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Single Case

Delayed Presenting Gastric Duplication Cyst Mimicking a Left Adrenal Cyst in a Young Female: A Case Report with a Literature Review

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Abstract
Introduction: Gastric duplication cyst (GDC) is a rare congenital anomaly of the gastrointestinal tract. Though GDC is often misdiagnosed, misidentification as an adrenal cyst has rarely been reported. Herein, we report a case of GDC in a young female mimicking an adrenal cyst. ***Case Presentation:*** A 17-year-old female presented with chronic epigastric pain, nausea, and intermittent vomiting. Physical examinations revealed mild tenderness in the epigastric region. Esophagogastroduodenoscopy showed no abnormality. Ultrasound, contrast-enhanced computed tomography scan, and MRI of the abdomen and pelvis showed an oval-shaped left adrenal cystic lesion measuring 33 × 26 mm. Preoperative blood investigations and hormonal assessments were normal. Laparoscopy showed that the cyst originated from the greater curvature of the stomach. The left adrenal gland was normal. After an intraoperative consultation with a gastrointestinal surgeon, a wedge resection of the cyst was performed. Histopathology confirmed the gastric duplication cyst. ***Conclusion:*** GDCs are rare congenital malformations that may become symptomatic during adulthood. They can mimic adrenal cysts and lead to misdiagnosis.

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Introduction

Gastric duplication cyst (GDC) is a rare congenital anomaly of the gastrointestinal tract (GIT), which comprises only 4% of GIT duplications [1]. It is a globular muscular structure with all gastric layers, found anywhere along the GIT, communicating with or without the organ of origin [2, 3]. This anomaly is primarily diagnosed in childhood, often within the first year of life, but may go undetected until adulthood [2, 4]. Based on age and lesion features, GDC can be associated with abdominal pain, hematemesis, vomiting, and weight loss. Although it may be asymptomatic in adults and pose challenges for preoperative diagnosis [5], GDC is often misdiagnosed with several diseases, like gastrointestinal stromal tumors and other gastric and endocrine tumors [1, 6]. Its association with ectopic tissues of gastrointestinal organs or mimicking other anomalies, such as lymphangiomas, has also been reported [4, 6]. GDC simulating an adrenal cyst, on the other hand, is an exceedingly rare incidence [3, 7, 8]. Herein, we report a case of GDC in a young female mimicking an adrenal cyst. Additionally, we reviewed several similar cases in the literature (Table 1) [3, 7–9]. The study has been written according to the CARE checklist, and all references have been checked to avoid citing non-peer-reviewed information [10, 11]. The CARE Checklist has been completed by the authors for this case report and attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000537972).

Case Presentation

Patient Information

A 17-year-old female presented with chronic epigastric pain, nausea, intermittent vomiting, and food intolerance for 2 years. Along with the family history, her past medical history was negative for any chronic GIT disease. She was taking proton pump inhibitors, antacids, and prokinetic drugs.

Clinical Findings

On physical examination, there was no pallor, jaundice, or skin lesions. However, she had mild tenderness in the epigastric region without abdominal distention, organomegaly, or striae. The blood examination results were normal.

Diagnostic Assessment

Esophagogastroduodenoscopy (OGD) showed non-significant findings apart from mild antral erosions. Transabdominal ultrasound (U/S) revealed a well-defined, hypoechoic left adrenal cystic lesion measuring 40 × 35 mm. A contrast-enhanced computed tomography (CT) scan of the abdomen and pelvis showed an oval-shaped cystic lesion on the left adrenal gland measuring 33 × 26 mm (Fig. 1). Abdominal magnetic resonance imaging (MRI) demonstrated a well-defined T1 hypointense 21 × 22 mm left adrenal cystic lesion (Fig. 2). All indications pointed to an adrenal lesion, prompting referral to an endocrinologist for further assessment. Hormonal evaluation, including a dexamethasone suppression test (9.51 nmol/L) and metanephrine (30.7 pg/mL), were all normal.

Therapeutic Intervention

Considering the investigation results and the benign radiological features of the cyst, conservative treatment was initiated. However, the parents of the patient preferred surgical excision. A laparoscopy through three ports was performed in the right lateral position. The

Table 1. Review of the cases of gastric duplication cysts mimicking adrenal mass

Authors	No. cases	Age, years	Gender	Symptoms/ complaints	Cyst size, cm	Laterality	Location	Diagnostic tools	Management	Adrenal gland preservation	Histopathology	Follow-up
Zhao et al. [9] (2023)	1	15	Female	Elevated blood pressure, and intermittent right flank pain	3.5	Right	The mass was situated within the retroperitoneum, positioned between the right adrenal gland and the kidney. It exhibited no connections to these organs and other adjacent structures	MRI	Complete excision	N/A	The cyst was lined by respiratory and gastric antral/oxyntic-type mucosa, surrounded by muscularis mucosa, submucosal tissue, muscularis propria, and pericentric tissue	No complications after 7 months of follow-up
Al Awfi et al. [3] (2021)	1	2	Female	Abdominal pain	5.2	Left	The cyst was attached to the pancreas tail, splenic vessels, and the abdominal posterior wall	CT scan	Laparoscopic adrenalectomy	No	The examination of the specimen disclosed a cyst wall containing all four layers of the stomach, encompassing specialized gastric mucosa, submucosa, muscularis propria, and adventitia	No complications
Castillo-Fernández et al. [7] (2018)	1	Prenatal	Female	Incidental	2.9	Left	The cyst was located between the upper pole of the left kidney and the adrenal gland	U/S and MRI	Laparotomy	No	Histopathology revealed a gastric duplication wall-cyst with three layers of mucosa, muscular, and serosa	N/A
Terry et al. [8] (2007)	1	75	Female	Abdominal pain, decreased appetite, weight loss, and chronic shortness of breath	5	Left	The cyst was adherent to the lateral-most portion of the adrenal gland	CT scan, MRI, and colonoscopy and OGD	Laparoscopic adrenalectomy	No	Microscopic examination indicated a non-malignant adrenal gland with no signs of nodularity, neoplasia, or genuine hyperplasia. The adjacent adipose tissues displayed abundant blood vessels and peripheral nerve fibers. Additionally, there was a genuine cyst lined by tall, columnar, ciliated cells, resembling an upper respiratory tract-type epithelium. The underlying wall of the cyst contained bundles of the smooth muscle	N/A
Bapir et al. (current report)	1	17	Female	Chronic epigastric pain, nausea, intermittent vomiting, and food intolerance	3.7	Left	The cyst was located on the adrenal gland with no connections	OGD, U/S, CT scan, and MRI	Laparoscopy and wedge resection of the cyst	Yes	The cyst wall was lined by pseudostratified ciliated columnar epithelial cells, with the underlying smooth muscle wall	No complications after 2 months of follow-up

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surgeon observed that the cyst did not originate from the left adrenal gland but instead arose from the greater curvature of the stomach (Fig. 3). After an intraoperative consultation with a gastrointestinal surgeon, a wedge resection of the cyst was performed. The cyst contained thick, gelatinous, and white-yellowish mucus. Histopathology confirmed a gastric duplication cyst (Fig. 4).

Follow-Up

The postoperative outcome was satisfactory, and the patient was discharged on the first postoperative day. A follow-up U/S 2 months postoperatively revealed a small collection at the surgical site, managed conservatively.

Discussion

Alimentary tract duplications are rare congenital anomalies affecting any part of the alimentary canal, from the mouth to the anus [12, 13]. Duplications of the stomach, duodenum, and rectum are the rarest forms. Gastric duplications are classified into cystic and tubular forms. The cystic form, constituting 80%, has no communication with the gastric lumen, while the latter, representing 20% of gastric duplications, communicates with the gastric lumen [5]. GDCs account for only 4% of GIT duplications, with an incidence of 17/1,000,000 individuals [14].

GDCs are most commonly found along the greater gastric curvature, with occasional findings in the cardia, pylorus, and both the anterior and posterior walls of the stomach [12]. Most cases are diagnosed in childhood, usually within the first 3 months and rarely after the age of twelve [12]. No consensus exists on gender distribution. Some authors reported a female predominance with a ratio of 8:1, while others observed a male predilection [12, 14]. In another study, Li et al. [5] found equal gender distribution.

GDCs can present with diverse symptoms, ranging from none to abdominal pain, epigastric pain, nausea, vomiting, weight loss, bloody stools, and feeding difficulties. Symptoms may manifest before the age of 2 and remain asymptomatic in adults. Due to non-specific symptomatology, the disease may be misdiagnosed, or even complications like infection, ulceration, bleeding, perforation, fistula formation, and compression can occur [12, 13].

GDC misdiagnosis with different abnormalities has been discussed in the literature, such as pancreatic cysts, renal cysts, and stomach muscle adenomas [15]. Ren et al. [15] reported a misdiagnosed case initially thought to be a choledochal cyst on MRI, later confirmed as a GDC intraoperatively. Another study mentioned that because GDCs are commonly found on the greater curvature of the stomach, they may compress adjacent organs and appear to originate from them [16].

Despite all of that, the mimicking of GDCs as an adrenal mass has rarely been reported [3]. Al Awfi et al. [3] reported a case of a 2-year-old girl with abdominal pain. The preoperative diagnosis based on imaging suggested a retroperitoneal supra-adrenal cyst. However, the histopathological examination after laparoscopic resection revealed a GDC. In another study, Castillo-Fernández et al. [7] reported a case of retroperitoneal gastric duplication that mimicked an adrenal cyst in a prenatal baby girl. Terry et al. [8] reported a retroperitoneal foregut duplication cyst in an elderly woman, presenting as an adrenal mass preoperatively. The patient also reported a 25-kg weight loss and decreased appetite.

Our case was a young lady who presented with chronic epigastric pain, nausea, intermittent vomiting, and food intolerance. The condition was preoperatively misdiagnosed as an adrenal mass. The literature notes that due to their rarity, non-specific presentation, and proximity to or compression of adjacent organs, GDCs are prone to misdiagnosis with

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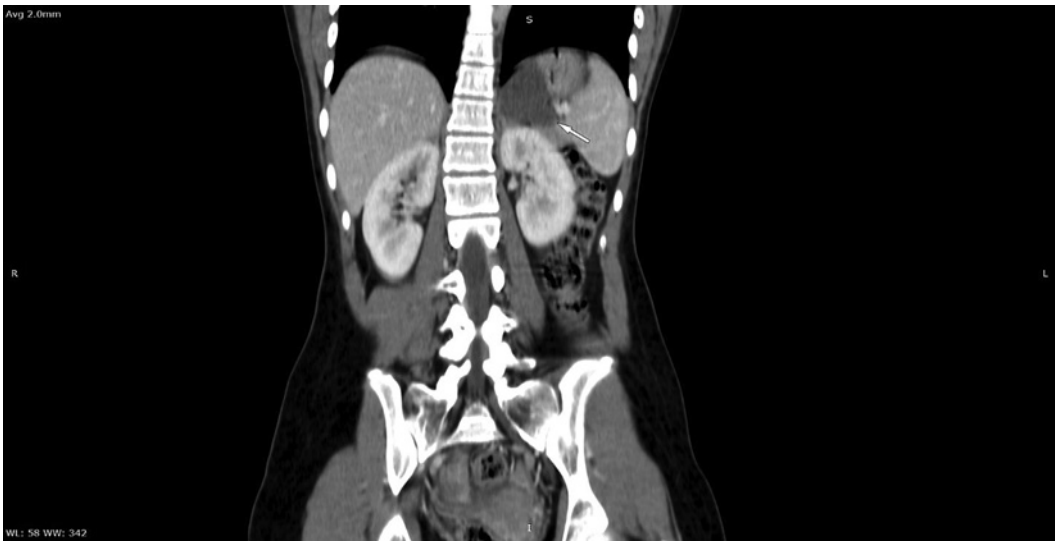

Fig. 1. Abdominal CT with IV contrast, coronal section, shows a cystic lesion between the stomach and upper pole of the left kidney (white arrow).

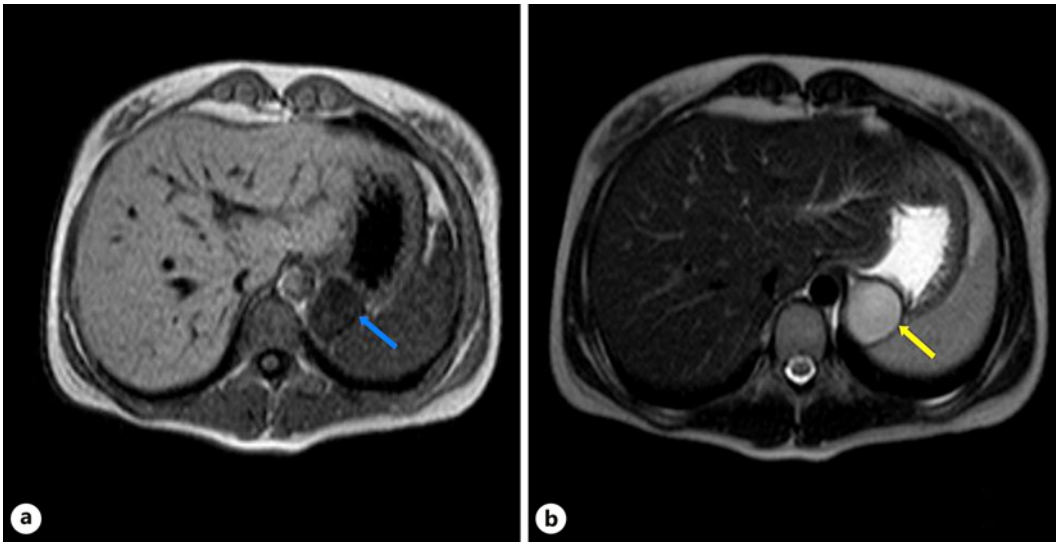

Fig. 2. a Axial section of the upper abdomen, T1 opposed phase MRI, shows a well-defined hypointense lesion in the region of the left adrenal (blue arrow). **b** Axial section of the upper abdomen, T2 sequence MRI, shows a fluid signal lesion between the fundus of the stomach and spleen, in the region of the left adrenal (yellow arrow).

other diseases and phenomena [15, 17]. The GDC in our case was located on the posterior part of the greater curvature of the stomach, and this can be the explanation for the misdiagnosis.

The major characteristics for diagnosing GDC include the following: the cyst wall is continuous with the stomach; the cyst is surrounded by a smooth muscle shared with the stomach; both share the same blood supply; and the cyst wall is lined by a benign epithelium [17]. Smooth muscle, on a cellular level, is identified as an involuntary muscle displaying a non-striated pattern and comprising thick and thin filaments without organizing into sarcomeres. Under microscopic

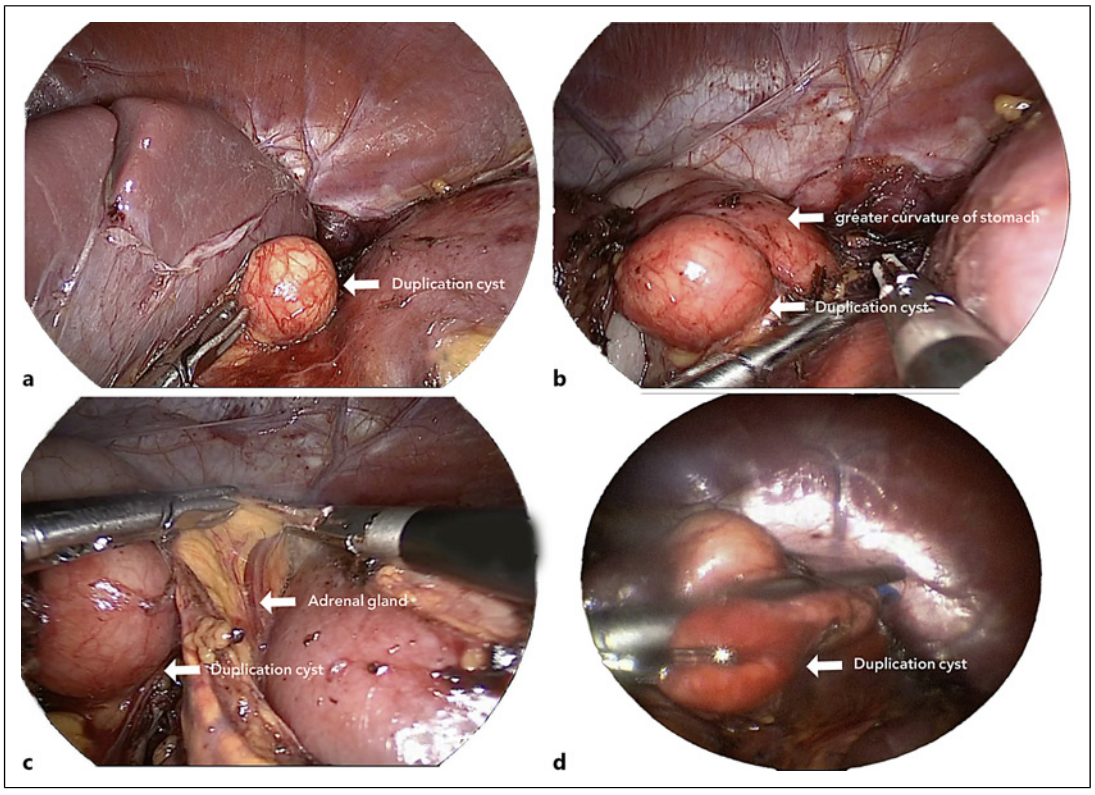

Fig. 3. Intraoperative laparoscopic view. **a** A cystic mass between the spleen and upper pole of the left kidney. **b** The cyst is attached to the greater curvature of the stomach. **c** Normal appearance of the left adrenal. **d** Stapler wedge resection of the stomach.

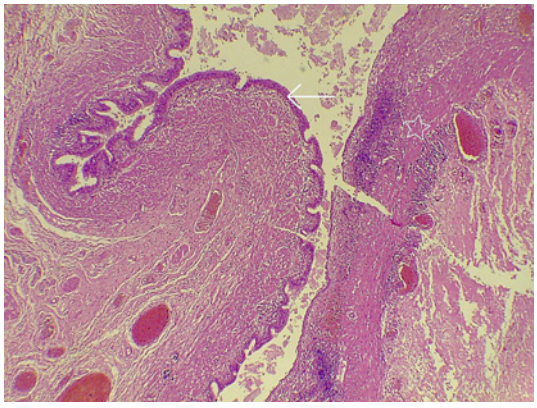

Fig. 4. The section shows that the cyst wall is lined by pseudostratified ciliated columnar epithelial cells (arrow), with the underlying smooth muscle wall (star).

examination, its appearance is homogenous. The cytoplasm of smooth muscle is notably rich in both actin and myosin [18]. The same was reported in the histopathological results of the current case. The preoperative diagnosis of GDCs is difficult, and there is no gold-standard examination. U/S or radiographic modalities can be used as the initial options, although they are not specific. OGD is valuable for identifying ulcers and strictures and defining the anatomy of upper GIT duplication cysts, both preoperatively and intraoperatively, though it is not typically diagnostic. CT and MRI scans can also be used in complicated cases or when the U/S finding is unclear [12, 13]. In the current case, OGD revealed normal findings except for mild antral erosion. U/S, CT, and MRI

results all indicated an adrenal cyst, leading to the preoperative consideration of an adrenal cyst diagnosis. Surgical resection is suggested for symptomatic GDC cases, but controversy surrounds asymptomatic ones. Some scholars also recommended surgery for asymptomatic cases due to the potential for neoplastic transformation [12, 19]. Partial gastrectomy or cystectomy, whether through open surgery or laparoscopy, is often the preferred approach for managing patients with GDC [20]. Laparotomy has also been suggested for the management of complicated GDCs [13]. Laparoscopy significantly aids in diagnosing GDCs and is linked to fewer traumatic injuries and improved postoperative recovery [15]. In this case, laparoscopy revealed that the cyst originated from the posterior part of the greater curvature of the stomach. Following consultation with a gastrointestinal surgeon, a wedge resection of the cyst was performed. The histopathology examination confirmed a GDC.

In conclusion, GDCs are rare congenital malformations that may become symptomatic during adulthood. They can be easily misdiagnosed as adrenal masses, and complications may progress if left untreated.

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Dana Gharib, Hoshmand Asaad, and Ismaeel Aghaways were not available to confirm co-authorship, but the corresponding author, Fahmi Kakamad, affirms that they contributed to the paper, had the opportunity to review the final version to be published, and guarantees their co-authorship status and the accuracy of the author contribution and conflict of interest statements.

Statement of Ethics

Ethics approval was not required for this case in accordance with the national guidelines. Written informed consent was obtained from the parent of the patient for publication of the details of her medical data and any accompanying images.

Conflict of Interest Statement

All the authors declare that there is no conflict of interest regarding the publication of this article.

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Author Contributions

Rawa Bapir and Hemn Ali, surgeons, managed the case and majorly contributed to the study, literature review, and final approval of the manuscript. Soran Tahir, a radiologist, performed the radiological assessment of the case. Ari Abdullah, a pathologist, performed the histological analysis of the case specimen, a major contributor to the study, and critically revised the manuscript. Deedar Qader critically revised the manuscript and gave final

approval of the manuscript. Dana Gharib, Hoshmand Asaad, Shaho Ahmed, Sasan Ahmed, and Ismaeel Aghaways were involved in the literature review, design of the study, revision of the manuscript, and processing of the figures. Fahmi Kakamad and Hiwa Abdullah contributed to the literature review, writing of the manuscript, and final approval of the manuscript.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author.

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