ISSN 2307-8960 (online)

World Journal of *Clinical Cases*

World J Clin Cases 2024 May 6; 12(13): 2138-2292

Published by Baishideng Publishing Group Inc

World Journal of Clinical Cases

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ABOUT COVER

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RESPONSIBLE EDITORS FOR THIS ISSUE

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Thrice Monthly

PUBLICATION DATE

May 6, 2024

World Journal of Clinical Cases

Submit a Manuscript: https://www.f6publishing.com

World J Clin Cases 2024 May 6; 12(13): 2254-2262

DOI: 10.12998/wjcc.v12.i13.2254

ISSN 2307-8960 (online)

CASE REPORT

Young patient with a giant gastric bronchogenic cyst: A case report and review of literature

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Specialty type: Medicine, research and experimental

Provenance and peer review: Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's scientific quality classification

Grade A (Excellent): 0
Grade B (Very good): 0
Grade C (Good): C
Grade D (Fair): 0
Grade E (Poor): 0

P-Reviewer: Utano K, Japan

Received: December 17, 2023
Revised: February 9, 2024
Accepted: April 2, 2024
Published online: May 6, 2024

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Abstract

BACKGROUND

Gastric bronchogenic cysts (BCs) are extremely rare cystic masses caused by abnormal development of the respiratory system during the embryonic period. Gastric bronchial cysts are rare lesions that were first reported in 1956; as of 2023, only 33 cases are available in the PubMed online database. BCs usually have no clinical symptoms in the early stage, and imaging findings also lack specificity. Therefore, they are difficult to diagnose before histopathological examination.

CASE SUMMARY

A 34-year-old woman with respiratory distress presented at our hospital. Endoscopic ultrasound revealed an anechoic mass between the spleen, left kidney and gastric fundus, with hyperechogenic and soft elastography textures and with a size of approximately 6.5 cm × 4.0 cm. Furthermore, a computed tomography scan demonstrated high density between the posterior stomach and the spleen and the left kidney, with uniform internal density and a small amount of calcification. The maximum cross section was approximately 10.1 cm × 6.1 cm, and the possibility of a cyst was high. Because the imaging findings did not suggest a malignancy and because the patient required complete resection, she underwent laparotomy surgery. Intraoperatively, this cystic lesion was found to be located in the posterior wall of the large curvature of the fundus and was approximately 8 cm × 6 cm in size. Finally, the pathologists verified that the cyst in the fundus was a gastric BC. The patient recovered well, her symptoms of chest tightness disappeared, and the abdominal drain was removed on postoperative day 6, after which she was discharged on day 7 for 6 months of follow-up. She had no tumor recurrence or postoperative complications during the follow-up.

CONCLUSION
This is a valuable report as it describes an extremely rare case of gastric BC. Moreover, this was a very young patient with a large BC in the stomach.

Key Words: Bronchogenic cyst; Stomach; Endoscopic ultrasound-guided fine needle aspiration; Endosonography; Case report

Core Tip: Gastric bronchogenic cysts (BCs) represent uncommon congenital anomalies, often manifesting as indistinct cystic formations on preoperative evaluations. Herein, we document a noteworthy instance of a sizable gastric BC occurring in a young female patient. The definitive diagnosis of gastric BC was established through histopathological examination following laparotomy resection. The analysis of the reported cases revealed that gastric BC often mimics gastrointestinal stromal tumors on preoperative imaging. We recommend elective radical surgical resection for young patients with large cysts, as they might progress to malignancies.

Citation: Lu XR, Jiao XG, Sun QH, Li BW, Zhu QS, Zhu GX, Qu JJ. Young patient with a giant gastric bronchogenic cyst: A case report and review of literature. *World J Clin Cases* 2024; 12(13): 2254-2262
URL: https://www.wjgnet.com/2307-8960/full/v12/i13/2254.htm
DOI: https://dx.doi.org/10.12998/wjcc.v12.i13.2254

INTRODUCTION

Bronchogenic cysts (BCs) are uncommon developmental malformations that result from the aberrant development of the primitive tracheobronchial tube, leading to the formation of cystic lesions. These lesions are typically congenital, meaning they are present at birth, and can cause various symptoms depending on their location and size[1]. The lesions can be divided into mediastinal, intrapulmonary, and ectopic types according to their location[2]. The tumor is primarily located in the mediastinum if it occurs early in gestation, as opposed to the thoracic cavity if it arises later in development[3]. Abdominal BCs, especially those situated within the gastric wall, are exceptionally uncommon occurrences[4]. Dewing *et al*[5] were the pioneers in describing gastric bronchial cysts in 1956, and as of 2023, only a few cases have been described in reports available in the PubMed online database. For patients with BCs, there is a wide range for the age at diagnosis, which has ranged from 17 to 81 years, and it has a higher prevalence in females, with a median age of development of 43 years[1]. BCs of the gastric region typically manifest along the posterior wall of the gastric body and the lesser curvature of the stomach[6]. In previous reports, it has been observed that a significant proportion of patients were asymptomatic [7]. However, among those who exhibited symptoms, epigastric pain and vomiting were the most prevalent[8]. Most gastric BCs are easily misdiagnosed as gastrointestinal stromal tumors (GISTs) before surgery[9]; however, fortunately, their prognosis is good[10]. In the current investigation, we have documented a rare instance of a gastric BCs occurring in a 34-year-old patient. Furthermore, we have undertaken a thorough examination of the existing literature (Table 1) to delve into the clinical manifestations associated with these cysts, aiming to contribute to a deeper understanding of their nature and incidence.

CASE PRESENTATION

Chief complaints

A 34-year-old Chinese woman presented to the gastrointestinal surgery clinic with a complaint of respiratory distress for 5 d.

History of present illness

A 34-year-old Chinese female patient presented with chest tightness and shortness of breath for 5 d with no nausea, vomiting, sour regurgitation, belching, dysphagia, melena, or weight loss.

History of past illness

She had a history of psoriasis.

Personal and family history

Her father died of lymphoma. In addition, the patient denied any family history of other malignancies.

Table 1 Thirty-three cases

Ref.	Age	Gender	Size (cm)	Location	Symptom	Preoperative diagnosis	Intervention
Braffman *et al*[24], 1988	64	Female	15 × 8	Posterior wall	Epigastric pain		Resection
Matsubayashi *et al*[25], 2003	62	Male	10 × 3 × 3	Posterior wall		Lymphangioma/benign	Resection
Hedayati *et al*[26], 2003	59	Female	7 × 5	Posterior wall		Adrenocortical cancer	Laparoscopic
Rubio *et al*[27], 2005	26	Male			Epigastric pain		
Melo *et al*[12], 2005	39	Female	4 × 2.5 × 1	Gastric fundus	Rib pain	GIST	Laparoscopic resection
Song *et al*[28], 2005	62	Female	1.7	Lesser curvature		Benign stromal tumor	Resection
Wakabayashi *et al*[29], 2007	37	Male	5	Lesser curvature	Epigastric pain	A duplication cyst	Exploration
Sato *et al*[19], 2008	60	Female	3			Benign cyst of the gastric submucosa	
Shibahara *et al*[17], 2009	43	Male	9 × 4	Lesser curvature	Epigastric pain	Gastric cancer	
Jiang *et al*[30], 2010	25	Female	3 × 2.5 × 2.0	Gastric fundus	Epigastric pain	GIST	Laparotomy
Tan *et al*[10], 2010	30	Female	5.1 × 3.6 × 4.6	Posterior wall of the stomach			Laparoscopic wedge resection
Kurokawa *et al*[31], 2013	71	Male	3.2	Gastric cardia	Throat discomfort	Cyst	Laparoscopic
Yang and Guo[6], 2013	50	Male	8.0 × 6.0	Gastric fundus		Retroperitoneal mass	Laparotomy
Yang and Guo[6], 2013	37	Female	10 × 6	Posterior wall of the stomach		GIST	Wedged gastrectomy
Sun *et al*[32], 2015	67	Male	4.1 × 3.2	Gastric fundus	Dull epigastric pain	GIST	Laparotomy
Tu *et al*[1], 2016	17	Female	3 × 2.5	Gastric cardia	Epigastric pain	Cyst	Laparotomy
Chhaidar *et al*[33], 2017	65	Female	7 × 8	Gastric cardia	Epigastric pain	GIST	Total gastrectomy
Xiao *et al*[13], 2020	62	Female	6.4 × 4.9	Lesser curvature of the gastric cardia	Right lower abdominal pain	GIST	Laparoscopic
He *et al*[15], 2020	55	Female	7 × 6 × 2	Gastric cardia	Intermittent epigastric pain	Benign cyst of the gastric submucosa	Da vinci robotic-assisted laparoscopic
Sun *et al*[7], 2020	68	Male	10 × 8 × 8	Fundus of stomach		Retroperitoneal mass	Laparotomy
Erbenová *et al*[34], 2021	30	Female		Gastric cardia		Tumor of gastric cardia	Laparoscopic
Lou *et al*[8], 2022	38	Female	5 × 2.6	Gastric fundus	Upper abdominal pain	Lymphatic cyst	Laparoscopic
Wang *et al*[35], 2022	76	Male	3.1 × 3.0	Gastroesophageal junction	Abdominal pain and distension	GIST	Laparoscopic
Li *et al*[36], 2022	35	Male		Posterior wall of the gastric cardia		Abdomystic Lymphangioma	Laparotomy
Lv *et al*[37], 2023	23	Male		Greater curvature	Abdominal discomfort		
Qian and Xu[16], 2023	50	Male	2 × 1.5	Posterior gastric fundus wall		Schwannoma or low-grade stromal tumor	Gastroscope
Qian *et al*[38], 2023	45	Female	3 × 2	Gastric cardia	Upper abdominal pain	GIST	Laparoscope

Ma *et al*[39], 2023	46	Female	5 × 4.5 × 3.5	Gastric body	Abdominal pain	GIST	
Lin and Cao[40], 2023	47	Male	8 × 7	Posterior wall of the fundus	Pain in the right chest		
Liu and Li[41], 2023	65	Male	4 × 3	Gastric cardia	Blech		
Terayama *et al*[42], 2023	37	Female	3	Gastric cardia	Asymptomatic	Bronchogenic cyst	Resection
Terayama *et al*[42], 2023	47	Male	5	Gastric cardia	Asymptomatic	Gastric bronchogenic cyst	Resection
Terayama *et al*[42], 2023	37	Male	3.5	Gastric cardia	Asymptomatic		Resection

GIST: Gastrointestinal stromal tumor.

Physical examination

Her abdomen was smooth and flat, and there was no gastrointestinal type or mild upper abdominal tenderness without rebound pain or an overt mass. The bowel sounds were normal.

Laboratory examinations

No abnormalities were found in routine blood or urine analyses or liver or kidney function tests.

Imaging examinations

Endoscopic ultrasound (EUS) revealed a 65 mm × 40 mm single cyst. The images also showed that the cyst was located at the bottom of the gastric wall. Anechoic masses were detected between the spleen, left kidney, and gastric fundus with punctate hyperechogenicity. Elastography revealed a soft texture. These findings suggested the presence of a cyst within the stomach (Figure 1). Additionally, an enhanced computed tomography (CT) scan revealed a cystic mass measuring 101 mm × 61 mm in size. There was no contrast enhancement, and the mass was located within the posterior wall of the gastric fundus, spleen and left kidney with regular and smooth outlines. The mass appeared to have a slightly high and uniform density with a small, calcified shadow in the posterior gastric region. No septation was observed. It also showed an extraluminal growth pattern with an obvious border, and the gastric wall was affected by pressure (Figure 2). Moreover, no significant enlargement of lymph nodes was observed in the vicinity of the stomach or retroperitoneal region. Before surgery, an EUS examination was performed to determine which layer of the gastric wall the cyst had originated from. However, because the cyst wall was evaluated by CT, because there was calcification on the cyst wall, and because the contents of the cyst were mainly liquid components according to the density, there was concern that fine needle aspiration (FNA) may cause rupture of the cyst before surgery and would increase the risk of infection. Second, we suspected that the patient's chest tightness was caused by compression of the diaphragm muscle by the large cyst. To eliminate the patient's symptoms and because the patient strongly desired surgery, after consultation and discussion with many experts, we decided to prudently remove the cyst and obtain a complete pathological specimen so that a safe postoperative examination could be performed to obtain the most accurate diagnosis. Based on these two points, we did not use preoperative FNA.

FINAL DIAGNOSIS

After treatment, the patient's symptoms of chest tightness resolved. We suspected that the chest tightness was due to the large cyst exerting pressure on the diaphragm. Based on the obtained specimen, this mass was approximately 80 mm × 50 mm × 40 mm in size. Under microscopic examination, the cyst lining exhibited pseudostratified ciliated columnar epithelial cells, while the cyst wall displayed smooth muscle and small salivary gland tissue. Immunohistochemical staining revealed the following results: CK7 (+) TTF-1 (partial +), NapsinA (+), CK20 (-), Villin (-), SMA (+), Desmin (+), P63 (+), and elastic fiber (+) (Figure 3). The pathologists conclusively confirmed that the cystic mass located in the fundus was indeed gastric BCs.

TREATMENT

Given the young age of the patient, the large cyst with prominent gastric wall compression and chest tightness could have been associated with the cystic mass, and given that the patient wished to have the lesion completely removed, the patient underwent intra-abdominal mass resection under general anesthesia and nerve block anesthesia. Intraoperative observations of this cystic mass revealed it to be a smooth, single-port cyst originating from the posterior wall of the gastric fundus and extending along the greater curvature. Surgical exploration revealed no intra-abdominal ascites or

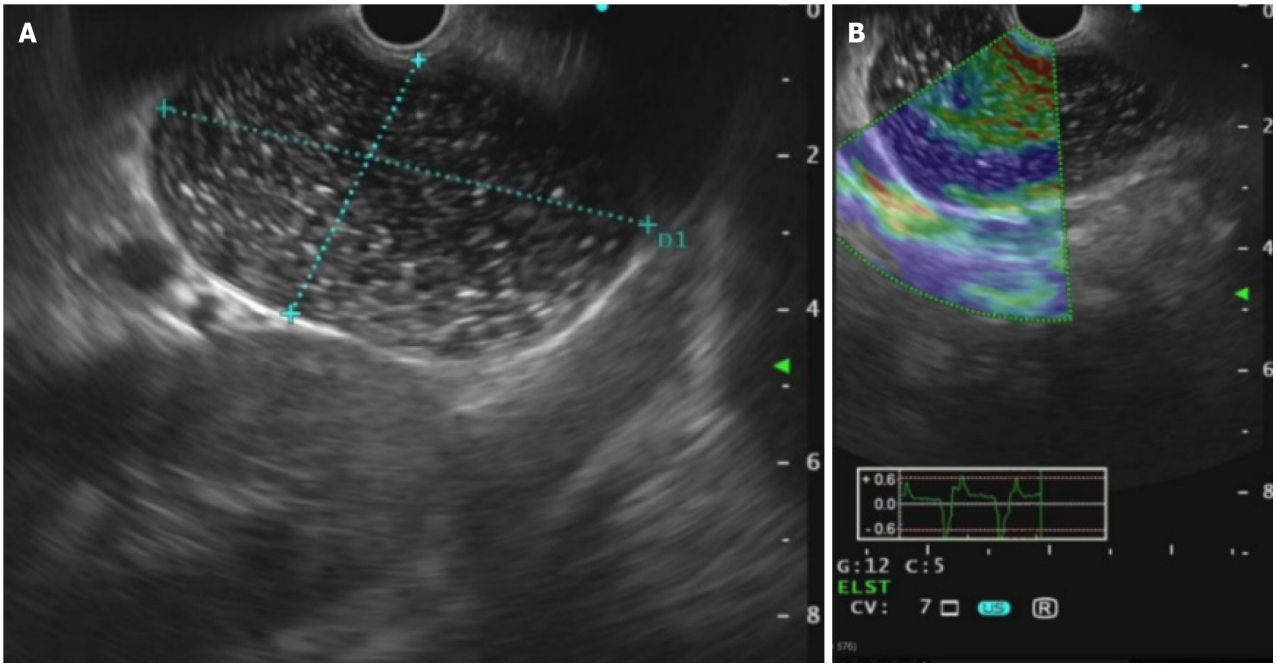

Figure 1 Imaging at admission. Endoscopic ultrasound revealed a 6.5 cm × 4.0 cm single cyst. The cyst, without any echoes or color flow signals, was located in the posterior wall of the gastric body. These results indicated the possibility of a cyst of the stomach.

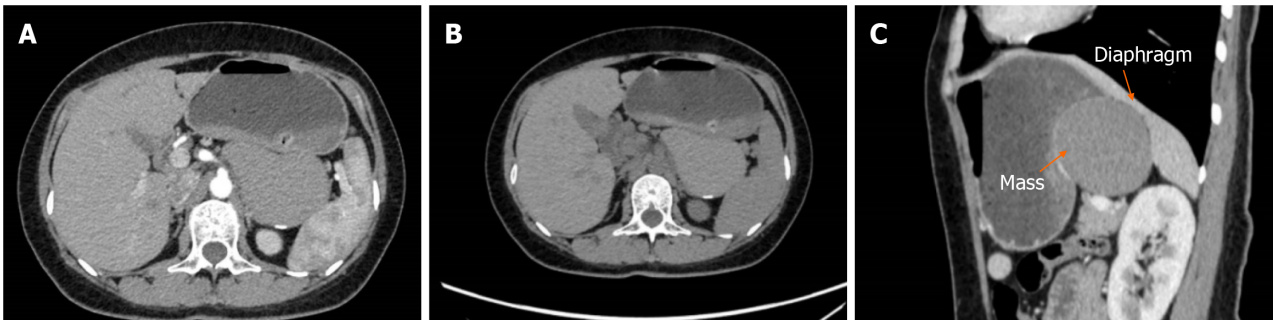

Figure 2 Imaging at admission. A: An enhanced computed tomography scan demonstrated a 10.1 cm × 6.1 cm mass. The mass was present in the posterior part of the stomach between the spleen and the left kidney; a large, slightly high-density swelling shadow with uniform internal density was shown; and a small calcification shadow could be seen at the rear. The computer tomography (CT) value was approximately 70 HU, and the gastric wall was compressed and changed; B: Noncontrast CT image; C: It can be clearly seen from the sagittal position that the diaphragm was compressed by the mass.

obvious abnormalities in the liver, abdominal wall, pelvic cavity, omenta, or mesentery. The cystic mass was then completely dissected from the stomach. It is worth noting that surgeons need to avoid cyst rupture during these types of surgery.

OUTCOME AND FOLLOW-UP

The patient recovered well, her symptoms of chest tightness completely disappeared, and the abdominal drain was removed on postoperative day 6, after which she was discharged on day 7 for 6 months of follow-up. The patient had no tumor recurrence or postoperative complications that occurred after surgery.

DISCUSSION

To conduct a comprehensive study on BCs of the stomach, a systematic literature review was undertaken using the PubMed database. The search was focused on articles published in English and employed the keyword "gastric BCs" to identify relevant studies. The final date for data collection was set as December 2023. The inclusion criteria stipulated that all patients must have a confirmed diagnosis of gastric BCs through pathological examination. Additionally, patients exhibiting imaging characteristics typical of gastric BCs were included, irrespective of age and sex. Conversely, patients

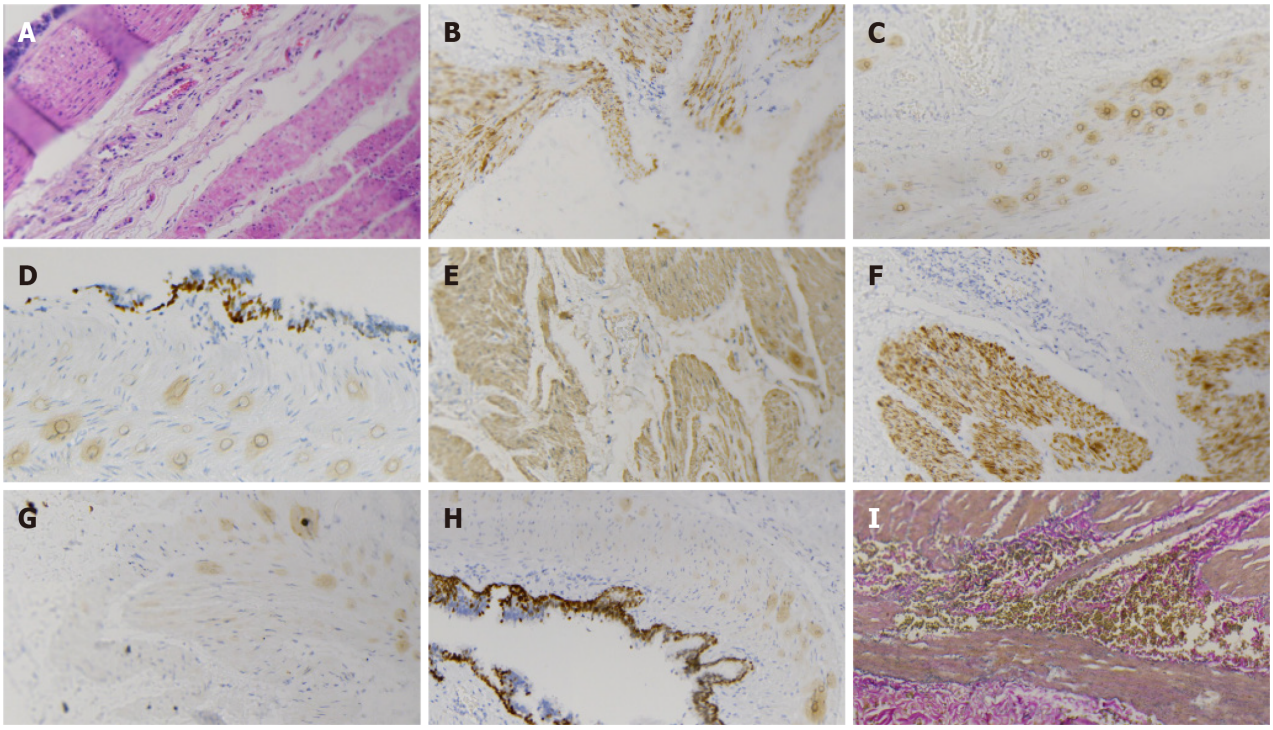

Figure 3 Histopathological analysis and immunohistochemical examination of the resected specimen. A: Microscopically, pseudostratified ciliated columnar epithelial cells could be observed in the cyst lining, and smooth muscle and small salivary gland tissue could be observed in the cyst; B: Napsin A (+); C: CK20 (-); D: P63 (+); E: SMA (+); F: Desmin (+); G: CK7 (+); H: TTF-1 (+); I: Elastic fiber (+).

lacking typical pathological or imaging features were excluded from the study. Based on our knowledge cutoff of December 2023, a total of 33 cases of gastric BCs were reported, meeting the specified search criteria. These cases are comprehensively listed in Table 1.

BCs are primitive-foregut-derived congenital cystic abnormalities[11]. Migration of BCs may ensue when their attachments to the trachea or esophagus fail to persist, resulting in their potential displacement within the anatomical structures of the body[12]. The primary sites of occurrence for BCs predominantly involve the thoracic region, notably within the mediastinum. However, on rare occasions, they may also manifest in the subdiaphragmatic region. BC of the stomach appears to be a disease detected at all ages (from 17 to 76 years of age), and there was no apparent sex difference (17 females and 16 males). The dimensions of BCs exhibited considerable variability, ranging from 1.7 to 15 cm, as indicated by available data from our cases and referenced literature. However, the majority of cyst diameters fell within the range of 3 to 7 cm. Regarding localization, our investigation revealed a predilection for cysts to be situated in the gastric cardia or posterior wall of the fundus. A large proportion of patients with BC have clinical manifestations of epigastric pain, while others generally experience nonspecific symptoms, which may be due to local tumor compression and infection[13]. Notably, four patients with gastric BC had elevated tumor marker levels. Elevated CA19-9 levels were present in two of the patients[10,14], and elevated CA72-4 levels were also present in patients with elevated CA72-4 levels [15,16]. Interestingly, these elevated tumor markers returned to normal after surgery, suggesting that there is a direct relationship between benign BCs and elevated tumor marker levels. However, the relationship between tumor markers and BC needs further study. It is worth noting that BCs of the stomach have been associated with the presence of gastric carcinoma. Chronic inflammation of the gastric mucosa, stemming from BCs, may have contributed to the development of these adenocarcinomas in the stomach, as reported in previous studies[17]. While the current investigation illustrates that EUS and other imaging modalities can aid in localizing the lesion, they are limited in their ability to offer qualitative diagnostic insights. Accurate preoperative diagnosis is challenging, and most patients are easily misdiagnosed with GIST. In summary, preoperative diagnosis of gastric BCs is challenging due to the absence of specific clinical manifestations, as well as inconclusive findings from laboratory tests and imaging studies. Moreover, the rarity of these lesions further complicates their diagnosis.

BCs are commonly detected through CT and magnetic resonance imaging (MRI). However, relying solely on imaging techniques to differentiate them from other types of cysts can be challenging due to the presence of similar radiological features among various cystic lesions. Ubukata *et al*[18] demonstrated that far greater clarity was achieved when using MRI than when using CT, especially for identifying the contents of the cystic lesions. In the present case, since the patient had metal dental implants, this patient was not suitable to undergo MRI. EUS is commonly employed to ascertain the specific layer of the gastric wall from which the lesion originates and to delineate its approximate location within the gastrointestinal tract[15]. Imaging alone cannot usually distinguish between nonneoplastic lesions and benign or malignant neoplasms. In clinically warranted situations, EUS-FNA biopsy has been previously established as a valuable tool for the unequivocal diagnosis of gastric BCs[19]. Its effectiveness is further emphasized by its sensitivity range of 86%-93%[20], diagnostic accuracy spanning 82%-95%, and an exceptionally low complication rate of merely 1%-3%[21].

Although the probability of complications is extremely low, there are risks of ulceration, infection and hemorrhage. EUS-FNA provides a new diagnostic and treatment approach for asymptomatic gastric BC patients. We recommend that EUS-FNA be performed first for elderly patients, patients in poor physical condition, asymptomatic patients, and patients who cannot undergo surgical treatment for various reasons in the short term; then subsequent treatment options should be discussed based on the results.

Through our systematic review of existing cases, we conclude that GIST is the most common preoperative diagnosis of gastro BCs. The two conditions are difficult to distinguish by imaging, and an accurate preoperative diagnosis can be obtained *via* EUS-FNA before surgery. However, there are risks such as infection and bleeding. During the operation, these masses can be distinguished by observing their nature. GISTs are brittle and prone to bleeding, while gastro BCs are composed mainly of cystic components[9]. Second, due to the rarity of BCs, clinicians lack understanding of this disease, which is also one of the reasons for the failure to obtain an accurate preoperative diagnosis of this disease.

The ultimate diagnosis typically hinges on a histopathological analysis of specimens obtained postoperatively. A review of the literature revealed that surgical resection was the most common option. Surgical removal will improve the symptoms of cyst compression and reduce the risk of BC transforming into a malignant tumor[22]. The findings from the current literature review indicate that asymptomatic patients harboring small masses require careful monitoring. Conversely, for symptomatic patients, particularly those who are young as exemplified in this case report, surgical resection is advisable. In the case presented, the patient's respiratory distress was attributed to the growing mass exerting pressure on the diaphragmatic muscle. In addition to routine laparoscopic resection, Lee *et al*[23] proposed endoscopic mucosal resection (EMR) for the treatment of gastric BC. They proposed that when a lesion is suspected to be a solid tumor on the basis of EUS and CT investigations and if there is a positive cushion sign, the differential diagnosis of a developmental cyst should be considered, and EMR could be used for curative treatment. Regardless of the operation method, care should be taken to avoid intraoperative cyst rupture and postoperative infection complications. Due to the large size of the cyst in this case, open surgery was chosen to obtain a sufficient surgical field of view and ensure complete resection.

Although gastric BCs are a very rare disease, when comparing our case with those reported in PubMed, it can be seen that there are no specific clinical manifestations or laboratory indicators associated with BC. Elevated tumor markers have been reported in some cases; however, the sample size was insufficient to support an association. Early detection of suspected lesions is a favorable factor affecting patient survival. In addition, CT, MRI, and EUS are popular methods for detecting gastric BCs. Surgical removal is the most common way to relieve symptoms; however, the recommendation of surgical intervention for asymptomatic patients remains controversial. Through our diagnosis and treatment and the postoperative follow-up of this patient, we would like to show that surgical resection is recommended for young patients with large cysts and clinical symptoms to eliminate symptoms and the uncertainty of transformation of gastro BCs into malignant tumors.

CONCLUSION

Although gastric BCs are a very rare disease, when comparing our case with those reported in PubMed, it can be seen that there are no specific clinical manifestations or laboratory indicators associated with BC. Elevated tumor markers have been reported in some cases; however, the sample size was insufficient to support an association. Early detection of suspected lesions is a favorable factor affecting patient survival. In addition, CT, MRI, and EUS are popular methods for detecting gastric BCs. Surgical removal is the most common way to relieve symptoms; however, the recommendation of surgical intervention for asymptomatic patients remains controversial. Through our diagnosis and treatment and the postoperative follow-up of this patient, we would like to show that surgical resection is recommended for young patients with large cysts and clinical symptoms to eliminate symptoms and the uncertainty of transformation of gastro BCs into malignant tumors.

FOOTNOTES

Author contributions: Qu JJ was responsible for the overall project progress, paper revision and submission; Lu XR and Jiao XG contributed to manuscript writing and editing and data collection; Sun QH and Zhu QS contributed to the data analysis; Li BW and Zhu GX contributed to the conceptualization and supervision. All the authors read and approved the final manuscript.

Supported by Weifang Municipal Health Commission Scientific Research Project, No. WFWSHKK-2021-028; and Shandong Province Medical Health Science and Technology Project, No. 202304010544.

Informed consent statement: Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Conflict-of-interest statement: The authors declare that they have no conflicts of interest to disclose.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Country/Territory of origin: China

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S-Editor: Liu H
L-Editor: A
P-Editor: Zhao S

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