

Case Report

Laparoscopic resection of intra-abdominal esophageal duplication cyst near spleen: a case report

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Abstract: Esophageal duplication cysts (EDCs) are congenital malformations of the posterior primitive foregut and often within the thoracic esophagus. Here we describe a rare case of intra-abdominal EDC near spleen in a 20-year-old female patient with a complaint of an asymptomatic abdominal mass for 5 years. The diagnosis of intra-abdominal EDC was confirmed by the Ultrasonography (US) and Magnetic resonance imaging (MRI) as well as Histological examination. Then the patient was received the laparoscopic resection and recovered well after the operation. We conclude that the laparoscopic resection is considered to be feasible and a reasonable treatment for intra-abdominal esophageal duplication cyst.

Keywords: Laparoscopic Resection, esophageal duplication cysts (EDCs), spleen

Introduction

Esophageal duplication cysts (EDCs) are rare malformations of the gastrointestinal (GI) tract, comprising 4% of all cases and 10-15% of all foregut duplication cysts [1], which have a two-layered muscular wall and may be lined by any epithelium of aero digestive origin [2]. EDCs were previously categorized as a type of esophageal cyst, owing to the duplication of the sub-mucosal and muscular portions of the esophagus [3]. Being the second most common benign posterior mediastinal lesion in children, the incidence rate of EDC is 1 in 8,200, with a two-fold male prevalence over women [1]. Most of esophageal duplication cysts (EDCs) always appear within the mediastinum, but sometimes it occurs in abdominal cavity [4]. Intra-abdominal EDCs are rare and usually near the intra-abdominal esophagus [5]. Although the pathogenic mechanisms of EDC are unknown, it is thought to be associated with abnormal esophageal development occurring in the fifth to eighth week of gestation, when the posterior primitive foregut coalesces to form a single esophageal lumen [3]. Patients may present with respiratory or digestive symptoms due to

complications such as esophageal stenosis, respiratory system compression, rupture, infarction, or malignancy [6]. For diagnostic purposes, ultrasound and endoscopy are the preferred tools in detecting cystic lesions (if located in the upper abdomen) [7]. Traditional treatment involves complete surgical resection of the cysts via thoracotomy or peritoneotomy, even in asymptomatic [8, 9], however, endoscopic therapy may advantage over complete surgical resection for its minimal invasiveness and expedited postoperative recovery [1]. In this case study, we present a rare case of intra-abdominal EDC near spleen in a young female patient without any symptoms and successfully resected by laparoscopic therapy.

Case report

A 20-year-old female patient with an asymptomatic abdominal mass for 5 years was admitted to our hospital. She denied any history of gastrointestinal disturbances including dysphagia and epigastric pain.

Physical examination was unremarkable. Laboratory test showed the following concentra-

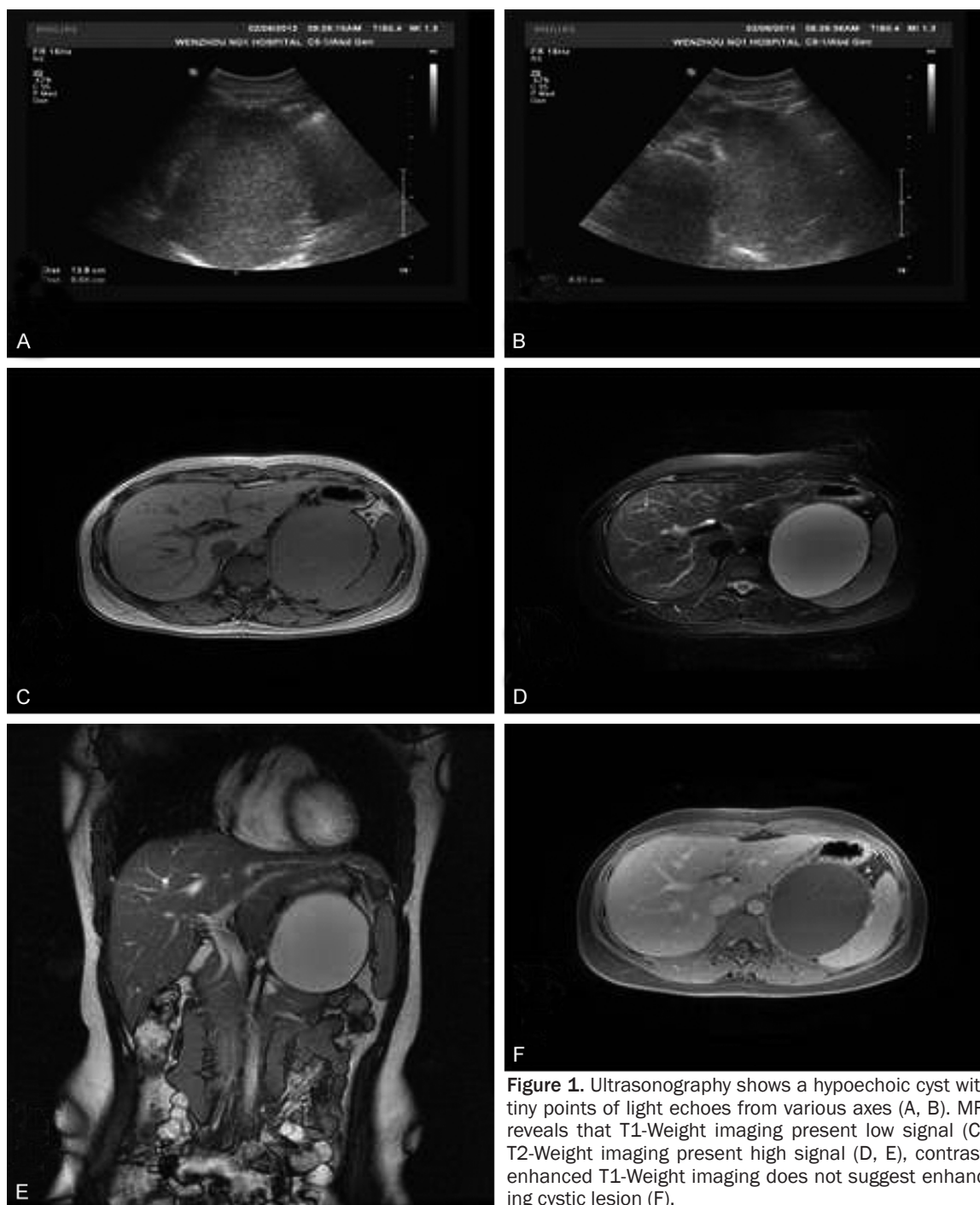


Figure 1. Ultrasonography shows a hypoechoic cyst with tiny points of light echoes from various axes (A, B). MRI reveals that T1-Weight imaging present low signal (C), T2-Weight imaging present high signal (D, E), contrast-enhanced T1-Weight imaging does not suggest enhancing cystic lesion (F).

tions: white-cell count $5.85 \times 10^9/L$, erythrocyte count $4.58 \times 10^{12}/L$, hemoglobin 142 g/L , platelet count $200 \times 10^9/L$, total bilirubin $18 \text{ } \mu\text{mol/L}$, albumin 45.7 g/L , alanine aminotransferase 18 U/L , creatine kinase 61 U/L , blood amylase 56 U/L , urine amylase 574 U/L , prothrombin time 12.5 s , prothrombin activity 105% , activated partial thromboplastin time 36.4 s .

Ultrasonography (US) demonstrated a hypoechoic cyst, size of $138 \times 96 \times 85 \text{ mm}$, with tiny points of light echoes (**Figure 1A, 1B**). Magnetic resonance imaging (MRI) indicated a mass, with clear boundary, on the right side of the spleen for which T1-Weight imaging presented low signal, T2-Weight imaging presented high signal (**Figure 1C-E**) and contrast-enhanced

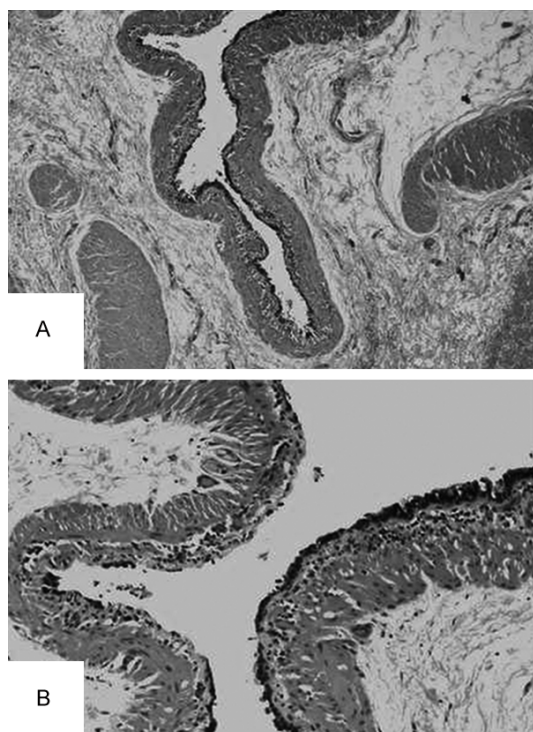


Figure 2. This cyst arose from esophagus since its wall was lined by epithelium and was composed of two smooth muscle layers (hematoxylin and eosin stain [H&E], A with original magnification $\times 40$, B with original magnification $\times 100$).

T1-Weight imaging revealed a large non-enhancing cystic lesion (**Figure 1F**).

It was suspected for a cyst. It also demonstrated distinct compression of left kidney, spleen and stomach. Laparoscopic resection was performed with general anesthesia. During the operation, we found a cyst adhered to the greater curvature side of the stomach without any fistula. Therefore, a drainage tube was placed in the lesser omentum. After operation, she received antibiotics and acid inhibitors therapy, along with parenteral nutrition. She recovered well after operation. On postoperative day 7, the removal of drainage was performed. She was discharged on postoperative day 8. Histological examination showed that the cyst arose from esophagus as its wall was lined by epithelium and was composed of two smooth muscle layers (**Figure 2A, 2B**). It strongly suggested intra-abdominal esophageal duplication cyst.

Discussion

Esophageal duplication cysts are rare congenital anomalies of foregut origin [10], classifica-

tion of which is based on embryologic origin, microscopic [11]. In esophageal duplication cysts, respiratory distress is usually present because of airway compression (due to enlarging mass), or no symptoms with a thoracic mass found incidentally on chest radiograph [12]. They are prone to develop complications (gastrointestinal bleeding, obstruction, diverticulitis, and umbilical abnormalities etc.) [3].

To be diagnosed as an esophageal duplication cyst, a lesion must meet the following criteria: 1) the cyst in the esophageal wall, 2) the cyst is covered with a thick muscularis propria, generally of two layers, and 3) the cyst has an epithelial lining consistent with that of the 4-wk embryo, which maybe columnar or pseudostratified columnar, and maybe ciliated [13].

Several imaging methods can be used for the diagnosis of a duplication cyst, the most valuable tool among which is endoscopic ultrasonography (EUS) that can define the intramural and extramural relationship with the gastrointestinal tract as well as distinguish between solid and cystic lesions [14]. EUS is considered to be an effective adjuvant to the radiographic and MR evaluation of EDC, and often shows EDCs as anechoic or hypoechoic cysts [15]. We found a hypoechoic cyst by the EUS, size of $138 \times 96 \times 85$ mm, with tiny points of light echoes in our case.

Histological examination is the best diagnostic tool of EDC, the pathological criteria of which are as follows [16]: the cyst is attached to the esophageal wall, is communicating with the epithelia originating from the GI tract, and is underlain by two layers of muscularis propria. EDC can be lined by pseudostratified columnar epithelium, gastric mucosal epithelium, and squamous epithelium. In our case, the histological examination showed that the cyst arose from esophagus with its wall lined by epithelium and composed of two smooth muscle layers, which exactly matched those criteria above.

Surgical resection of the cyst is the treatment of choice. Even though some esophageal duplication cysts are often asymptomatic at the time of diagnosis, surgical excision is also suggested since definitive diagnosis is better done on the surgical specimen and most patients

with untreated esophageal duplication will suffer dysphagia or develop complications such as bleeding or aspiration [9, 17]. In addition, malignant degeneration of esophageal duplication cysts is a very rare event [18]. The surgical approach (transthoracic or transabdominal) depends upon the location of the cyst, surgeon's preference, and presence of abnormal reflux [14]. Owing to the recent advances in minimally invasive surgery, endoscopic resection is believed to be as effective as the conventional open procedure with additional minimal invasiveness and expedited postoperative recovery [1]. In most cases thoracoscopy provides such an excellent exposure that it is the preferred approach [14]. When the cyst is located in the distal esophagus, however, laparoscopy plays a role particularly when abnormal reflux is present for a fundoplication can be added [14].

Summary, as intra-abdominal EDCs are rare it should be kept in mind that the intra-abdominal cysts maybe originated from esophagus when confronted with an abdominal mass. Versleijen MW et al. [19] followed-up an EDC patient with conservative management for 13 years, during that period, no growth of the cyst was detected and the patient presented no symptoms. They claimed that such asymptomatic patients may minimize the need for surgery if put under close observation. But we disagree with them, according to Martin, N.D et al. [5] as described in the preview, the cyst may have the potential risk of compressing adjacent organs even without symptoms. Complete surgical excision should be performed for all EDCs. Laparoscopic procedure may be a recommended therapy.

Disclosure of conflict of interest

None.

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