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Adult Vallecular Vascular Malformation - A Case Report

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Abstract

Vascular malformations are one of the most common forms of congenital benign lesions. The majority of them arise in the head & neck region and they are rare in adults. Laryngeal vascular malformations mostly involve supraglottis and glottis. We are reporting this case of vascular malformation, involving vallecula, a rare site reported in the literature.

Keywords: Adult vascular malformations, Vallecular Vascular Malformation.

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Introduction

Vascular malformations including hemangiomas are one of the most common benign infantile tumours/lesions. The commonly involved region is the head and neck with incidence reported to be 14-65%.[1] The oral cavity is the most often affected area in the head and neck.[2] A laryngeal vascular malformation is a benign lesion and a rare entity. History of the first description of a laryngeal vascular malformation which was a hemangioma, dates back to 1871, by Mac Kenzie.[3] In 1921, Sweetser was the first to classify these into the commoner infantile type and the rarer adult type.[4] Hayden described the first case of adult laryngeal venous malformation in 1924.[2] In 2004, vascular abnormalities were divided into vascular tumours and vascular malformations by the International Society for the Study of Vascular Anomalies (ISSVA). The latter was further subdivided into malformations that were simple, combined, malformations with large named vessels and those that were linked to other anomalies.[5] The incidence of laryngeal vascular malformations in adults is not known as there are very few reported cases.[6] Mostly, this benign lesion involves glottic and supraglottic regions in adults, and the commoner gender which is affected is male.[7] We are reporting this case of vallecular vascular malformation which mimicked the submucosal malignancy. In this case, we faced a problem in diagnosis preoperatively which might have changed our surgical approach to the patient.

Case Report

A 55-year-old male presented to the Otorhinolaryngology outpatient department, with complaints of moderate intensity, intermittent throat pain and odynophagia for 2 months. There was no complaint of any hoarseness, dyspnea or history of laryngeal trauma. The patient had a history of pulmonary tuberculosis 15 years back for which the patient had taken anti-tubercular treatment. The patient had a history of tobacco chewing and bidi smoking for the last 25 years. On clinical examination, there was smooth multilobulated swelling involving the vallecula and lingual surface of the epiglottis [Figure 1]. The rest of the supraglottis, glottis and hypopharynx were normal without any airway compromise. Since this patient was a chronic smoker and had recent onset of symptoms, the preliminary diagnosis of Submucosal Vallecular growth with epiglottis involvement was made. The patient underwent contrast-enhanced computerized tomography (CECT) of the neck, which suggested a homogenous soft tissue lesion involving vallecula [Figure 2]. Since the authors had a suspicion of malignancy, the patient was planned for direct laryngoscopy and biopsy from the lesion, under general anesthesia. Peroperatively, there was gross oedema of the epiglottis, and multiple pinkish-grey cystic lesions in the vallecula and lingual surface of the epiglottis were encountered. The lesions were excised with cold instruments, and there was moderate bleeding. The postoperative outcome was uneventful, and the patient started oral feeds after 48 hours. The specimen was sent for histopathological examination, and the report suggested polypoidal mucosal fragments

lined by focally hyperplastic squamous epithelium with the presence of submucosal hemorrhage and dilated vascular channels, which were congested and lined by a single layer of flattened endothelium; with overlying epithelium showing no signs of dysplasia. Pathologically, a diagnosis of simple capillary malformation was made [Figure 3].

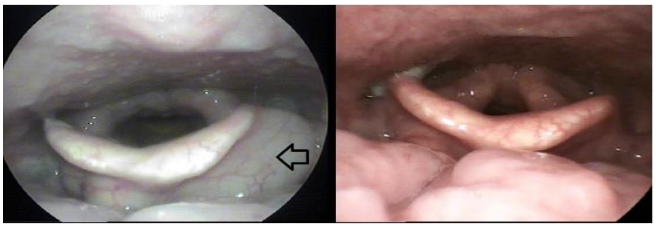

Figure 1: Laryngeal Examination Preoperative and Postoperative follow up after 3 Months

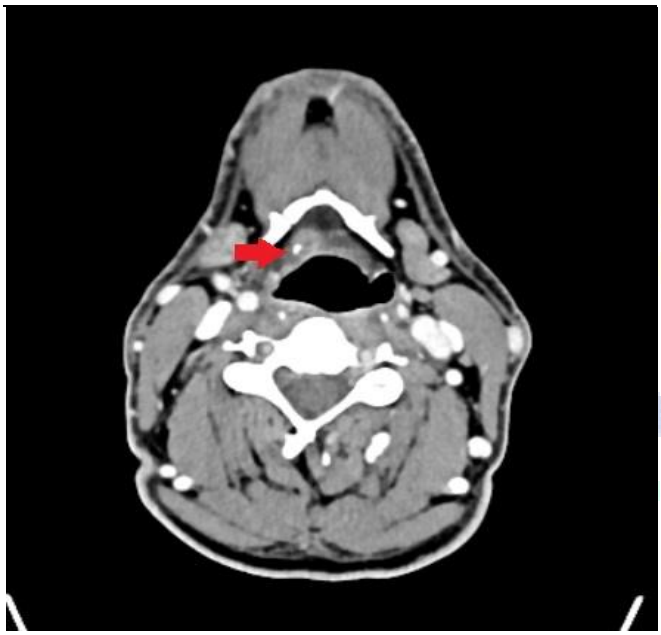

Figure 2: Contrast enhanced CT scan showing Mild enhancing lesion in vallecula with calcification within the lesion

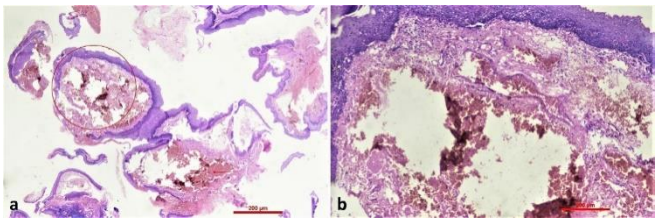

Figure 3: Postoperative Histopathology

The patient is under regular follow-up with periodic laryngeal examination, and 3 monthly follow up show no recurrence.

Discussion

Laryngeal vascular malformation in the adult population is a rare entity as compared to its infantile counterpart. [6] Most of these cases have been reported to involve supraglottis followed by glottis. In the present report, the patient had a rare site of involvement of vallecula and epiglottis. Involvement of vallecula has been reported in a study of lymphatic malformations of the head and neck. [7]

Some of the proposed causative factors include cigarette smoking, laryngeal trauma and vocal abuse. [8] Our patient was a chronic smoker and a vendor by occupation; a history of vocal abuse was also present. In our case, the most likely hypothesis is chronic trauma due to both patient's profession and the patient's smoking habit, which can be likely the cause of the simple capillary malformation.

Symptoms associated with vascular malformations are vague and not confirmatory, which include hoarseness, throat discomfort, dyspnoea and dysphagia in larger lesions. Radiological investigations like contrast-enhanced magnetic resonance imaging (CEMRI) would help in diagnosis, but in the present case, as strong suspicion was of a malignant lesion, we got only a CECT scan done instead of CEMRI.

Vascular malformations don't usually regress spontaneously. The stage of the lesion determines the best course of treatment for vascular malformations. The most common forms of treatment are sclerotherapy, surgery, laser therapy, and embolization.[9]

Malignant transformation in these vascular malformations is infrequent, but the possibility should be kept in mind, and the patient should be kept under regular follow-up.[10]

Conclusion

Although the incidence of adult laryngeal vascular malformation is very low, one must keep them as a differential diagnosis in cases with submucosal lesions involving laryngeal subsites.

References

1. Kobayashi K, Nakao K, Kishishita S, Tamaruya N, Monobe H, Saito K, et al. Vascular malformations of the head and neck. Auris Nasus Larynx. 2013;40(1):89-92. doi: 10.1016/j.anl.2012.02.002.
2. Kumar A, Subash A, Singh A, Patro S, Bakshi J. Coblation-assisted excision of hypopharyngeal venous malformation: a case report. Egypt J Otolaryngol. 2021;37(1):57.
3. Berkes B, Sente M. Adult laryngeal hemangioma. Med Pregl. 1998;51(11-12):547-50.
4. Sweetser TH. Hemangioma of the larynx. Laryngoscope. 1921;31(10):797–806.
5. Steiner JE, Drolet BA. Classification of Vascular Anomalies: An Update. Semin Intervent Radiol. 2017;34(3):225-232. doi: 10.1055/s-0037-1604295.
6. Kilcline C, Frieden IJ. Infantile hemangiomas: how common are they? A systematic review of the medical literature. Pediatr Dermatol. 2008;25(2):168-73. doi: 10.1111/j.1525-1470.2008.00626.x.
7. Lee DH, Yoon TM, Lee JK, Lim SC. Surgical Treatment Outcomes of Head and Neck Lymphatic Malformations in Patients With a Variety of Ages and Unusual Sites. J Craniofac Surg. 2016;27(3):602-4. doi: 10.1097/SCS.0000000000002504.

8. Mahmud KA, Zakaria R, Azman M, Mat Baki M. Adult Transglottic Haemangioma with Upper Airway Obstruction: Preservation of Airway and Voice. ORL J Otorhinolaryngol Relat Spec. 2021;83(4):295-298. doi: 10.1159/000515424.
9. Rutt A, Karatayli Ozgursoy S, Paz-Fumagalli R. Laryngeal Venous Malformation. Ear Nose Throat J. 2020;99(6):367-368. doi: 10.1177/0145561319840136.
10. Tanaka Y, Seike S, Tomita K, Ikeda JI, Morii E, Isomura ET, et al. Possible malignant transformation of arteriovenous malformation to angiosarcoma: case report and literature review. J Surg Case Rep. 2019;2019(12):rjz375. doi: 10.1093/jscr/rjz375.

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