficacy rate of 95%.¹⁴ The laser technology offers several advantages, including its ease of manipulation, ability to be applied repeatedly, and its effectiveness in integrated coagulation and tissue division. Laser treatment is widely regarded as a minimally invasive and safer therapeutic approach due to its abbreviated surgical and recovery durations, as well as its favorable outcomes.⁷ In this patient we applied DL radiation and incision between the normal mucosa and the margin of hemangioma to prevent intraoperative bleed.

The presence of a hemangioma characterized by a vascular matrix, along with a delicate and thin mucosal layer, can potentially lead to significant intraoperative bleeding, posing challenges during surgical procedures.⁷ The performance of a biopsy procedure has the potential to result in significant hemorrhaging, which could potentially lead to a fatal outcome. Consequently, in situations where a hemangioma is suspected, it is advised against to conduct a pre-operative biopsy.⁹ After the fully excision of hemangioma, we performed the histopathological examination and the result indicated the hemangioma type is capillary cavernous or mixed type. From a histological perspective, the observed structure primarily consists of cavernous, capillary, or mixed types, characterized by a delicate and easily crumbled mucosa that

envelops the vascular stroma. Cavernous hemangioma is a non-malignant, yet occasionally locally invasive lesion characterized by the presence of enlarged and dilated vascular spaces. The color exhibited by an object is perceived as blue. Under microscopic examination, the identification of these tumors is based on the observation of large vascular channels with irregular sizes, which may exhibit a haphazard arrangement within the tissue. Capillary type hemangioma is characterized by a proliferation of small, capillary-sized blood vessels.⁴

One month after surgery, there was no complaints and the surgical wound was healed. The persistence of the recurrence remains a significant obstacle following surgical debulking. Tumor recurrence may arise as a consequence of incomplete excision, which can be attributed to factors such as the presence of a large tumor size or the diffuse or multifocal nature of the hemangioma. In cases where there is minimal residual hemangioma tissue, progression to tumor recurrence becomes a possibility.⁹

CONCLUSION

Adult laryngeal hemangiomas are very rare, they could come with absent clinical symptoms but also can occur with airway obstruction. Microlaryngoscope surgery with diode laser is a best choice for treatment of adult laryngeal haemangioma because it is the safest and most effective. In cases accompanied by shortness of breath that causes by upper airway obstruction, a tracheostomy could be conducted.

REFERENCES

1. Laohakittikul C, Srirompotong S. Adult Vocal Fold Hemangioma: A Case-Series Study and Review of Literature. *J Voice*. 2021 Mar;1-5.
2. Rafie A, Jolly K, Darr A, Thompson S. Adult cavernous haemangioma of the vocal cords with a unique presentation of acute respiratory distress: A case report. *Ann R Coll Surg Engl*. 2020;102(7):E152-4.
3. Olsen GM, Nackers A, Drolet BA. Infantile and congenital hemangiomas. *Semin Pediatr Surg*. 2020 Oct;29(5):150969.
4. Mesolella M, Allosso S, Mansueto G, Fuggi M, Motta G. Strategies and Controversies in the Treatment With Carbon Dioxide Laser of Laryngeal Hemangioma: A Case Series and Review of the Literature. *Ear, Nose Throat J*. 2022 Jun 13;101(5):326-31.
5. Rafie A, Jolly K, Darr A, Thompson S. Adult cavernous haemangioma of the vocal cords with a unique presentation of acute respiratory distress: a case report. *Ann R Coll Surg Engl*. 2020 Sep;102(7):e152-4.
6. Mender, Ines Chang, Sousa, Patricia Melo, Barros E. Vocal cord hemangioma. *Heal Sci*. 2022;2(2):111-4.
7. Shim HK, Kim MR. Potassium-titanyl-phosphate (Ktp) laser photocoagulation combined with resection using an ultrasonic scalpel for pharyngolaryngeal hemangioma via a transoral approach: Case report and literature review. *Am J Case Rep*. 2021;22(1):1-10.

8. Liu F, Xiao Y, Wang J. Therapeutic efficacy of intralesional bleomycin injection for laryngopharyngeal haemangioma in adults. *Acta Otolaryngol.* 2019 Dec 2;139(12):1117–21.
9. Xu S, Yu Y, ElHakim H, Cui X, Yang H. The Therapeutic Effect of the Combination of Intratumor Injection of Bleomycin and Electroresection/Electrocautery on the Hemangiomas in Hypopharynx and Larynx Through Suspension Laryngoscopy. *Ann Otol Rhinol Laryngol.* 2019;128(6):575–80.
10. Kaushal S, Shakeel M, Manickavasagam J. Adult Laryngeal Haemangioma Misdiagnosed as Obstructive Sleep Apnoea. 2020;3(2):26–32.
11. Cipolla F, Ragusa M, Andaloro C, Bonanno A, Grillo C, Basile A, et al. Superselective embolization and transoral ultrasonic surgery of laryngeal hemangioma: a case report. *B-ENT.* 2020;16(4):240–3.
12. Kazikdas KC, Yalcinozan ET, Tinazli R, Safakogullari H, Safak MA. Vocal Fold Hemangioma. *Ear, Nose Throat J.* 2019;98(5):257–8.
13. Orgain, C, Rothholtz V, Wong B. Lasers in Dermatology and Medicine. Nouri K, editor. *Laser/Light Applications in Otolaryngology.* Cham: Springer International Publishing; 2018. 1–241 p.
14. Rong Y, Wu C, Huang Y. *Advanced Materials, Structures and Processing Technologies Based on Pulsed Laser.* Advanced Materials, Structures and Processing Technologies Based on Pulsed Laser. 2022.

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