

CASE REPORT

AN UNUSUAL CASE OF LARYNGEAL INFLAMMATORY MYOFIBROBLASTIC TUMOUR INVOLVING LEFT ARYEPIGLOTTIC FOLD PRESENTING WITH LATERAL NECK SWELLING AND EXCISION BY LARYNGOFISSURE APPROACH: A CASE REPORT

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ABSTRACT: Benign tumors of larynx in adults are common and are often presents with hoarseness of voice or respiratory difficulty with stridor. Papilloma constitutes the major benign tumor of larynx in adults and usually presents with hoarseness of voice or stridor. A unique case of inflammatory myofibroblastic tumour of left aryepiglottic fold in a 19 year old female presenting with lateral neck swelling is presented. MRI revealed a polypoidal left aryepiglottic fold mass with caudal extension into the glottis and changing its position into the left PFS on e-phonation. The diagnosis of inflammatory myofibroblastic tumour remained elusive till it was proved histopathologically, after excision by laryngofissure approach. We present this rare case so that this condition and its surgical approach can be better recognized and understood.

KEYWORDS: Magnetic Resonance Imaging, Myofibroblastic tumour, aryepiglottic fold, laryngofissure.

INTRODUCTION: Benign laryngeal tumours are common in adults and are usually presents with hoarseness of voice or respiratory difficulty. Papilloma constitutes the major benign tumour and usually they are treated by variety of surgical approach. Cases of chondroma, paraganglioma and benign schwannoma are also reported and they also usually presents with hoarseness of voice or respiratory difficulty.

Inflammatory myofibroblastic tumour is a newly described entity of neoplasm which usually follows a benign course and is a locally recurrent neoplasm having distinct histological appearance. It is a benign fibroinflammatory mass of an unknown origin.^[1] The term has been used to describe the entity known as Inflammatory pseudo tumour.^[2] IMFT has many synonyms: plasma cell granuloma, inflammatory pseudotumor, xanthogranuloma, histiocytoma, and myofibrohistiocytic proliferation.^[3]

We present here a pathologically confirmed IMFT in a 19-year-old female patient. We also review the imaging, clinical and pathologic aspects of this unusual disease entity.

CASE PRESENTATION: A 19 year old girl presented to the Department of ENT in the month of February 2014 with swelling in left side of neck for 4 years and occasional mild difficulty in swallowing for 2 years. On examination of the patient, we noted a smooth, firm, non-tender swelling of size 5x 3x 2 cm over upper aspect of left side of neck which was mobile in nature but not in relation to deglutition and breathing. On 70 degree laryngoscopic examination we found a smooth mass arising from left aryepiglottic fold (AEF) with a broad pedicle (Figure 1).

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MRI was done. It revealed a smooth margin T2 hyperintense polypoidal lesion involving the left aryepiglottic fold plunging into the larynx causing significant narrowing of the same, with caudal extension up to the level of glottis (Figure 2 A, B). After e- phonation the lesion appeared to change its position and is seen extending along the lateral aspect of the aryepiglottic fold into the left PFS reaching up to the apex of the PFS (Figure 2 C, D). On post contrast study, the lesion showed delayed but intense enhancement with few non-enhancing areas (Figure 2 E, F). There was no diffusion restriction. MR angiography showed no connection with great vessels. The lesion measured approximately 2.98 x 2.3 x 4.8 cm in CS & CC dimensions. The patient was taken up for surgery.

An elective tracheostomy was performed and under general anesthesia, a laryngofissure approach was performed by splitting the thyroid cartilage, and the mass was found attached with a broad base pedicle to the left aryepiglottic fold on its medial aspect and left ventricle. The entire mass was removed in piece meal and sent for histopathological examination. The pharyngeal mucosa was repaired. Post operatively the tracheostomy stoma was closed after 15 days and a 70 degree laryngoscopy was done. The operative area showed healthy slough over the left AEF and left PFS. Vocal cord movements were normal.

The histologic examination of the surgical specimens demonstrated multifocal infiltrations of lymphocytes, histiocytes, plasma cells, and eosinophils. Between the inflammatory cells, spindle cells with eosinophilic cytoplasm and oval to elongated nuclei were seen which was suggestive of inflammatory myofibroblastic tumour (Figure 3). The patient has been discharged and kept under follow up (Figure 4).

DISCUSSION: IMFT more commonly affects the lung of children and adolescents,^[4] the gastrointestinal tract and orbit. However the omentum, retroperitoneum, hepatobiliary system, genitourinary tract, soft tissues of the trunk and extremities^[5,6] can also be affected. These lesions most commonly recur locally and rarely metastasize to distant sites. In 1991, Meis and Enzinger^[7] published results of a study involving 38 patients with inflammatory fibrosarcoma. Among the 27 patients with clinical follow-up results, 10 (37%) had local recurrence and three (11%) had metastatic disease.^[7] Laryngeal IMFT is extremely rare^[8] and it may easily be misinterpreted as a tumor because of its clinical findings, severe symptoms, and rapid proliferation.

The origin of this tumour is still unclear. Several theories have been proposed including infectious and immunological factors. Overall this kind of tumour represents less than 1% of all tumour in the entire respiratory tract and up to date only 31 cases have been reported in the larynx in literature.^[9] Weing et al first reported case series of 8 cases on 1995 in larynx.^[10] Among the 31 cases reported the most common site is true vocal fold constituting 16 cases of the total cases and the least commonest site is supraglottis constituting only 3 cases among the total cases reported.^[9] Coming to the presentation of these tumours, the most common presentation was voice change followed by stridor. Presentation with neck swelling has not been reported. Concentrating on treatment option, most of the cases have undergone endoscopic excision. Excision by open laryngofissure approach has not been reported in literature.

Reports of the imaging findings of IMFT are rarer.^[11] The lesion showed mixed high and low signal intensity on T2- weighted images. It was a well-enhanced lesion on gadolinium-enhanced T1-weighted fat suppressed images. Presumably the portion of the high signal intensity region of the tumour on T2-weighted images of our case reflects the myxoid, vascular, or active inflammatory area and the area of the low signal intensity on T2- weighted images may be a dense fibrous area.

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Although its histopathologic nature is benign, it may be difficult to differentiate this lesion from a malignant tumor because of its local invasiveness and its tendency to recur. Recurrence among laryngeal inflammatory myofibroblastic tumour can occur from 8 to 18% of patients and it usually occurs within one year post operatively.^[9] The current study does not show the long term follow up and whether this patient will have recurrence or not.

CONCLUSION: We report a rare case of inflammatory myofibroblastic tumour of larynx presenting with neck swelling. We did not recognize the condition until radiological investigation and surgery was performed and proven histopathologically. The diagnosis of IMFT should be considered for a patient with a solid sub mucosal laryngeal mass that especially shows good enhancement and mixed high and low signal intensity on T2-weighted images. However, the imaging characteristics of IPT are nonspecific since it has a wide range of imaging presentations, often mimicking tumors.

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Fig. 1: Picture showing the mass preoperatively.

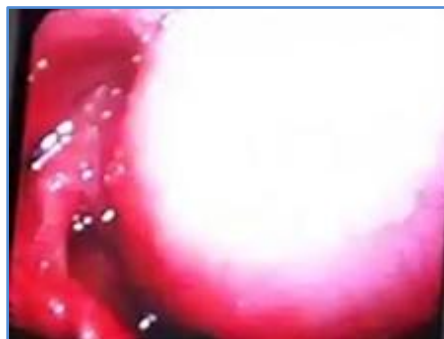


Fig. 1

Fig. 2: T2 weighted axial images at the level of supraglottis and glottis showing a broad based polypoidal mass with base attached to the left AEF, protruding into the laryngeal airway causing its narrowing (arrows) (A, B). T2 weighted axial and coronal images with e-phonation; the lesion changed position on e-phonation to lie lateral to the AEF in the pyriform fossa (arrows) (C, D). T1 post contrast fat sat axial and coronal images showing: intense delayed post contrast enhancement with few non-enhancing areas within (arrows) (E, F).

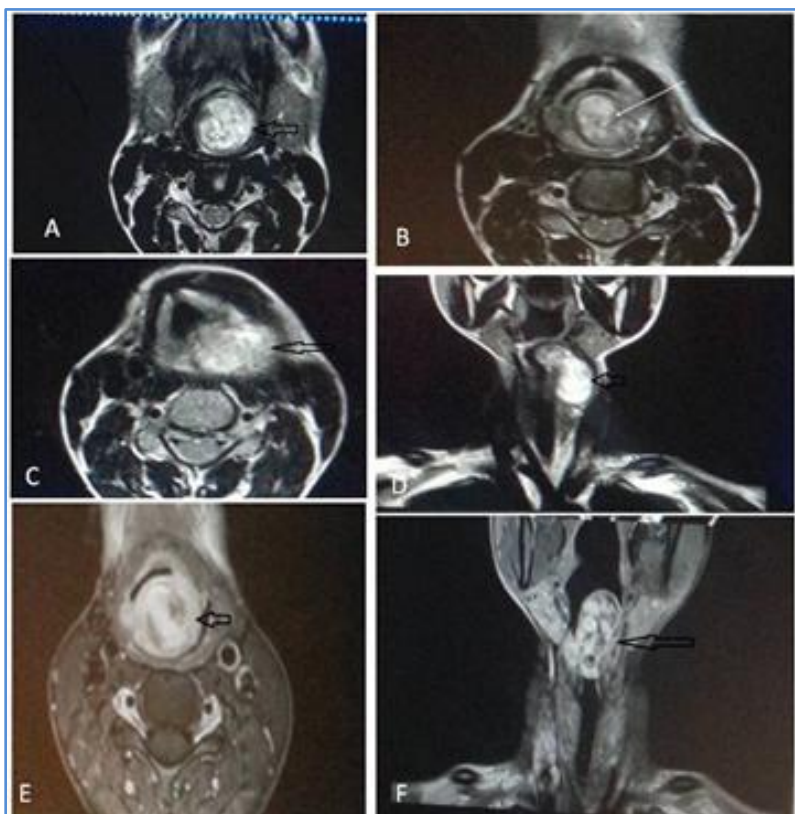


Fig. 2

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Fig. 3: Photomicrograph showing inflammatory myofibroblastic tumour (Hematoxylin & Eosin stain, ×10).

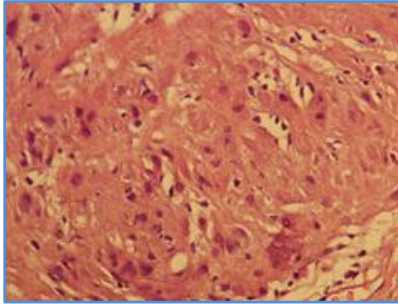


Fig. 3

Fig. 4: Post-Operative Photograph: Day 15 laryngoscopic findings.

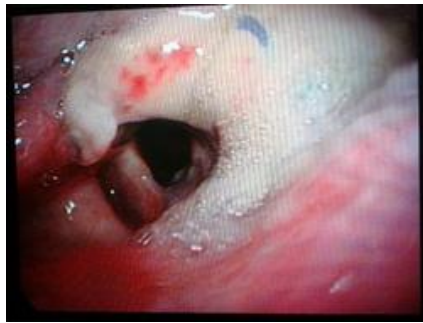


Fig. 4

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