Esophageal Duplication Cyst Presenting With Gastric Hemorrhage and Perforation in an Elderly Patient

Ann. Ital. Chir., 2026 97, 7: 1127–1130
https://doi.org/10.62713/aic.4678

Ali Yildirim[1], Uğur Topal[1], Tuğba Toyran[2], İshak Aydin[1], Serdar Gümüş[1], Cem Kaan Parsak[1]

[1]Department of General Surgery, Balcali Hospital, Cukurova University Faculty of Medicine, 01330 Adana, Türkiye
[2]Department of Pathology, Balcali Hospital, Cukurova University Faculty of Medicine, 01330 Adana, Türkiye

To the Editor

Gastrointestinal duplication cysts are rare congenital anomalies that can arise most frequently in the ileum, esophagus, colon, jejunum, and stomach [1,2]. Esophageal duplication cysts (EDCs) represent only 0.5%–2.5% of all esophageal masses, making them an exceedingly rare clinical entity [3].

EDCs are cystic structures with a double-layered smooth muscle wall. Ectopic gastric mucosa, present in 20%–30% of cases, may cause peptic ulceration, hemorrhage, and perforation [3–5]. Preoperative diagnosis remains challenging when cysts present atypically, especially in acute settings where imaging findings may mimic other acute abdominal pathologies.

The most common symptoms were dysphagia, chest pain, and weight loss [6]. Hematemesis and melena have been described in cysts containing ectopic gastric mucosa, but life-threatening acute hemorrhage and perforation have been reported only in a handful of cases [5,7].

While most EDCs are diagnosed in younger adults with chronic dysphagia or chest pain, life-threatening complications such as massive hemorrhage and perforation are exceedingly rare, particularly in elderly patients with multiple comorbidities [5,7]. Such catastrophic presentations pose significant diagnostic and therapeutic challenges, as emergency settings preclude comprehensive preoperative workup and necessitate urgent surgical intervention. Herein, we report an exceptionally rare case of an EDC in an elderly patient who presented with acute abdomen, massive intraperitoneal hemorrhage, and perforation, and was diagnosed after emergency wedge resection. The report adheres to the SCARE 2023 guideline [8].

A 76-year-old male presented to an outside facility with a 2-day history of epigastric pain, numbness in the hands and feet, generalized weakness, and syncope. His medical history was significant for coronary artery disease (coronary artery bypass grafting 10 years earlier), congestive heart failure, hypertension, and a prior operation for a splenic cyst approximately 30 years earlier. His medications included acetylsalicylic acid, irbesartan, atorvastatin, carvedilol, and vasodilator agents. He denied hematemesis, melena, or hematochezia.

Non-contrast abdominal thoracoabdominopelvic computed tomography (CT) at the referring center demonstrated a 12 cm intragastric lesion with central cystic-necrotic and peripheral solid components, a hyperdense area consistent with intracystic and intraluminal hemorrhage, and perihepatic, perisplenic, and pelvic free fluid, suggestive of intraperitoneal hemorrhage, prompting transfer to our institution for suspected gastrointestinal bleeding with possible perforation.

On admission, he was hypotensive (blood pressure 95/53 mmHg), tachycardic (heart rate 130 beats/min), with a respiratory rate of 20/min and SpO_2 of 93%. The abdomen was diffusely tender without guarding or rebound. Laboratory results showed C-reactive protein 150 mg/L, creatinine 1.55 mg/dL, leukocytes 16.75 $\times$ 10^3/µL, and hemoglobin 13.6 g/dL with a progressive hematocrit decline (43% $\rightarrow$ 39% $\rightarrow$ 36%), suggestive of ongoing hemorrhage.

Abdominal CT angiography revealed a 16 $\times$ 12 cm mass originating from the gastroesophageal junction and extending into the gastric lumen markedly distending the stomach, with a heterogeneous hyperdense collection consistent with intracystic and intraluminal hemorrhage and no active extravasation; the radiological impression was gastric carcinoma (Fig. 1). Upper gastrointestinal endoscopy demonstrated normal esophageal mucosa and a friable intraluminal mass with its base at the cardia-fundus region immediately below the gastroesophageal junction, extending along the greater curvature of the gastric corpus, without active bleeding.

Submitted: 4 April 2026 Revised: 21 May 2026 Accepted: 10 June 2026 Published: 14 July 2026
Correspondence to: Ali Yildirim, Department of General Surgery, Balcali Hospital, Cukurova University Faculty of Medicine, 01330 Adana, Türkiye (e-mail: aliyildirim@cu.edu.tr).
Editor: Weihua Gong

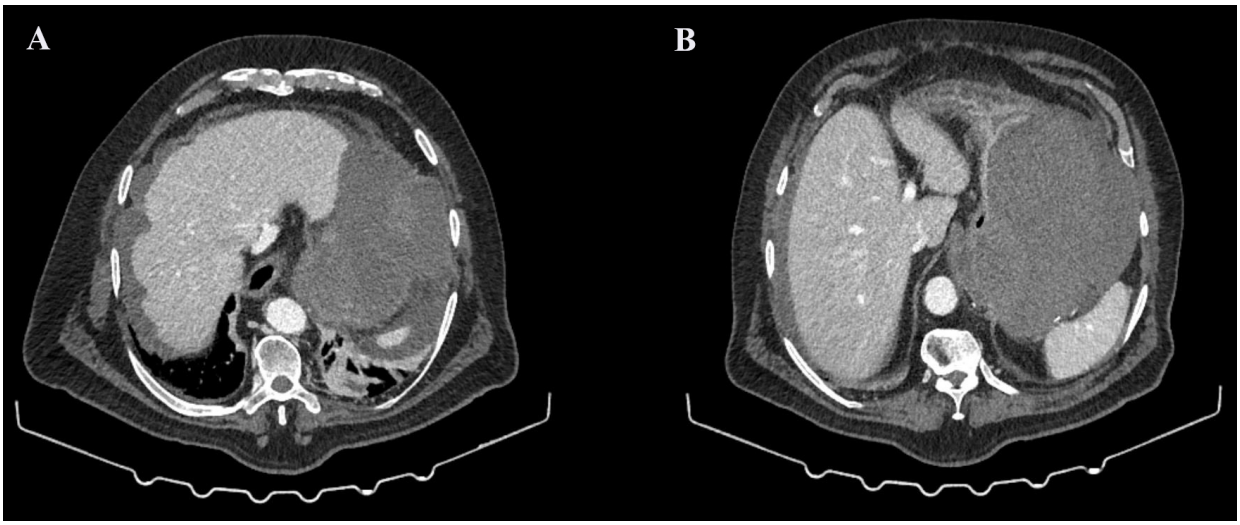

Fig. 1. Axial abdominal thoracoabdominopelvic computed tomography (CT) angiography images demonstrating a large intraluminal mass approximately 16 × 12 cm in size, markedly distending the gastric lumen, with an intraluminal hyperdense collection consistent with hemorrhage. (A) Upper-level axial section showing the mass filling the gastric lumen. (B) Lower-level axial section demonstrating the extent of the lesion.

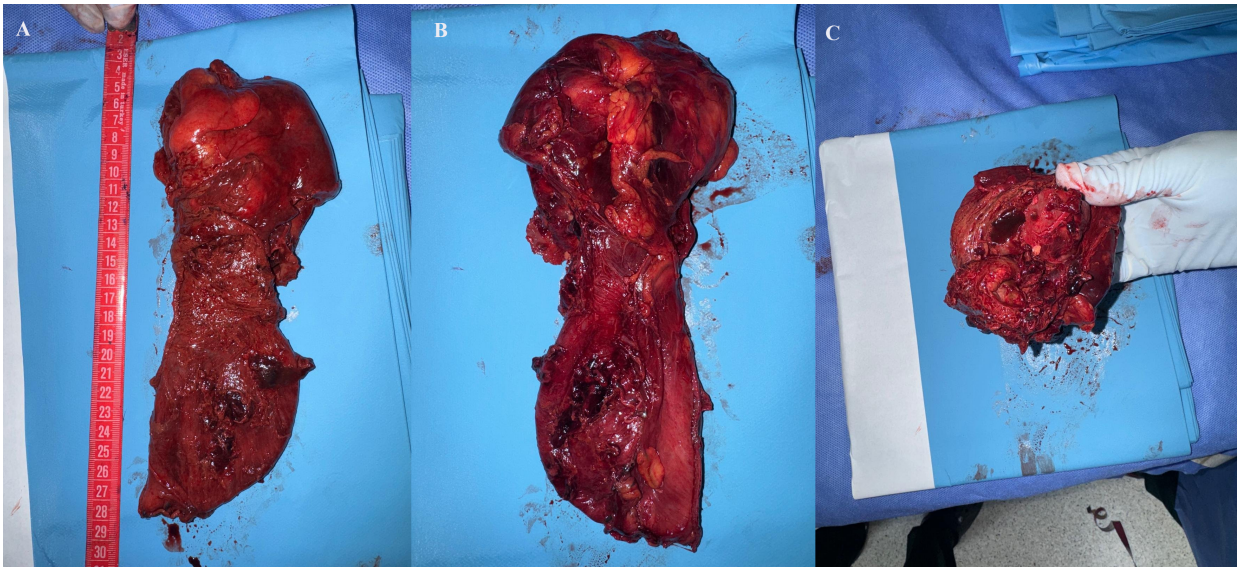

Fig. 2. Gross specimen photographs. (A) Resected specimen measured with a ruler; approximately 28 cm in length, including surrounding gastric tissue, revealed an 18 × 11 × 7 cm cystic lesion adherent to the gastric wall with ulcerated and hemorrhagic areas. (B) Serosal surface demonstrating cystic-ulcerated areas and hemorrhagic changes. (C) Close-up view of the cystic lesion with the perforation site and hemorrhagic content.

Because of progressive hemodynamic instability, emergency laparotomy was performed. Approximately 1.5–2 L of hemorrhagic fluid was evacuated from the peritoneal cavity. Exploration revealed a large cystic lesion attached to the gastric wall at the level of the gastroesophageal junction, with the bulk of the lesion extending into and distending the gastric corpus along the greater curvature. The cyst was adherent to the gastric serosa with a perforation site identified on the cyst wall. Wedge resection was performed along the greater curvature using a linear stapler, and a single abdominal drain was placed.

Gross pathology of the 18 × 11 × 7 cm wedge specimen revealed a cystic lesion adherent to the gastric wall with ulcerated and hemorrhagic areas (Fig. 2). The cyst dimensions correlated with the 16 × 12 cm lesion identified on preoperative CT imaging. Microscopy showed a cyst adjacent to the gastroesophageal junction, lined by squamous and mucinous columnar epithelium with pseudostratified foci and intestinal metaplasia, bounded by a smooth muscle layer. Active chronic ulcerative inflammation, inflammatory granulation tissue, hyalinization, calcification, cholesterol clefts, and hemorrhage were present within the cyst

wall. The definitive diagnosis was esophageal duplication cyst with active chronic ulcerative inflammation.

Postoperatively, the patient was monitored in the surgical intensive care unit. On postoperative day 3, a methylene blue leak test was negative, oral feeding was initiated and gradually advanced, and the patient was discharged uneventfully on a regular diet.

EDCs are among the rarest congenital anomalies of the gastrointestinal tract. In the systematic review by Gonzalez-Urquijo *et al*. [6], the mean age at diagnosis was 42.3 years, the mean duration of symptoms was 17.3 months, and the mean hospital stay was 8.6 days. Our patient, diagnosed at 76, is among the oldest reported cases in the literature, demonstrating that this rare entity can present even in the elderly and that chronologic age should not a priori exclude EDC from the differential diagnosis of an intragastric or periesophageal mass.

The most exceptional feature of this case is the acute presentation with perforation, massive intra-abdominal hemorrhage, and hemodynamic instability. Imaging demonstrated both intragastric hemorrhage and intraperitoneal free fluid, while operative findings confirmed a dual hemorrhage pathway: bleeding from the ulcerated cyst wall extended both into the gastric lumen and through the perforation site into the peritoneal cavity, resulting in 1.5–2 L hemoperitoneum. Although gastrointestinal bleeding has been described in EDCs harboring ectopic gastric mucosa—Cioffi *et al*. [9] emphasized peptic ulceration secondary to acid secretion and Chaudhry *et al*. [7] reported sudden intracystic hemorrhage as a rare cause of acute dysphagia—perforation with 1.5–2 L of free intraperitoneal hemorrhagic fluid and shock is, to our knowledge, exceedingly rare. This acute presentation also made preoperative workup inevitably limited and rendered endoscopic ultrasound, the imaging gold standard for cyst wall characterization [10], unfeasible.

The pathological spectrum encountered—squamous, mucinous columnar, and pseudostratified epithelia with intestinal metaplasia, along with granulation tissue, hyalinization, calcification, and cholesterol clefts—suggests a long-standing inflammatory process. Although gastric-type mucosa was not definitively identified on histology, the chronic ulcerative inflammation may reflect secretory activity of the mucinous epithelium, consistent with ectopic gastric mucosa reported in 20%–30% of EDCs [4,5]. Intestinal metaplasia may reflect chronic mucosal adaptation and, together with reports of malignant transformation within EDCs, underscores the rationale for elective resection when EDCs are identified, even in asymptomatic patients [3–5].

Differential diagnosis among foregut duplication cysts was essential given the intragastric location. The predominant squamous epithelial lining, double-layered smooth muscle wall, and absence of cartilage or bronchial glands established the diagnosis of esophageal duplication cyst. Bronchogenic cysts contain cartilage and bronchial glands, while gastric duplications are lined exclusively by gastric mucosa [9]. The anatomical origin at the gastroesophageal junction further confirmed esophageal rather than gastric origin.

Preoperative diagnosis of EDCs is notoriously challenging. On CT, typical EDCs appear as well-defined, homogeneous, low-density cystic masses; however, mucinous fluid, hemorrhage, or proteinaceous content may produce high attenuation and mimic a solid neoplasm [10]. In our case, the combination of intracystic hemorrhage and an atypical intragastric location led to a preoperative radiological impression of gastric carcinoma. Although imaging suggested an intragastric mass, microscopic examination confirmed the cyst originated from the gastroesophageal junction with extension into the gastric lumen, a pattern typical of distal EDCs. The esophageal origin was established by the predominantly squamous epithelial lining and attachment at the gastroesophageal junction, distinguishing it from gastric duplication. This diagnostic pitfall is well recognized and highlights the importance of including duplication cysts in the differential diagnosis of complex intra-abdominal masses, particularly when imaging features are discordant with typical malignancy patterns.

Surgical excision is the mainstay of treatment for symptomatic EDCs. In the systematic review by Gonzalez-Urquijo *et al*. [6], 84.5% of patients underwent surgery, and video-assisted thoracoscopic surgery was associated with shorter hospital stays than thoracotomy. Minimally invasive and robotic approaches are increasingly preferred for elective cases and have been shown to offer comparable oncologic and functional outcomes with reduced morbidity [11]. However, in patients presenting with acute abdomen and hemodynamic instability, as in our case, emergency laparotomy with wedge resection is the life-saving approach. Stapled wedge resection along the greater curvature achieved complete excision, avoided the need for gastric or esophageal reconstruction, and was well tolerated despite the patient's comorbidities.

Advanced age (76 years) and significant comorbidities—coronary artery disease, heart failure, and chronic antiplatelet therapy—were additional risk factors in our patient. The use of acetylsalicylic acid may have contributed to the magnitude of hemorrhage, and the cardiac and renal reserves complicated perioperative fluid management. Careful hemodynamic resuscitation and close postoperative monitoring in the intensive care unit were essential to the favorable outcome.

Although management of asymptomatic EDCs remains debated, prophylactic excision has been recommended in fit patients given the risks of hemorrhage, perforation, and malignant transformation [3–5,11]. This case illustrates the potential for catastrophic complications: a presumably asymptomatic EDC presented decades later with a catastrophic perforation in an elderly patient with multiple comorbidities, a scenario that carries substantially higher pe-

rioperative risk than elective resection. Limitations of this report include the single-case design, the inability to perform EUS due to the emergency setting, the lack of a preoperative definitive diagnosis, and the absence of long-term follow-up data.

Despite their rarity, esophageal duplication cysts can cause life-threatening complications such as perforation and massive hemorrhage, even in elderly patients with significant comorbidities. Preoperative diagnosis can be challenging when atypical radiological features such as intracystic hemorrhage are present. EDCs should be considered in the differential diagnosis of atypical intra-abdominal masses, and emergency wedge resection may be a feasible option in emergency settings. Prophylactic excision should be considered in asymptomatic cases to prevent potentially fatal complications.

Availability of Data and Materials

All data supporting the findings of this study are contained within the manuscript.

Author Contributions

AY: Conceptualization, data collection, writing—original draft and project administration. UT: Study design. TT: Histopathological analysis and interpretation. İA: data analysis and interpretation. SG: conception, design and data acquisition. CKP: conception, design and supervision. All authors contributed to the critical revision of the manuscript for important intellectual content. All authors read and approved the final manuscript. All authors have participated sufficiently in the work and agreed to be accountable for all aspects of the work. All authors read and approved the final manuscript.

Ethics Approval and Consent to Participate

This study was conducted in accordance with the Declaration of Helsinki. According to the regulations of Çukurova University Faculty of Medicine Non-Interventional Clinical Research Ethics Committee, retrospective single short report based on routine clinical care do not require formal ethics committee approval. Written informed consent was obtained from the patient for publication of this report and accompanying images.

Acknowledgment

The authors thank the patient for granting permission to publish this report.

Funding

This research received no external funding.

Conflict of Interest

The authors declare no conflict of interest.

Declaration of Generative AI and AI-Assisted Technologies in Manuscript Preparation

We used an AI-assisted tool (Claude Opus 4.8) to improve the language quality and take responsibility for the accuracy, integrity, and originality of this article.

References

[1] Macpherson RI. Gastrointestinal tract duplications: clinical, pathologic, etiologic, and radiologic considerations. Radiographics. 1993; 13: 1063–1080. https://doi.org/10.1148/radiographics.13.5.8210590.

[2] Holcomb GW, 3rd, Gheissari A, O'Neill JA, Jr, Shorter NA, Bishop HC. Surgical management of alimentary tract duplications. Annals of Surgery. 1989; 209: 167–174. https://doi.org/10.1097/00000658-198902000-00006.

[3] Wahi JE, Safdie FM. Esophageal duplication cysts: a clinical practice review. Mediastinum. 2023; 7: 1. https://doi.org/10.21037/med-22-33.

[4] Khoury T, Rivera L. Foregut duplication cysts: a report of two cases with emphasis on embryogenesis. World Journal of Gastroenterology. 2011; 17: 130–134. https://doi.org/10.3748/wjg.v17.i1.130.

[5] Zhang Z, Jin F, Wu H, Tan S, Tian Z, Cui Y. Double esophageal duplication cysts, with ectopic gastric mucosa: a case report. Journal of Cardiothoracic Surgery. 2013; 8: 221. https://doi.org/10.1186/1749-8090-8-221.

[6] Gonzalez-Urquijo M, Hinojosa-Gonzalez DE, Padilla-Armendariz DP, Saldaña-Rodriguez JA, Leyva-Alvizo A, Rodarte-Shade M, *et al*. Esophageal Duplication Cysts in 97 Adult Patients: A Systematic Review. World Journal of Surgery. 2022; 46: 154–162. https://doi.org/10.1007/s00268-021-06325-8.

[7] Chaudhry IUH, A Alsuhaimi M, M Al Ghamdi A, Almalali M, Almalali F, A Alqahtani Y, *et al*. A sudden hemorrhage in the esophageal duplication cyst: A rare cause of acute dysphagia in an adult. Annals Of Medicine And Surgery. 2022; 80: 104199. https://doi.org/10.1016/j.amsu.2022.104199.

[8] Sohrabi C, Mathew G, Maria N, Kerwan A, Franchi T, Agha RA, *et al*. The SCARE 2023 guideline: updating consensus Surgical CAse REport (SCARE) guidelines. International Journal of Surgery. 2023; 109: 1136–1140. https://doi.org/10.1097/JS9.0000000000000373.

[9] Cioffi U, Bonavina L, De Simone M, Santambrogio L, Pavoni G, Testori A, *et al*. Presentation and surgical management of bronchogenic and esophageal duplication cysts in adults. Chest. 1998; 113: 1492–1496. https://doi.org/10.1378/chest.113.6.1492.

[10] Ronkainen E, Barner-Rasmussen N, Volmonen K, Färkkilä M, Arkkila P, Tenca A. Comparison of Endoscopic Ultrasound and CT Scan in the Diagnosis of Esophageal Duplication Cysts. Digestive Diseases and Sciences. 2024; 69: 4133–4139. https://doi.org/10.1007/s10620-024-08655-8.

[11] Kunisaki C. Esophageal duplication cyst. Annals of Laparoscopic and Endoscopic Surgery. 2022; 7: 31. https://doi.org/10.21037/ales-22-43.