Case Report
A cervicothoracic esophageal duplication cyst in an infant and review of literature

Cai-Yun Long, Yu-Zuo Bai

Department of Pediatric Surgery, Shengjing Hospital, China Medical University, Shenyang, Liaoning, PR China
Received October 9, 2015; Accepted November 20, 2015; Epub January 1, 2016; Published January 15, 2016

Abstract: Cervical esophageal duplication cysts are uncommon congenital malformations, and those with air-fluid level are rarely reported. We describe here a case of cervicothoracic esophageal duplication cyst manifesting air-fluid level on Computed Tomography (CT) in a 6-month-old girl. The preoperative diagnosis was suspected as esophageal duplication or abscess. The postoperative pathological diagnosis was esophageal duplication cyst with infection. This report is to provide insights that esophageal duplication cysts should be included in the differential diagnosis of cervical cystic lesions.

Keywords: Esophageal duplication, cyst

Introduction
Duplication of esophagus is uncommon, and rarer is cervical esophageal duplication cyst. Usually, the lesion shows up as fluid-filled cystic mass on CT, and sometimes communication with esophagus can be found. A 6-month-old infant with cervicothoracic esophageal duplication cyst manifesting air-fluid level on CT is described here.

Clinical data
A 6-month-old girl was found with superior mediastinal neoplasm in the 4th antenatal examination. She remained asymptomatic after birth. At the age of 6-month, the patient was reexamined by CT showing the mass larger than before. The radiographic imaging revealed a mass about 2.4*1.6 cm located in superior mediastinum and neck with an air-fluid level on CT (Figure 1). To explore the origin of the lesion and its relation with esophagus, upper gastrointestinal contrast was executed then, but the result presented no abnormal signs. The diagnosis was not confirmed but cervical duplication cyst was suspected.

Cystectomy was performed in cervical approach. The mass originated from pharyngeal esophagus down into mediastinum at the level of suprasternal fossa. There was a communication between the cyst and esophagus. During operation, the cyst was found containing air, which was consistent with the manifestation of air-fluid level on CT. Finally, the lesion was completely resected (Figure 2).

Histological examination revealed that the excised tissue was lined by squamous epithelium. Smooth muscle was found in the wall. Hyperplasia of interstitial tissue and inflammatory tissue were also embedded on pathological sections (Figure 3). The final diagnosis was confirmed as esophageal duplication cyst with infection. The patient recovered well. No complications occurred postoperatively and during 12-month follow-up period.

Discussion
Esophageal duplication cyst is a rare congenital anomaly occupying 8.3% of all alimentary tract duplications with a male predominance of 2:1 [1-3]. Frequently, the cysts are located in mid to distal esophagus [2]. Those located in the upper 1/3 of esophagus are second rarest [3]. The final diagnosis is made by histological findings as follows: a well-developed coat of smooth muscle; an epithelial lining representing some portion of the alimentary tract (unless destroyed
Cervicothoracic esophageal duplication cyst

The majority of patients with esophageal duplication cysts are asymptomatic and some are found incidentally or prenatally by radiographic examinations [5]. However, those located in neck are likely to be relative to clinical symptoms such as dysphagia, respiratory distress, cervical mass, or even stridor based on the location and size of the lesions [2, 3, 6, 7]. In our case, the patient was found with a lesion in neck prenatally but remained asymptomatic in her 6-month life after birth.

Imaging examinations may contribute to the clinical diagnosis. Chest radiograph can show space occupying lesions and deviation of trachea [8]. Esophagography, ultrasonography, CT and magnetic resonance imaging (MRI) are

Figure 1. Enhanced CT revealed that a cystic mass with air-fluid level occupied superior mediastinum and neck (A and B). Because of the horizontal position during the examination, the air-fluid level cannot be reflected on the coronal images (C).

Figure 2. General observation of specimen (A and B): the lesion is cystic with a smooth outer wall and a coarse inner wall.

by infection); an intimate attachment to some portion of the tract [4].
Cervicothoracic esophageal duplication cyst

also commonly used during the procedure of diagnosis. CT and MRI can better demonstrate the size, exact anatomic location of the lesion, and the relationship with adjacent tissues [7]. Upper gastrointestinal contrast may detect a cystic mass communicating with esophagus which highly indicates the existence of esophageal duplication cyst, or show extrinsic compression on the esophagus [9]. With the improvement of accuracy and precision, ultrasonography was reported to be effective in detecting esophageal duplication cysts [10]. But the use of ultrasonography in esophageal duplication cysts located in larynx or neck seems limited [9].

Esophageal duplication cysts can be intraluminal or external of esophageal cavity. About 80% esophageal duplication cysts have no communication with the esophageal lumen [3]. Most lesions manifest cystic mass on images. And because of the secretion of the wall or infection of the mass, the overwhelming majority of esophageal duplication cysts are fluid-filled on CT or MRI.

Air-fluid level in esophageal duplication cyst as in our case is extremely rare. Till now, we found only two reports in which esophageal duplication cysts with air-fluid level on CT were presented [9, 11]. In Wootton’s case, air-fluid level on CT and outpouching of contrast into cyst on contrast esophagram were thought to be due to communication with the lumen of esophagus. What is different in our case is that although air-fluid level was revealed on CT and communication with esophagus was confirmed during operation, contrast esophagram showed no abnormal sign. We speculated that it was because the inlet of the lesion was too skinny or epiglottis blocked the entrance, which prevented contrast agent going into the cyst.

Generally, cervical lesions with air-fluid level are considered to be abscess, infected piriform fossa fistula or tracheal cyst. For cervical esophageal duplication cyst with air-fluid level, misdiagnosis can be easily made and wrong disposition may be taken. For example, puncture or drainage of abscess may lead to satisfactory effect, but for esophageal duplication cyst, treatments mentioned above are inappropriate or even lead to severe consequence. Considering that there is a tendency of occurrence of complications (such as perforation, infection, bleeding, structural compression) and malignant progression, once the esophageal duplication cysts were found, complete resection is recommended [4, 12, 13]. Thoracotomy, antero-superior cervico-thoracic, “trap door” incision, and thoracoscopy are surgery approaches. For those located in thoracic inlet which extends into neck and superior mediastinum, cystectomy in cervical approach is more applicable for better exposure and one-time complete excision [8, 14]. However, Lee thought that thoracoscopic approach should be considered when the cyst extends to upper mediastinum [15].

In conclusion, for cervical mass with air-fluid level, esophageal duplication cyst should be taken as a differential diagnosis and operation in time should be performed.

 Disclosure of conflict of interest

None.

 Address correspondence to: Dr. Yu-Zuo Bai, Department of Pediatric Surgery, Shengjing Hospital, China Medical University, Sanhao Avenue, Shenyang 110004, Liaoning, PR China. Tel: 189-4025-1660; Fax: +8624-8395-5092; E-mail: baiyz@sj-hospital.org

 References

Cervicothoracic esophageal duplication cyst


