



Case Report:

Gastric duplication cyst lined by pseudostratified columnar ciliated epithelium: a case report and literature review

Wu JIANG¹, Bo ZHANG², Yan-biao FU³, Jia-wei WANG⁴,
 Shun-liang GAO^{†‡2}, Su-zhan ZHANG¹, Yu-lian WU²

¹Department of Surgical Oncology, the Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou 310009, China)

²Department of General Surgery, the Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou 310009, China)

³Department of Pathology, the Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou 310009, China)

⁴Department of Radiology, the Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou 310009, China)

[†]E-mail: gaoshunliangzju@163.com

Received Apr. 8, 2010; Revision accepted Oct. 8, 2010; Crosschecked Dec. 17, 2010

Abstract: Gastric duplication cyst (GDC) lined by pseudostratified columnar ciliated epithelium (PCCE) is an uncommon lesion stemming from a foregut developmental malformation. Its clinical and radiological presentation is usually nonspecific. In this study, we reported a 76-year-old man who presented with an incidentally found perigastric mass. An exploratory laparotomy revealed a non-communicating cyst below the gastroesophageal junction, measuring 4 cm×4 cm in size. Microscopically, the gastric cyst was lined merely by PCCE. Although rare, GDC lined by PCCE should be included in the differential diagnosis of gastric wall masses. Surgical intervention is warranted in patients who have clinical symptoms, or who are aged more than 50 years.

Key words: Gastric duplication cyst (GDC), Foregut, Pseudostratified columnar ciliated epithelium (PCCE), Endoscopic ultrasound-guided fine needle aspiration (EUS-FNA)

doi:10.1631/jzus.B1000130

Document code: A

CLC number: R573

1 Introduction

Gastric duplication cyst (GDC) is an uncommon congenital malformation and is usually lined by gastrointestinal mucosa. GDC lined by pseudostratified columnar ciliated epithelium (PCCE) is extremely rare, and only 17 cases have been reported up to Mar. 2010 (Table 1). Due to the nonspecific clinical and radiological presentation, GDC lined by PCCE is easily misdiagnosed as a gastrointestinal stromal tumor (GIST). In this paper, we describe a unique case of GDC lined merely by PCCE, and the available English literature is reviewed.

2 Case report

A 76-year-old man was referred to our hospital for evaluation of a perigastric mass found incidentally on abdominal ultrasonography. He had been in a good state of health. Physical examination findings and laboratory studies were within normal ranges. An abdominal computed tomography (CT) demonstrated a homogeneous, low-density mass, measuring 4 cm×4 cm, on the lesser curvature of stomach without contrast enhancement (Fig. 1). Gastroscopy revealed a submucosal type elevation with overlying normal mucosa below the gastroesophageal junction. Endoscopic ultrasound (EUS) showed this to be a hypoechoic lesion, arising within the tunica muscularis (Fig. 2). Preoperative diagnosis was a GIST and surgical excision was performed. The patient had an uneventful recovery and remained healthy to date.

[†] Corresponding author

Table 1 Summary of gastric duplication cyst lined by pseudostratified columnar ciliated epithelium

References	Age (year)	Gender	Complaint	Location	Size (cm)
Mardi <i>et al.</i> , 2010	42	M	Left lumbar pain	AGIJ, LC	4.5×5.2
Jiang <i>et al.</i> , 2010	25	F	Epigastric pain	Gastric fundus	3.0×2.5×2.0
Shibahara <i>et al.</i> , 2009	43	M	Epigastric pain	Cardia, LC	NA
Sato <i>et al.</i> , 2008	60	F	No	Cardia, LC	3
Murakami <i>et al.</i> , 2008	72	F	No	Middle body, LC	2.0×1.5
Wakabayashi <i>et al.</i> , 2007	37	M	Epigastric pain	NGEJ, LC	4×4
Hall <i>et al.</i> , 2007	40	M	Epigastric discomfort	NGEJ, LC	6×5
Theodosopoulos <i>et al.</i> , 2007	46	F	Vomiting	(1) PW of fundus; (2) Gastrosplenic ligament	8×5.5 3
Lee <i>et al.</i> , 2006	38	F	No	Cardia, LC	7×5
Cunningham <i>et al.</i> , 2006	63	F	Fever, abdominal pain	PW of fundus	10×7.6
Melo <i>et al.</i> , 2005	39	F	No	Fundus	4×2.5×1
Rubio <i>et al.</i> , 2005	26	M	Epigastric pain	NA	NA
Song <i>et al.</i> , 2005	62	F	No	NGEJ, LC	3.5×2.5×1.5
Hedayati <i>et al.</i> , 2003	59	F	No	PW of stomach, LC	7×5
Kim <i>et al.</i> , 2000	35	M	Epigastric pain	NGEJ, LC	7×6×5
Takahara <i>et al.</i> , 1996	25	M	No	PW of fundus	6.5×5×5
Gensler <i>et al.</i> , 1966	46	F	No	NGEJ, GC	6×8
Present	76	M	No	NGEJ, LC	4×4

M: male; F: female; AGIJ: anterior of gastrointestinal junction; NGEJ: near gastroesophageal junction; LC: lesser curvature; GC: greater curvature; PW: posterior wall; NA: not available



Fig. 1 Abdominal contrast-enhanced computed tomography (CT) scan demonstrating a homogeneous, low-density mass on the lesser curvature of the stomach (arrow)

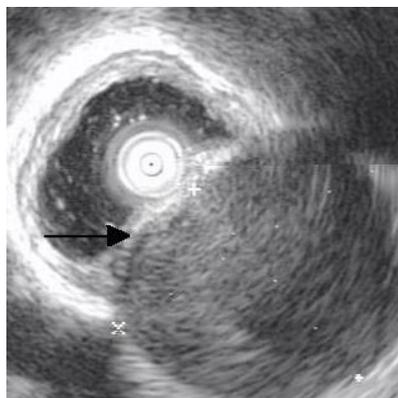


Fig. 2 Endoscopic ultrasound (EUS) showing a hypoechoic lesion, arising within the tunica muscularis (arrow)

Pathologic examination reported that the cyst was embedded within the gastric muscular layer, and did not communicate with the gastric lumen. Microscopically, the cyst wall was lined merely by PCCE, with two or three complete layers of smooth muscle bundles. However, cartilaginous tissue, seromucous gland, or gastric epithelium was not identified (Fig. 3).

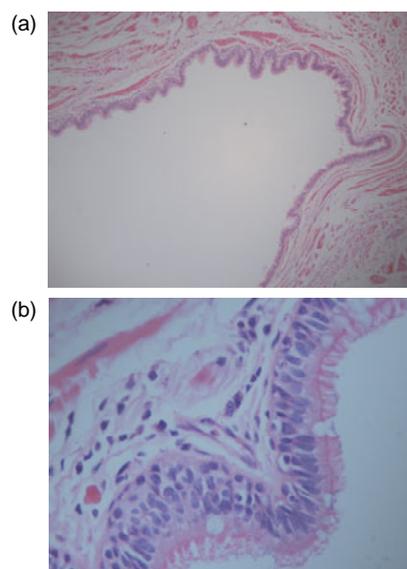


Fig. 3 Microscopic sections of the gastric duplication cyst Cystic structures are lined merely by pseudostratified columnar ciliated epithelium (PCCE), with two or three complete layers of smooth muscle bundles (hematoxylin and eosin staining; original magnification: (a) 100×; (b) 400×)

3 Discussion

Gastric duplication cyst is an uncommon lesion that accounts for 2%–8% of all alimentary duplications (Kim *et al.*, 2000). It is suggested that GDC lined by PCCE is derived from the caudad-most portion of the laryngotracheal outgrowth, which remains attached to the portion of the primitive foregut destined to become the stomach (Gensler *et al.*, 1966), and migrates into the abdominal cavity before the fusion of the pleuroperitoneal membrane in the 7th week of gestation (Song *et al.*, 2005). When PCCE is seen in a cyst, we designate those containing cartilage or seromucous respiratory glands as bronchogenic cysts, those containing two well-developed layers of smooth muscle without cartilage as esophageal cysts, and those without these distinguishing features as foregut cysts (Murakami *et al.*, 2008). Moreover, it is advocated that the nomenclature for these duplicated cysts should involve the location as the prefix and the differentiation of the cyst suggested by the epithelium as the suffix (Sharma *et al.*, 2009). Our present case includes none of those distinguishing features mentioned above. Therefore, we describe it as gastric duplication cyst with foregut differentiation.

GDC lined by PCCE is a late onset disease (from 25 to 76 years in age) with little difference between genders (10 women and 8 men). The most common location is on the lesser curvature, near the gastroesophageal junction. It is usually asymptomatic and is discovered incidentally as a gastric wall mass. However, some patients complain of epigastric pain which may be caused by compression on adjacent structures or ulceration due to the ectopic gastric mucosa in the cysts (Kim *et al.*, 2000). Malignant change of GDC lined by PCCE is rare. Shibahara *et al.* (2009) reported a case of bronchogenic cyst of the stomach located at the opposite side wall to the gastric adenocarcinoma, and attributed the adenocarcinoma to the chronic inflammation from the bronchogenic cyst to the gastric mucosa. Worth mentioning, because of persistent exposure to carcinogenic substances, alimentary duplication cyst should be recognized as a lesion at risk for malignant transformation in patients aged more than 50 years (Fukumoto *et al.*, 2008).

CT can detect the presence of abdominal mass, but it frequently fails to identify the cystic nature of the GDC lined by PCCE due to the thick, proteina-

ceous cyst fluid (Lee *et al.*, 2006). EUS is helpful in identifying the intramural or extramural relation of the gastrointestinal tract (Eloubeidi *et al.*, 2004). Moreover, EUS-guided fine needle aspiration (EUS-FNA) provides cytological diagnosis of the GDC. It is believed that the presence of detached ciliary tufts and absence of neoplastic cells confirm the benign nature of the cyst (Eloubeidi *et al.*, 2004). In addition, $^{99}\text{Tc}^{\text{m}}$ -pertechnetate scintigraphy can diagnose ectopic functioning gastric mucosa, which is often associated with complications like ulceration in the GDC (Kumar *et al.*, 2005). However, in our case, we misdiagnosed the mass as a GIST, which is much more common than a congenital cyst, and it is not advised to perform biopsy to confirm the diagnosis of a resectable GIST as it can lead to tumor dissemination or hemorrhage (Chaudhry and DeMatteo, 2009). Therefore, we failed to make an accurate diagnosis before operation.

The recommended management of symptomatic GDC is complete excision of the cyst without violation of the gastric lumen. If this is not possible, segmental gastrectomy is an alternative (Holcomb *et al.*, 1989). When malignant change is seen in the cyst, a surgical procedure without rupture of the cyst is required (Shibahara *et al.*, 2009). Management of asymptomatic cases remains controversial. Eloubeidi *et al.* (2004) suggested watchful waiting after confirming the benign nature of these cysts by EUS-FNA. However, considering the potential for complications and malignant change, we believe, for asymptomatic cases, surgical intervention is warranted in patients aged more than 50 years, especially when ectopic functioning gastric mucosa was detected.

4 Conclusions

In summary, GDC lined by PCCE is an uncommon lesion stemming from foregut developmental malformation. Its clinical presentation is usually nonspecific and it is easily misdiagnosed as a GIST radiologically. EUS-FNA can provide cytological diagnosis and confirm the benign nature of the cyst. Although rare, GDC lined by PCCE should be included in the differential diagnosis of gastric wall mass. Surgical intervention is warranted in patients with clinical symptoms, or who are aged more than 50 years.

References

- Chaudhry, U.I., DeMatteo, R.P., 2009. Management of resectable gastrointestinal stromal tumor. *Hematol. Oncol. Clin. North Am.*, **23**(1):79-96. [doi:10.1016/j.hoc.2009.01.001]
- Cunningham, S., Hansel, D., Fishman, E., Cameron, J., 2006. Foregut duplication cyst of the stomach. *J. Gastrointest. Surg.*, **10**(4):620-621. [doi:10.1016/j.gassur.2005.04.004]
- Eloubeidi, M.A., Cohn, M., Cerfolio, R.J., Chhieng, D.C., Jhala, N., Jhala, D., Eltoun, I.A., 2004. Endoscopic ultrasound-guided fine-needle aspiration in the diagnosis of foregut duplication cysts: the value of demonstrating detached ciliary tufts in cyst fluid. *Cancer Cytopathol.*, **102**(4):253-258. [doi:10.1002/cncr.20369]
- Fukumoto, K., Suzuki, S., Sakaguchi, T., Morita, Y., Oishi, K., Suzuki, A., Inaba, K., Kamiya, K., Miura, K., Konno, H., 2008. Adenocarcinoma arising from gastric duplication: a case report with literature review. *Clin. J. Gastroenterol.*, **1**(4):148-152. [doi:10.1007/s12328-008-0024-1]
- Gensler, S., Seidenberg, B., Rifkin, H., Rubinstein, B., 1966. Ciliated lined intramural cyst of the stomach: case report and suggested embryogenesis. *Ann. Surg.*, **163**(6):954-956. [doi:10.1097/0000658-196606000-00018]
- Hall, D.A., Pu, R.T., Pang, Y., 2007. Diagnosis of foregut and tailgut cysts by endosonographically guided fine-needle aspiration. *Diagn. Cytopathol.*, **35**(1):43-46. [doi:10.1002/dc.20573]
- Hedayati, N., Cai, D., McHenry, C., 2003. Subdiaphragmatic bronchogenic cyst masquerading as an "adrenal incidentaloma". *J. Gastrointest. Surg.*, **7**(6):802-804. [doi:10.1016/S1091-255X(03)00134-3]
- Holcomb, G., Gheissari, A., O'Neill, J., Shorter, N., Bishop, H., 1989. Surgical management of alimentary tract duplications. *Ann. Surg.*, **209**(2):167-174. [doi:10.1097/0000658-198902000-00006]
- Jiang, L., Jiang, L.S., Cheng, N.S., Yan, L.N., 2010. Bronchogenic cyst of the gastric fundus in a young woman. *Dig. Liver Dis.*, **42**(11):826. [doi:10.1016/j.dld.2009.06.014]
- Kim, D.H., Kim, J.S., Nam, E.S., Shin, H.S., 2000. Foregut duplication cyst of the stomach. *Pathol. Int.*, **50**(2):142-145. [doi:10.1046/j.1440-1827.2000.01008.x]
- Kumar, R., Tripathi, M., Chandrashekar, N., Agarwala, S., Kumar, A., Dasan, J.B., Malhotra, A., 2005. Diagnosis of ectopic gastric mucosa using ⁹⁹Tc^m-pertechnetate: spectrum of scintigraphic findings. *Br. J. Radiol.*, **78**(932):714-720. [doi:10.1259/bjr/16678420]
- Lee, S., Park, D., Park, J., Kim, H., Park, S., Kim, S., Oh, M., 2006. Endoscopic mucosal resection of a gastric bronchogenic cyst that was mimicking a solid tumor. *Endoscopy*, **38**(Suppl. 2):E12-E13. [doi:10.1055/s-2006-944862]
- Mardi, K., Kaushal, V., Gupta, S., 2010. Foregut duplication cysts of stomach masquerading as leiomyoma. *Indian J. Pathol. Microbiol.*, **53**(1):160-161. [doi:10.4103/0377-4929.59214]
- Melo, N., Pitman, M.B., Rattner, D.W., 2005. Bronchogenic cyst of the gastric fundus presenting as a gastrointestinal stromal tumor. *J. Laparoendosc. Adv. Surg. Tech.*, **15**(2):163-165. [doi:10.1089/lap.2005.15.163]
- Murakami, S., Isozaki, H., Shou, T., Sakai, K., Toyota, H., 2008. Foregut duplication cyst of the stomach with pseudostratified columnar ciliated epithelium. *Pathol. Int.*, **58**(3):187-190. [doi:10.1111/j.1440-1827.2007.02209.x]
- Rubio, C.A., Orrego, A., Willén, R., 2005. Congenital bronchogenic cyst in the gastric mucosa. *J. Clin. Pathol.*, **58**(3):335.
- Sato, M., Irisawa, A., Bhutani, M.S., Schnadig, V., Takagi, T., Shibukawa, G., Wakatsuki, T., Imamura, H., Takahashi, Y., Sato, A., et al., 2008. Gastric bronchogenic cyst diagnosed by endosonographically guided fine needle aspiration biopsy. *J. Clin. Ultrasound*, **36**(4):237-239. [doi:10.1002/jcu.20425]
- Sharma, S., Nezakatgoo, N., Sreenivasan, P., Vanatta, J., Jabbour, N., 2009. Foregut cystic developmental malformation: new taxonomy and classification—unifying embryopathological concepts. *Indian J. Pathol. Microbiol.*, **52**(4):461-472. [doi:10.4103/0377-4929.56119]
- Shibahara, H., Arai, T., Yokoi, S., Hayakawa, S., 2009. Bronchogenic cyst of the stomach involved with gastric adenocarcinoma. *Clin. J. Gastroenterol.*, **2**(2):80-84. [doi:10.1007/s12328-008-0042-z]
- Song, S.Y., Noh, J.H., Lee, S.J., Son, H.J., 2005. Bronchogenic cyst of the stomach masquerading as benign stromal tumor. *Pathol. Int.*, **55**(2):87-91. [doi:10.1111/j.1440-1827.2005.01788.x]
- Takahara, T., Torigoe, T., Haga, H., Yoshida, H., Takeshima, S., Sano, S., Ishii, Y., Furuya, T., Nakamura, E., Ishikawa, M., 1996. Gastric duplication cyst: evaluation by endoscopic ultrasonography and magnetic resonance imaging. *J. Gastroenterol.*, **31**(3):420-424. [doi:10.1007/BF02355033]
- Theodosopoulos, T., Marinis, A., Karapanos, K., Vassilikostas, G., Dafnios, N., Samanides, L., Carvounis, E., 2007. Foregut duplication cysts of the stomach with respiratory epithelium. *World J. Gastroenterol.*, **13**(8):1279-1281.
- Wakabayashi, H., Okano, K., Yamamoto, N., Suzuki, Y., Inoue, H., Kadota, K., Haba, R., 2007. Laparoscopically resected foregut duplication cyst (bronchogenic) of the stomach. *Dig. Dis. Sci.*, **52**(8):1767-1770. [doi:10.1007/s10620-006-9580-8]