

CASE REPORT

Pseudocyst Formation in Gastric Ectopic Pancreas

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ABSTRACT

Context It is rare for ectopic pancreatic tissue to cause symptoms or require treatment, however diseases of normal pancreas may also occur in ectopic pancreas tissue.

Case report This report describes the clinical, endoscopic, radiologic and histologic features of a pseudocyst occurring in gastric ectopic pancreas in a 19-year-old man. The difficulty and implications of making an accurate pre-operative diagnosis are highlighted.

Conclusion Ectopic pancreatic tissue, although rare, should be considered in the

differential diagnosis of a submucosal gastric mass.

INTRODUCTION

Ectopic pancreatic tissue is a relatively rare form of hamartoma, which is usually an incidental finding at post-mortem examination. We report a case of aberrant pancreatic tissue presenting as an antral gastric tumour in a young male.

CASE REPORT

A 19-year-old male presented with a two-year history of post-prandial dyspepsia and occasional haematemesis, refractory to treatment with proton pump inhibitors. He had never smoked and drank alcohol only occasionally.

Gastroscopy showed a submucosal mass in the gastric antrum (Figure 1). The mucosa appeared normal throughout the stomach. Multi-slice spiral computerised tomography (MSCT), with oral and intravenous contrast in the portovenous phase showed a 2.5 cm diameter well-defined mass lesion in the posterior wall of the stomach with no evidence of local invasion (Figure 2). A smooth-walled non-enhancing cystic structure, not containing debris, was present within the mass.

We performed a wedge resection of the greater curvature of the stomach including the tumour, for symptom relief and to obtain

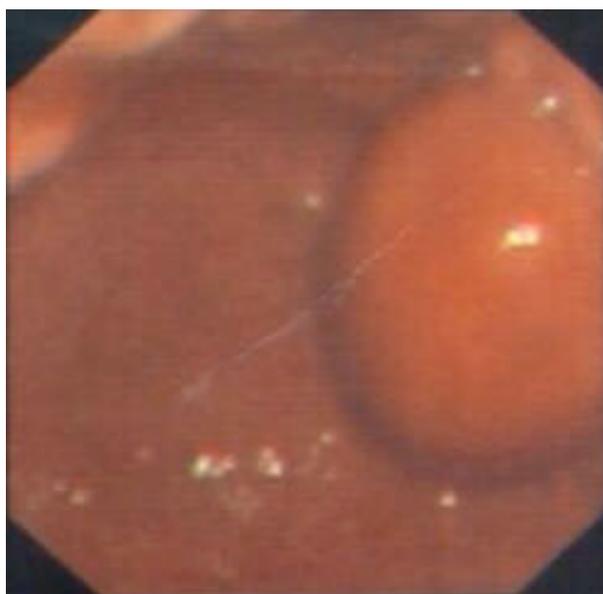


Figure 1. Gastroscopic view of antral submucosal mass.

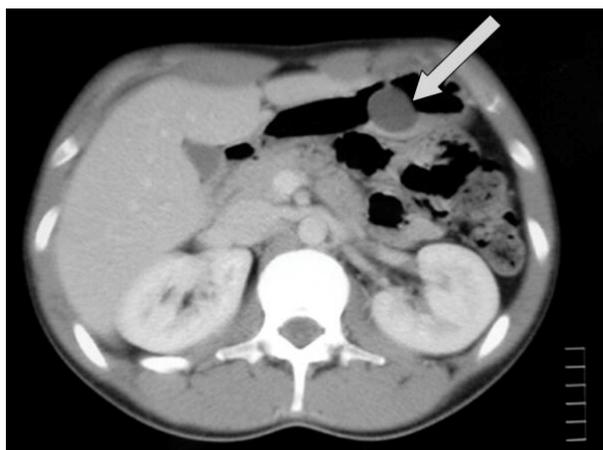


Figure 2. Multi-slice CT scan of abdomen (oral and portovenous phase IV contrast) demonstrating a gastric mass lesion with a spherical cystic component (arrow) and posterior enhancement.

histological diagnosis (Figure 3). No coexisting intra-abdominal pathology was detected at laparotomy. Histological examination showed well-differentiated glandular tissue within the *muscularis propria* with evidence of pancreatic acinar formation, duct development and the presence of islets of Langerhans. Chronic inflammatory change was present in the fibrous wall of a pseudocyst containing clear fluid.

The most likely cause of his symptoms, radiologic and histopathologic findings was acute pancreatitis with pseudocyst formation in gastric ectopic pancreatic tissue. Our patient had not previously been diagnosed as having an episode of acute pancreatitis, but several instances in his history were compatible with such an event. After an uneventful post-operative recovery he remains well and symptom free two years later.

DISCUSSION

Ectopic pancreas is defined as pancreatic tissue that lacks either anatomical or vascular communication with the normal body of the pancreas [1], and possesses all of the following histological features: pancreatic acinar formation, duct development and islets of Langerhans.

It is rare that ectopic pancreas causes symptoms requiring treatment, and is usually

an incidental finding, with an incidence at laparotomy of 0.2% [2, 3]. Up to 13% of post-mortem examinations report the presence of ectopic pancreas [3]. The most commonly reported sites for pancreatic ectopy are the stomach (24-38%), the duodenum (9-36%) and the jejunum (0.5-27%) [1, 4, 5]. Other sites include Meckel's diverticulum, biliary tract, liver, spleen and mesentery.

Ectopic pancreatic tissue may present, as in our case, with abdominal pain, nausea, vomiting or gastrointestinal bleeding. Depending upon the anatomical location, symptoms may originate from the mass effect of the tumour, such as gastric outlet obstruction in a pre-pyloric rest or obstructive jaundice in a bile duct focus [4]. The ectopic tissue can also undergo the complications and pathological changes that occur in normal pancreatic tissue [2, 3, 4, 5, 6]. Acute pancreatitis in ectopic pancreas can cause abdominal pain with elevation of serum pancreatic enzyme levels such as amylase or lipase. Malignant transformation has been reported, although it is difficult to determine its true incidence, as there have been only fifteen documented cases in the literature.

Cystic formation in ectopic pancreas could result from retention of exocrine secretions in the absence of a communication between the glandular epithelium and the gastric lumen.



Figure 3. Gross specimen. Note: intact overlying mucosa.

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 as in this case. Measurement of the amylase content of the cystic fluid will not differentiate between retention cyst and pseudocyst as both will be elevated.

Radiological diagnosis of gastric ectopic pancreatic tissue is difficult, however double contrast barium meal may characteristically show a focally raised mucosal area with associated superficial ulceration within the gastric antrum [5, 7]. In this case, multi-slice spiral CT (Figure 2), with oral and portovenous phase IV contrast, demonstrated a lesion arising from within the gastric wall excluding an exophytic lesion. Leiomyoma and lymphoma were unlikely due to the cystic nature of the mass, but leiomyoblastoma, a stromal cell tumour with malignant potential was an important differential diagnosis. Dual-phase spiral CT with oral contrast is sensitive to the detection of the submucosal, ectopic tissue, which enhances similarly to the normal pancreas. It is, however, difficult to distinguish ectopic pancreas tissue from other submucosal tumours [8].

Endoscopic findings are usually of a broad-based, umbilicated, submucosal nodule [9] with superficial biopsies non-diagnostic in the vast majority of cases [5]. The primary role for endoscopic visual assessment is exclusion of more common pathology such as peptic ulceration or adenocarcinoma. Other differential diagnoses include gastrointestinal stromal tumour (GIST), gastrointestinal autonomic nerve tumour (GANT), carcinoid or lymphoma [6]. Definitive diagnosis is by histological examination but endoscopic ultrasound combined with fine needle aspiration cytology has recently been reported to be of benefit [6]. If an accurate diagnosis can be made in an asymptomatic patient, conservative management is appropriate, as the risk of malignant change is extremely low [3]. Surgical excision, either endoscopically or at open operation [1, 2, 6], provides

symptomatic relief and is recommended if diagnostic uncertainty remains.

In conclusion, this case report highlights the successful management of an unusual pathological entity causing upper gastrointestinal symptoms. Ectopic pancreatic tissue, although rare, should be considered in the differential diagnosis of a submucosal gastric mass. We have demonstrated the clinico-pathologic findings and radiologic appearance of a typical case, and highlighted the rare occurrence of pseudocyst formation in ectopic pancreatic tissue.

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Keywords Choristoma; Pancreas; Pancreatic Pseudocyst

Abbreviations GANT: gastrointestinal autonomic nerve tumor; GIST: gastrointestinal stromal tumor; MSCT: multi-slice spiral computerised tomography

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