

Adult Glottic Hemangioma: A Case Report



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ABSTRACT

Glottic hemangioma is a benign vascular tumor that is rarely seen in the adult population. We report a rare Filipino case of glottic hemangioma in a 65-year-old female presenting with 2 weeks history of hoarseness and a smooth, pedunculated, bluish mass at the anterior one-third of the right vocal cord in flexible laryngoscopy. Direct suspension laryngoscopy showed a pedunculated mass that was paler-looking, similar to the color of the surrounding mucosa, exhibiting the Phonation sign of Menzel. The patient underwent microlaryngeal excision and histopathology showed findings consistent with cavernous hemangioma.

Keywords vocal cord; hemangioma; adult; cavernous

INTRODUCTION

Hemangiomas are the most common congenital benign tumors diagnosed, and approximately 60% arise in the head and neck region.[1] However, its occurrence in the larynx is rare. In pediatric patients, it is self-limited and resolves to approximately half

by 5 years of age and resolves further with age.[1] It occurs more frequently in girls and usually in the subglottic region.[2,3]

Its occurrence in adult patients is even more rare with only 16 cases reported from 1979 to 2017.[4] They are more frequent in males and occur in the supraglottic region.[1,5] Detailed history, fiberoptic laryngoscopy and imaging are essential for the diagnosis but it may only be confirmed through biopsy.[1] Depending on the site of lesion, it presents with dysphagia, dysphonia and shortness of breath. Fiberoptic laryngoscopy may reveal a bluish, discoloured, mucosa-covered mass. Doppler ultrasound, computed tomography, technetium imaging and plain radiographs can play a role in determining the dimensions and extent of hemangiomas.[3] Angiography and MRI may be used for extensive cases to confirm the vascular nature and determine its extent.[3]

Unlike in the pediatric population, it does not seem to show the tendency to spontaneously regress. [1] Due to limited reported cases, there has been no consensus and a well-established treatment protocol for the management of these cases. Treatment options that were documented include corticosteroid injection, laser ablation with CO₂ or KTP lasers, cryosurgery, radiation therapy, and excision with microlaryngoscopic techniques.[3]

CASE

The patient was an otherwise healthy 65-year-old female who sought consult due to a 2-week history of hoarseness. It was not accompanied by cough, dyspnea, dysphagia, odynophagia, hematemesis or aspiration episodes. Hoarseness was non-progressive, but persistent, prompting consultation. The patient was a known diabetic for 14 years, maintained on

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insulin and metformin + sitagliptin, and hypertensive for 10 years, maintained on telmisartan. She had no asthma, COPD, pulmonary tuberculosis, known coagulopathy or autoimmune disease. She was a non-smoker, not an alcoholic beverage drinker, with occasional intake of caffeine and dairy products and adequate water intake. She had no previous head and neck surgeries but was intubated once, however, hoarseness was not noted after the procedure. She had no allergies to food or medications. She was a businesswoman and was an averagely talkative person(5) based on the Voice Talkativity Scale. She had no other voice demands aside from work.

Clinical Findings

Aural examination showed anterior rhinoscopy and posterior rhinoscopy findings to be unremarkable. She had moist buccal mucosa, non-hyperemic posterior pharyngeal wall with non-enlarged and non-hyperemic tonsils. She had a supple neck, with no palpable neck mass, no cervical lymphadenopathy or limitation in neck movement. The rest of the physical examination findings were unremarkable.

Diagnostic Focus and Assessment

Visualization through flexible laryngoscopy showed non-hyperemic and non-edematous epiglottis and arytenoids. However, as seen in Figure 1a, there was

a smooth, pedunculated bluish mass from the ventricle at the anterior one-third of the right vocal cord partially obstructing the glottic opening. The patient did not undergo any imaging study as the lesion appeared isolated and restricted to the right vocal cord under flexible laryngoscopy. Impression at this time was a hemorrhagic cyst and she was advised surgery.

Therapeutic Intervention

On direct suspension laryngoscopy, there was a smooth, pedunculated mass from the ventricle at the anterior one-third of the right vocal cord partially obstructing the glottic opening, as seen in Figure 1b. This time, the mass was noted to be paler-looking, similar to the color of the surrounding mucosa. The patient underwent microlaryngeal excision of the pedunculated mass. A 0.5 x 0.5 cm well-defined, smooth-bordered mass, seen in Figure 2a, was removed from the ventricle of the right vocal cord.

After removal of the mass, the true vocal cords have maintained their smooth borders with minimal bleeding at the area of the surgical site as seen in figure 2b. A gross specimen consisted of a cream white to light red, firm, polypoid tissue measuring 0.8 x 0.8 x 0.5 cm.

The specimen was sent for histopathology and microsections disclosed fibrocollagenous tissue showing surface epithelium erosion and proliferation of large cavernous vascular spaces partly filled with

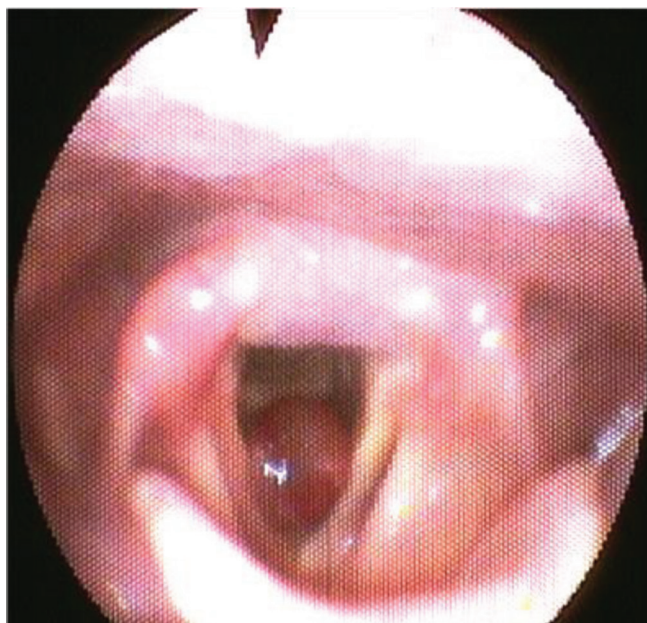


Figure 1. (a) Firm, deep colored, pedunculated mass seen flexible laryngoscopy (b) Paler looking mass seen on direct suspension laryngoscopy

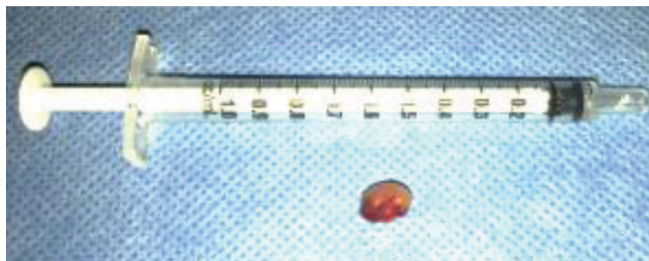


Figure 2. (a) Glottic mass removed through microlaryngeal surgery (b) Glottic mass removed through microlaryngeal surgery leaving the true vocal cord with smooth mucosa

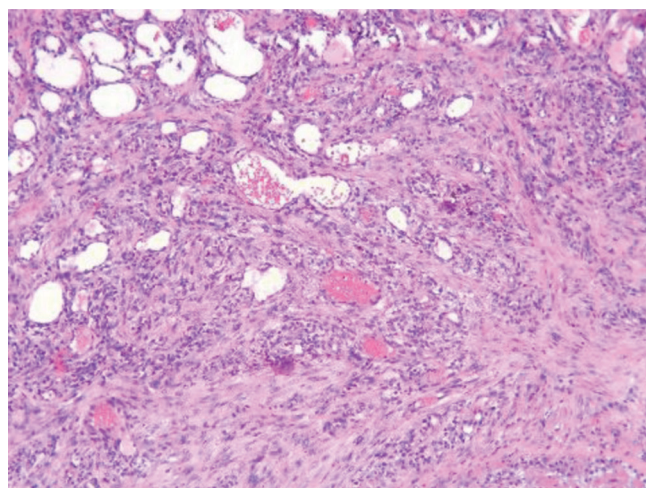
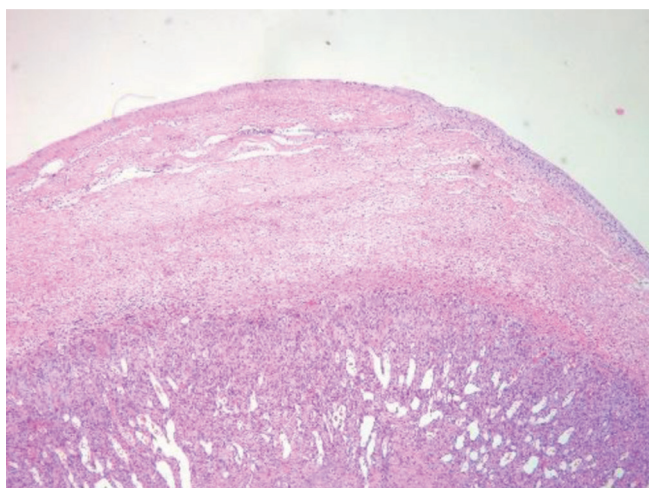


Figure 3. (a) Low power field, H&E stain, 40x fibrocollagenous tissue septa of varying spaces with blood-filled channels in between (b) high power field, H&E stain, 100x proliferation of large cavernous vascular spaces partly filled with red blood cells

red blood cells separated by scant connective tissue stroma, consistent with cavernous hemangioma, as seen in Figure 3.

Follow-up and Outcome

Four months postoperatively, the patient had no hoarseness, dyspnea, and aspiration. Repeat flexible nasopharyngolaryngoscopy showed smooth mucosa of the true vocal cords with no signs of recurrence of hemangioma as seen in Figure 4. The rest of the findings were unchanged.



Figure 4. True vocal cords showed no signs of recurrence of hemangioma

DISCUSSION

Among benign neoplasms of the larynx, about 95% are papillomas while others include oncocytic tumors, pleomorphic adenomas, lymphangiomas, neurofibromas, fibromatosis, paragangliomas, rhabdomyomas and hemangiomas.[1] With the history and character of hoarseness, laryngologic examination and computerized scanning, benign nature of the lesion may be elicited but can only be confirmed through biopsy.[1] The extent of the lesion and its dimensions may be identified through Doppler ultrasound, computed tomography, technetium imaging and plain radiographs.[2] Histologically, hemangiomas are composed of large, irregular, blood-filled channels lined with a single layer of endothelial cells between loose fibrous tissue septa of varying thickness.[2]

Laryngeal hemangioma is common in the pediatric population and is often associated with the cutaneous type. It can be seen more frequently among the female population and mostly are found in the subglottic region.[3] It usually manifests with respiratory symptoms, which include biphasic stridor, fluctuating respiratory distress and pseudocroup, especially during periods of venous engorgement.[2,3] It was postulated that the formation of hemangioma is due to an imbalance of positive and negative vasculogenic factors. In the first five months, there may be expansion, which is the proliferative phase, followed by the involution phase, manifesting with regression.[1] Failure of the lesion to involute causes persistence of hemangioma and marks as the base for uninhibited proliferation.

Among the adult population, laryngeal hemangioma is rare such that the incidence is not known due to the little number of cases.[1] Currently, there have been 16 reported cases for four decades since the first case in 1979 until 2017,[4,6,7] confirmed through literature searches. There is only one reported case in southeast Asia[6] and none in the Philippines. Based on the reported cases, it is usually in the cavernous form, where vascular channels are larger, less well-circumscribed and usually deeper in submucosal tissues.[3] They are also found to be more common among men. It usually involves the supraglottic region followed by glottis and subglottis.[1,5] There are no causative or predisposing factors that have been compellingly cited but some considered vocal abuse, cigarette

smoking and laryngeal trauma.[3,8] Symptoms vary and include dysphagia, dysphonia and shortness of breath[1] and vague symptoms such as hoarseness, cough, hemoptysis, dyspnea, and a lump sensation.[2] Compared to those of congenital hemangioma, it appears to be covered by thinner mucosa and grossly appears as a bluish, discolored mucosa-covered mass on direct laryngoscopy. With administration of epinephrine, shrinkage and compressibility on palpation can be noted. The phonation sign of Menzel is of diagnostic value and described as increased firmness, erection and deepening color of the growth during phonation.[3]

The treatment of adult laryngeal hemangiomas is influenced by patient's age, type, size and localization of the tumor.[2] Small hemangiomas can be managed conservatively, while large hemangiomas can be treated either by systemic steroid, injection of corticosteroids or ethanol, surgical excision, cryosurgery, radiation therapy and CO2 laser excision.[3] Some authors advise that it may be left alone if at all possible, especially if asymptomatic or if there is no involvement of surrounding structures. For cases where the hemangioma shows a tendency to progressively involve additional parts of the larynx, corticosteroid therapy or radiotherapy is recommended by some authors. CO2 laser excision may be used in limited or pedunculated supraglottic cavernous hemangioma, but it is ineffective in extended lesions and large vessels with significant bleeding.[2,9] Excision under microlaryngoscopy is a common surgical technique that is considered a safe, simple and effective option in the management of small lesions.[2] The surgical outcome is favorable and similar to other treatment options. Due to the small number of cases reported, there is no conclusive data regarding recurrence rates and transformation to malignancy.

Hemorrhagic polyp is commonly encountered in the practice. Although rare, glottic hemangioma should always be considered as an important differential.

CONCLUSION

Laryngeal hemangioma is a benign, slowly progressing vascular tumor that is rarely seen in the adult population with only 16 cases of adult laryngeal hemangioma reported from 1979 to 2017. As far as we know, this will be the first

reported case in the Philippines, second in southeast Asia, and seventeenth in the world. Although rare, it is an important differential for vascular tumors in the glottis as procedures such as biopsy may cause bleeding. Microlaryngeal excision using microscissors is a simple, safe and effective technique in the management of glottic hemangioma with favorable outcomes.

PATIENT ANONYMITY, CONSENT AND CONFIDENTIALITY

A printed informed consent was obtained from the patient for the writing and publication of this case report. They were also informed that the laryngoscopic and histopathologic findings will be included in this material. All personal information regarding the patient (name, geographic location, date of birth, contact number, etc.) will be kept confidential. Patient anonymity

will be maintained when the study is for presentation or publication. A breach of confidentiality may occur if the information is used in any other way.

ETHICS APPROVAL

This case report must acquire granted approval from the Institutional Review Board of the University of Santo Tomas Hospital prior to presentation or publication of this material as required by our institution.

CONFLICT OF INTEREST

No conflict of interest relevant to this material was reported by the principal investigator and co-author, which may interfere with the presentation and publication of this case report.

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Nothing to declare

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