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CLINICAL REPORT

Combined Laryngocele Secondary to Localized Laryngeal Amyloidosis

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Abstract Laryngocele is a benign condition due to abnormal dilatation of the laryngeal saccule. Localized amyloidosis causing laryngocele is a rare entity with few reports in the literature. We present a young male patient with a large combined laryngocele secondary to laryngeal amyloidosis.

Keywords Laryngocele · Amyloidosis · Combined laryngocele · Laryngeal amyloidosis · Neck swelling · Laryngeal saccule

Introduction

Laryngocele is a benign condition due to abnormal dilatation of the laryngeal saccule. Localized laryngeal amyloidosis is an uncommon condition presenting as diffuse or nodular deposition of fibrillar protein in the endolarynx. Amyloidosis causing laryngocele is a rare entity with few reports in the literature. We present a young male patient with a large combined laryngocele secondary to laryngeal amyloidosis.

Case Report

A 25-year-old male patient, non-smoker presented with an insidious onset swelling in the right upper neck of 10 months duration. No history of chronic cough or playing wind instrument. He gave history of increase in swelling on coughing and straining and reduction on pressure. He also gave history of hoarseness of 1 month duration. On clinical examination, there was a 6 × 4 cm soft, non-tender swelling situated in the right upper neck anterior to the sternocleidomastoid muscle. The swelling increased by valsalva maneuver and it could be reduced on pressure. It moved on deglutition but not on protrusion of tongue. Indirect laryngoscopy revealed bulge in the right vallecula, aryepiglottic fold, and false cord with normal and mobile true vocal cords with adequate airway.

Plain X-ray of neck showed a right air filled space (Fig. 1). Ultrasound of neck revealed a right neck cystic mass attached to the larynx. CT neck confirmed the diagnosis of a large combined laryngocele (Fig. 2).

The laryngocele was approached by an external approach. The fundus of laryngocele was dissected and the neck followed down through the thyrohyoid membrane taking care to avoid injury to the superior laryngeal neurovascular bundle. Upper 1/3rd of right thyroid ala was excised to access the internal component which was excised completely (Fig. 3). The patient was extubated after 24 h made a good recovery.

Histopathology of the specimen revealed a cyst with squamous lining and chronic inflammation with air filled stroma. Congo red showed deep red staining with apple green birefringence on polarization strongly suggesting amyloid deposition (Fig. 4). He was further investigated for clinical findings and markers for systemic amyloidosis which was negative. The patient was identified as a case of combined laryngocele secondary to localized laryngeal amyloidosis.

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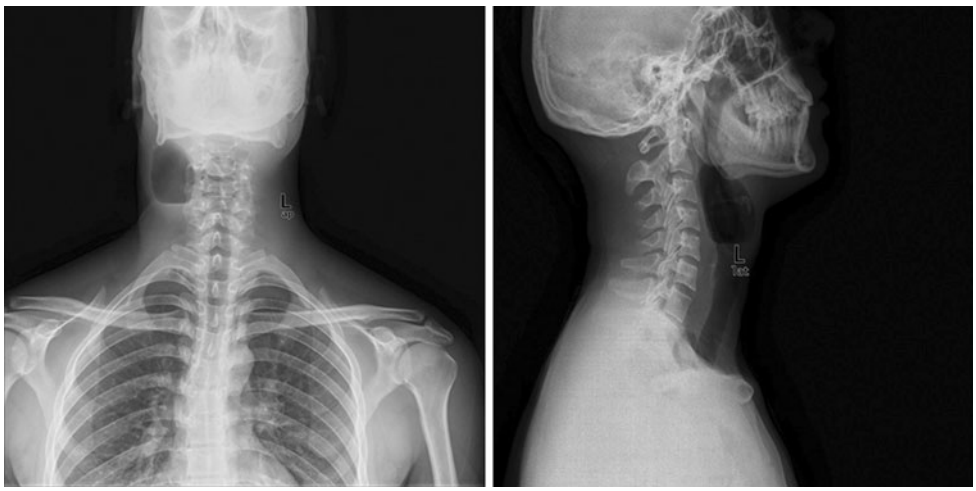

Fig. 1 Plain X-ray neck showing air filled space in the soft tissue neck on right side

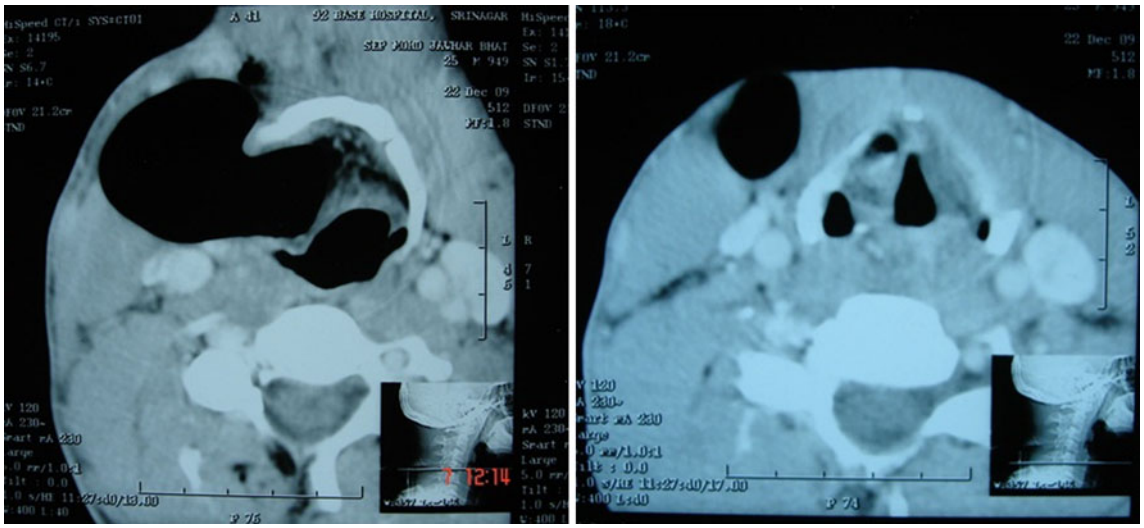

Fig. 2 CT scan revealed air filled space in connection to the endolarynx

Discussion

Laryngocele has been described as an abnormal saccular dilatation of the appendix of laryngeal ventricle of morgagni. Phylogenetically laryngeal saccule is a remnant of lateral laryngeal air sacs of the higher anthropoid apes and is present as an appendage in most human larynxes. The laryngeal appendage arises from the lateral recess in the anterosuperior aspect of the ventricle and extends superiorly through the paralaryngeal space with the thyroid cartilage laterally and ventricular fold medially.

Laryngoceles are uncommon, benign, abnormal cystic dilatation of the laryngeal saccule lined by ciliated pseudostratified cylindrical epithelium with varied goblet cells on a thin basal membrane connected to the ventricle. They represent 5% of all benign laryngeal abnormalities [1, 2].

Laryngoceles are congenital or acquired and may present at any age with peak incidence in 6th decade in males (5:1) [2]. Congenital laryngocele is due to presence of a large ventricular appendix whereas acquired causes can be due to either increased intraglottic pressure caused by excessive coughing, playing wind instrument, etc., or due to blockage of appendicular ostium [1].

They are usually unilateral and Lindsay classified them as internal, external, and combined. Internal laryngoceles may remain small and localized, confined to the interior of the larynx or extend posterosuperiorly into the false vocal cord and the aryepiglottic fold, upper border of the thyroid cartilage to the epiglottis and even into the base of the tongue causing reduction in supraglottic space and appear as a smooth swelling of the supraglottis. In 20% of cases, it penetrates the thyrohyoid membrane at the site of entry of superior laryngeal nerve and vessels and extends as a soft swelling in the

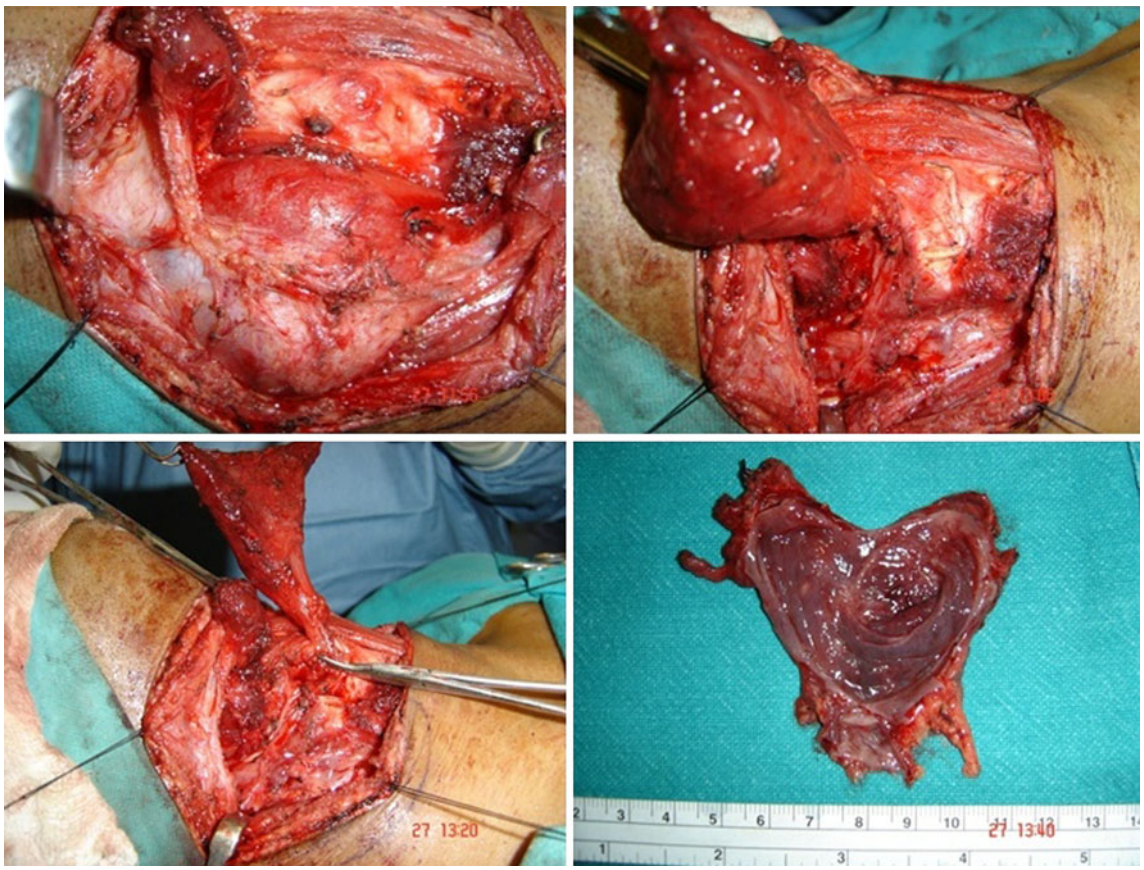

Fig. 3 Surgical excision of the combined laryngocele included removal of part of thyroid ala

subcutaneous tissues of neck at the level of hyoid bone anterior to the sternocleidomastoid muscle as an external laryngocele. Pure external laryngocele do not exist as they are extension of internal laryngocele and when both components are present they are called combines laryngocele.

Laryngoceles secondary to local pathology in the larynx causes a flap-valve mechanism allowing only air entry into the saccule during increased pressure, or retention of fluid secondary to continued mucus production in the distended saccule. Local pathology described in the literature are amyloidosis, ankylosing spondylitis, oncocytic cysts, recurrent respiratory papillomatosis, and unsuspecting laryngeal carcinoma [3, 4].

Laryngocele are usually asymptomatic and may be discovered incidentally on radiographic studies. The main symptoms of laryngocele are recurrent coughing spells, globus sensation, hoarseness to dysphagia, and airway obstruction. In case of infection a laryngocele is formed which may also cause pain, odynophagia, and stridor. An external laryngocele typically presents with a soft, cystic mass in the upper neck which is reducible on pressure with a gurgling sound (Bryce sign) and can be increased on valsalva maneuvre.

Plain radiography can identify large laryngocele but CT scan provides accurate spatial orientation of laryngocele along with identification of any coexisting pathology. Small asymptomatic incidental laryngocele may be observed. Surgery is the treatment of choice and the options include endoscopic and external approach. CO_2 laser has been described for endoscopic approach in deroofing a small internal laryngocele [5]. External approach is for larger, external or combined laryngocele and should involve careful dissection to prevent injury to superior laryngeal pedicle.

Amyloidosis consists of an idiopathic group of disorders in which extracellular deposition of insoluble fibrillar protein (amyloid) occurs in various tissues and organs. Localized amyloidosis is a rare and benign disease presenting in the 4–6th decade of life with a male preponderance (3:1) affecting abdominal organs and structures of the head and neck. In the head and neck, laryngeal amyloidosis is a localized and primary disease commonly affecting ventricles, vocal folds, aryepiglottic folds and subglottis [6]. The lesions are discretely nodular or commonly present as diffuse intralaryngeal subepithelial deposition [6, 7]. Laryngeal amyloidosis leading to

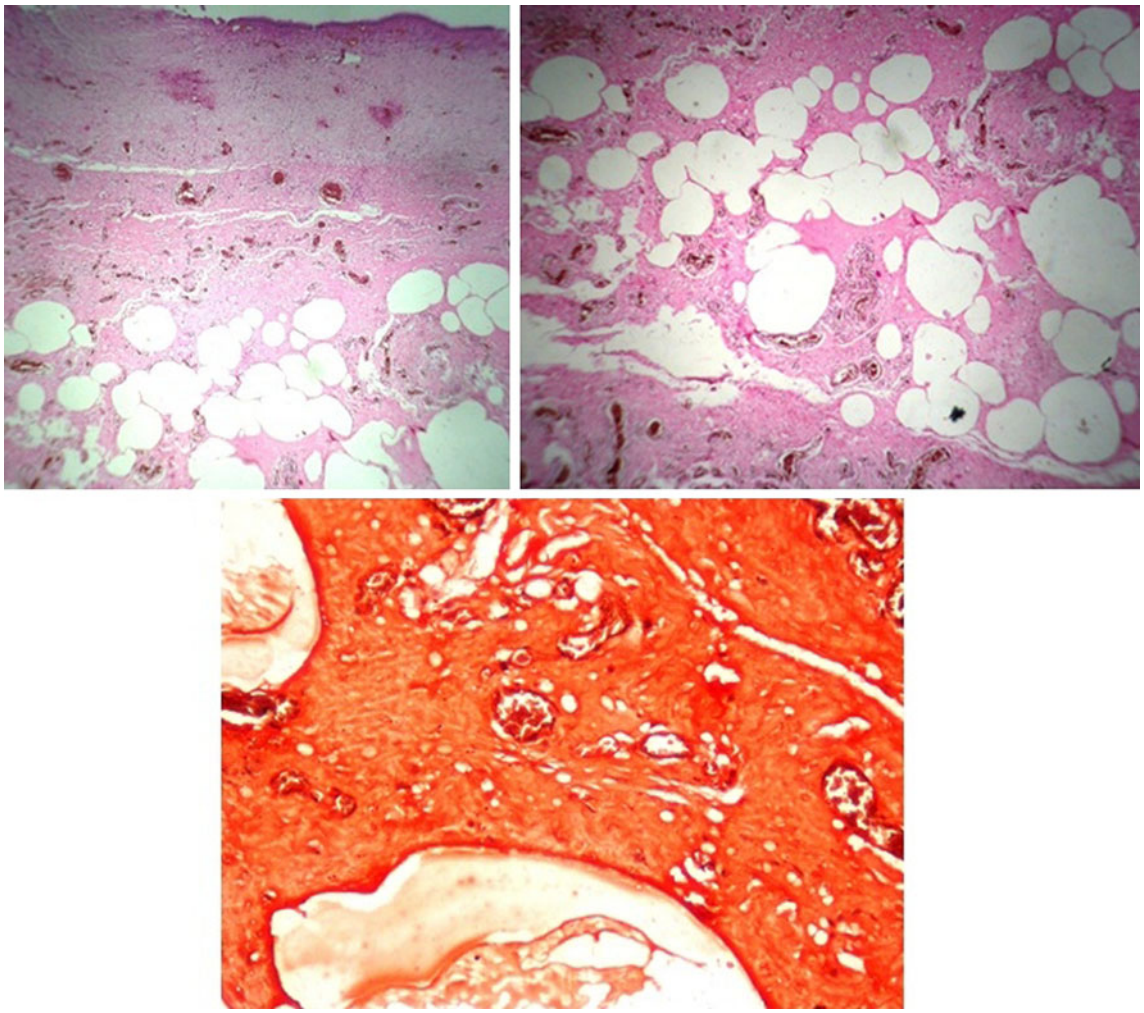

Fig. 4 H&E section shows squamous lining with air filled stroma. *Congo red* stain showed *dark red* staining with *apple green* birefringence on polarized light

blockage of ventricular appendage causing laryngocele is uncommon with our case reports being the fifth reported in the literature [4, 7–9]. Our patient reported with a large combined laryngocele managed by external approach with excellent results.

The case has been reported for its rarity and the necessity of high index of suspicion in cases of laryngocele to rule out local laryngeal inflammatory and neoplastic pathology.

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