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CASE REPORT

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Sudden Death from Saccular Laryngeal Cyst

ABSTRACT: Laryngeal cysts are benign, uncommon lesions of the larynx that have been reported on rare occasions to cause sudden death in infants and adults by acute airways obstruction. In this report, we document the sudden death of a 36-year-old woman from a previously undiagnosed, asymptomatic laryngeal saccular cyst that presented with acute, and consequent fatal, airway obstruction. Difficulty during intubation, both in theater and in emergency settings, is a frequent presenting problem. This can have significant medicolegal implications in determining possible negligence. The diagnosis, classification, and management of such cysts, and their importance to both the forensic pathologist and clinicians are discussed.

KEYWORDS: forensic science, laryngeal cyst, sudden death, airway obstruction

Case Report

The decedent was a 36-year-old morbidly obese woman (BMI = 41 kg/m^2) who had given birth 5 months ago. Her pregnancy and delivery were uncomplicated. She had no other significant medical history and had reportedly been well since the birth. On the day of her death, she had a 5-h history of a sore throat and palpable lump on the right side of her neck. She developed a sudden onset of severe dyspnea and hemoptysis before losing consciousness. Paramedics were called, but she could not be resuscitated. It was noted that an endotracheal tube could not be inserted during the resuscitation attempt.

Autopsy

At autopsy, the decedent was found to have a 40 × 15 mm multicolored mass obstructing the lumen of the larynx in the supraglottis (Fig. 1). The mass originated from the lateral wall of the supraglottis, and contained areas of recent hemorrhage, edematous tissue, and pultaceous material. The remainder of the larynx was normal and there were no injuries to the hyoid bone, thyroid cartilage, or cervical spine.

Microscopic examination of the larynx showed a large polypoid cystic lesion with a lining of squamous epithelium and mucinous material in its lumen (Fig. 2). There were extensive acute inflammatory cells and hemorrhage within the cyst's wall, which is consistent with the patient's acute deterioration (Fig. 3). There was no evidence of atypia or neoplasia. The features were those of a saccular cyst of the epiglottis with inflammatory change.

Other abnormalities noted at postmortem included pulmonary edema, features of hypertensive disease, mild atherosclerosis, and hepatomegaly with normal histology.

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Discussion

Laryngeal cysts are relatively uncommon lesions that can occur at any age. The peak incidence is during the sixth decade of life, with no gender predominance (1,2). While most cysts are managed through observation or simple surgical removal, they have, on rare occasions, been reported to cause sudden death. Reported deaths have occurred predominantly in the infant population due to the small caliber of their airways, whereas adult fatalities are rare (3).

Clinically laryngeal saccular cysts can be asymptomatic and picked up as an incidental finding or they can present with a spectrum of symptoms that depend largely on the size of the cyst and the age of the patient. In infants, inspiratory stridor and dyspnea are frequently noted (4), whereas symptomatic adults more commonly present with hoarseness, throat pain, dysphagia, and milder respiratory symptoms (1–3). Both infants and adults can develop complete airway obstruction requiring emergency

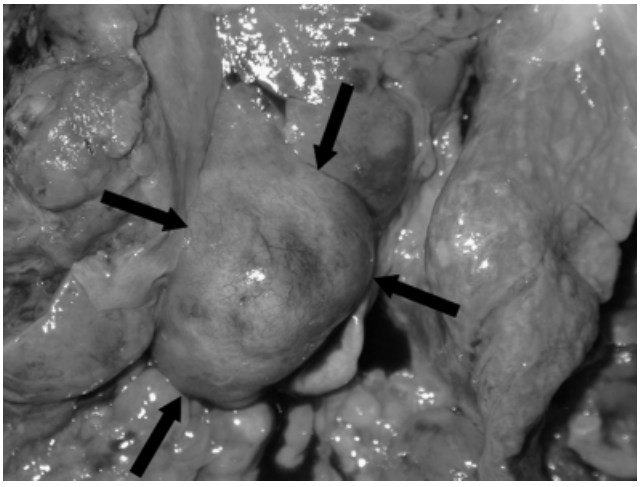

FIG. 1—*Laryngeal cyst obstructing the larynx (arrows).*

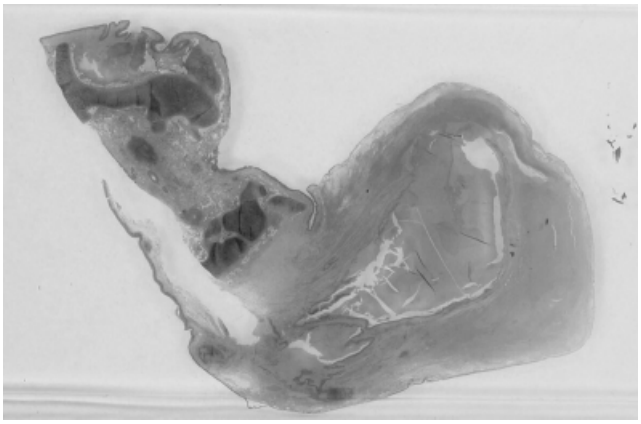

FIG. 2—*Whole-mount section of laryngeal cyst and adjacent laryngeal structures. H&E staining.*

surgical intervention. Because of this spectrum of symptomatology, it follows that the clinical signs may also be quite variable.

In the case we present, the patient had no previous documented symptoms, indicating that the cyst was asymptomatic. It is possible that the cyst was too small to manifest typical symptoms until just prior to death when it rapidly enlarged as a result of an inflammatory process. However, it is also possible that she could have experienced symptoms that were masked by her obesity. For example, it is possible that the patient could have had a background level of dyspnea associated with obesity, which, if progressive, may not have caused any alarm to the patient as she could have attributed it to her weight gain.

Laryngeal cysts are generally classified using the system proposed by DeSanto et al. (5), which is based on the site, size, content, and relationship with the laryngeal mucosa. DeSanto et al. divide laryngeal cysts into ductal and saccular; a third type, the thyroid cartilage foraminal cyst, was described as an isolated case. Ductal cysts are the most common laryngeal cysts and they can occur anywhere within the larynx where mucosal glands are present. These cysts arise as a consequence of mucus retention within the collecting ducts. They are generally small lesions, usually less than 1 cm in diameter, and quite superficial (3).

In contrast, laryngeal saccular cysts are a rare anomaly. These cysts arise as mucosal dilations of the saccula larynges. The communicating orifice between the cyst and the laryngeal ventricle is either completely obstructed or severely stenosed. This is an important characteristic as it distinguishes it from a laryngocele. The etiology is not completely understood but it is thought that it involves congenital and acquired factors. Acquired factors include obstruction of the orifice caused by inflammation, infection, or neoplasm, whereas proposed congenital factors include embryological malformations (6). The cyst itself is typically lined with columnar epithelium, but can have a squamous lining, which contain seromucinous glands in the submucosa and as a consequence the saccular cyst is filled with mucus.

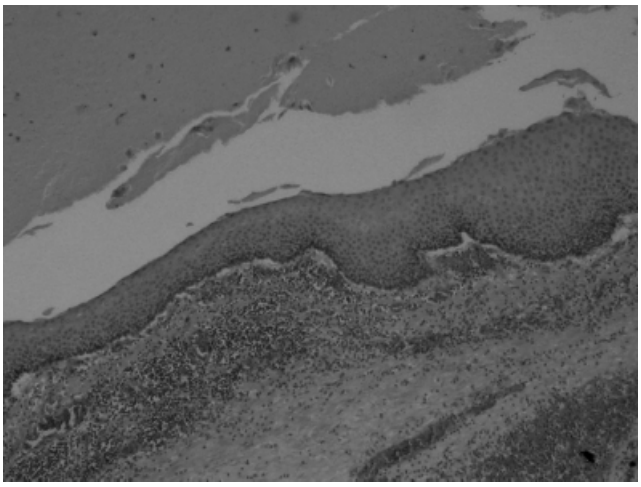

FIG. 3—*Photomicrograph of the wall of cysts. H&E staining.* × *4 objective.*

The saccular cysts can be classified further based on anatomical location (5). Lateral saccular cysts, being more common, extend into the region of the false cords and aryepiglottic fold. These can be quite large in size. Anterior saccular cysts are typically located at the anterior ventricle near the orifice and are characteristically quite small.

Laryngoceles are evaginations of the mucous membrane of the laryngeal saccule. They differ from saccular cysts in that they are air-filled cavities and they communicate directly with the laryngeal ventricle.

While DeSanto et al.'s classification system is widely accepted, a more recent classification has been proposed by Newman et al. (1). This system divides laryngeal cysts into: tonsillar cysts, epithelioid cysts (subgroups being saccular and ductal), and oncocytic cysts. Tonsillar cysts have abundant follicular lymphoid tissue with squamous-lined crypt-like structures. Epitheliod cysts have a prominent epitheliod component and focal follicular lymphoid aggregates. Oncocytic cysts demonstrate metaplasia of the larynx with cyst formation that is associated with a high rate of recurrence. Newman et al.'s system proposes recognizing the presence of lymphoid tissue and its possible involvement with the pathogenesis of laryngeal cysts. The system also allows classification of a cyst when there has been disturbance of the relationship between the cyst and surface mucosa such as during removal, an element needed for classification with DeSanto et al.'s system. In the present case, lymphoid tissue was not prominent and there was no oncocytic change.

The diagnosis of laryngeal cysts relies on the clinician having an awareness of the condition and a degree of suspicion, particularly in cases of acute dyspnea. A complete history is necessary as is examination. The cysts can occasionally be palpated; however, visualization using indirect or direct laryngoscopy and CT/MRI imaging is required for diagnosis.

At autopsy, the diagnosis of laryngeal cysts equally relies on the pathologist's knowledge of the condition and a thorough postmortem examination being conducted. Dada and colleagues illustrate the importance of this particularly in forensic cases.

Dada (2) describes the case of a 32-year-old woman who was sexually assaulted and found dead the following day. Investigations led to the arrest of two men charged with sexual assault and murder. Initial postmortem examination found "no cause of death." Sexual assault was confirmed on vaginal smears. A subsequent postmortem examination found the cause of death to be laryngeal obstruction secondary to an epiglottic cyst. These findings exonerated the two men from homicide charges.

Harruf and Bell (3) describe the case of a 36-year-old woman who died suddenly and was found in a street. There were suspicious circumstances surrounding her cause of death. The only reported medical history was an admission to an emergency department for the treatment of asthma 2 years back. At autopsy, no evidence of asthma was found and the cause of death was found to be asphyxia secondary to an obstructing laryngeal

cyst. This eliminated any sinister involvement in the death of the deceased.

Both these cases highlight that careful examination of the neck and larynx should be conducted in cases of sudden death.

As seen with our patient, there was difficulty with intubation—this also has medico-legal implications in determining medical negligence in cases where there is difficulty in intubations during the induction of anesthesia or accidental rupture.

Laryngeal saccular cysts that are asymptomatic are often managed by observation in order to assess for any size changes and also because of the increased incidence of dilated saccules reported in patients with laryngeal cancer (7).

Symptomatic saccular cysts in adults can be managed by endoscopic marsupialization of the dome of the cyst with laser ablation of the remaining cyst wall, or by external excision. If there is severe airway obstruction, then an urgent tracheotomy is required before excision. Recurrences, however, are noted to occur with marsupialization and there is increased morbidity associated with the external approach. CO_2 laser excision is a third management option available. It is well recognized that marsupialization or simple aspiration alone often leads to recurrences (8). For larger or recurrent cysts, the treatment usually requires a submucosal resection through an external approach; however, Hogikyan and Bastian (9) challenge this, reporting the use of CO_2 laser excision for cysts of this nature.

In summary, we present a case of sudden death in a morbidly obese 36-year-old woman from an acute airway obstruction by a laryngeal saccular cyst. We have discussed classification, diagnosis, and management and highlighted the importance of the condition to both the forensic pathologist and the clinician, as it is a rare cause of sudden death that is amenable to surgical intervention.

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