Hypopharyngeal lymphoepithelial cyst in a patient with a history of difficult intubation

Introduction

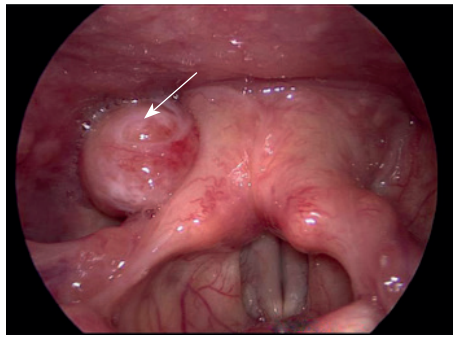
Most hypopharyngeal tumours are malignant, and over 95% of hypopharyngeal cancers are squamous cell carcinomas (Muir and Weiland, 1995). Benign tumours in the hypopharynx are rare, and the two most common are fibrolipoma and leiomyoma (Choi et al, 2014). Hypopharyngeal lymphoepithelial cysts have seldom been reported in the literature, and their incidence has never been explored because of their rarity. In the head and neck region, the frequency of lymphoepithelial cysts is greatest in the parotid gland and the lateral aspect of the neck.

Laryngoscopic examination may help distinguish benign hypopharyngeal lesions from squamous cell carcinoma. However, differentiating lymphoepithelial cysts from other benign hypopharyngeal lesions remains a challenge for the treating physician. Because benign hypopharyngeal lesions look macroscopically like retention cysts, the precise diagnosis is usually made by pathological analysis (Jungehulsing et al, 2000).

Discussion

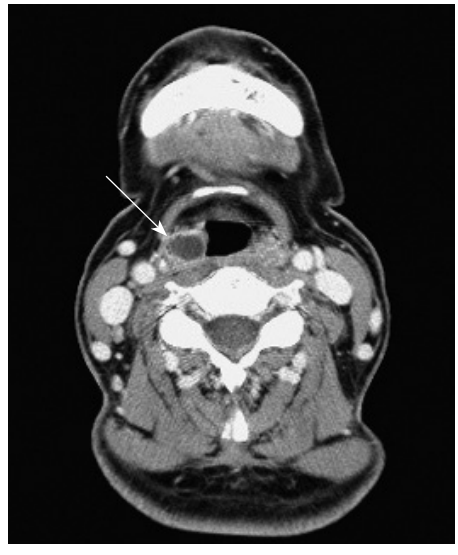
The timing and selection of management strategies for hypopharyngeal lymphoepithelial cysts remain controversial. There is no gold standard for treating hypopharyngeal lymphoepithelial cysts. Given their benign nature, observation of patients without symptoms resulting from lymphoepithelial cysts might be acceptable. Surgical intervention might be indicated for patients with symptoms related to lymphoepithelial cysts. The main treatment for hypopharyngeal cysts is endoscopic total excision. In some cases, giant obstructing cysts may require open field surgery for complete resection (Kawaida et al, 1995). In the current case transoral carbon dioxide laser-assisted endoscopic surgery was performed. The patient's symptom of foreign body sensation resolved 3 weeks after the operation and had not recurred at 7 months follow up.

Figure 1. Endoscopic image of the hypopharyngeal mass. The 70° indirect rigid laryngoscope revealed a smooth-surfaced round mass in the hypopharynx (arrow).



The pathogenesis of lymphoepithelial cysts is not clear. The most widely accepted two theories are epithelial entrapment in lymph nodes and tonsillar crypt obstruction. In a series of studies, Bhaskar concluded that lymphoepithelial cysts were epithelium inclusions located in the lymphoid tissue with cystic alteration (Bhaskar and Bernier,

Figure 2. Computed tomography of the neck showed a cystic mass (arrow), sized about 1.5 x 1.5 cm in diameter, in the hypopharynx.



CASE REPORT

A 53-year-old woman with a medical history of uterine myoma and difficult intubation attended the authors' otolaryngology clinic, complaining of the sensation of something being caught in her throat, a feeling that had progressively worsened over 2 weeks. The patient had undergone a myomectomy under general anaesthesia in February 2015, and noted the lumping feeling 3 months later in May 2015. She did not have dysphagia, choking, dyspnoea, history of smoking or gastro-oesophageal reflux disease.

Flexible nasendoscopy revealed a well-circumscribed mass with smooth mucosa lining in the right pyriform sinus (*Figure 1*). Both vocal cords were freely movable. Computed tomography of the neck showed a well-defined, round cystic mass in the right pyriform sinus, approximately 1.5 cm in diameter (*Figure 2*). There was neither enlarged lymph nodes, a visible tract communicating with the hypopharyngeal mass, nor continuity with the laryngeal ventricle.

Transoral carbon dioxide laser-assisted endoscopic surgery for the hypopharyngeal mass was performed under direct laryngoscopy. During the operation, the authors noticed that the mass was attached to the right lateral pharyngeal wall with a pedicle. Histopathological examination showed a cystic cavity lined by stratified squamous epithelium with an abundance of surrounding lymphoid tissue including germinal centres (*Figure 3*).

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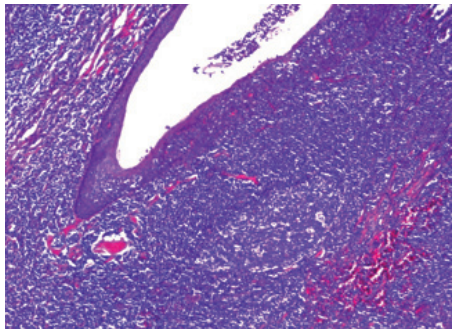
Figure 3. Histopathological findings of the lymphoepithelial cyst: stratified squamous epithelium with abundant surrounding lymphoid tissue, including germinal centres (haematoxylin-eosin, original magnification x100).



1959; Bhaskar 1966). These findings were supported by Vickers and von der Muhll (1966) with surgical autogenous transplantation of buccal epithelium into lymph nodes of hamsters. In contrast, Knapp (1970) proposed that oral lymphoid tissue had epithelial-lined crypts, and obstruction of the crypt caused crypt dilation leading to the eventual formation of a cystic cavity. The lining epithelium of lymphoepithelial cysts has striking similarities with the crypt epithelium of palatine tonsils (Crocker and Jenkins, 1985), and this observation provided evidence to support Knapp's theory. Baba et al (2010) reported a patient with chronic pharyngolaryngitis having an incidental finding of tonsillar tissue in the hypopharynx, which may be the precursor of hypopharyngeal lymphoepithelial cysts.

In the current case, the patient had a history of difficult endotracheal intubation before the diagnosis of hypopharyngeal lymphoepithelial cyst. The formation of her cyst could be partially attributed to repeated attempts at intubation, causing implantation of sloughed epithelium into the nearby lymphoid tissue. In an animal study Vickers and von der Muhll (1966) showed that implantation of epithelial cells in lymph nodes led to formation of lymphoepithelial cysts.

The symptoms caused by benign hypopharyngeal tumours usually have an insidious onset and may vary depending on the location and the size of the lesions. Because patients with hypopharyngeal cysts are usually asymptomatic when the cysts are small, the incidence is possibly underestimated. This case highlights that mucosal injury, especially after an endotracheal intubation, might be correlated with the occurrence of hypopharyngeal lymphoepithelial cysts. Further studies are needed to elucidate the association of lymphoepithelial cysts and endotracheal intubation. This case suggests that if patients who have a history of endotracheal tube intubation are found to have a hypopharyngeal mass, lymphoepithelial cyst should be included in the differential diagnosis. **BJHM**

LEARNING POINTS

- If a patient has a persistent sensation of a foreign body in the throat after endotracheal tube intubation, a hypopharyngeal lymphoepithelial cyst should be included in the differential diagnosis.
- The optimal treatment of hypopharyngeal lymphoepithelial cysts is endoscopic total excision. Open field surgery for complete resection may be indicated in some cases of giant cysts.

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