

Case Report

Multiple Bronchogenic and Gastroenteric Cysts Arising from the Stomach in a Patient with Abdominal Pain

Maykong Leepalao¹ and Jessica Wernberg²

¹Department of General Surgery, Marshfield Clinic, Marshfield, WI 54449, USA

²Department of Surgical Oncology, Marshfield Clinic, Marshfield, WI 54449, USA

Correspondence should be addressed to Maykong Leepalao; leepalao.maykong@marshfieldclinic.org

Received 12 May 2015; Accepted 23 June 2015

Academic Editor: Francesco Petrella

Copyright © 2015 M. Leepalao and J. Wernberg. This is an open access article distributed under the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

Bronchogenic cysts arising from the stomach are uncommon. We discuss a young female patient with presumed enteric duplication cysts who was found to have three bronchogenic and gastroenteric cysts upon pathologic review. We discuss the pathophysiology of bronchogenic cysts and their malignant potential.

1. Introduction

Bronchogenic cysts arising from the stomach are a relatively rare entity. There have been reported cases of intra-abdominal bronchogenic cysts as early as fifty years ago with less than 25 published reports in the literature. This report highlights a rare case of three bronchogenic cysts arising in the gastric wall of an adult patient.

2. Background

A 29-year-old female who had experienced severe left upper quadrant pain during pregnancy presented to our clinic. During her pregnancy, she underwent imaging which demonstrated several large cystic structures felt to be arising from the stomach. These were presumed to be enteric duplication cysts. Due to the symptomatic nature, diagnostic insecurity, and concern over malignant potential of these cysts, resection was recommended.

She did not have any other pertinent medical history. She ran cross country in high school and was healthy. She had no other issues with her prior pregnancies. She was a nonsmoker.

On physical exam, she had palpable fullness in her left upper quadrant but was otherwise nondistended, soft, and nontender. Pertinent preoperative laboratory evaluation revealed no abnormal findings.

CT imaging showed three benign appearing well-demarcated thin-walled simple cystic masses in the left upper abdomen all having a mass effect on the stomach. The cysts measured 9.2×6.6 cm, 1.8×1.7 cm, and 3.0×2.8 cm and were located along the posterior aspect of the upper stomach, anterolateral upper abdomen, and greater curvature of the stomach, respectively (Figures 1 and 2). There had been a slight interval growth from a CT one year earlier.

Patient proceeded to the operating room where she underwent wedge resection via an upper midline incision of all three cysts with no complications. Intraoperatively, the cysts did not communicate with the gastric lumen but arose from the gastric wall. They were all soft and filled with crystalline, particulate-laden fluid.

Pathology demonstrated benign developmental, thin-walled cysts with a smooth muscle wall. These were lined by respiratory ciliated and mucinous glandular epithelium resembling the epithelium of the stomach and respiratory system consistent with bronchogenic and combined bronchogenic/gastroenteric cysts (Figures 3–5).

Given the pathology, a CT chest was completed and showed left lower lobe partial bronchial agenesis. There were no pulmonary cystic lesions. The patient, however, remained asymptomatic with no signs of respiratory difficulty or hypoxia and no further workup was done.



FIGURE 1: Computed tomography of intra-abdominal cysts. Arrows point to multiple cysts arising from the stomach wall (axial).

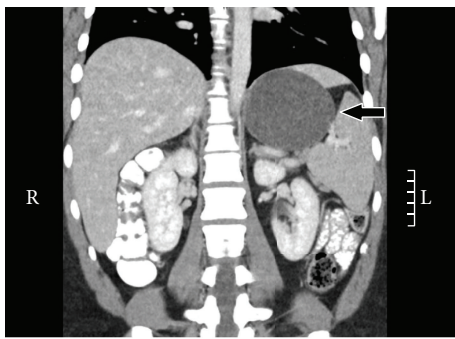


FIGURE 2: Computed tomography of intra-abdominal cysts. Arrow points to bronchogenic cyst arising from the stomach wall (coronal).

3. Discussion

There is a paucity of reported cases of intra-abdominal bronchogenic cysts. Our case outlines a unique case of three symptomatic bronchogenic or mixed bronchogenic/enteric gastric cysts. Gensler et al. reported the first case of an intramural gastric cyst in 1966 that was composed of ciliated pseudostratified columnar epithelium with focal squamous metaplasia. Since then, review of the literature reveals less than thirty case reports of single bronchogenic cysts located in the gastric mucosa [1–3].

Bronchogenic cysts typically arise from the foregut during embryological development in the 3rd to 7th week of life [4–10]. Esophageal epithelium undergoes a transient stage of cilia formation during the tenth week of gestation [5, 11] before differentiating into the usual squamous epithelium. This could potentially explain the pathophysiological mechanism for the presence of respiratory epithelium in the proximal gastrointestinal tract [5]. Congenital bronchogenic anomalies are more commonly found in the mediastinum, typically esophagus, or retroperitoneal space [12–16]. Bronchogenic cysts have also been reported to have been found on the skin [17] and diaphragm [18–23] and within the pericardium [24].

Patients have been reported to present with symptoms ranging from reflux to abdominal pain with some having no symptoms at all [6, 25]. Treatment has ranged from observation to aspiration to resection [4, 21, 26]. Patients have

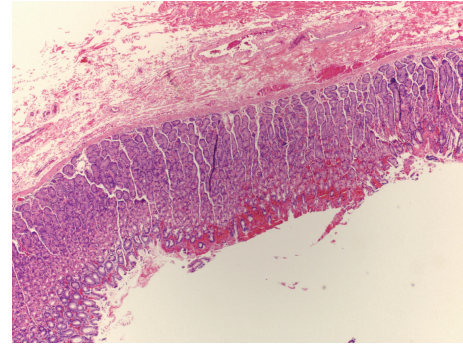


FIGURE 3: Histological H&E stain. Cystic lesion from greater curvature of stomach showing benign developmental cyst with smooth muscle wall and lined by respiratory ciliated epithelium. 40x magnification.

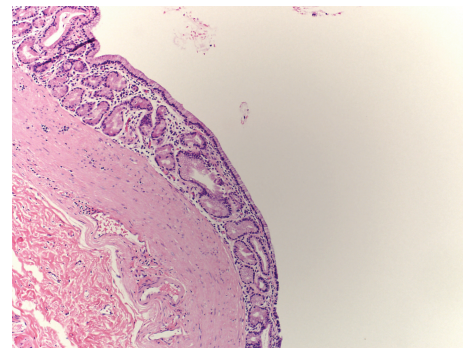


FIGURE 4: Histological H&E stain. Cystic lesion near the greater curvature of the stomach demonstrating benign developmental cyst lined by mucinous glandular epithelium resembling the epithelium of the stomach and respiratory epithelium. 100x magnification.

reported recurrence of cysts after aspiration [17]. Regardless, the majority of patients appeared to have undergone resection. The reported patient experienced abdominal pain during pregnancy with resolution after delivery, possibly due to mass effect. A hormonal component could not be excluded.

There have been a few published case reports of bronchogenic cysts involved with adenocarcinoma [26], bronchioloalveolar carcinoma, neuroblastoma [27], and rhabdomyosarcoma; however, there is minimal data in the literature to suggest oncologic potential for bronchogenic cysts [28, 29]. Most bronchogenic cysts are found incidentally and resected at the time. Vazquez et al. describe a case of a bronchogenic cyst that was found at the same time as a neuroblastoma in a pediatric patient. These were resected at separate procedures. They discuss the genetic basis for this association with speculations on oncogene mutations [27]. Sullivan et al. reported a case of adenocarcinoma arising from a retroperitoneal bronchogenic cyst. In that case, ciliated columnar epithelium was not present. Furthermore, some studies have suggested that loss of epithelial lining is associated with malignancy [26]. Our case study did have ciliated respiratory epithelium with no evidence of malignancy. However, the association between malignancy and bronchogenic cysts remains unclear.

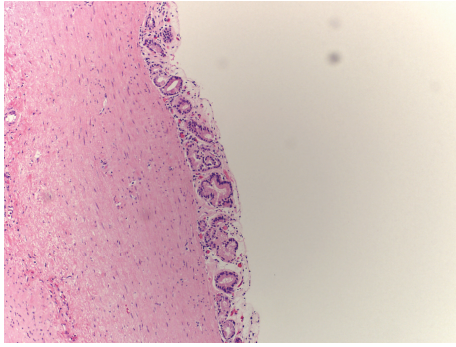


FIGURE 5: Histological H&E stain. Cystic lesion from the gastric cardia showing benign developmental cyst lined by mucinous columnar and respiratory epithelium with smooth muscle in the cyst wall. 100x magnification.

This case highlights a rare finding of multiple bronchogenic cysts arising from the gastric wall. Clearly, more investigation needs to be done to further understand the pathophysiology of these congenital bronchogenic cysts. Symptomatic or incidentally discovered cystic lesions in the foregut are generally felt to be benign. Symptomatic lesions probably warrant resection, especially if there is any diagnostic insecurity. There are occasional reports of bleeding, ulceration, or obstruction [5, 24, 30–34], and, depending on the clinical situation, resection rather than continued observation may be appropriate.

Conflict of Interests

The authors declare that there is no conflict of interests regarding the publication of this paper.

References

- [1] H. Ubukata, T. Satani, G. Motohashi et al., “Intra-abdominal bronchogenic cyst with gastric attachment: report of a case,” *Surgery Today*, vol. 41, no. 8, pp. 1095–1100, 2011.
- [2] C. A. Rubio, A. Orrego, and R. Willén, “Bronchogenic gastric cyst. A case report,” *In Vivo*, vol. 19, no. 2, pp. 383–385, 2005.
- [3] S. Gensler, B. Seidenberg, H. Rifkin, and B. M. Rubinstein, “Ciliated lined intramural cyst of the stomach: case report and suggested embryogenesis,” *Annals of Surgery*, vol. 163, no. 6, pp. 954–956, 1966.
- [4] L. Jiang, L. Jiang, N. Cheng, and L. Yan, “Bronchogenic cyst of the gastric fundus in a young woman,” *Digestive and Liver Disease*, vol. 42, no. 11, p. 826, 2010.
- [5] M. K. Liang and J. L. Marks, “Congenital bronchogenic cyst in the gastric mucosa,” *Journal of Clinical Pathology*, vol. 58, article 1344, 2005.
- [6] J. Matsubayashi, T. Ishida, T. Ozawa, T. Aoki, Y. Koyanagi, and K. Mukai, “Subphrenic bronchopulmonary foregut malformation with pulmonary-sequestration-like features,” *Pathology International*, vol. 53, no. 5, pp. 313–316, 2003.
- [7] E. Vlodaysky, B. Czernobilsky, Y. Bar, and B. Lifschitz-Mercer, “Gastric mucosa in a bronchogenic cutaneous cyst in a child: case report and review of literature,” *The American Journal of Dermatopathology*, vol. 27, no. 2, pp. 145–147, 2005.
- [8] X. Yang and K. Guo, “Bronchogenic cyst of stomach: two cases report and review of the English literature,” *Wiener Klinische Wochenschrift*, vol. 125, no. 9–10, pp. 283–287, 2013.
- [9] H. Shibahara, T. Arai, S. Yokoi, and S. Hayakawa, “Bronchogenic cyst of the stomach involved with gastric adenocarcinoma,” *Clinical Journal of Gastroenterology*, vol. 2, no. 2, pp. 80–84, 2009.
- [10] C. Endo, T. Imai, H. Nakagawa, A. Ebina, and M. Kaimori, “Bronchioloalveolar carcinoma arising in a bronchogenic cyst,” *Annals of Thoracic Surgery*, vol. 69, no. 3, pp. 933–935, 2000.
- [11] H. F. Krous and C. L. Sexauer, “Embryonal rhabdomyosarcoma arising within a congenital bronchogenic cyst in a child,” *Journal of Pediatric Surgery*, vol. 16, no. 4, pp. 506–508, 1981.
- [12] J. J. Murphy, G. K. Blair, G. C. Fraser et al., “Rhabdomyosarcoma arising within congenital pulmonary cysts: report of three cases,” *Journal of Pediatric Surgery*, vol. 27, no. 10, pp. 1364–1367, 1992.
- [13] S. M. Sullivan, S. Okada, M. Kudo, and Y. Ebihara, “A retroperitoneal bronchogenic cyst with malignant change,” *Pathology International*, vol. 49, no. 4, pp. 338–341, 1999.
- [14] R. Castro, M. I. Oliveira, T. Fernandes, and A. J. Madureira, “Retroperitoneal bronchogenic cyst: MRI findings,” *Case Reports in Radiology*, vol. 2013, Article ID 853795, 3 pages, 2013.
- [15] G. F. Orellana, R. Cárdenas, M. E. Manríquez, H. Ríos, L. Suárez, and D. Videla, “Retroperitoneal bronchogenic cyst: report of one case,” *Revista Médica de Chile*, vol. 135, no. 7, pp. 924–931, 2007.
- [16] K. H. Kim, J. I. Kim, C. H. Ahn et al., “The first case of intraperitoneal bronchogenic cyst in Korea mimicking a gallbladder tumor,” *Journal of Korean Medical Science*, vol. 19, no. 3, pp. 470–473, 2004.
- [17] S. Msika, R. Kianmanesh, P. Jouet et al., “Bronchogenic cyst of the right hemidiaphragm mimicking a hydatid cyst of the liver,” *Gastroenterologie Clinique et Biologique*, vol. 24, no. 12, pp. 1224–1226, 2000.
- [18] H. Cerwenka, M. Uggowitz, H. Bacher, G. Werkgartner, A. El-Shabrawi, and H. J. Mischinger, “Bronchogenic cyst appearing as a hepatic mass,” *Abdominal Imaging*, vol. 25, no. 1, pp. 86–88, 2000.
- [19] J. Mouroux, A. Bourgeon, D. Benchimal et al., “Bronchogenic cysts of the esophagus. Classical surgery or video-surgery?” *Chirurgie Paris*, vol. 117, no. 7, pp. 564–568, 1991.
- [20] Y. Katayama, H. Kusagawa, T. Komada, S. Shomura, and H. Tenpaku, “Bronchopulmonary foregut malformation,” *General Thoracic and Cardiovascular Surgery*, vol. 59, no. 11, pp. 767–770, 2011.
- [21] K. Inaba, Y. Sakurai, Y. Umeki, S. Kanaya, Y. Komori, and I. Uyama, “Laparoscopic excision of subdiaphragmatic bronchogenic cyst occurring in the retroperitoneum: report of a case,” *Surgical Laparoscopy, Endoscopy and Percutaneous Techniques*, vol. 20, no. 6, pp. e199–e203, 2010.
- [22] M. Sato, A. Irisawa, M. S. Bhutani et al., “Gastric bronchogenic cyst diagnosed by endosonographically guided fine needle aspiration biopsy,” *Journal of Clinical Ultrasound*, vol. 36, no. 4, pp. 237–239, 2008.
- [23] D. A. Hall, R. T. Pu, and Y. Pang, “Diagnosis of foregut and tailgut cysts by endosonographically guided fine-needle aspiration,” *Diagnostic Cytopathology*, vol. 35, no. 1, pp. 43–46, 2007.
- [24] N. Melo, M. B. Pitman, and D. W. Rattner, “Bronchogenic cyst of the gastric fundus presenting as a gastrointestinal stromal

- tumor,” *Journal of Laparoendoscopic & Advanced Surgical Techniques, Part A*, vol. 15, no. 2, pp. 163–165, 2005.
- [25] S. Y. Song, J. H. Non, S. J. Lee, and H. J. Son, “Bronchogenic cyst of the stomach masquerading as benign stromal tumor,” *Pathology International*, vol. 55, no. 2, pp. 87–91, 2005.
- [26] B. N. Vazquez, J. Mira, C. Navarro et al., “Neuroblastoma and bronchogenic cyst: a rare association,” *European Journal of Pediatric Surgery*, vol. 10, no. 5, pp. 340–342, 2000.
- [27] K. K. Nobuhara, Y. C. Gorski, M. P. La Quaglia, and R. C. Shamberger, “Bronchogenic cysts and esophageal duplications: common origins and treatment,” *Journal of Pediatric Surgery*, vol. 32, no. 10, pp. 1408–1413, 1997.
- [28] M. J. Haddon and A. Bowen, “Bronchopulmonary and neurenteric forms of foregut anomalies. Imaging for diagnosis and management,” *Radiologic Clinics of North America*, vol. 29, no. 2, pp. 241–254, 1991.
- [29] K. Ohno, T. Miyamoto, H. Murata, K. Kaku, S. Maeda, and K. Yamashita, “Intrapericardial bronchogenic cyst—a report of two surgical cases,” *Nihon Kyobu Geka Gakkai Zasshi*, vol. 38, no. 4, pp. 660–666, 1990.
- [30] B. Braffman, R. Keller, E. S. Gendal, and S. I. Finkel, “Subdiaphragmatic bronchogenic cyst with gastric communication,” *Gastrointestinal Radiology*, vol. 13, no. 4, pp. 309–311, 1988.
- [31] M. E. Keohane, I. Schwartz, J. Freed, and R. Dische, “Subdiaphragmatic bronchogenic cyst with communication to the stomach: a case report,” *Human Pathology*, vol. 19, no. 7, pp. 868–871, 1988.
- [32] E. Anagnostou, V. Soubasi, E. Agakidou, C. Papakonstantinou, N. Antonitsis, and M. Leontsini, “Mediastinal gastroenteric cyst in a neonate containing respiratory-type epithelium and pancreatic tissue,” *Pediatric Pulmonology*, vol. 44, no. 12, pp. 1240–1243, 2009.
- [33] D. C. Salyer, W. R. Salyer, and J. C. Eggleston, “Benign developmental cysts of the mediastinum,” *Archives of Pathology and Laboratory Medicine*, vol. 101, no. 3, pp. 136–139, 1977.
- [34] H. Linder, “Intrathoracic gastroenteric cysts,” *Surgery*, vol. 25, no. 6, pp. 862–868, 1949.