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WORD COUNT	2967	TIME SUBMITTED	22-MAR-2024 11:59AM
		PAPER ID	107744473

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Name of Journal: *World Journal of Clinical Cases*

Manuscript NO: 90924

Manuscript Type: CASE REPORT

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

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 6.5×4.0 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 10.1×6.1 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 8×6 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 bronchogenic cyst in the stomach.

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

Xuren L, Xuguang J, Qihang S, Li B, Qingshun Z, Guangxu Z, Qu J. Young patient with a giant gastric bronchogenic cyst: A case report and literature review. *World J Clin Cases* 2024; In press

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. These diseases should be included in the differential diagnosis when dealing with an intramural gastric lesion close to the fundus or cardia of the stomach. We recommend elective radical surgical resection for young patients with large cysts, as they might progress to malignancies. Endoscopic ultrasound-guided fine needle aspiration (EUS-FNA) holds promise as a means to attain a definitive diagnosis of gastric bronchogenic cysts (BCs) prior to surgical intervention, yet further investigation is warranted to validate its efficacy in this regard.

INTRODUCTION

Bronchogenic cysts 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 bronchogenic cysts, 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]. Bronchogenic cysts (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 before surgery[9]; however, fortunately, their prognosis is good[10]. In the current investigation, we have documented a rare instance of a gastric bronchogenic cyst 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 days.

History of present illness

A 34-year-old Chinese female patient presented with chest tightness and shortness of breath for 5 days 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.

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 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 a gastric bronchogenic cyst.

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 obvious abnormalities in the liver, abdominal wall, pelvic cavity, omentum, 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 type 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 bronchogenic cysts 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 bronchogenic cysts" 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 bronchogenic cyst through pathological examination. Additionally, patients exhibiting imaging characteristics typical of gastric bronchogenic cysts were included, irrespective of age and sex. Conversely, patients 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 bronchogenic cysts were reported over a 10-year period, meeting the specified search criteria. These cases are comprehensively listed in Table 1.

Bronchogenic cysts are primitive-foregut-derived congenital cystic abnormalities[11]. Migration of bronchogenic cysts 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

bronchogenic cysts 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 bronchogenic cysts (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 3.0 to 7.0 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 bronchogenic cysts of the stomach have been associated with the presence of gastric carcinoma. Chronic inflammation of the gastric mucosa, stemming from bronchogenic cysts, 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 gastrointestinal stromal tumors. In summary, preoperative diagnosis of gastric bronchogenic cysts (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 computed tomography (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. Endoscopic ultrasound (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-fine needle aspiration (FNA) biopsy has been previously established as a valuable tool for the unequivocal diagnosis of gastric bronchogenic cysts[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 gastrobronchogenic cysts. 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 gastrobronchogenic cysts are composed mainly of cystic components[9]. Second, due to

the rarity of bronchogenic cysts, 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 gastrobronchogenic cysts 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 gastrobronchogenic cysts into malignant tumors.

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