

## Case Report

# An unusual case of angioleiomyoma of the vocal cord in a patient with oral cancer

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## ABSTRACT

Angioleiomyomas, as the name suggests, are tumors arising from smooth muscle of vascular tissue. We report a case of angioleiomyoma arising from the right vocal cord in a 45-year-old male with a history of squamous cell carcinoma (SCC) of the oral cavity who had restricted mouth opening owing to previous surgery and adjuvant radiation therapy because of which accessing the vocal cord tumour was difficult by conventional laryngoscopy. Initial bronchoscopy guided excision of the tumour was done. After a month, an emergency tracheostomy had to be performed as he presented with stridor due to recurrence of tumour. Laryngofissure was performed and tumour was excised. The patient is on regular follow up with no recurrence of tumour. We present in this case report, the clinical presentation, diagnosis, management options, pathological features and outcomes of this rare entity known as laryngeal angioleiomyoma.

**Keywords:** Vocal cord, Angioleiomyoma, Benign, Management

## INTRODUCTION

Angioleiomyoma is a rare benign tumour and presents mostly within the uterus, extremities and gastrointestinal tract.<sup>1</sup> The incidence of this tumour in the head and neck region is extremely rare with few case reports in literature. Within the larynx, supraglottic region has been the most common site of occurrence. The presenting symptoms usually depend on the site of involvement and include hoarseness of voice, dyspnoea, dysphagia, foreign body sensation in the throat and treatment involves complete excision of the tumour. We present a case of vocal cord angioleiomyoma in a patient with history of oral cancer.

## CASE REPORT

A 45-year-old male, known case of carcinoma of the buccal mucosa, treated with surgery and adjuvant radiotherapy in 2017, presented to us in January 2022 with hoarseness of voice. A fiberoptic laryngoscopy showed a globulated mass with a broad base extending from the anterior commissure to the middle of the right vocal cord, occluding 70% of the glottic opening. There was no stridor. Patient had Grade III trismus attributable to his previous treatment because of which a fiberoptic bronchoscopy and biopsy of the lesion was performed under local anesthesia. Biopsy was suggestive of hyperplastic lesion, with no definite evidence of malignancy. In view of vascularity of the lesion and risk

of bleeding during excision of the tumor, he was planned for bronchoscopy guided laser excision with thulium laser (5 watt power) under general anesthesia. After initial debulking and assessing vascularity of the lesion, anesthesia was continued through IGEL supra-glottic airway and the rest of the mass excised using a cryobiopsy probe.

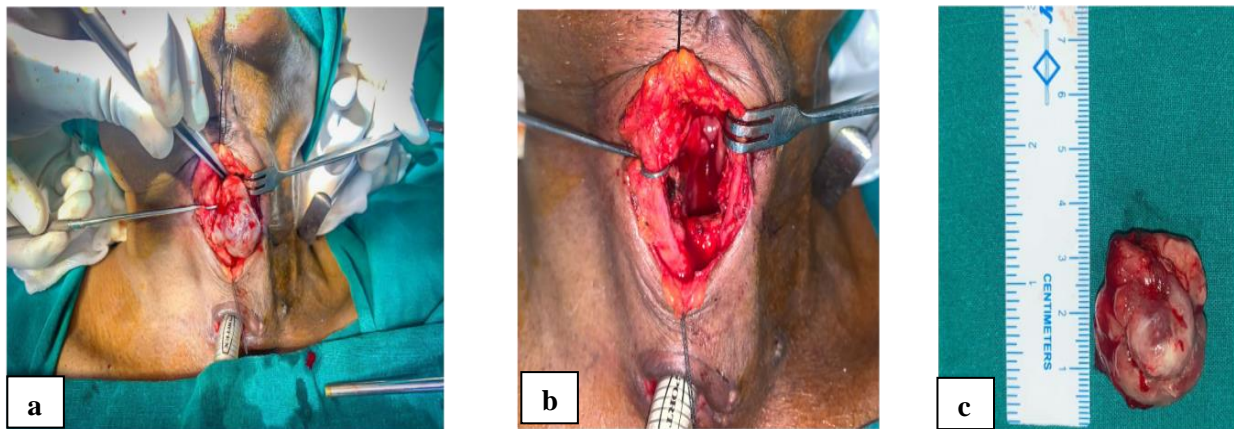
A month later, patient presented to the emergency room of our hospital with moderate to severe stridor. An emergency tracheostomy was performed to secure the airway. CECT of the head and neck revealed a mass lesion in the larynx almost completely obstructing the glottis. Patient was counseled for an open surgical procedure. Laryngo-fissure (Midline thyrotomy) was performed, tumor was exposed and excised en bloc. Intra-operatively, a tumour approximately 2 CMS in size was

seen to be arising from the right true vocal cord without any sub-glottic extension (Figure 1a-c).

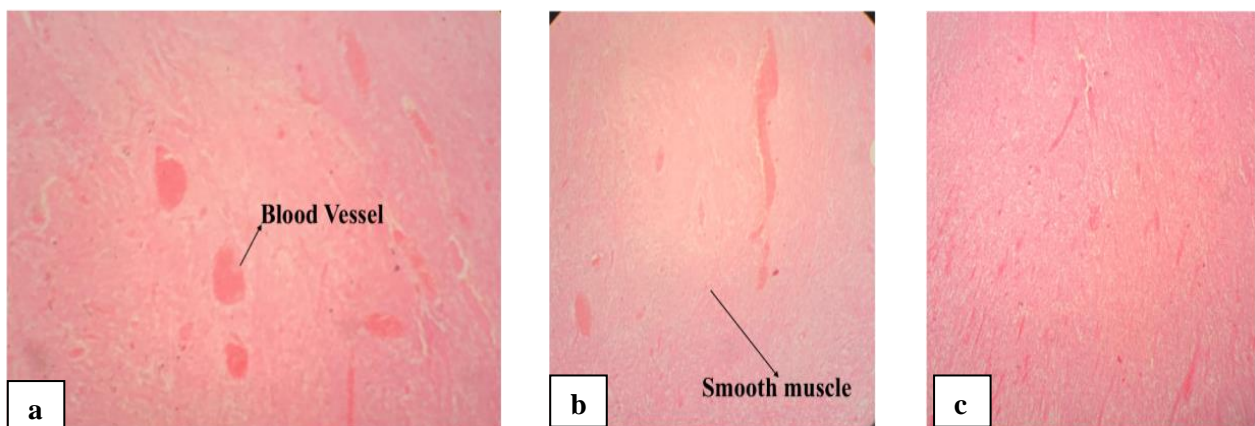
Post-operative recovery was uneventful. On day 5, video laryngoscopy showed bilateral mobile vocal cords, no post-operative oedema and adequate glottic opening. Patient was decannulated.

On pathologic gross examination, the tumor was solid, encapsulated measuring 2.0×1.5×1.0 cm. Microscopic examination showed stratified squamous epithelium with deeper stroma showing interlacing fascicles of spindle cells containing spindle shaped nuclei and scanty to moderate eosinophilic cytoplasm and the diagnosis of Angioleiomyoma was confirmed (Figure 2a-c).

At 6 months follow up, the patient has no complaints and is doing well with no signs of recurrence.



**Figure 1: (a) Intra-operative view of tumor arising from right vocal cord, (b) post excision of tumor and (c) specimen.**



**Figure 2 (a-c): Microscopic images.**

## DISCUSSION

Angioleiomyomas are benign tumours arising due to the proliferation of vascular smooth muscle cells (tunica media).<sup>2</sup> These rare tumors usually do not afflict the head

and neck region. An extensive Medline literature search showed few cases reported in the oral cavity arising from the palate and buccal mucosa.<sup>3</sup> Laryngeal angioleiomyomas more often have been reported in supraglottic, subglottic regions.<sup>4,5</sup> Reports on glottic angioleiomyomas are lacking.

**Table 1: Previous publications.**

Year	Author	Site	Gender	Age (in years)	Treatment
1979	Ebert et al <sup>1</sup>	Glottis	M	65	External approach
2000	Anderson and Weinstein <sup>10</sup>	Right Vocal cord	M	39 Recurrence at 51	Micro laryngeal surgery
2000	St John et al <sup>1</sup>	Left Vocal cord	M	66	Denied surgery
2008	Xu et al <sup>9</sup>	Left Vocal cord	M	53	Micro laryngeal surgery
2018	Youssef Khafateh et al <sup>11</sup>	Left vocal cord	M	73	Micro laryngeal surgery

The etiology of these tumors is unknown with possible theories proposed for their occurrence. While the role of sex hormones has been attributed to formation of uterine angioleiomyomas, the role of parathyroid-hormone-related peptide in creating a microenvironment suitable for angioleiomyoma by autocrine and/or paracrine mechanism has been suggested for head and neck angioleiomyomas.<sup>6,7</sup>

Histologically, these tumours have been classified into three types: solid, venous, and cavernous by Morimoto et al.<sup>8</sup> The treatment plan however is not affected by the type of variant.

Our histopathology report suggested the possibility of it being a venous sub-type.

The most common presenting symptoms reported are hoarseness of voice, dyspnoea, or a sensation of foreign body in the throat.<sup>9</sup> Our patient presented with hoarseness of voice initially and developed dyspnoea later.

Laryngocele, amyloidosis, papilloma may be considered in the differential diagnosis of these tumors, at initial evaluation. Clinical evaluation with laryngoscopy and imaging with CT/MRI aid in evaluating the size, extent of lesion and the vascularity of the lesion. Complete surgical excision is the treatment of choice for this tumor and the recurrence rates are low when excised completely. There is one report of recurrence in glottic angioleiomyoma in literature, which recurred after 12 years of original excision.<sup>10</sup>

Laryngeal angioleiomyomas may be excised either by external surgical approach or by an endoscopic approach. The choice of access to tumor is influenced by various factors such as the site, size and vascularity of the lesion. Reports on use of Co2 laser for excision of laryngeal angioleiomyoma have been described.<sup>1,10</sup>

In our case, due to trismus, an open thyrotomy approach was planned.

A review of English literature of exclusive glottic angioleiomyoma identified only 5 cases reported till date. (Table 1).

## CONCLUSION

We would like to add another unique case to the existing literature. Our case report is unusual in that the patient is a treated case of oral cancer who received adjuvant radiotherapy to the primary and neck 5 years earlier. Whether radiation therapy may be attributable to the development of this tumor is a question which requires further studies.

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