

Recurrent Inflammatory Myofibroblastic Tumor of the Larynx: Presentation and Management

Nupur K Nerurkar¹, Jahnvi², Enu M Kukreja³

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ABSTRACT

Background: Inflammatory myofibroblastic tumor (IMT) is an uncommon lesion that usually involves the lungs and mostly affects young adults. Head and neck IMTs account for 14–18%. These benign tumors are locally aggressive; hence, an accurate diagnosis is vital for the treatment plan.

Case description: A young male presented with hoarse voice for 9 months with a history of three previous laryngeal surgeries. Stroboscopy revealed a multilobulated vascular lesion, attached to the left vocal process. Laser-assisted wide excision of the lesion with an adjuvant treatment of botulinum toxin injection was performed. Histopathology revealed spindle cell tumor, immunohistochemistry was suggestive of myoepithelioma. On 6-month postoperative follow-up, stroboscopy revealed good healing.

Conclusion: Laryngeal IMTs have a tendency to recur unless excised with a wide margin. Histopathology along with immunohistochemistry is essential for diagnosis. Tumors attached to the vocal process may benefit from botulinum toxin injection. The patient is advised for a close and prolonged follow-up.

Keywords: Botulinum toxin, Case report, Immunohistochemistry, Inflammatory myofibroblastic tumor, Stroboscopy.

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INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) is a rare lesion usually involving the lungs and mostly affects young adults.^{1,2} The most commonly affected areas in the head and neck region include the paranasal sinuses and orbit.³ IMTs in head and neck region account for 14–18% of all lesions.^{4,5} Only a few cases of laryngeal IMTs have been documented in the literature. These tumors, although benign, tend to be locally aggressive, progress slowly or rapidly, and usually present with progressive symptoms referable to mass effect.⁴ Here, we present a case of recurrent laryngeal IMT.

CASE DESCRIPTION

A young male student from North India was referred to us with a history of a hoarse voice for 9 months and three previous laryngeal

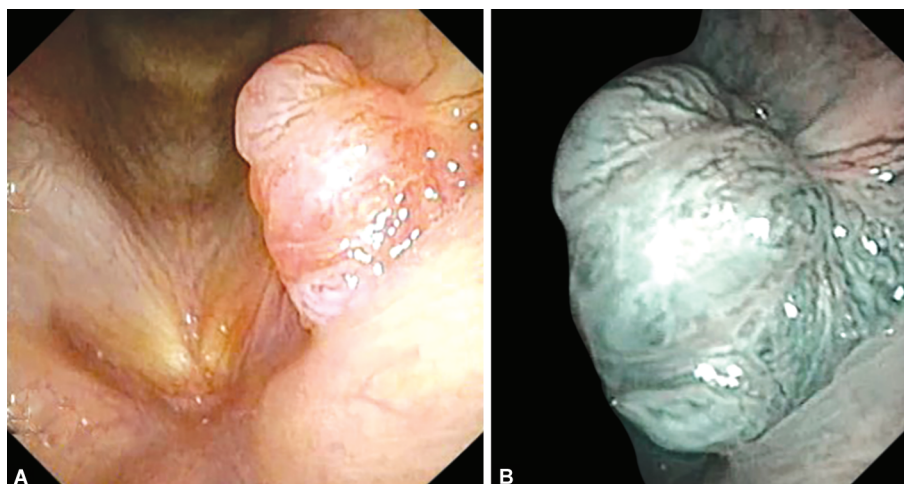
^{1–3}Department of Voice and Swallowing Center, Bombay Hospital & Medical Research Center, Mumbai, Maharashtra, India

Corresponding Author: Nupur K Nerurkar, Department of Voice and Swallowing Center, Bombay Hospital & Medical Research Center, Mumbai, Maharashtra, India, Phone: +91 9821034085, e-mail: nupurkapoor@yahoo.com

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Figs 1A and B: (A) Flexible laryngoscopic image revealing a multilobulated vascular lesion, attached to the left vocal process (B) and NBI of the same lesion revealing a leash of meandering blood vessels

surgeries in that time duration. He had been diagnosed twice as a polyp and as a benign tumor after the third surgery.

this peer review handled independently of this editorial board member and her research group.

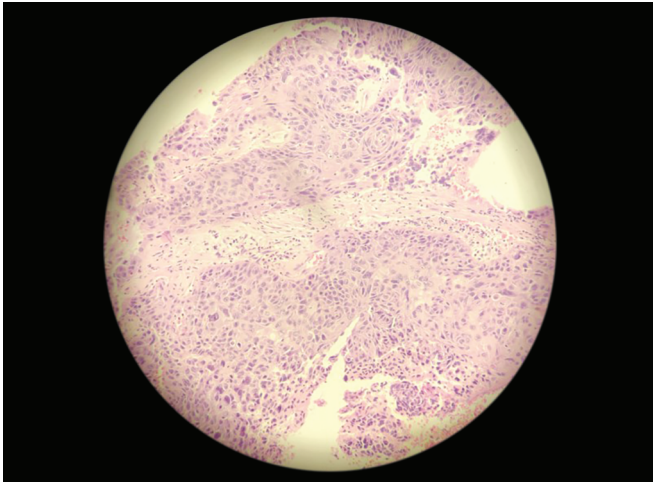


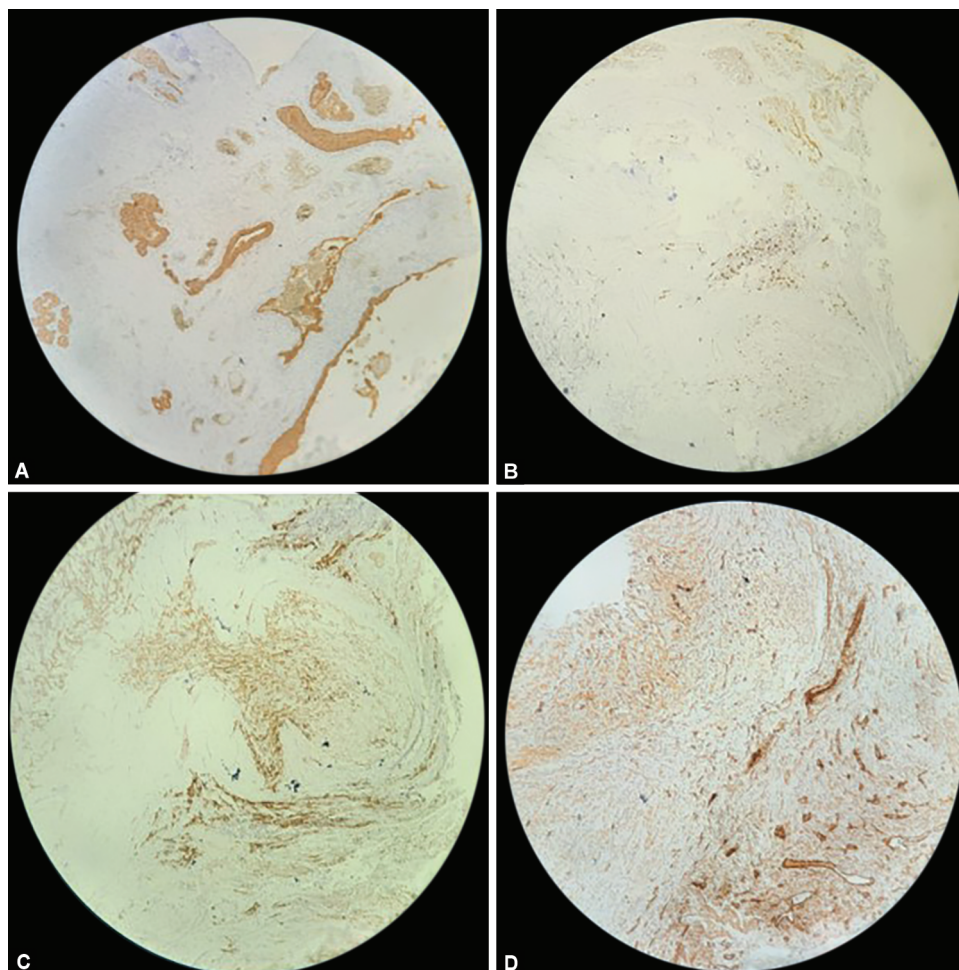
Fig. 2: Histopathology (hematoxylin-eosin stain, 100x magnification), showing spindle cells loosely arranged in fascicles within myxoid stroma. The cells possess scanty cytoplasm and contain uniform elongated nuclei

On presentation to us, stroboscopic evaluation revealed a left multilobulated vascular lesion, attached to the vocal process with a marked reduction in amplitude of the left mucosal wave, probably consequent to the phonatory gap caused by the lesion (Fig. 1A). Narrow band imaging revealed a meandering pattern of a leash of blood vessels (type 2 Ni classification) (Fig. 1B).⁶

A contrast-enhanced computer tomography of the neck was performed which revealed a mildly enhancing lesion arising from left vocal fold with mild diffuse thickening of the rest of the anterior fold with no paraglottic infiltration.

Laser-assisted wide excision of the lesion was performed and since the lesion was confirmed during surgery to be attached to the left vocal process and arytenoid, an area more prone to frictional injury due to phonation, adjuvant treatment of botulinum toxin injection into the left thyroarytenoid muscle was performed. A total dose of 7.5 mu of botulinum toxin was injected during the surgery into the left thyroarytenoid muscle.

Histopathological examination revealed a spindle cell tumor (Fig. 2) and immunohistochemistry was positive for pan-cytokeratin (pan-CK), epithelial membrane antigen (EMA), smooth muscle



Figs 3A to D: Immunohistochemistry revealing positive staining for pan-CK (A), Ki-67 (B), EMA (C), SMA (D)

actin (SMA), and in Ki-67 it showed 10–11% of high proliferation suggestive of myoepithelioma (Fig. 3).

Our patient's 6-month postoperative follow-up stroboscopy revealed good healing and he has been advised regular follow-up.

DISCUSSION

The IMTs are also known as plasma cell granulomas and were first described in the larynx in 1986.^{3,4} They are defined as distinctive neoplasms composed of myofibroblastic and fibroblastic spindle cells with inflammatory infiltrate of lymphocytes, plasma cells, and eosinophils.^{7–9} The myxoid/vascular pattern, compact spindle cell pattern, and hypocellular fibrous pattern are the three histologic types that are described. These three patterns can be mixed in a single tumor.^{7,8} As a result, diagnosis may be difficult, especially in rare sites such as the vocal folds. Conventional immunostaining with keratin, CD34, EMA, SMA, S100, and desmin lacks specificity is usually inconclusive, and sometimes may be misleading.^{7–9} Our patient had the spindle cell variant of IMT and immunohistochemistry revealed pan-CK, EMA, SMA, and Ki-67 suggestive of myoepithelioma.

Since many differential diagnoses may be considered, such as malignant tumors, sarcomatoid carcinoma or sarcoma, immunostaining, and special stains are very helpful in reaching a conclusion.⁷ Presentations of these tumors vary but according to the 30–40 cases which have been discussed in literature, the most common presentation is voice change in 74% of cases, stridor in 29%, dyspnea and shortness of breath in 22.5% and globus sensation in 16% of cases.⁵

The recurrence rate of laryngeal IMT is 8–18%,^{5,10,11} typically within 2–12 months.⁵ Our patient had history of three recurrences within 2 months of surgery prior to presenting to us. These recurrences may have been a combination of residual lesion left behind due to limited excision as well as due to the location involved, that is, the vocal process. Vocal process lesions are more prone to phonotrauma following excision due to the exposed cartilage. Botulinum toxin injection in the thyroarytenoid muscle causes vocal fold paresis which prevents phonotrauma in the healing phase thus decreasing chances of recurrence of lesions such as contact granulomas.¹²

CONCLUSION

Laryngeal IMTs have a tendency to recur unless excised aggressively with a wide margin. Immunohistochemistry is essential to identify these rare tumors. Tumors attached to the vocal process of the

arytenoid may benefit during the healing period with botulinum toxin injection of the adductor muscles. A close and prolonged follow-up is advisable for these benign but locally aggressive tumors.

ORCID

Nupur K Nerurkar  <https://orcid.org/0000-0002-4127-1590>

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