Bilateral Vocal Fold Lobular Capillary Haemangioma Mimicking Glottic Carcinoma in a Chronic Smoker

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Introduction

Laryngeal haemangioma is a rare, benign neoplasm with an unknown incidence and etiology.[1] Fechner et al classified laryngeal haemangioma into adult and infantile forms, with the latter being more common.[2] The proposed etiologies of the adult-form haemangioma include previous laryngeal trauma (i.e. intubation), hormonal shifts (i.e. in pregnancy granuloma), viral oncogenes, infection and stress.[2–4] The available literature includes only 13 case reports of adult glottic haemangioma since the first report in 1979, and these were mainly on a unilateral vocal fold[5]. Here, we present and discuss a rare case of adult bilateral vocal fold haemangioma that was initially suspected to be a glottic carcinoma in a chronic smoker.

Case Report

A 50-year-old male presented with hoarseness, persistent dry cough and intermittent dyspnoea for 3 months. The symptoms had progressively worsened for 2 weeks prior to presentation. The patient had no history of fever, neck swelling, weight loss, night sweat, haemoptysis, contact with tuberculosis (TB) patients or any laryngopharyngeal reflux (LPR) symptoms. He

was a chronic smoker for 30 years (90 pack years) and worked as a mechanic with some extent of voice abuse (frequent shouting).

A general examination was unremarkable except for the hoarse voice. The severity of the hoarseness was graded subjectively using the GRBAS scale (Grade of hoarseness, Roughness, Breathiness, Asthenia, and Strain) proposed by Hirano in 1981.[6] The patient had overall hoarseness of grade 3 (G3, severe hoarseness) with a main component of roughness (R3). The maximum phonation time (MPT) was 7 seconds, which is a reduced time.

Flexible endoscopy of the larynx revealed an irregular, pinkish mass at the middle third of bilateral true vocal fold. The edge of the mass had whitish lesions. The bilateral vocal fold was mobile on phonation and inspiration. A chest radiograph showed normal findings.

The provisional diagnosis was glottic carcinoma, based on the patient's age, symptoms and smoking habit and the irregular appearance of the masses. The patient therefore underwent endolaryngeal microsurgery with a type 1 cordectomy (subepithelial excision of the mass) under general anaesthesia with a diagnostic and curative intent. Intraoperatively, the masses at the middle third of the vocal fold were not adherent to the vocal ligament and did not involve the anterior commissure (Figure 1).

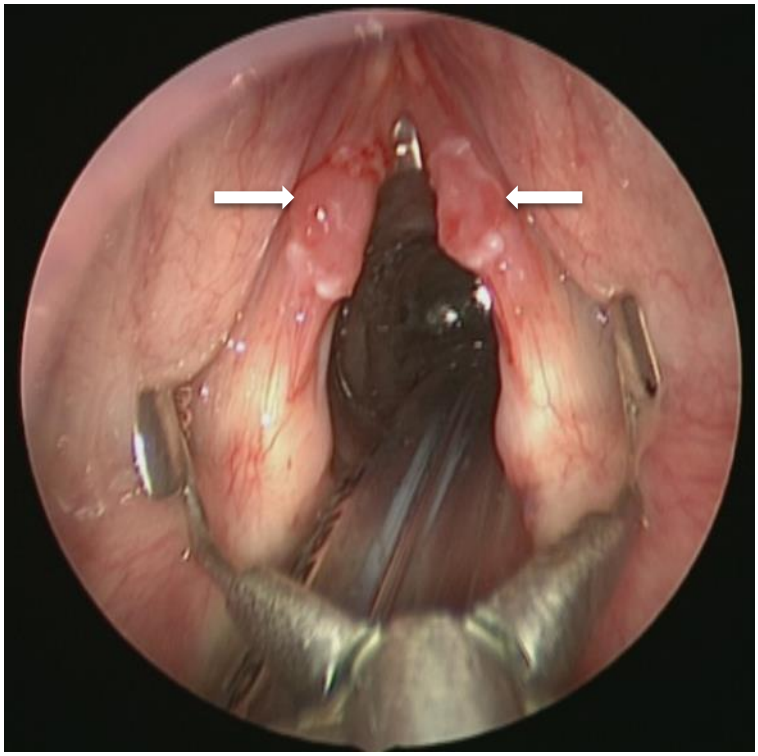

Figure 1: A direct laryngoscopy view showing pinkish, irregular surface lesion arising from middle third of bilateral vocal folds (arrows). An endotracheal tube is seen posteriorly.

Histopathological examination (HPE) showed lobular proliferation of small capillary-sized blood vessels, lined by endothelial cells that were positive for CD31 and contain red blood cells (Figure 2). Contrary to the initial diagnosis, no evidence of malignancy was revealed by the HPE. Hence, a diagnosis was made of bilateral vocal fold LCH. At 10 months post-surgery, the patient's voice improved, with overall dysphonia of grade 1 (G1, mild hoarseness), a main component of roughness (R1) and an MPT of 15 seconds, which is normal. A repeat flexible endoscopy of the larynx showed mobile vocal folds with normal mucosa.

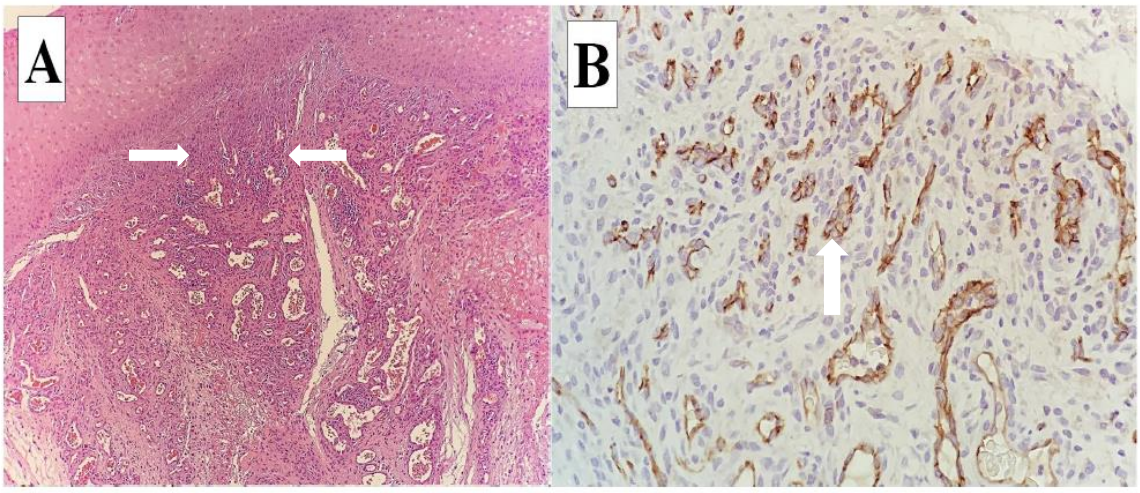

Figure 2: A) Microscopic view of glottic LCH composed of lobular proliferation of small capillary sized blood vessels (arrows) lined by endothelial cells and contain red blood cells. The overlying epidermis is acanthotic (hematoxylin and eosin stain, magnification = 10x). **B)** The endothelial cells are positive toward CD31 immunohistochemistry (arrow) (magnification = 40x).

Discussion

Benign tumours of the larynx are rare. Overall, 95% are papillomas, while the remaining 5% is divided among chondroma, rhabdomyosarcoma, neurofibroma, paraganglioma, haemangioma and others.[7] Laryngeal haemangioma is truly rare, especially in adults, with an unknown incidence due to the scarce literature.[1] Adult laryngeal haemangioma usually occurs in men (60%) with the commonest subsites being the supraglottis and glottis. However, haemangioma of the subglottis and trachea has been reported.[1,5,7–9] The clinical presentations vary depending on the location and size of haemangioma; nonetheless, hoarseness is the most commonly reported symptom, followed by dyspnoea, dysphagia and, rarely, haemoptysis.

Our patient presented with chronic progressive hoarseness, dyspnoea and persistent dry cough, which can be manifestations of many laryngeal pathologies, such as laryngeal tuberculosis (TB), papillomas, haemangiomas, fungal infection and laryngeal carcinoma.[7,10–12] Based on the patient's symptoms, the endoscopic appearance of the laryngeal masses and certain predisposing factors, a clinician should be able to list the possible differential diagnoses. Laryngeal TB is a good mimicker of carcinoma; hence, TB work-ups, such as an erythrocyte sedimentation rate, Mantoux test and chest x-ray, are necessary to determine its possibility, especially in patients with a history of contact with persons with TB.[12] A diagnosis of laryngeal carcinoma is highly suspicious in a chronic smoker with irregular, whitish, fungating or ulcero-fungating laryngeal masses.[12]

The existing literature describes an adult laryngeal haemangioma as a smooth-surfaced, lobulated or fleshy, reddish to purplish unilateral mass, which is typical for a haemangioma.[5,9,13,14] However, in the present case, the lesion involved the middle third of the bilateral vocal fold, with an irregular mucosal surface and a whitish lesion at the edge. The absence of the typical features of a haemangioma, the presence of a predisposing factor for malignancy and the normal chest radiograph findings led us to the initial suspicion of malignancy.

In this case, computed tomography (CT) of the neck was unnecessary because the masses were small and localised at the true vocal fold, as evidenced by their normal mobility and the absence of the typical appearance of a vascular tumour. In glottic carcinoma, the vocal fold mobility is affected when the paraglottic space, recurrent laryngeal nerve or cricoarytenoid joint are heavily infiltrated by the tumour. For that reason, a CT of the neck is indicated for staging purposes.[12] A large laryngeal vascular mass may require an angiogram to determine the presence or absence of feeding vessels and to confirm the diagnosis of haemangioma.

Apart from the adult and infantile forms, laryngeal haemangioma can be further classified into 3 histological types: cavernous (common in adults), capillary (mostly in infants) and mixed.[7,15] Lobular capillary haemangioma (LCH), also known as pyogenic granuloma, is a benign vascular lesion of skin and mucosa that commonly occurs in the head and neck region. However, laryngeal LCH is uncommon and is usually prevalent in children, predominantly in the subglottic region.[16–18] Glottic LCH in adults is extremely rare and, to our knowledge, no cases of bilateral glottic LCH have been reported, either in adults or in children.[2,16–18] Glottic LCH was described as multilobulated, polypoidal, pedunculated mass or raised nodule.[2,16–18] Diagnosis of LCH is made histologically by its distinct lobular growth pattern of capillary proliferation, fibromyxoid stroma and ulceration-based inflammation.

A literature review on glottic LCH in both adult and paediatric cases reported successful endolaryngeal surgical excision using cold instruments, a coblator or a CO_2 laser.[16–18] Alternative treatments for glottic LCH include intralesional steroid injection and medical therapy, such as proton pump inhibitors, steroids, antibiotics and speech therapy; these alternatives give best results when combined with surgery.[16–18] The alternative treatments are less invasive, but they require multiple sessions or long term therapy to achieve a cure. Thus, they may not be suitable for large LCH masses with airway obstruction.

Laryngeal biopsy using a channelled flexible laryngoscope under local anaesthesia is feasible in a cooperative patient who has glottic fungating mass. In the present case, this was not done because the glottic masses appeared superficial and biopsy under local anaesthesia had a risk of injury to the lamina propria of the vocal fold. Endolaryngeal subepithelial excision (type 1 cordectomy) of the bilateral glottic masses using a direct laryngoscope, cold instruments and a microscope was executed here to achieve both diagnostic and curative therapy, with the main aim of preserving vocal fold function. This surgery with precision technique respects the layers

of the vocal fold and prevents injury to the lamina propria, thereby avoiding life-long hoarseness due to vocal fold scarring.

Conclusion

Adult bilateral glottic LCH is a rare benign lesion of larynx that may present with an atypical appearance that mimics glottic carcinoma. An endolaryngeal microsurgery with subepithelial excision is a surgical option for both diagnostic and therapeutic purposes.

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References

1. Akhtar S, Shamim AA, Ghaffar S, Ahmed MS, Ikram M. Adult laryngeal haemangioma; a rare entity. J Pak Med Assoc 2012 Feb; 1:173–174.

2. Fechner RE, Cooper PH, Mills SE. Pyogenic granuloma of the larynx and trachea: A causal and pathologic misnomer for granulation tissue. Arch Otolaryngol Head Neck Surg 1981 Jan;107(1):30–32.

3. Wuyts FL, Bodt MSD, Molenberghs G, Remacle M, Heylen L, Millet B, et al. The Dysphonia Severity Index: An objective measure of vocal quality based on a multiparameter approach. J Speech Lang Hear Res. 2000 Jun;43(3):796–809.

4. Al Washahi MK, Al Killidar A, Razek YA, Khamis T. Endonasal endoscopic complete excision of unilateral nasal haemangioma in a child: case report and technical note. Oman Med J. 2019 Mar;34(2):156–159.

5. Salahuddin NA, Hitam S, Hassan NE, Azman M, Abdullah MA, Ab.llah NA. Glottic haemangioma – a rare entity in adult. Rawal Med J 2017;42(1):130–132.

6. Hirano M. Clinical examination of voice. Disord Hum Commun 1981;5:1–99.

7. Grant DG, Birchall MA, Bradley PJ. Surgery for benign tumours of the adult larynx. In: Remacle M, Eckel HE, editors. Surgery of Larynx and Trachea [Internet]. Berlin, Heidelberg: Springer Berlin Heidelberg; 2009 [cited 2020 Jun 15]. p. 91–111.

8. Xu Q, Yin X, Sutedjo J, Sun J, Jiang L, Lu L. Lobular capillary haemangioma of the trachea. Arch Iran Med 2015 Feb;18(2):127–129.

9. İriz A, Durmaz E, Akmansu H, Dağli M, Albayrak L, Eryilmaz A. Vocal cord haemangioma; A rare localization in adults. Turk J Med Sci. 2009 Apr 1;39(2):305–307.

10. Ulkumen B. Primary laryngeal tuberculosis presenting as a laryngeal carcinoma. J Clin Case Rep [Internet]. 2014; 04. 10.4172/2165-7920.1000393.

11. Mäkitie AA, Bäck L, Aaltonen L-M, Leivo I, Valtonen M. Fungal infection of the epiglottis simulating a clinical malignancy. Arch Otolaryngol Head Neck Surg 2003 Jan 1;129(1):124.

12. Marioni G, Marchese-Ragona R, Cartei G, Marchese F, Staffieri A. Current opinion in diagnosis and treatment of laryngeal carcinoma. Cancer Treat Rev 2006 Nov;32(7):504–515.

13. Yılmaz MD, Aktepe F, Altuntaş A. Cavernous haemangioma of the left vocal cord. Eur Arch Otorhinolaryngol 2004 Jul;261(6):310–311.

14. Prasad SC, Prasad KC, Bhat J. Vocal cord haemangioma. Med J Malaysia. 2008 Dec;63(5):419–420.

15. Shirazi N, Bist SS, Selvi TN, Harsh M. Spectrum of sinonasal tumours: A 10-year experience at a tertiary care hospital in North India. Oman Med J. 2015 Nov 10;30(6):435–440.

16. Ozturk B, Gunduz FK, Altun E. Lobular capillary haemangioma of vocal cord in an adult. Surg Oncol 2018;3:3.

17. Egilmez O, Uzun L, Ozkanli S, Kalcioglu M, Tekin M. A troublesome lesion of the larynx: Lobular capillary haemangioma. Scr Sci Med 2015 Dec 8;47(4):56.

18. Walner DL, Parker NP, Kim OS, Angeles RM, Stich DD. Lobular capillary haemangioma of the neonatal larynx. Arch Otolaryngol Head Neck Surg. 2008 Mar 1;134(3):272–277.