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Esophageal Duplication Cyst Presenting with Dysphagia in a Middle-Aged Male: A Case Report

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ABSTRACT

Esophageal duplication cysts (EDCs) are rare congenital foregut anomalies often presenting with nonspecific gastrointestinal (GI) symptoms in childhood. Diagnosis requires a high index of suspicion and comprehensive work-up. We report a 56-year-old male with type 2 diabetes mellitus and benign prostatic hyperplasia. He presented with a one-month history of intermittent, burning epigastric pain exacerbated by eating and accompanied by nausea. Physical examination revealed mild epigastric tenderness. Upper GI endoscopy showed a bulging lesion in the lower esophagus, a large cardia polyp, a small gastric body polyp, and antral erythema. Endoscopic ultrasound (EUS) identified a well-defined, homogeneous, round lesion (33 × 20 mm) demonstrating acoustic enhancement in the lower esophagus. Contrast-enhanced abdominopelvic computed tomography (CT) revealed an intraluminal, non-enhancing lesion (25 × 26 mm) at the lower esophageal sphincter (LES), suggestive of a duplication cyst. Thoracic imaging showed no abnormalities. Due to persistent symptoms, the patient underwent successful video-assisted thoracoscopic surgery without complications; no recurrences occurred during follow-up. EDCs should be considered in the differential diagnosis of submucosal or intraluminal esophageal lesions, even in adults despite their rarity. EUS yields high accuracy in characterizing these lesions. Thoraco-abdominal CT scans delineate relationships to surrounding structures. For symptomatic lesions, video-assisted thoracoscopic surgery demonstrates a successful outcome with low complication rates.

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Introduction

Dysphagia is a common gastrointestinal (GI) symptom with a broad spectrum of etiologies, ranging from benign to malignant causes, including iatrogenic, infectious, neurologic, metabolic, myopathic, and structural disorders [1]. Gastrointestinal duplication cysts (GIDCs) are rare congenital anomalies, among which esophageal duplication cysts (EDCs) represent the second most common type, arising from aberrations in foregut development [2,3]. The clinical presentation of EDCs is variable, ranging from asymptomatic cases to those with gastrointestinal, respiratory, or even cardiac symptoms. Dysphagia and epigastric pain are common manifestations in symptomatic individuals [4]. Most EDCs are asymptomatic and discovered incidentally during imaging studies [5]. These lesions typically become symptomatic in childhood and are rarely diagnosed in adults [6]. In patients presenting with dysphagia, esophagogastroduodenoscopy (EGD) is a primary diagnostic tool used to evaluate structural abnormalities or strictures. On EGD, EDCs typically appear as well-defined, round submucosal lesions with smooth borders. Suspicion for EDC is often raised by characteristic features on imaging modalities, while definitive diagnosis is established through histopathological examination. Surgical excision remains the treatment of choice, particularly in symptomatic cases [7].

Here, we present the case of a 56-year-old man who developed non-radiating epigastric pain and dysphagia. Imaging findings were characteristic of a duplication cyst. The patient underwent successful surgical resection and remained asymptomatic postoperatively.

Case presentation

We report the case of a 56-year-old male with a medical history of type 2 diabetes mellitus (DM II) and benign prostatic hyperplasia (BPH), who presented with a one-month history of intermittent, burning epigastric pain. The pain was non-radiating, exacerbated by food intake, and accompanied by nausea. It was not related to physical activity, and the patient denied anorexia or weight loss.

On physical examination, the patient appeared well and non-toxic. Vital signs were within normal limits: blood pressure 125/75 mmHg, pulse rate 80 beats per minute, respiratory rate 16 breaths per minute, and body temperature 37.2°C. Conjunctivae were not pale, and sclerae were anicteric. Examination of the oral cavity and oropharynx was unremarkable. There was no cervical lymphadenopathy, tracheal deviation, mass lesion, or elevated jugular venous pressure. Pulmonary auscultation was clear bilaterally, and no cardiac murmurs were detected. The abdomen was soft, non-distended, and non-guarded, with normal bowel sounds. Mild tenderness was noted in the epigastric region. There was no peripheral edema, cyanosis, or clubbing. All peripheral pulses were symmetrical and of normal strength. Neurological examination of the limbs revealed normal muscle strength and tone.

Initial laboratory tests revealed the following results: white blood cell (WBC) count of 7,000/mm^3 with a differential of 55% neutrophils and 35% lymphocytes; red blood cell (RBC) count of 4.8 million/mm^3; hemoglobin (Hb) level of 13 g/dL; hematocrit 40%; mean corpuscular volume (MCV) 83 fL; mean corpuscular hemoglobin (MCH) 27 pg; and red cell distribution width (RDW) 14%. Platelet count was 205,000/mm^3. Renal function tests were within normal limits: serum creatinine 1.1 mg/dL and blood urea nitrogen (BUN) 14 mg/dL. Serum electrolytes were sodium 135 mmol/L and potassium 4.2 mmol/L. Pancreatic enzymes were normal, with an amylase level of 40 U/L and a lipase level of 20 U/L. Liver function tests showed aspartate aminotransferase (AST) at 20 U/L, alanine transaminase (ALT) at 30 U/L, and alkaline phosphatase (ALP) elevated at 270 U/L. Inflammatory markers included an erythrocyte sedimentation rate (ESR) of 20 mm/hour and a C-reactive protein (CRP) level of 4 mg/dL.

Upper GI endoscopy revealed a bulging lesion in the lower third of the esophagus, a large polyp (<10 mm) in the cardia, and a small polyp (<5 mm) in the gastric body. Additionally, the antral mucosa appeared erythematous (Figure 1).

EUS revealed a round, well-circumscribed lesion with acoustic enhancement located in the lower third of the esophagus. The lesion exhibited five distinct wall layers and measured 33 × 20 mm. It appeared homogeneous with regular borders and demonstrated peripheral vascularity on color Doppler imaging (Figure 2).

Further imaging was performed due to suspicion of a mediastinal or duplication cyst (Figure 3). Spiral chest CT revealed no additional abnormalities. Contrast-enhanced abdominopelvic CT identified an intraluminal, fluid-density lesion at the level of the LES, measuring 25 × 26 mm. The lesion demonstrated no post-contrast enhancement. These

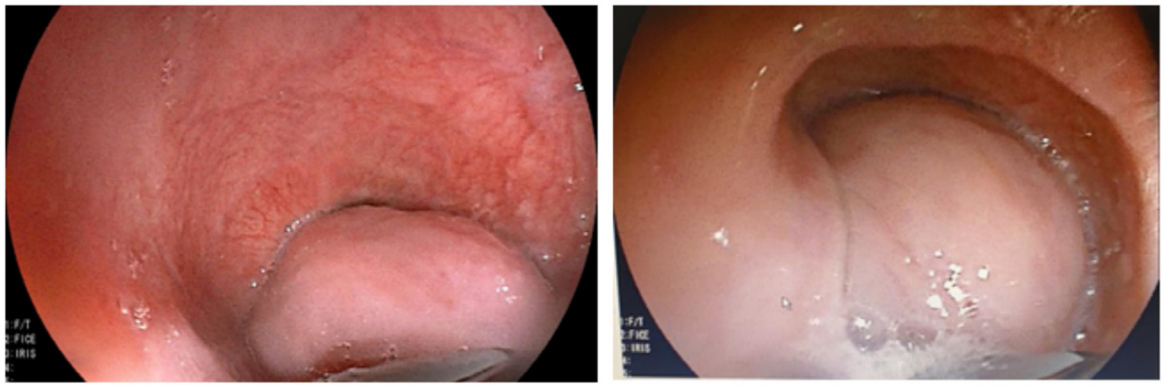

Fig. 1. Upper GI endoscopy demonstrating a round, well-defined, smooth-bordered lesion in the lower third of the esophagus.

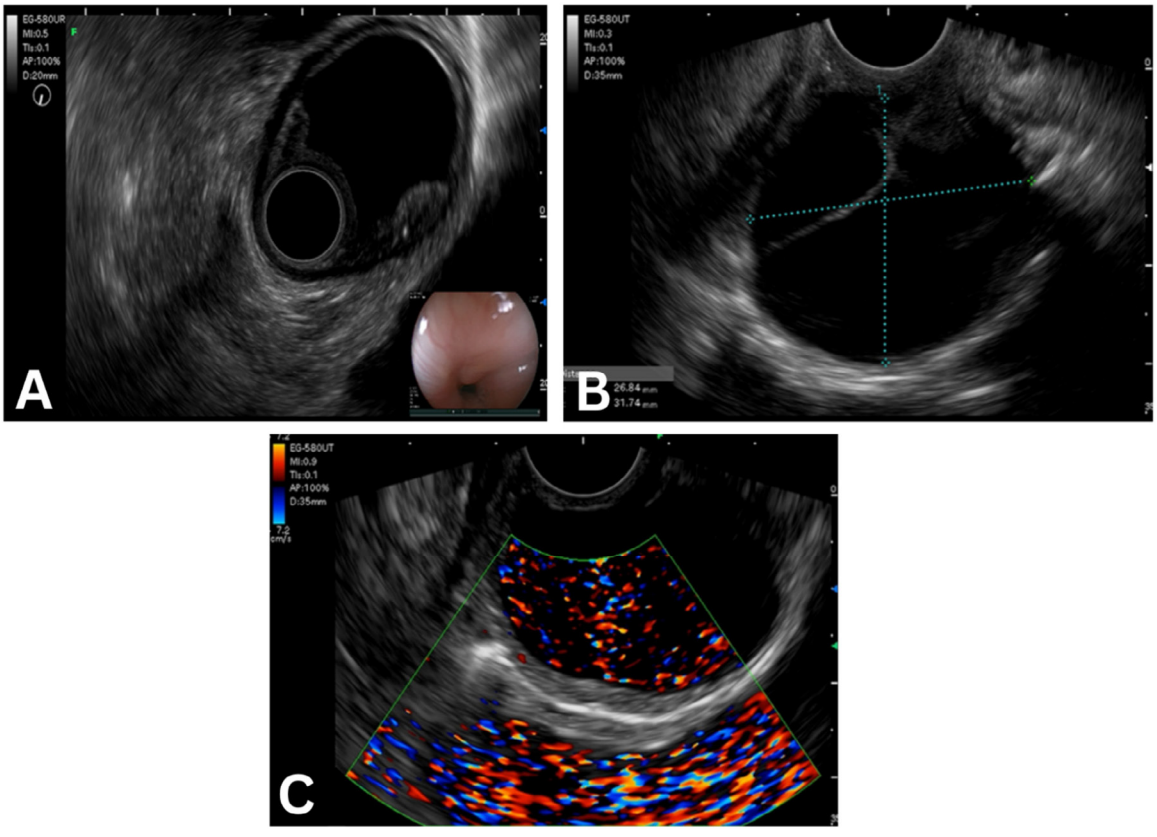

Fig. 2. (**A, B**) EUS images showing a round, homogenous, well-circumscribed lesion measuring 33 × 20 mm with acoustic enhancement in the lower third of the esophagus. (**C**) Color Doppler image demonstrating vascular flow within the lesion.

imaging characteristics were highly suggestive of an esophageal duplication cyst. Given the persistence of symptoms despite conservative management, the patient was scheduled for surgical intervention.

To optimize surgical exposure and reduce the risk of intraoperative complications, a right thoracoscopic approach was selected to access the lower third of the esophagus. The procedure was performed under general anesthesia with the use of a double-lumen endotracheal tube to facilitate single-lung ventilation of the left lung. The right lung was deflated to enhance visualization of the operative field. Following thoracoscopic entry, the azygos vein arch was identified, ligated, and divided to improve access to the posterior mediastinum. Meticulous dissection was then performed to mobilize the esophagus and separate it from adjacent mediastinal structures, including the pericardium, with careful preservation of surrounding tissues. To clearly delineate the margins

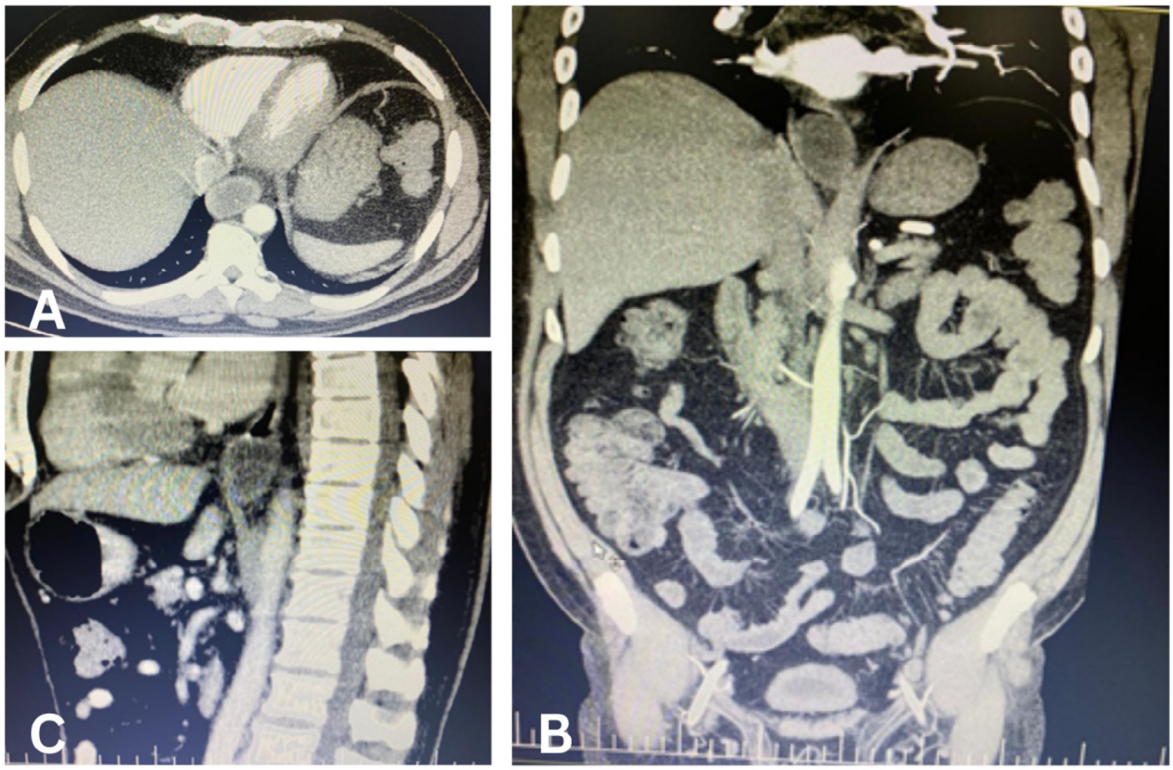

Fig. 3. Multiplanar contrast-enhanced abdominopelvic CT images demonstrating a posterior-lateral esophageal duplication cyst with rightward deviation. (A) Axial view showing an intraluminal, fluid-density lesion (25 × 26 mm) at the level of the lower esophagus near the gastroesophageal junction. (B) Coronal reconstruction confirming the cyst's posterior-lateral position adjacent to the esophagus, with no evidence of surrounding tissue invasion or mediastinal abnormalities. (C) Sagittal view illustrating the cyst's relationship to the esophageal lumen and its smooth, non-enhancing appearance, consistent with a duplication cyst.

of the cyst, the esophageal lumen was insufflated with air via a nasogastric tube. The lesion was sharply dissected and excised in its entirety, ensuring preservation of esophageal wall integrity. Hemostasis was achieved, and a thoracic drain was placed before closure.

The patient was discharged on postoperative day three in stable condition and remained asymptomatic. Follow-up assessments at two weeks, three months, and one year post-discharge revealed no complications or evidence of recurrence.

Discussion

EDCs are uncommon congenital anomalies within the spectrum of GIDCs. Since the initial unifying concept proposed by Ladd and Gross in 1940, these lesions have been recognized as sharing common embryologic and structural characteristics despite their diverse anatomical presentations [8]. Among GIDCs, the ileum is most frequently involved, while the esophagus represents the second most common site [2,8–10]. Although EDCs constitute only 0.5–2.5% of all esophageal cysts, they account for approximately 10–20% of GIDCs and are considered the second most common benign tumor of the distal esophagus [2,5,8–11].

EDCs predominantly present in childhood, with 70–80% of cases becoming symptomatic before the age of two; however, a smaller proportion (20–30%) are diagnosed in adulthood, often incidentally [4,11]. Autopsy studies estimate a prevalence of approximately 1 in 8,200 individuals, with a male predominance of nearly 2:1 [2]. Adult presentations, as illustrated in the current case, are therefore relatively rare and may pose diagnostic challenges due to nonspecific gastrointestinal symptoms that mimic more prevalent disorders such as peptic disease or functional dyspepsia [3,12].

The embryogenesis of EDCs remains incompletely understood, though most theories implicate abnormal budding or aberrant recanalization of the primitive foregut between the third and eighth weeks of gestation [13–15]. Disruption of vacuole formation and coalescence during foregut development may result in cystic structures that become incorporated within the esophageal wall

and enveloped by the muscularis propria [4,15,16]. Histologically, the Palmer criteria define EDCs by their attachment to the esophageal wall, epithelial lining, and enclosure by two muscular layers, although not all criteria are invariably present, particularly in ectopic or intra-abdominal lesions [2,17].

Clinically, most EDCs are asymptomatic, with symptomatology largely dependent on cyst size, location, and the presence of heterotopic tissue [7,13]. In adults, gastrointestinal manifestations such as dysphagia, epigastric pain, and retrosternal discomfort are more common than respiratory symptoms, which predominate in pediatric populations [3,12]. Complications, although infrequent, can be significant and include infection, hemorrhage, perforation, fistula formation, and mechanical obstruction [3,6,14,16,18]. Malignant transformation has been reported rarely, most commonly as adenocarcinoma, though the true incidence remains unknown due to limited case numbers [11,13,14].

Imaging plays a central role in the evaluation of suspected EDCs. Among available modalities, EUS is considered the most sensitive tool for characterizing mediastinal and periesophageal lesions [5]. Typical EUS findings include a homogeneous, hypoechoic, well-circumscribed lesion with a characteristic multilayered wall structure [19–22]. Cross-sectional imaging with CT further aids in defining lesion size, anatomical relationships, and exclusion of alternative mediastinal pathologies, usually demonstrating a well-defined, non-enhancing, fluid-density mass along the esophageal wall [4,5]. Despite advances in imaging, definitive diagnosis ultimately relies on histopathological confirmation following excision [4].

Surgical management remains the cornerstone of treatment for symptomatic EDCs, as medical therapy has no established role [5,12,18]. While open surgery has traditionally been regarded as the gold standard, minimally invasive approaches such as thoracoscopy, video-assisted thoracoscopic surgery, and robotic techniques have gained increasing acceptance due to favorable perioperative outcomes, reduced morbidity, and shorter hospital stays [5,7,14,18]. The management of asymptomatic lesions, however, remains controversial. Although prophylactic resection may prevent future complications, conservative surveillance with periodic EUS has been shown to be a viable alternative in select cases, particularly in patients with high surgical risk [3,4,16,23].

Conclusion

EDCs are rare congenital anomalies with variable clinical presentations, particularly in adults. Accurate diagnosis requires a high index of suspicion and integration of endoscopic and imaging findings. Surgical excision remains the definitive treatment for symptomatic patients, whereas individualized management strategies are essential for asymptomatic cases, balancing potential surgical risks against the likelihood of future complications.

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Ethical Considerations

Ethic Approval

This study has been approved by the Ethical Committee of Hormozgan University of Medical Sciences (IR.HUMS.REC.1402.443) and has been conducted in accordance with the principles of the Declaration of Helsinki (1964) and its subsequent amendments.

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Conflict of Interests

The authors have no conflict of interest to declare.

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