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Huge saccular cyst of the larynx: a case report

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Abstract
Lateral laryngeal saccular cysts of the larynx that herniate through the thyrohyoid membrane are quite rare. We present a case of a huge midcervical cyst which occurred as a result of extreme neglect of a lateral saccular cyst. This case provided us with a unique opportunity to characterize the late stages of this pathology. The differential diagnosis of cervical cysts and treatment approaches are discussed.

Key words: Cysts; Larynx

Introduction

The laryngeal saccule is a small diverticulum arising out of the laryngeal ventricle and extending postero-superiorly between the vestibular fold and the thyroid cartilage. The saccular disorders, laryngocele and saccular cyst, involve an abnormal dilatation of the laryngeal saccule. Although the limit of extension of the saccule is normally the upper border of the thyroid cartilage (Donegan, 1993), the relative size of the saccule varies with age. Bartels (1962) found that 25 per cent of newborns have laryngeal saccules that extend beyond the upper border of the thyroid cartilage; Broyles (1959) noted only seven per cent of such extensions in adults. Therefore, some authors define only symptomatic saccules as pathological.

DeSanto (1974) differentiated between laryngoceles, which freely communicate with the laryngeal lumen and are filled with air, and saccular cysts, which have no communication with the laryngeal lumen and are filled with mucus. Three types of laryngocele have been described: internal, external and combined. Saccular cysts are of two types: anterior, which is the smaller of the two and tends to bulge medially, obscuring the anterior portion of the vocal fold, and lateral, which is the larger of the two and tends to expand in a more superior and lateral direction, into the vestibular, vocal and aryepiglottic folds. If the latter type of saccular cyst enlarges sufficiently, it can herniate through the thyrohyoid membrane, similar to a laryngocele, and appears in the neck. In such cases the mass can be palpated at the level of the thyrohyoid membrane, somewhat off the midline. This is then referred to as a combined type saccular cyst.

Most symptomatic patients with a laryngocele or saccular cyst, seek medical attention at an early stage of their disease. The common symptoms are dyspnoea, hoarseness and appearance of a neck mass. We report on a 70-year-old woman with a huge cervical cyst and progressive dyspnoea.

Case report

A 70-year-old woman was admitted to the emergency room of Beilinson Medical Center with progressive dyspnoea and several episodes of syncope which had occurred during the previous few weeks. She had a mass in the lower anterior neck which had developed gradually over the past 30 years. The rest of her medical history was uneventful. On physical examination her temperature was

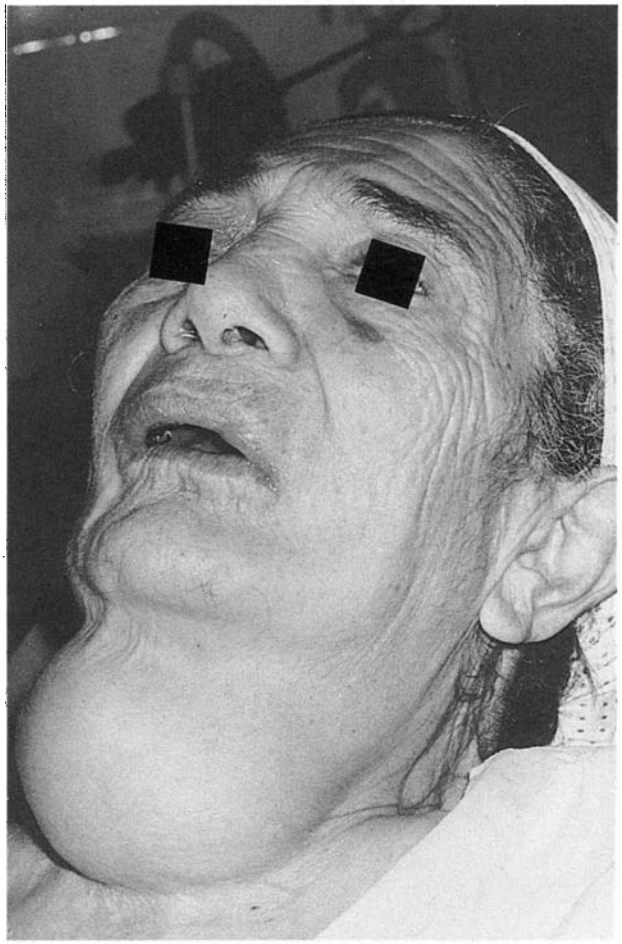

FIG. 1
Patient with a huge midcervical mass.

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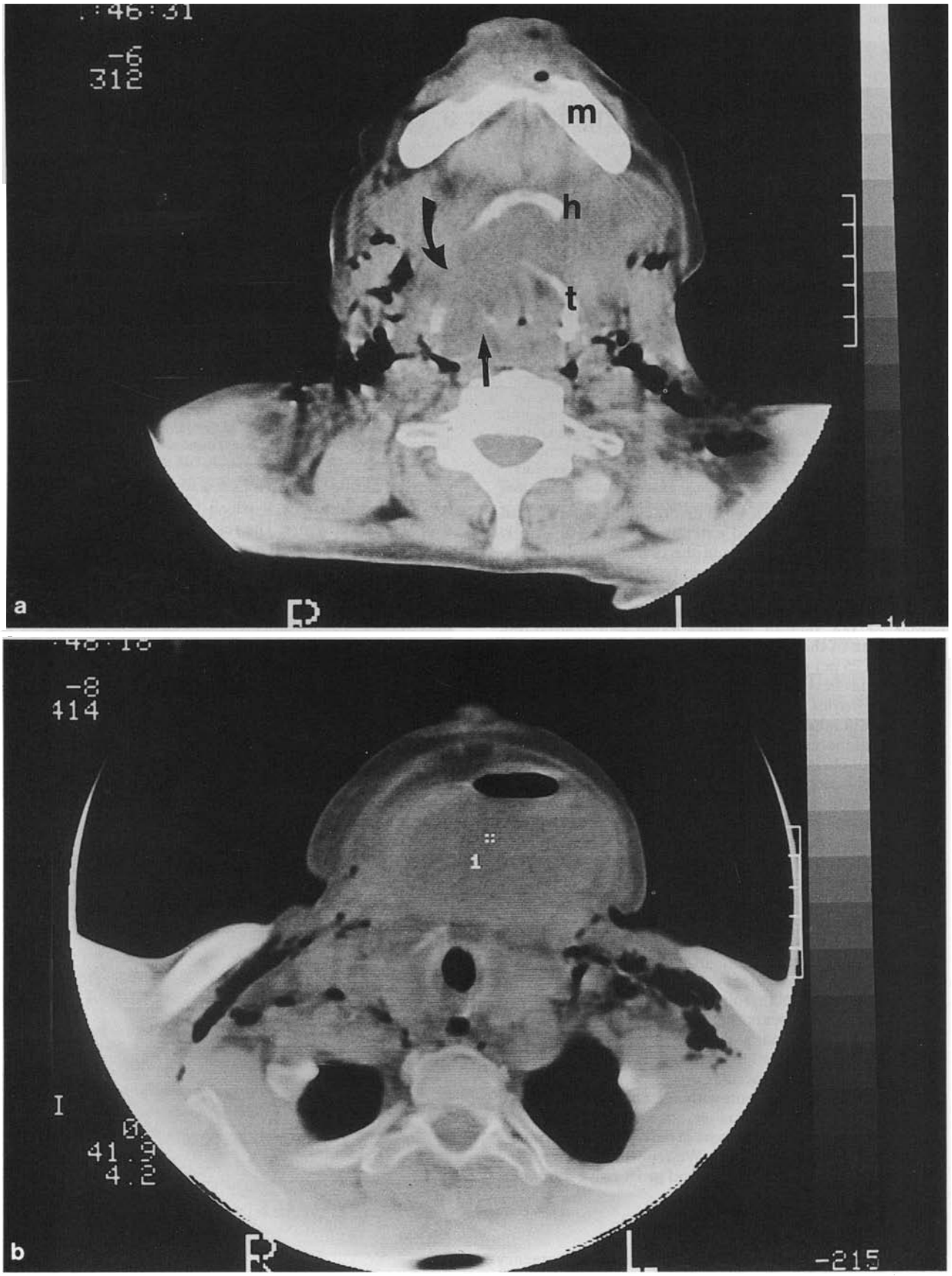

FIG. 2

(a) The external and internal parts of the mass connected through the thyrohyoid membrane (curved arrow). The internal part obstructs the laryngeal vestibule. Mandible (m); hyoid bone (h); thyroid cartilage (t). (b) The external component of the mass has a well defined wall slightly more hyperdense than the content of the mass and has a post-aspiration air-fluid level. The measured attenuation in the cyst is relatively high, possibly due to the viscous content of the cyst.

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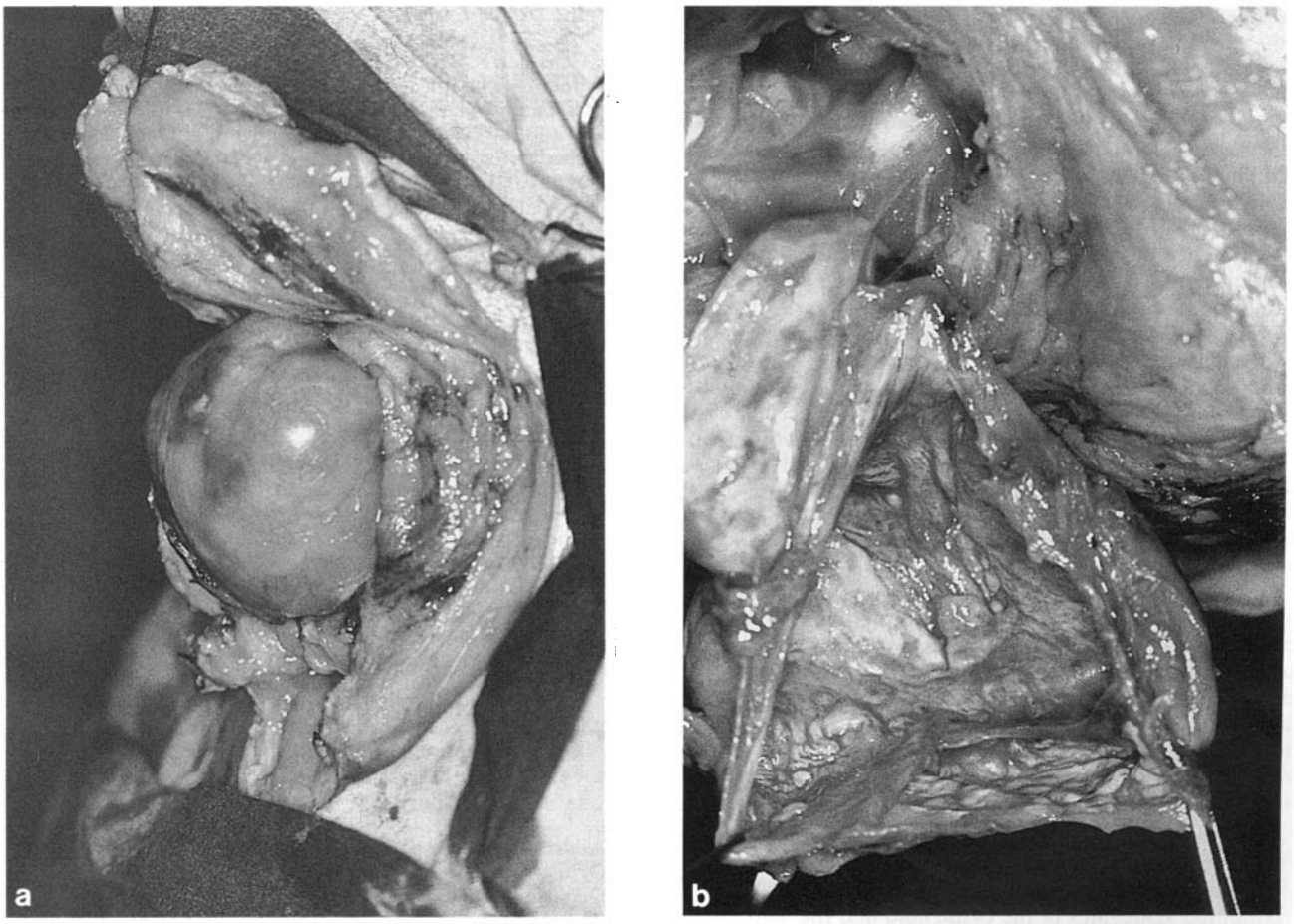
normal: the patient became dyspnoeic on exertion, but had no stridor. There was a nontender cystic mass in the anterior neck measuring 10 × 15 cm, which spready symmetrically to both sides (Figure 1). No other masses were palpated in the neck. Indirect laryngoscopy revealed a large cystic mass in the right aryepiglottic fold, obstructing the laryngeal inlet. The mucosa covering the mass was normal in appearance. The glottis could not be seen. Leucocyte count was normal. Ultrasound of the neck showed a large bilobed cystic mass with an external component occupying the anterior neck, and an internal component filling and obstructing the supraglottic lumen. From the computed tomography (CT) study, after partial aspiration of the cyst, two selected axial sections (Figure 2a and b) characterize the mass and its relationship with the larynx.

The laryngeal mass was aspirated through the mouth, and yielded 40 cc of brown fluid. Both the internal and external components appeared to have shrunk after the aspiration. The following day the patient again became dyspnoeic and indirect laryngoscopy showed that the cyst had returned to its previous size. An additional aspiration yielded 100 cc of the same brown fluid. There was no improvement in dyspnoea after this procedure. The following evening the patient's temperature rose to 38.5°C and she was moderately dyspnoeic. A tracheostomy was performed under local anaesthesia followed by intravenous antibiotic therapy. The next day the patient underwent exploration of the neck through an extended anterior lower neck approach. A large cyst was easily detached from the overlying strap and sternocleidomastoid muscles (Figures 3a and b). It had a stalk which penetrated the thyrohyoid membrane. The stalk was followed through the membrane, and the laryngeal component was excised without thyrotomy. The cyst was filled with a thick fluid. The thyroid gland had a normal appearance. The lymph nodes were not enlarged. Histological examination showed a connective tissue walled cyst, 7.5 × 5 × 3 cm, lined with columnar ciliated epithelium, containing cholesterol crystals and lymphocytes.

Post-operatively, a slow but persistent shrinkage of the supraglottic bulge was noted. The vocal folds were normal and freely mobile. Three days post-operatively the patient was decannulated. At follow-up one year post-operatively she was well, with no evidence of recurrence.

Discussion

Holinger *et al.* (1978) stated that the presence of a laryngocele or a saccular cyst is probable if: (1) a large saccule becomes symptomatic; (2) swelling can be palpated in the neck; (3) swelling is observed by laryngoscopy; and (4) an air-filled sac is shown on a radiography. Most laryngoceles are of the combined type. Close *et al.* (1987) showed a high incidence of an internal laryngocele in asymptomatic patients and suggested that when small, they are confined to the laryngeal lumen. Saccular cysts with both an internal and external component are very rare; only 10 cases have been reported in the literature (Wansa *et al.*, 1990). All these patients presented long histories of



(a) (b)

FIG. 3

Photographs taken during exploration of the neck, showing: (a) cyst detached from the surrounding tissue; (b) excised cyst, opened in order to gain better access to the stalk.

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hoarseness and cervical swelling. Laryngoscopic findings showed swelling of the vestibular and aryepiglottic folds.

The external component of the saccular cyst is palpable superior and lateral to the thyroid lamina. It is expected that if left untreated, the mass would expand in the same antero-lateral plane. The mass we found was located symmetrically around the lower midline of the neck, reminiscent of neglected goitre.

The differential diagnosis of a cyst in the neck includes many pathological conditions. Branchial cleft cyst, cystic hygroma, and cystic metastases are typically more laterally located. Midline or near-midline cysts are: (a) thyroglossal duct cyst and dermoid cyst, which are usually located in the upper two-thirds of the neck and occur in a much younger age group; (b) rare cases of parathyroid or thymic cysts; (c) rare anterior neck lipoma (Ramakantan and Shah, 1989); and (d) thyroid cyst.

The diagnosis of saccular cyst of the combined type is generally based on laryngeal and neck examination combined with operative findings. This diagnostic approach was applied in our case.

Stell and Maran (1975) estimated that about eight per cent of laryngoceles become infected, and most of these were of the combined type. In our case the patient's cyst was infected secondary to the repeated drainage attempts. Once infection occurs, symptoms usually progress quite rapidly.

The aetiology, in most cases, remains unclear. A few cases have been associated with occupations involving increased intraglottic pressure, such as glass blowing or playing musical wind instruments. Saccular cysts may be congenital, caused by atresia of the saccular orifice. This is the current explanation for cysts occurring in childhood (Wansa *et al*, 1990). Cysts in adults are said to be acquired by obstruction of the saccular orifice secondary to trauma, neoplasia, and inflammation. Laryngoceles occur with increased frequency in patients with squamous cell carcinoma of the larynx (4.9–28.8 per cent) (Broyles, 1959; Canalis *et al.*, 1977; Holinger *et al.*, 1978; Micheau *et al.*, 1978; Close *et al*, 1987; Celin *et al.*, 1991). Only one study (DeSanto *et al.*, 1970) associated saccular cysts with laryngeal cancer. In their report two out of 29 patients with anterior saccular cyst had coexistent carcinomas.

Treatment is surgical. Some internal saccular cysts, especially anterior ones, can be removed transorally by endoscopic marsupialization (DeSanto *et al.*, 1970; Holinger *et al.*, 1978; Donegan *et al.*, 1980). Large saccular cysts are approached externally through the thyrohyoid membrane, usually without disturbing the thyroid cartilage (DeSanto *et al.*, 1970; Holinger *et al.*, 1978; Wansa *et al.*, 1990). Another approach, less popular today, is midline or lateral thyrotomy (Booth and Birck, 1981). The decision regarding an endoscopic or an external approach depends on the classification and size of the cysts and individual patient factors. In our patient we used the external approach through the thyrohyoid membrane without opening the thyroid cartilage. As endotracheal intubation can result in rupture and aspiration of cyst content and possible post-operative laryngeal oedema, a temporary tracheotomy is usually performed. In our case, the progressive dyspnoea was an additional justification for following this procedure.

The histological appearance of laryngeal cysts is of little help in their diagnosis, unlike the structural findings (DeSanto, 1974). The presence of cholesterol crystals and lymphoid aggregates in the cystic fluid and the wall of the cyst is nonspecific and has also been associated with thyroglossal, thyroid, and thymic cysts.

The adult larynx can bear masses of up to about 4 cm; with larger cysts, dyspnoea usually ensues. External cysts tend to be diagnosed earlier. DeSanto (1974) presented a laryngocele that extended from the thyroid ala to the tail of the parotid gland. He assumed the patient had had the cyst for many years.

With the recent tendency toward early surgical intervention, we seldom observe the fully extended, natural history of many pathological conditions. The outcome of a long protracted course may cause a change in the clinical presentation and lead to misdiagnosis. The above rare case provided an opportunity to study the natural history and outcome of a longstanding combined-type saccular cyst.

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