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Cyst of the gastric wall arising from heterotopic pancreas: report of a case

Stefano Cecchini[1], Federico Marchesi[1], Pietro Caruana[2], Francesco Tartamella[1], Maria Teresa Mita[1], Francesco Rubichi[1], Luigi Roncoroni[1]

[1] Dipartimento di Scienze Chirurgiche, sezione di Clinica Chirurgica Generale e Terapia Chirurgica, Università degli studi di Parma, Parma, Italia; [2] Dipartimento di Patologia e Medicina di Laboratorio, Sezione di Anatomia Patologica, Università degli studi di Studi di Parma, Parma, Italia

Summary. Heterotopia of pancreatic tissue is a common developmental anomaly, affecting predominantly the gastrointestinal tract. The case of a symptomatic cyst arising from the posterior gastric wall in a 40-year-old man is presented, undergoing laparoscopic gastric wedge resection. Pathology report described a cyst of the gastric wall lined by ductal pancreatic epithelium. (www.actabiomedica.it)

Key words: pancreatic heterotopia, gastric cyst, laparoscopy, gastrectomy, wedge, resection

Introduction

Heterotopic pancreas (HP) is defined as the presence of pancreatic tissue outside the usual anatomical location of the pancreas, typically asymptomatic. HP occasionally presents symptomatically and the manifestations vary depending on the location of the lesion. HP also may present with symptoms related to complications similar to those normally associated with the diseases of pancreas. We describe the first reported case of gastric HP which presents as a cystic lesion mimicking a gastric duplication cyst.

Case report

The case of a symptomatic cyst arising from the posterior gastric wall in a 40-year-old man is presented. The patient presented to our Institution with a 6 months history of dyspepsia and recurrent left upper quadrant abdominal pain, uncorrelated to mealtimes and chest-abdominal movement. The physical examination was unremarkable. Laboratory blood testes were unremarkable. Gastroscopy showed a mild antral hyperemia without ulceration or erosion. CT scan revealed an exophytic cystic mass raising from the posterior wall of the stomach. The inferior pole of this lesion was sited dorsally to the body of the pancreas (Fig. 1). CT findings were compatible with the diagnosis of gastric diverticulum or gastric duplication cyst. Contrast swallow showed a normal esophago-gastro-duodenal transit, ruling out the diagnosis of gastric diverticulum (Fig. 2). Therefore MRI scan was performed, showing a 6 cm-cyst of the posterior gastric wall not-communicating with the lumen of the stomach, suspected to be a gastric duplication cyst (Fig. 3). EUS confirmed the report of CT scan whereas FNAB showed rare squamous cells and histiocytes consistent with serum cyst. Because of persistent painful symptoms, unresponsive to medical therapy the patient consented to the surgical therapy. Laparoscopic exploration of the abdominal cavity was unremarkable. The gastro-colic ligament was dissected and fibrous adhesions between the posterior gastric wall and the ventral surface of the body

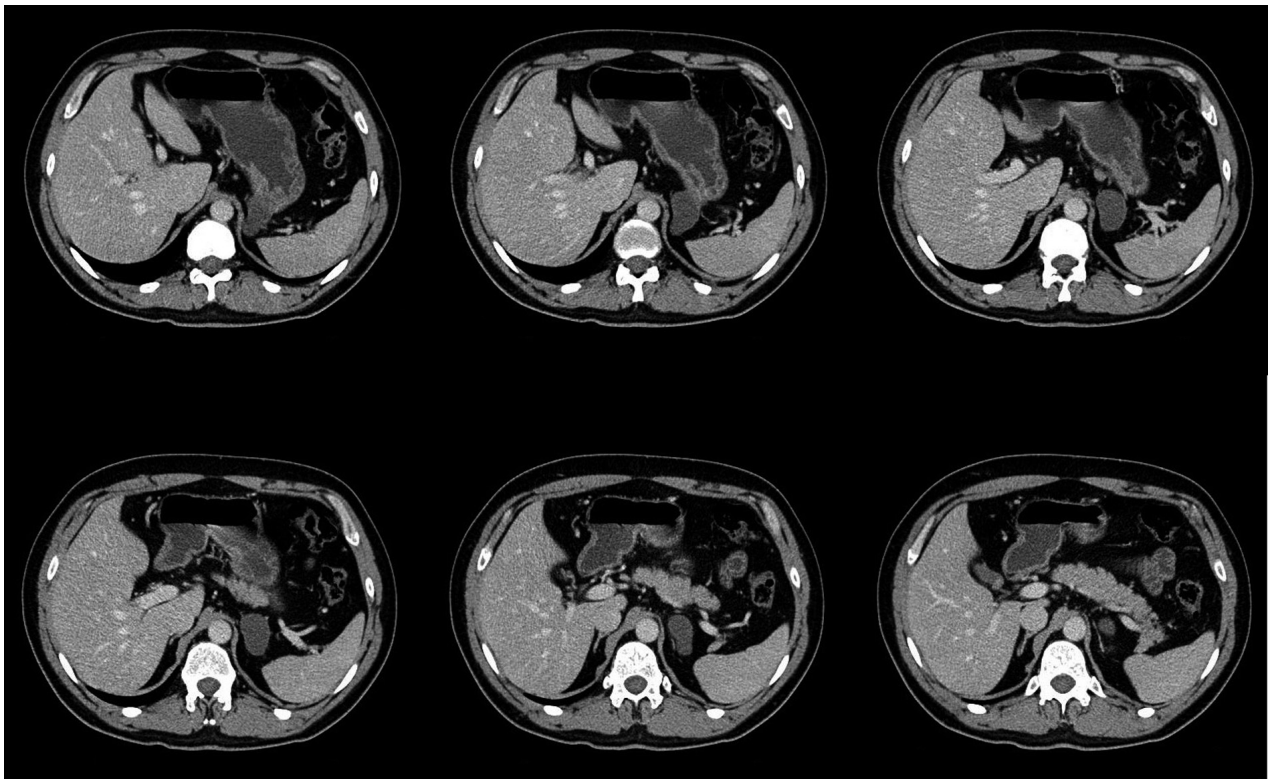

Figure 1. CT scan

of the pancreas were found. After dissecting the adhesions, an intramural gastric cyst was clearly identified, raising from the posterior wall of the stomach, extending caudally and dorsally to the body of the pancreas. A laparoscopic gastric wedge resection was performed, after division of the short gastric vessels, including the gastric fundus.

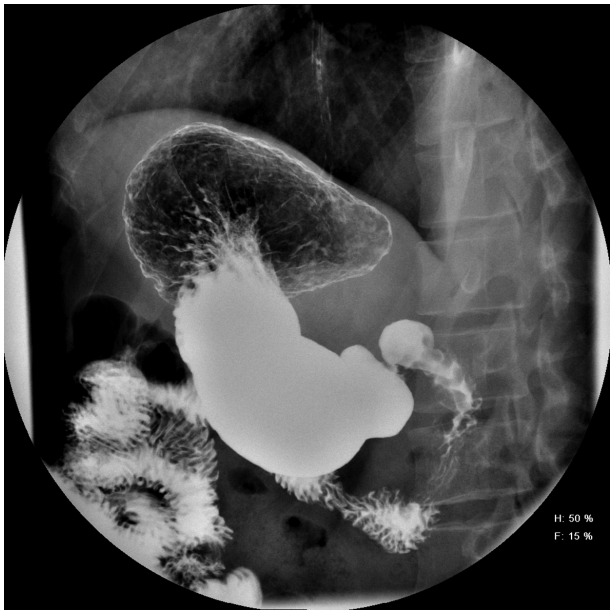

Figure 2. Preoperative contrast swallow

The post-operative course was uneventful and the patient was discharged in post-operative day 4. A contrast swallow was performed on postoperative day 3 (Fig. 4). Pathology report described a cyst of the gastric wall lined by ductal pancreatic epithelium, confirmed by immunochemistry staining positive for CK7 and CK19 (Fig. 5).

At 3 month follow up the patient reported a partial resolution of symptoms, but at 6 month follow up, after proton pump inhibitor therapy interruption, the patient's symptoms recurred as much as complained before the surgery. A full work-up, including laboratory blood analysis, CT scan, EGDS, esophago-gastric pH-manometry was performed, revealing only a mild non-specific distal gastritis, as preoperatively demonstrated. Noteworthy, the patient reported a weight-loss

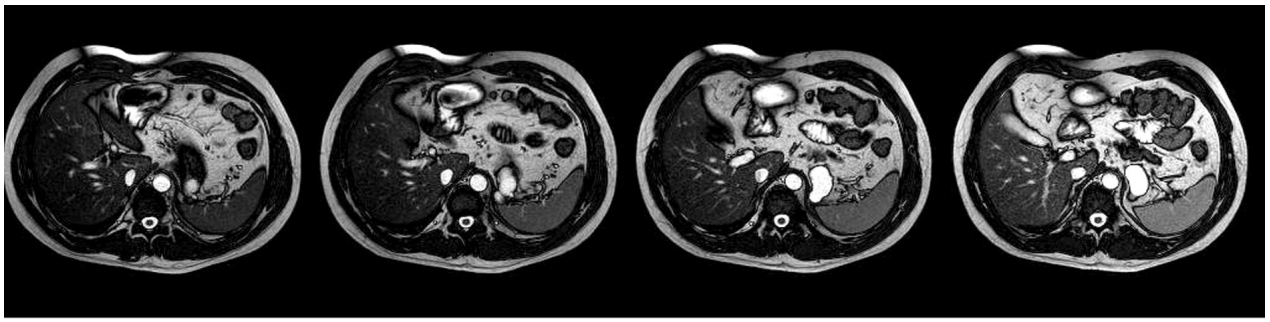

Figure 3. MRI scan

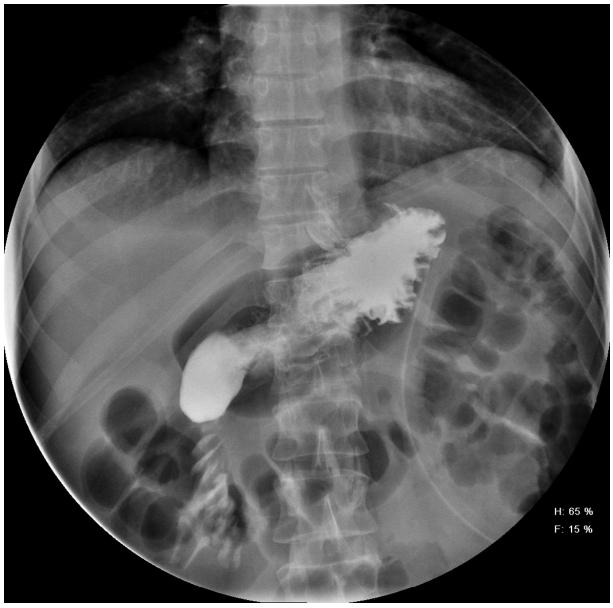

Figure 4. Postoperative contrast swallow PO day 3

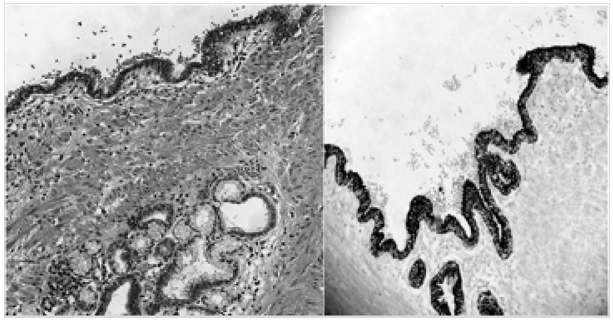

Figure 5. Histopathological slides with positive CK7 and CK19

of about 20 kg, due to the gastric resection. At 1 and 2 year follow up visits the patient had no symptoms with a stable body-weight of 62 kg and normal appetite.

Discussion

Heterotopic pancreas (HP) is defined as the presence of pancreatic tissue outside the usual anatomical location of the pancreas. Although HP can occur throughout the entire gastrointestinal tract, it is most commonly found in the stomach, duodenum and jejunum (1). The reported frequency of this finding during laparotomy is 0.5% and at autopsy is 1.7% (2). Despite the relatively frequent occurrence of HP, the vast majority of these cases are asymptomatic. When present, symptoms vary depending on the anatomical location and size of the lesion. Abdominal pain, nausea, vomiting and gastrointestinal bleeding are the most commonly reported symptoms and are most likely to be seen with lesions greater than 1.5cm in diameter (3). Pain associated with HP may be related to the local secretion of hormones and enzymes resulting in tissue inflammation or chemical irritation (1). Pain may also be related to mechanical obstruction of the intestinal lumen, especially when associated with nausea or vomiting. Gastric lesions are the most likely to be symptomatic, presenting with either epigastric pain or symptoms of gastric outlet obstruction due to a prepyloric mass (4). Other causes must be ruled out before the symptoms can be attributed to the HP, even if diagnosis and a clear association between HP and symptoms is rarely demonstrable preoperatively. HP also may present with symptoms related to complications similar to those normally associated with the pancreas, such as pancreatitis (5,6) pseudocyst formation (7,8) or malignant transformation (9,10).

Gastric HP presenting as cystic lesion is extremely rare. Few cases of retention cysts raised from het-

erotopic pancreas of the stomach (11,12) and only one case of granulomatous partially transformed cyst in heterotopic pancreas intramurally of the gastric corpus (13) were reported. However, to our knowledge, gastric HP presenting as serum cyst has never been reported.

Evaluation of cystic lesions of the stomach should consider cystic presentation of HP in differential diagnosis, along with gastric duplication cyst and gastric HP presenting as pseudo-cyst. Gastric duplication cyst is usually diagnosed at a median age of 3 years and it presents typically as a not-communicating cystic lesion located along the greater curve or posterior aspect of the stomach. This case showed clinical and radiographic findings compatible with a gastric duplication cyst, therefore in our opinion gastric duplication cyst was the most likely preoperative diagnosis to rule out albeit it is unfrequent in adult patients (14). True pseudocyst formation is extremely rare in ectopic pancreas. Histologically, it is defined as a collection of pancreatic juice enclosed by fibrous or granulation tissue with inflammatory cell infiltration (15). The persistent symptoms unresponsive to medical therapy, the absence of further pathological conditions able to explain the symptoms along with a potential for malignant transformation and cells seeding as also demonstrated for colon cancer (16,17) led us to offer the patient a mini-invasive surgical resection of the cystic lesion, which allow for a better short term postoperative outcome as already demonstrated for most standard laparoscopic resections (18,19) along with a reduced incisional hernia rate (20). Unfortunately symptoms recurred within 6-months from the surgery as much as complained preoperatively. Although Schmitz et al. reported a complete resolution of symptoms after surgical removal of a granulomatous partially transformed cyst, in our case surgical resection failed to relieve patient's symptoms.

This case leads us to speculate that this malformation could be asymptomatic itself, independently on the size of the cyst. We didn't identify any other associated pathological condition able to explain the patient's symptoms, unless a mild non-specific distal gastritis unresponsive to medical therapy. Pathogenesis, clinical significance and consequently the correct management of HP presenting as gastric cyst remains unclear. More advances are needed to improve the preoperative differential diagnosis and to better identify the association between clinical symptoms and radiographic, cytological and pathological findings of gastric cystic malformations. Surgical resection should be kept as the last therapeutic option and surgical candidates should be informed on the risk of failure to relieve their symptoms and of gastrectomy-related weight-loss.

In conclusion heterotopic pancreatic tissue is an extremely rare condition and it could present as serum cyst. This malformation should be included in the differential diagnosis of the cystic lesions of the stomach. Whether ectopic pancreatic cyst could be the cause of gastrointestinal symptoms remains unclear and surgical resection should be kept as the last therapeutic option.

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Correspondence:
Stefano Cecchini M.D.
Department of Surgical Sciences
Parma University
Via Gramsci 14 - 43100 Parma, Italy
Tel. +39.347.8103759
Fax +39.0521.940125
E-mail: stefano.cecchini@unipr.it