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Case Report

Benign fibrous histiocyoma of larynx

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Introduction

Benign fibrous histiocyoma (BFH) is a benign neoplasm of mesenchymal origin composed of histiocytes and fibroblasts. It is a common neoplasm found in the sun-exposed skin of extremities and rarely presents as a non-cutaneous lesion especially in the head and neck. We report the clinical and pathological aspects of a rare case of BFH of the larynx treated at our centre.

Case Report

A 42-year-old female patient presented with progressive dysphagia of eight months duration. She also gave history of mild aspiration of one-month duration especially to liquids. There was no history of hoarseness or difficulty in breathing. She was a vegetarian, non-smoker and non-alcoholic with no other addictions.

On evaluation, 90° Hopkins rod lens evaluation revealed a 3 cm × 2 cm submucosal smooth lesion in the right aryepiglottic fold with bulge in the medial wall of right pyriform fossa and supraglottis. The arytenoid and bilateral vocal cords were normal and mobile. There was no cervical lymphadenopathy.

She was evaluated by contrast enhanced CT scan, which revealed a mild enhancing soft tissue lesion (2.6 cm × 2.7 cm × 3.3 cm) in the right aryepiglottic fold and adjoining epiglottis. The pre-epiglottic and paraglottic space were normal. The thyroid cartilage was not involved and there was no significant cervical lymphadenopathy (Fig. 1). She was planned for a direct laryngoscopy evaluation and biopsy. Direct laryngoscopy confirmed the earlier findings, and a biopsy was taken for histopathological examination.

Histopathological examination revealed nodular cellular proliferation of spindle cells in storiform pattern with few xanthoma cells. There was no cellular pleomorphism or mitotic activity (Fig. 2a and b). Immunohistochemistry (IHC) revealed positive for CD 68 and Vimentin with negative test for Desmin and S-100. Ki 67 index was less than 5%. The diagnosis

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Fig. 1 – Axial contrast enhanced CT scan of larynx.

was a case of BFH of larynx. She underwent transoral CO₂ laser assisted endoscopic excision of the lesion. The lesion was found to be well encapsulated and peeled of the surrounding tissue. Final histopathology confirmed BFH without any evidence of malignant changes. Postoperatively, she had mild aspiration, which improved, and she was asymptomatic for six months.

The patient reported back with complaints of dysphagia and aspiration and on evaluation was found to have recurrence of lesion in the right aryepiglottic fold. In view of the previous histology and its high propensity for recurrence, the patient was then planned for resection of the lesion by open approach for adequate visualization and resection of the lesion with adequate margins. A transcervical lateral pharyngotomy approach with extension to vallecula was used to expose the pyriform fossa and supraglottis. The lesion in the aryepiglottic fold, adjacent epiglottis and false vocal cord was excised with adequate margins. Postoperatively, the patient again had mild aspiration with no change in voice, which improved by two months. She is asymptomatic on regular follow-up for the past three years with no evidence of disease (Fig. 3a and b).

Discussion

BFH is a benign tumour of mesenchymal origin, which is usually found to be of cutaneous origin and its presence in non-cutaneous deep organs of the body is rare with few isolated reports in literature.¹⁻³ It has been referred to as fibrous xanthoma, dermatofibroma, xanthogranuloma and fibroxanthoma and was first described as a separate clinical entity in the 1960.¹⁻³

Cutaneous BFH is relatively common in areas of sun exposed dermis and superficial subcutaneous tissue of the extremities which are attributed to reactive proliferation of histiocytes due to sun exposure, repeated trauma and chronic infection. The presence of BFH in deep organs, especially in the head and neck, is rare and the exact aetiology is not understood. In the head and neck region, the commonest region involved is the orbit and there are only rare isolated reports of involvement of oral cavity, nasal cavity, ethmoids, neck and larynx.⁴⁻⁸

In the head and neck region, they usually present as painless well-circumscribed firm lesions causing no specific symptom. The symptoms develop later due to pressure effects depending on their size and site of involvement. They may present as nasal obstruction, epistaxis, proptosis, stridor, dysphagia and dysphonia.^{1,2,5,6,8} Our patient presented with dysphagia and features of aspiration, which was of insidious onset and gradually increased in severity.

Clinically, it is difficult to diagnose BFH due to its histological features resembling various other mesenchymal tumours. The diagnosis of BFH is challenging and rests on characteristic histopathologic and immunohistochemical features. The classical histological pattern is a neoplasm of histiocytic origin with dual cell population of histiocytes and fibrocytes.⁹ On microscopic examination, it is predominantly composed of fibroblastic cells arranged in a storiform pattern or interlacing fascicles, supported in a collagenous stroma and admixed with varying numbers of histiocytes, foam cells, siderophages and inflammatory cells. On IHC, it is positive for CD 68 & Vimentin and negative for SMA & S-100 (muscle and neurogenic origin). Histologically, BFH needs to be differentiated from aggressive conditions like malignant fibrous histiocytoma, which typically has a pleomorphic pattern of cells with mitotic figures, giant cells and areas of haemorrhage and necrosis.¹⁰

Fig. 2 – (a and b) Histopathology depicting spindle cells in storiform pattern (H&E), (a) low power (5X), (b) high power (20X).

Fig. 3 – (a and b) 90° Hopkins rod lens evaluation of the larynx, (a) preoperatively, (b) 6 month postoperatively.

Surgical excision is the mainstay of treatment with no specific role of radiotherapy or chemotherapy. Macroscopically, the lesion is well circumscribed and as it has a benign histology which may be the rationale for enucleation of the lesion by various surgeons as an adequate surgical option. The recurrence rate of 44–73% suggests a high degree of recurrence with surgical excision especially with close margins.¹¹ In our case, initially we performed a transoral CO₂ laser assisted excision of the lesion with close margins and the patient presented with recurrence within 6 months of surgery. So in the second instance, we performed an open partial laryngectomy to facilitate adequate exposure and wide surgical margins and the patient is disease-free for the past 3 years.

There have been only a few case reports of BFH of larynx^{6,8} and our case typically highlights the need for IHC for diagnosis of this condition and the requirement of wide surgical margins to prevent recurrence.

Conflicts of interest

The authors have none to declare.

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