CASE REPORT

Adult bronchogenic cyst of the neck presenting as large neck abscess

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Summary Bronchogenic cysts are rare congenital malformations originating from the ventral foregut during its development. Cervical bronchogenic cysts are often asymptomatic and are generally found in childhood. We report a case of bronchogenic cyst with abscess formation in a 67-year-old woman who presented with progressive dysphagia. The cyst was completely excised. To the best of our knowledge, this is the third documented case of bronchogenic cyst leading to neck abscess in an adult.

1. Introduction
Bronchogenic cysts are congenital anomalies believed, from an embryological aspect, to originate from abnormal budding of the tracheobronchial component during foregut development. The final location is related to the timing of budding, and most bronchogenic cysts are located in the mediastinum and remain asymptomatic. Neck bronchogenic cysts are rare and are generally detected in childhood. They are often found incidentally at an early age, but may have symptoms including stridor in neonates, dyspnea on exertion, dysphagia, and recurrent infection. Only two cases of neck abscess complicating neck bronchogenic cyst have been reported previously. Here, we report a case of a large neck abscess arising from a bronchogenic cyst in an adult. The patient underwent surgical excision, and no recurrence was noted during a 12-month follow-up period.

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2. Case report

A 67-year-old woman was referred to our hospital for evaluation of neck pain with dysphagia. She had suffered from progressive dysphagia and sore throat for 5 days before admission. There was no history of any systemic disease. On examination, the body temperature was 37.3°C, the systolic and diastolic blood pressures were 124 and 82 mmHg, respectively, the pulse was 106 beats/minute, and the respiratory rate was normal. A protruding soft mass without obvious local heat measuring approximately 3 cm in diameter was located in the suprasternal area. The patient reported mild pain during palpation. There were no palpable neck lymph nodes or other oropharyngeal lesions. Leukocytosis (white blood cells: 17.76 \(10^3/\mu L\); segmented: 86.6%) was noted, but the remainder of the complete blood count, urinalysis, and serum levels of electrolytes were within normal limits. Plain radiographs of the chest showed a localized air–fluid interface in the upper mediastinum (Fig. 1), and a computed tomography (CT) scan revealed a 5-cm cystic lesion containing an air–fluid level located near the trachea and esophagus (Fig. 2A), extending from the neck to the thoracic inlet (Fig. 2B). The trachea and esophagus were displaced to the left side (Fig. 2C). Cervical abscess of unknown origin was suspected, and possible causes were esophageal leakage and infection of a diverticulum; however, a subsequent esophagography revealed no visible fistula or diverticulum formation (Fig. 2D). Echoguided needle aspiration was performed, and pus-like fluid was aspirated for cytology and culture examinations. A cytological examination yielded numerous polymorphonuclear leukocytes and scant histiocytes and ciliated columnar cells; Gram staining revealed only pus cell without any visible bacteria (Fig. 3A). The cyst was surgically removed through a paramidline incision with blunt dissection from the surrounding anatomical structures including the sternocleidomastoid muscle, common carotid artery, external jugular vein, and vagus and recurrent laryngeal nerves. A histopathological examination was performed, which revealed a bronchogenic cyst composed of fibrotic stroma lined by ciliated respiratory epithelium with a fibrinopurulent exudative coating (Fig. 3B). Acute and chronic inflammation, lymphoid follicles, hemorrhage, necrosis, and granulation tissue formation were also seen. Postoperatively, the patient suffered unilateral vocal cord palsy with voice change, but no swallowing disorder was noted on postoperative esophagography. The hoarseness resolved without treatment 10 months later.

3. Discussion

Bronchogenic cysts are rare congenital malformations arising from abnormal budding of the primitive tracheobronchial tube during the development of ventral foregut. Corresponding to the rostral to caudal development of the tracheobronchial tree, the earlier abnormal budding are located at higher levels, whereas the later ones are located peripherally.

Neck bronchogenic cysts are less common, and are mostly detected in childhood. Ustundag et al reported 45 cervical bronchogenic cysts, and only six of the patients were older than 25; furthermore, the cysts remained asymptomatic in most cases, which correlated with the location in the mediastinum or lung parenchyma. Although previous reports emphasized that bronchogenic cysts are usually asymptomatic, more recent series have revealed that the majority of bronchogenic cysts in adults ultimately become symptomatic. A fistulous opening that drains mucoid material may be observed, or the cysts may fluctuate in size and become infected, presenting as neck abscesses. However, only two adult cases have been reported previously.

Differential diagnoses for bronchogenic cyst are branchial cleft cyst, thyrmic cyst, thyroid cystic papillary carcinoma, thyroglossal duct cyst, cystic hygroma, and tracheal diverticulum. These conditions mostly present with cystic components with an air–fluid level on CT images, and a distinction among these lesions is primarily based on location and histology. Therefore, a histopathological examination is still needed for definitive diagnosis. Aspiration cytology is an alternative diagnostic tool for a neck mass, and diagnostic components in the bronchogenic cyst are ciliated columnar epithelial cells. In our case, these were found retrospectively after surgical resection and pathological confirmation. Therefore, sensitivity of aspiration cytology may be low because relatively few epithelial cells may be scattered in the cystic components. Previous studies also revealed a low yield of fine-needle aspiration (FNA), which may produce a false-negative result in pre-operative differential diagnosis.

Therapeutic aspiration may temporarily relieve the symptoms, but the recurrence rate is high. Surgical resection may be difficult in symptomatic cases because of
severe pericystic adhesion,\textsuperscript{2,6} as in our case, making prevention of intraoperative recurrent laryngeal nerve injury difficult, as was observed in all the three documented cases (Table 1). Despite the observed high complication rate, complete resection should be attempted to avoid recurrence following incomplete resection, and the patient should be informed about surgery-related vocal cord palsy preoperatively. In previous cases, postoperative vocal cord palsy was treated using Teflon injection into the true vocal cord,\textsuperscript{5} and the patient underwent conservative speech rehabilitation.\textsuperscript{3} However, Teflon is no longer used in laryngopasty, because of poor tissue compatibility, and permanent substances including hydroxyapatite, silicone, and hyaluronic acid are favored for vocal cord injection. Intraoperative nerve monitoring, which is widely used in thyroid surgery for recurrent laryngeal nerve detection, is also valuable for preventing such complications.

A malignancy arising from bronchogenic cysts is rare and mostly seen in intrathoracic cysts. Reported malignancies have included adenocarcinoma, squamous cell carcinoma,
anaplastic carcinoma, and various types of sarcomas. Only one documented malignancy has been reported arising from a cervical bronchogenic cyst; however, this was a poorly differentiated adenocarcinoma with thyroid invasion, which was initially misdiagnosed as thyroid papillary carcinoma by ultrasound-guided FNA.8

In conclusion, neck bronchogenic cyst should be considered as the origin of deep neck infection or neck abscess, particularly with the presence of thoracic extension. Surgical resection is the treatment of choice for neck bronchogenic cyst based on the risk of abscess formation and the possibility of malignant transformation.

References


