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CASE REPORT

Pancreatic and gastric heterotopic tissue presenting as a symptomatic gallbladder mass: A case report and literature review

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KEYWORDS
Pancreatic heterotopia;
Gastric heterotopia;
Intestinal metaplasia;
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Summary Pancreatic and gastric heterotopia is an uncommon congenital anomaly in which gastric and/or pancreatic tissue is found outside of its anatomic location. In the majority of patients, lesions are found incidentally because they tend to be asymptomatic. However, lesions may become symptomatic when inflammation, obstruction, and bleeding occurs. Depending on tissue size and location they can harvest a landscape of nonspecific symptoms, causing a delay in diagnosis. Heterotopic tissue of either gastric or pancreatic origin have been reported in literature, however the presence of combined gastric and pancreatic heterotopic tissue is exceedingly rare. We report a case of an 18-year-old female with polypoid gastric and pancreatic heterotopia and focal intestinal metaplasia of the gallbladder with clinical findings of acute cholecystitis. In this literature review, we synthesize the clinical significance, histopathological features, diagnosis and management of pancreatic and gastric heterotopic tissue.

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Introduction

Pancreatic and gastric heterotopias are defined as pancreatic and gastric tissue lying outside their anatomic location without vascular or anatomical continuity with the pancreas proper or the stomach [1—3]. These heterotopias have been described in the esophagus, stomach, duodenum, jejunum, and Meckel's diverticulum, however their presence in the gallbladder is very rare [3—6]. Its clinical significance varies from an asymptomatic incidental finding to a landscape of symptoms depending on tissue type, size, and location [2,3]. Gastric heterotopias tend to be more clinically significant due to acid secretion and polyp formation causing abdominal pain, ulceration, and bleeding [3]. Though usually benign, reports of malignant transformation, such as adenocarcinoma, carcinoid, and microinvasive carcinoma arising from heterotopia located in the intestines and gallbladder have been documented [1,3].

Heterotopias in the gallbladder can present with symptoms of acute and chronic cholecystitis, gallbladder neck obstruction, and even gallbladder perforation [2,6]. We report a case of an 18-year-old female with polypoid gastric and pancreatic heterotopia with focal intestinal metaplasia of the gallbladder with a clinical presentation of postprandial right upper quadrant pain leading to the clinical impression of chronic cholecystitis.

Case presentation

This is the case of an 18-year-old African American woman with history of ovarian cyst who presented with episodes of nausea, non-biliary emesis and right upper quadrant pain. Symptoms were most noticeable post prandial after she ate fatty food or vegetables with coconut oil, usually in the morning. She had no previous abdominal surgeries and was being treated symptomatically with ondansetron, promethazine, non-steroid anti-inflammatory drugs and sucralfate without relief. She denied a family history of colon cancer, biliary, liver disease, or inflammatory bowel disease. A previous esophagogastroduodenoscopy (EGD) was normal, and gastric and duodenal biopsies were negative for metaplasia and dysplasia. A right upper quadrant ultrasound showed a 9.6 mm polyp on the posterior wall of gallbladder without sludge or stones. The patient underwent a laparoscopic cholecystectomy and the gallbladder and liver looked unremarkable during the procedure. The gallbladder was sent for pathology evaluation.

Pathology

Grossly, the gallbladder was intact measuring $6.3 \times 2.2 \times 1.8$ cm. The cystic duct was unremarkable. Opening the gallbladder revealed a $0.9 \times 0.7 \times 0.6$ cm pink red rubbery polypoid nodule on the posterior wall of the body. No calculi were identified. Cut surfaces of the polypoid nodule was gray-pink and there was no invasion identified. Remaining mucosal surfaces are yellow-green and finely granular. The rest of wall is 0.1 to 0.2 cm in thickness and no other mucosal lesions or masses are identified. Representative sections were submitted for microscopic assessment. Histologically, the gallbladder nodule contained gastric foveolar epithelium and oxyntic glands composed of parietal cells and chief cells, pancreatic acini admixed with ducts present in the polypoid nodule (Fig. 1). No nests of islet cells were identified. No dysplasia, significant chronic inflammation or Helicobacter Pylori were identified. Helicobacter Pylori immunostains was negative. The remaining gallbladder had mild chronic cholecystitis with intestinal metaplasia and minimal cholesterolosis (Fig. 2). The final diagnosis of the gallbladder body nodule was polyploid gastric and pancreatic heterotopia.

Discussion

The incidence of heterotopic pancreas and gastric tissues in autopsy studies ranges from 0.1 to 13 percent in the general population [2,3,7]. Though the etiology is no yet well defined, theories suggest it results from an error during embryological development during rotation of the foregut when fragments of the pancreas and gastric tissue separate and deposit in ectopic sites [3,5,6,8]. To explain the occurrence of heterotopic pancreas in distant organs, it has been suggested that metaplasia of pluripotent endodermal cells of the embryonic foregut migrate to the submucosa during embryogenesis and transform into pancreatic tissue [6,9]. A recent theory asserts abnormalities in the Notch signaling system gives rise to pancreatic heterotopia through cell to cell communication and gene regulating mechanisms involved in the development of the pancreas [2]. The gallbladder arises from the hepatic primordium, which originates from the ventral surface of the foregut, slightly caudal to the developing stomach. Gastric heterotopia in the gallbladder could result either by entrapment of the primitive gastric tissue, by heterotopic differentiation within the primitive gallbladder or by metaplastic differentiation as described above [1,6,9].

The first reported case of pancreatic heterotopia was seen in Meckel's diverticulum but, almost half of heterotopic pancreas occurs in the proximal gastrointestinal tract, most frequently in stomach along the greater curvature of the gastric antrum or the duodenum [6,8—10]. Approximately 1% of all cases of pancreatic heterotopia are found in the liver, biliary tree, and/or gallbladder [6,9]. Grossly in the gallbladder, pancreatic heterotopia is usually a well demarcated, firm, yellow, and irregular lobular shaped intramural nodule [2,3,6,9]. Lesions may also present with central umbilication, representing an orifice of the ductal system [3,9]. Fifty percent of heterotopic pancreatic tissue found in the gallbladder, arise in the neck [2]. Most lesions are solitary and under 3.0 cm in size [6]. In our case, the polypoid nodule with both gastric and pancreatic heterotopia is present on the posterior wall of the gallbladder body. Histologically, pancreatic heterotopia is characterized as nodule or focus composed of at least two of three components from normal pancreas including acini, ducts and islets, which is defined as a mass covered with normal mucosa [6]. The most common presentation of pancreatic heterotopia is a submucosal nodule, seen in up to 73% of cases [2,4,6,9]. Less common presentations include tissue that spans the submucosa and muscularis propria, location in the muscularis

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