Adenocarcinoma in an Adult Gastric Duplication Cyst: A Case Report

[1]Maria Fleurdeliz R. Goco, M.D., [2]Jeffrey Cusapin, M.D., [3]Melchor M. Chan, M.D,
[4]Frederick T. Dy, M.D., [5]Rolando Lopez, M.D.
[1]Fellow, Gastroenterology, University of Santo Tomas Hospital
[2]Resident, Pathology, University of Santo Tomas Hospital
[3,4]Consultant, Gastroenterology, Univeristy of Santo Tomas Hospital
[5]Consultant, Pathology, University of Santo Tomas Hospital

ABSTRACT

Significance: Gastric duplication cysts (GDC) are relatively rare and mostly diagnosed in children, but GDCs that underwent malignant transformation are extremely rare, with only eleven cases reported in the literature to date.

Clinical presentation: We are presented with a case of a 52 year old female with dull abdominal pain and palpable mass at the epigastric region.

Management: Ultrasound of the abdomen, contrast enhanced CT imaging of the abdomen showed an extrapancreatic, extragastric cystic mass. Endoscopic ultrasound showed an anechoic cyst with a hyperechoic lesion within the cyst at the submucosal layer continuous with the muscularis propria of the stomach. Subtotal gastrectomy was done, with histopathologic confirmation of gastric duplication cyst with adenocarcinoma.

Recommendation: Gastric duplication cyst should be considered as a differential diagnosis of cystic lesions in the stomach. A high index of suspicion is warranted, and histopathologic confirmation is still necessary since malignant transformation has been reported.

Keywords: gastric duplication cyst, carcinoma

Introduction

Duplication cysts are congenital anomalies that may occur at any level of the gastrointestinal tract. Common sites of involvement are the small intestine (45%) and esophagus (19%) but occur most commonly at the ileocecal valve area. Gastric duplication cysts (GDC) are relatively rare, making up only 2% to 9% of gastrointestinal duplications[1,2]. It usually presents as a non-communicating saccular or elongated cystic mass attached to the greater curvature, as gastric duplications usually originate dorsal to the primitive intestine. Most GDCs are diagnosed during the first year of life[1-3], although it could also be detected in health screening of asymptomatic adults.[4] The following criteria are used in the diagnosis of gastric duplication cysts: (1) the wall of the cyst is contiguous with the stomach wall; (2) the cyst is surrounded by at least one coat of smooth muscle, which is continuous with the muscularis propria of the stomach; and (3) the cyst wall is lined by typical gastrointestinal mucosa.[5]

Case Report

This is a case of a 52-year old female who consulted due to dull abdominal pain with no other accompanying symptoms. On physical examination, a palpable, nontender, fixed, fluctuant mass was appreciated at the epigastric area. Ultrasound showed a well-defined septated cystic focus measuring 9.0 x 11.0 x 5.6 cm, adjacent to the pancreatic tail with a solid component at the inferior portion and exhibited vascularity on color doppler study. Layering medium level echoes were also appreciated at its inferior portion. A pancreatic pseudocyst versus a mesenteric cyst was considered.

Plain and dual-phase contrast-enhanced CT scan of the whole abdomen showed a lobulated, predominantly cystic mass with septations and solid nodular components in the right gastrocolic space, intimately related with and indenting the inferior wall of the gastric antrum, measuring 8 x 11.5 x 7.3 cm. This is associated with perilesional fat stranding and small lymphadenopathies.

The rest of the stomach, small and large intestinal loops show no focal abnormalities. The liver is normal-sized with intact parenchymal attenuation and smooth hepatic surface. There were thin-walled non-enhancing fluid-equivalent foci in the hepatic segments, the largest measuring 2.1 x 1.4 cm, which may represent hepatic cysts. The spleen, gallbladder, pancreas, and adrenal glands were normal. There was no ascites. The paraaortic, peripancreatic

Endoscopic ultrasound was done which showed an anechoic lesion at the submucosal layer with regular margins and minimal septations. A hyperechoic lesion was noted within the cyst at the inferior portion with noted irregular margins. The cyst was adjacent to the gastric body, contiguous with the muscularis propria of the gastric wall. Endoscpocially, a bulging mass was noted at the greater curvature with smooth and intact mucosa.

She then underwent exploratory laparotomy, subtotal gastrectomy and gastrojejunostomy. Intraoperatively, a 9 x 11 x 7 cm cystic, lobulated mesenteric mass with solid component was noted which was adherent to the gastric body with no cleavage plane for dissection. One 1 cm perigastric lymph node was seen. The liver was grossly normal.

Macroscopically, a bulging fluctuant mass measuring 8.5 x 7.5 x 7.0 was seen along the greater curvature. This has a smooth intact outer surface with minor irregularities. The inner surface is cream white to light brown and generally smooth (Figure 5). A fungating mass measuring 3.5 x 3.0 x 1.5 cm is seen at the inner wall of the larger locule. Sections of which show solid, cream white to yellow, firm with granular cut surfaces. It occupies 20% of the cystic mass (Figure 5). The gastric mucosa is generally smooth and intact, with portion of the mass covered with mucosa slightly bulging into the gastric lumen. Microsections of the cystic submucosal mass disclose a malignant neoplasm composed of tightly packed back to back glands with trabecular and nesting pattern (Figure 4). These are lined by low cuboidal to columnar epithelial cells (Figure exhibiting hyperchromatic to vesicular nuclei and prominent macronucleoli and granular to cleared out

cytoplasm. Tumor emboli are identified. The surgical margins are negative for tumor involvement. Two perigastric lymph nodes recovered are both negative for tumor metastasis. Sections of the uninvolved stomach wall show moderate lymphoplasmacytic and mild neutrophilic infiltrates at the lamina propria. Immunohistochemical staining showed positive for CK7 and negative for CK20, TTF-1, and Calretinin The diagnosis of primary adenocarcinoma was established.

Figure 1. Endoscpocially a bulging mucosa was noted at the greater curvature with smooth intac mucosa.

Figure 2. An anechoic lesion at the submucosal layer with regular margins and minimal septations was noted. A hyperechoic lesion was noted at the inferior portion with noted irregular margins. The cyst wall was noted to be contiguous with the muscularis propria of the gastric wall.

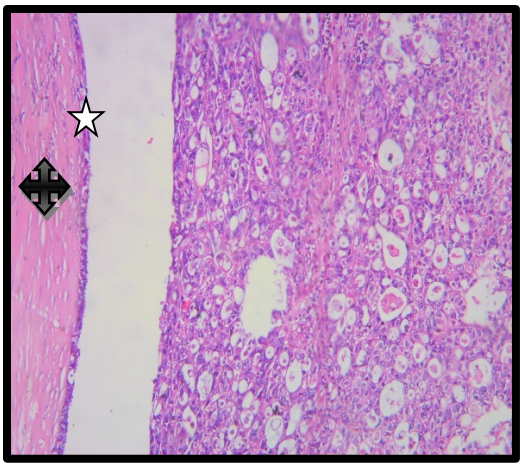

*Figure 3.*The mucosa (star) is lined by simple cuboidal to columnar epithelial cell. Surrounding the mucosa is a layer of smooth muscle that is continuous with the muscularis propria of the stomach (cross)

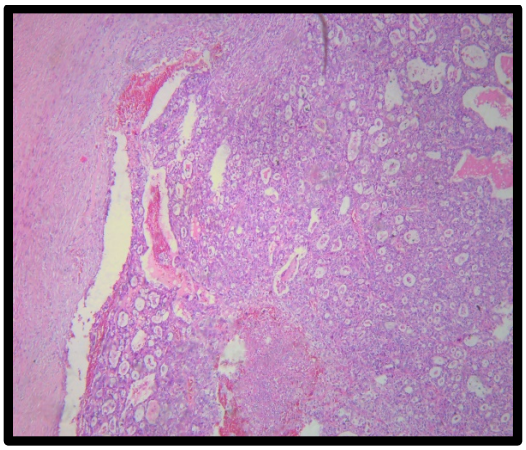

Figure 4. Malignant neoplasm composed of tightly packed back to back glands with trabecular and nesting pattern

Discussion

More than 50% of patients with GDCs may present with vomiting or as a palpable abdominal mass. Complications such as pain, obstruction, ulceration, and hemorrhage may also be present, causing associated symptoms. Only eleven cases of gastric duplications which underwent malignant transformation have been reported in the literature, making this case the twelfth. Adenocarcinoma is the most common histologic type seen in GDCs (nine cases), but squamous cell carcinoma (one case) and neuroendocrine carcinoma (one case) have also been documented.[1-4,6-11] Among the reported cases, six were noted to have invaded the adjacent

stomach wall, four were confined within the cyst wall, and one presented with peritoneal carcinomatosis after a failed attempt to do laparoscopic removal of a GDC.[1]

Differential diagnoses include cystic neoplasms or tumors with cystic degeneration, such as GIST, neuroendocrine tumor, or neurogenic tumor. One of the most important differential diagnosis is adenocarcinoma arising in gastritis cystica profunda (GCP). GCP is a benign neoplasm characterized by herniated or displaced normal gastric glands in the submucosa, which may sometimes show cystic dilatations. The major difference between a GDC and a GCP is that a GCP does not have a smooth muscle coat surrounding the lesion. CT scan or magnetic resonance imaging of the abdomen may aid in differentiating GDCs from other cystic lesions, but up to 70% of the cases can be misdiagnosed as solid lesions due to a high concentration of proteins in the cystic contents, hence some authors recommend histologic confirmation through endoscopic ultrasound-guided aspiration biopsy.[12]

REFERENCES

1) K Liu, X Lin, J Wu, M Meng, H Su, W Tai, H Chang. Peritoneal metastatic adenocarcinoma possibly due to a gastric duplication cyst: a case report and literature review Mar 2014; 14: 48
2) HJ Kang, SJ Jang, YS Park. Adenocarcinoma Arising in Gastric Duplication Cyst. The Korean Journal of Pathology 2014; 48: 159-161
3) K Kuraoka, H Nakayama, T Kagawa, T Ichikawa, W Yasui. Adenocarcinoma Arising from a Gastric Duplication Cyst with Invasion to the Stomach: A Case Report with Literature Review. Journal of Clinical Pathology Apr 2004; 57: 428-431
4) J Zheng, H Jing. Adenocarcinoma arising from a gastric duplication cyst. Surgical Oncology 2012; 21(2): e97-101
5) JT Rowling. Some observations on gastric cysts. The British Journal of Surgery Mar 1959; 46(199): 441-445
6) G Horne, C Ming-Lum, AW Kirkpatrick, RL Parker. High-grade neuroendocrine carcinoma arising in a gastric duplication cyst: a case report with literature review. International Journal of Surgical Pathology Apr 2007; 15(2): 187-191
7) ML Barussaud, G Meurette, E Cassagnau, B Dupasc, J Le Borgne. Mixed Adenocarcinoma and Squamous Cell Carcinoma arising in a gastric duplication cyst. Gastroenterologie Clinique et Biologique Feb 2008; 32(2): 188-191
8) J Singh, H Rajdeo, K Bhuta, J Savino. Gastric Duplication Cyst: Two Case Reports and Review of the Literature. Case Reports in Surgery 2013
9) DG Coit, C Mies. Adenocarcinoma arising within a gastric duplication cyst. Journal of Surgical Oncology Aug 1992; 50(4): 274-277

10) N Mamiya, Y Karasawa, N Kojima, T Takemoto, N Kondoh, S Kasai. A case of gastric duplication cyst containing papillary adenocarcinoma. The Japanese Journal of Gastroenterology Jan 1996; 93(1): 34-38
11) K Fukumoto, S Suzuki, T Sakaguchi, Y Morita, K Oishi, A Suzuki, K Inaba, K Kamiya, K Miura, H Konno. Adenocarcinoma arising from gastric duplication: a case report with literature review. Clinical Journal of Gastroenterology Dec 2008; 1(4): 148-152
12) S Seijo Rios, J Lariño Noia, I Abdulkader Nallib, A Lozano Leon, B Vieites Perez-Quintela, J Iglesias Garcia, JE Dominguez Muñoz. Adult gastric duplication cyst: diagnosis by endoscopic ultrasound-guided fine-needle aspiration (EUS-FNA). Revista Española de Enfermedades Digestivas: Organo Oficial de la Sociedad Española de Patologia Digestiva Sep 2008; 100(9): 586-90