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Case Report

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Synchronous presentation of giant anterior neck lipoma and interarytenoid hamartoma

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ABSTRACT

Giant lipomas are benign soft tissue tumor found rarely in the neck and are still rarer in the anterior part of neck. Laryngeal hamartoma (LH) is a very rare lesions and the number of reported cases is limited. Herein, we report a case of 68 years old male with a history of painless, slow growing swelling on anterior neck for past 8 years and also complaints of dyspnoea and stridor for past 6 months. FNAC from anterior neck swelling was suggestive of lipoma. USG and CT findings were suggestive of soft tissue lesions. Indirect and flexible laryngoscopy shows a large pedunculated, globular mass in interarytenoid region. A complete surgical excision of the anterior neck mass and interarytenoid polyp was done. Diagnosis of lipoma and hamartoma was confirmed on histopathology. Patient was followed up for 2 years with no evidence of recurrence.

Keywords: Anterior neck, Giant lipoma, Hamartoma, Larynx, Stridor

INTRODUCTION

Lipoma is a benign mesenchymal tumor of adult fat cells. A peak incidence of lipoma formation is noted in the fourth to sixth decades of life, and are common in obese individual. The incidence in head and neck being thirteen percent. Lipomas infrequently occur in head and neck. Of those lipomas that occur in head and neck region, posterior triangle is most common location while anterior neck lipoma is extremely rare. FNAC and sonography helps in making early diagnosis which can be supported with CT and confirmed with histopathology report. Surgical excision is the treatment of choice.

Hamartomas are rare benign developmental anomaly, characterized by the formation of a tumor like mass consisting of a focal, excessive growth of indigenous mature tissue present in wrong proportions without normal

arrangement tissue architecture.3 Hamartomas may occur in any organ, but are most commonly found in the lungs, liver, kidneys and intestines. Head and Neck hamartomas are extremely uncommon and those arising from the larynx are considered rare, presenting clinically with symptoms of upper respiratory tract obstruction, dysphonia, choking, hoarseness and progressive, persistent stridor. Hamartoma may present as a single lesion or in terms of a multiple hamartoma syndrome.⁴ Management consists conservative excision due to the benign nature of the disease. Partial or total laryngectomy should be reserved for those lesions involving so much of laryngeal framework. Recurrences are usually associated with incomplete removal. We report a case of synchronous presentation of giant anterior neck lipoma and interarytenoid hamartoma. Informed consent form was obtained.

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CASE REPORT

A 68 years old male presented to ENT clinic with complaints of painless, slowly progressive swelling on the anterior aspect of neck for past 8 years, but had enlarged rapidly over the past 4 months. The patient denied any recent weight loss or pain associated with the mass. Patient also complaints of dyspnoea, stridor for past 6 months. Dysphagia, hoarseness was not reported.

Vital signs demonstrated that the patient was afebrile and hemodynamically stable. Oxygen saturation in room air was 96%.

Physical examination revealed a globular swelling of 9×7 cm present on midline of neck more towards right, extending superiorly from 1 cm below symphysis menti and 3 cm above suprasternal notch, non-tender, soft in consistency, freely mobile in all directions with no movement on deglutition and protrusion of tongue (Figure 1). He had no lymphadenopathy or other palpable masses, and his oral cavity and oropharynx was normal.



Figure 1: Clinical photograph of the patient showing swelling arising anteriorly from the neck.

Routine investigations like complete blood count, liver function test, kidney function test, serum electrolytes, random blood sugar, ECG were all within normal limit. Chest radiograph was non-specific. Indirect laryngoscopy reveals a pedunculated mass arising from interarytenoid region and moves with respiration (Figure 2). Flexible laryngoscopy shows a large pedunculated, globular mass in interarytenoid region, smooth, well mucosalised, nonerythematous and move with respiration. It was also observed that there is enough space to fit endotracheal tube for intubation. FNAC from anterior neck swelling was suggestive of lipoma. CT revealed a 14×8 cm homogenous, well defined fat density lesion in the subplatysmal plane, trachea central in position with no compression.

The patient was planned for single stage surgery under general anaesthesia. Anterior neck was exposed and accessed via cervical collar incision (Figure 3). Platysma was divided, superior and inferior flaps were raised. Once a plane was identified between the platysma, tumor and underlying strap muscles, the entire tumor was delivered in toto (Figure 3).

Wound was closed in layers, drain was placed. For interarytenoid polyp, Kleinsasser laryngoscope was introduced and positioned to get the best view of polyp. Excision was done with endolaryngeal microscissor (Figure 3). Post-operative period was uneventful.

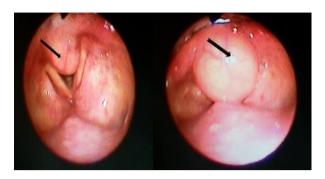


Figure 2: Indirect laryngoscopy shows a large pedunculated mass arising from interarytenoid region (black arrow) and moves with respiration.

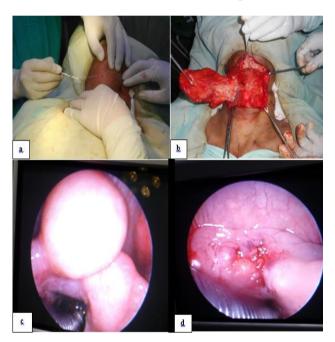


Figure 3: Intra-operative findings (a) cervical collar incision; (b) entire tumor was removed from the underlying strap muscles; (c) rigid endoscopic appearance of a large pedunculated mass arising from the interarytenoid region; and (d) following excision of inter-arytenoid polyp.

Gross examination of neck mass revealed a well encapsulated and soft yellowish tumor measuring $14\times7\times3$ cm (Figure 4). Microscopic examination of neck mass revealed clusters of mature adipocytes and fibrous stroma consistent with lipoma. Gross examination of polyp revealed a 2×2 cm mass (Figure 4). Microscopic examination of polyp revealed stratified squamous epithelium underneath of which were mature adipocytes, smooth muscle fibres admixed with mucous glands and

vascular structures consistent with hamartoma. Patient was followed up for 2 years with no evidence of recurrence.



Figure 4: Gross photograph of the excised neck specimen and inter-arytenoid polyp.

DISCUSSION

Lipomas are the most common benign adipose tumors of mesenchymal origin secondary to hamartomatous proliferation of mature fat cells. Lipomas make up approximately 5% of all benign tumors of the body, and they may occur anywhere on the surface of the body. Cheek is the most commonest site in head and neck region, followed by the tongue, floor of mouth, buccal sulcus, vestibule, lip, palate, gingiva and rarely occurring subcutaneously in the anterior neck region as was in our case.5 Based on location, they are classified as subcutaneous type, sub-fascial type or inter-muscular type. Lipomas usually present as solitary lesions as in our case, but multiple site involvement may be seen in alcoholics, diabetes mellitus and syndromes such as Madelung's disease and Kobberling-Dunnigan syndromes.⁶ The exact cause of lipoma is unclear, though there is an association with genetic mutation in chromosome 12q, 6p and 13q. Clinical features vary greatly depending upon the lesion size, location and rate of growth. Rarely lipoma reach to a size greater than 10 cm in one dimension, or weighing atleast 1000 g called as giant lipoma. In our case, size of lipoma was 14×7 cm and found in anterior triangle of neck, which is very rare both in terms of size and location. A rapid increase in size should always raise the suspicion of malignancy. Ultrasonography (USG) remains as the initial imaging modality in diagnosis of head and neck lipomas while fine needle aspiration cytology (FNAC) or computed tomography (CT) is indicated for confirmation of diagnosis.

Management is complete surgical excision. In our case, the tumour was removed completely through cervical collar incision. Complete excision with capsule should be performed to prevent recurrence. Liposuction is sometimes preferred in certain cases, but there is high chance of recurrence compared with excision.

The term hamartoma was derived from greek word 'hamartanein' means 'to go wrong' and introduced by

Albrecht in 1904, who distinguished between true neoplasm and tumor like lesions. Laryngeal hamartoma is a tumor like malformations composed of mature tissue elements normally located in the larynx, but with abnormal proportions or arrangements. 9

Hamartoma falls into two broad categories. The most common one is the mesenchymal hamartoma, composed of an overgrowth of mesodermal tissue without any epithelial component. The less common one is the epithelial or glandular hamartoma, composed of epithelial or glandular tissue admixed with the mesodermal elements. In infants, larvngeal hamartomas are typically associated with severe respiratory obstruction or feeding difficulties and cyanotic attacks. In adults, the signs and symptoms of laryngeal hamartomas are more diverse. Some adults experience hoarseness, dyspnoea and even an acute airway obstruction that may require tracheostomy. Other adults experience a long standing and sometimes nearly asymptomatic course of disease. ¹⁰ Patients having upper airway abnormality should undergo complete examination, radiological studies and examination of larynx with both direct and flexible techniques. Management of laryngeal hamartomas consists of conservative surgical excision. Endoscopic removal is usually adequate as in our case; however, larger lesions require open techniques for removal.¹¹

CONCLUSION

Lipomas in the anterior neck are rare, but can present as giant lipomas as in our case. The surgeon should be able to differentiate benign lipomas from liposarcomas. Surgical management of this tumor is challenging and should be performed by experienced surgeon due to the need of meticulous dissection with respect to underlying blood vessels and nerve. Laryngeal hamartomas are rare and should always be kept in mind as differential diagnosis of benign laryngeal lesions. The majority of LH develop either in early childhood or during the sixth decade of life. Two thirds of all patients are male. Upper airway compromise is the commonest symptom in children, and adults present with stridor, hoarseness and various degree of dyspnoea. Complete surgical removal is treatment of choice but should be limited in order to preserve laryngeal function, as the prognosis of patient with LH is excellent.

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