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Case report

Inflammation of Ectopic Pancreatic Tissue as Unusual Cause of Duodenal Perforation – A Case Report

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A B S T R A C T

Ectopic pancreatic tissue, also known as a pancreatic rest, is an uncommon congenital anomaly defined as extrapancreatic tissue located far from the pancreas and without any connection via vascular or anatomical means to it. Such tissue may occur throughout the GI tract but has a propensity to affect the stomach and the proximal small intestine. The majority of patients with pancreatic ectopia are asymptomatic, but when symptoms occur, they can be presented in a variety of ways. We report a patient with acute surgical abdomen due to a duodenal perforation caused by inflammation of ectopic pancreatic tissue in duodenum and stomach. Histology of the resected duodenum and stomach demonstrated heterotopic pancreatic tissue acute inflammation without atypia, suggesting »pancreatitis of the duodenum and stomach«. To date, there have been a few reports describing perforation of the stomach due to heterotopic pancreas. Therefore, the present case was considered to be a very rare case of this disorder. To conclude, heterotopic pancreas should always be considered in the differential diagnosis of acute abdomen.

Key words: *ectopic pancreas, acute abdomen, acute pancreatitis*

Introduction

Pancreatic ectopy (synonymous heterotopic, accessory, or aberrant pancreas etc.) is defined as the presence of pancreatic tissue lacking anatomic and vascular continuity with the main body of the gland[1–3]. It can be found anywhere along the foregut and proximal midgut and usually causes no symptoms[1–3]. About 75% of all pancreatic rests are located in the stomach, the duodenum, or the jejunum[1]. However, they have also been found in the ileum. Meckel's diverticulum, the gallbladder, the common bile duct, the splenic hilum, the umbilicus, the lung, and in perigastric and periduodenal tissue[2]. At autopsy, the frequency of ectopic pancreas is between 1–2% (range 0.55% to 13%); the rate of recognition at the time of laparotomy is 0.2%. In endoscopic practice ectopic pancreas is oftenly diagnosed and sometimes imposes differential diagnostic problems[1–3]. Here we present the case of a patient with acute duodenal perforation caused by inflammation of ectopic pancretic tissue located in the duodenal wall.

Case Report

A 51-year-old man presented with a 5-day history of upper abdominal pain of an increasing intensity associated with nausea. He denied any previous history of abdominal distress. Past medical history revealed diabetes mellitus type II and hyperlipoproteinaemia. Two years ago, the patient was treated for a typical pneumonia. Physical examination was normal, except for a moderate abdominal pain and tenderness in the epigastric area, showing no signs of peritonitis or palpable mass.

Blood tests showed leukocytosis (14,5x10^9/L), elevated C reactive protein (CRP 99,7–171,0 mg/L) and hyper-

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glycaemia (11,4 mmol/L). Blood and urine amylases were normal (37 U/L in blood and 123 U/L in urine). Laboratory data and reference value shown on Table 1. Abdominal X-ray was normal. Ultrasonography showed a thick gastric wall in antral region.

The patient was admitted to the Department of Gastroenterology. The upper endoscopy performed at day 2 showed a thick, edematosed gastric wall with inflammated gastric mucosa in the antral region and the duodenum. Hystological findings of mucosal biopsy in the antral region showed superficial gastritis. There were no serological or hystological signs of a Helicobacter pylori infection. Differential diagnosis, at this point, included different submucosal pathological proceses of antral region such as, lymphoma, leiomyosarcoma, sarcoma gastrointestinal stromal tumour (GIST), gastrointestinal autonomic nerve tumour (GANT), carcinoid etc. During days 1 and 2 the patient received intravenous fluids, electrolytes, insulin and pantoprazole (Table 2). The patient's condition was better. At day 3, the patient received a liquid diet. At day 5, the patient's condition worsened with clinical signs of acute surgical abdomen. Urgent CT showed a thick edematosed gastric wall with signs of the duodenal perforation (Figures 1a and 1b). The patient underwent urgent surgical treatment. Intraoperative findings showed purulent diffuse peritonitis, inflammation of the antral region and the duodenum with the posterior duodenal perforation. The Billroth II resection of stomach was performed with peritoneal lavage (Figures 2a and 2b).

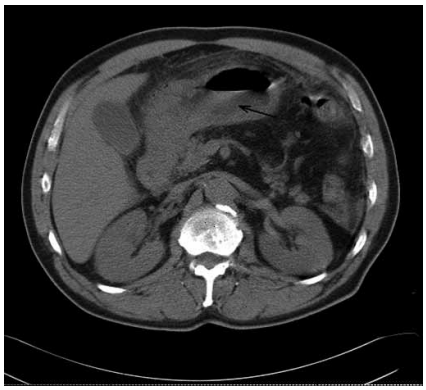
TABLE 1
SUMMARY OF LAB. DATA AT ADDMISION

Laboratory data	Value	Normal range
RBC	5.08	$4.34–5.72x10^{12}$/L
WBC	14.5	$3.4–9.7x10^{9}$/L
ESR	55/78	3–23 mm/3.6 ks
CRP	99.7–171.0	0.0–5.0 mg/L
Glucose	11.4	4.4–6.4 mmol/L
Amylase blood	37	23–91 U/L
Amylase urine	123	0–200 U/L

RBC (Red blood count), WBC (White blood count), ESR (Erythrocyte sedimentation rate), CRP (C reactive protein)

Histology of the resected duodenum and stomach showed acute and chronic inflammation of the entire gastric and duodenal wall. Within the inflammated duodenal and gastric tissue we found normal and inflammated heterotopic pancreatic tissue. Tissue samples were sent for a second opinion to the »Merkur« University Hospital, Zagreb and the final histology demonstrated heterotopic pancreatic tissue acute inflammation with-

a)



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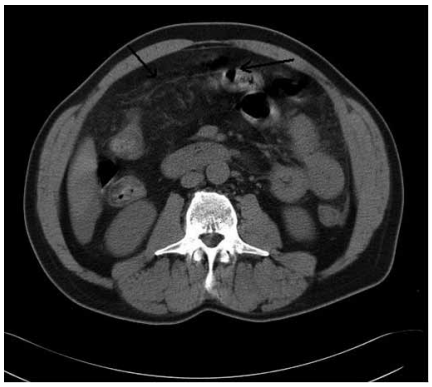
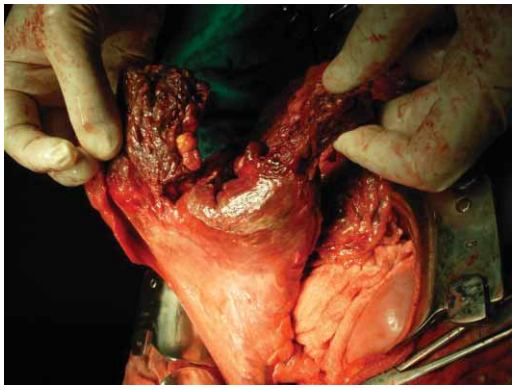


Fig. 1. a) Urgent CT scan showed thick edematosed gastric wall in antral region, b) CT scan: signs of peritonitis due to duodenal perforation.

a)



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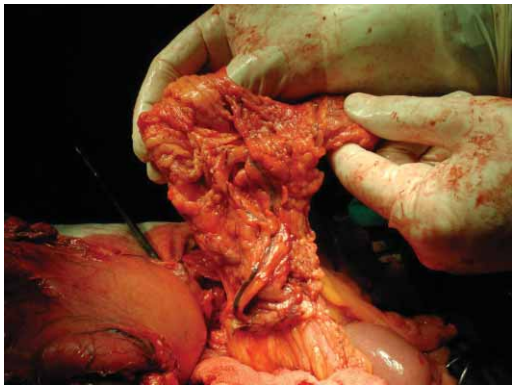
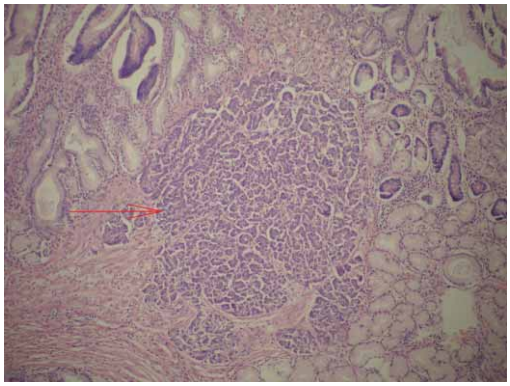


Fig. 2. a) Surgery: Billroth II resection of inflammated duodeum and stomach, b) Surgery: resection of inflammated omentum.

a)



b)

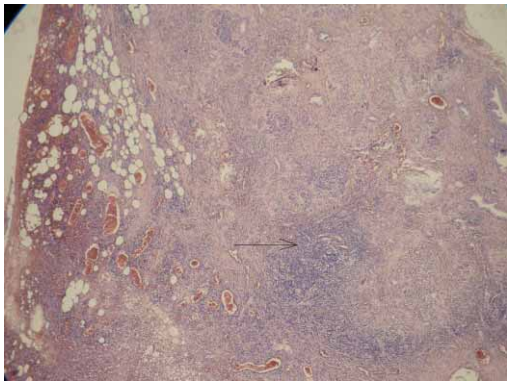


Fig. 3. a) Hystology of resected duodenum: ectopic pancreatic tissue, b) Hystology of resected duodenum: »pancreatitis in duodenum«.

out atypia, suggesting »pancreatitis of the duodenum and stomach« (Figures 3a and 3b). The postoperative period was uneventful and the patient was discharged on day 18.

Discussion

The pancreas develops from two primordial diverticula of the duodenum and becomes apparent at 4th week of gestation[3]. After axial rortation of the gut, the mesoduodenum fuses with the posterior peritoneum and forms the most part of the duodenum. Fusion of the two pancreatic buds occurs simultaneously. During pancreatic organogenesis, a number of embryological »mistakes« can occur.

Because of the close development of the embryonic pancreatic primordial buds to the foregut, it is not surprising that 70–90% of pancreatic ectopia occurs in the upper GI system. Most ectopic pancreas in the upper GI tract are found in the gastic antrum: 75% are in submucosa, 15% in the muscular layers, and 10% in the subserosa (4, 5, 6). However, heterotopic pancreatic tissue may be implanted extraintestinaly. e.g. gallbladder, common bile duct, liver, spleen, omentum, lungs, umbilicus, mediastinum etc. Microscopically, ectopic pancreas is composed of varyng amounts of pancreatic ducts, acini and

TABLE 2
SUMMARY OF THERAPY RECEIVED ON DAY 1–4

Therapy	Value
Normal saline solution	2000–1000 ml iv.
5% glucose-insulin potassium chloride (GIK) solution	1000 mL +8 U + 20 mEq iv.
Insulin	8–12 U/every 6 hours
Potassium chloride	40 mEq/ daily
Pantoprazole	2x40 mg iv.

islets of Langerhans[1]. In most cases this anomaly is an incidental finding at microscopic examination of tissue removed during endoscopy, surgery or autopsy, without requiring further investigations or management. Clinically, significant lesions are greater than 1.5 cm in diameter and are adjacent to or directly involve the mucosa[7]. Pancreatic rest appears as round, smooth submucosal filling defects with a central umbilication on barium contrast studies. Endoscopically, pancreatic rest appears as a round, sometimes yellow, well defined, firm, intramural lesion from 2 mm to 4 cm in diameter with a central depression which may ulcerate. Histological diagnosis of abberant pancreas is usually difficult when tissue specimens are obtained using standard endoscopic biopsy forceps. Tissue examination usually reveals normal pancreatic lobules with all pancreatic structural elements.

Symptoms occur between the fourth and the sixth decade of life. The most common clinical symptoms attributed to ectopic pancreas are abdominal (epigastric) pain, dyspepsia, and GI bleeding[1,7]. Pyloric obstruction by ectopic tissue, obstruction of the ampulla of Vater, the intestine, and the biliary tree were described as well[8–10]. Several cases of cancer occurring in ectopic pancreas have been already described[8]. Pancreatitis in the ectopic tissue, pseudocyst formation and inflammation with necrosis of the adjacent structures have been also reported[2,9–17]. Inflammation of the ectopic pancreatic tissue can cause variety of intraabdominal complications. There have been report of ectopic pancreas complicated by pancreatitis and pseudocyst formation mimicking jejunal diverticulitis[11]. Case report of Pedersen and al. decribed acute pancreatitis of ectopic pancreas presenting as small-intestine tumor[13]. Kaneda et al. reported case of ectopic pancreas in the stomach presenting as an inflammatory abdominal mass[14].

Despite the observation that most cases of heterotopic pancreas do not cause problems, the condition has been reported to lead to symptoms from inflammation (as in our case), obstruction, or even malignant transformation. A correlation has been established between the presence of symptoms, the size of the lesion and the extent of mucosal involvement[18].

Management of ectopic pancreas is somewhat controversial. As this anomaly is largely asymptomatic and rarely associated with clinical pathology, the incidental

findings of characteristic lesion do not necessarily warrant excision. Endoscopic removal is dangerous because of risk of perforation or bleeding since the lesion is submucosal. Operative treatment is reserved for complicated cases, when clinical symptoms are associated with its presence such as recurrent bleeding, obstruction, inflammation or malignant degeneration or when its appearance is difficult to differentiate from other lesions such as lymphoma, GITS or carcinoid.

Although the real reason for perforation in our case is speculative, it was considered that necrotising inflammation in the heterotopic pancreas tissue resulted in destruction of tissue and then perforation at the serosal site of duodenum.

To date, there have been a few reports describing perforation of the stomach due to heterotopic pancreas[15,17]. Therefore, the present case was considered to be a very rare case of this disorder. In addition to giving the patient's symptoms, the inflammation appeared to lead to a transmural and extraserosal involvement, simulating a gastric and duodenal submucosal tumor. To conclude, heterotopic pancreas should always be considered in the differential diagnosis of acute abdomen.

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UPALA EKTOPIČNOG TKIVA GUŠTERAČE KAO NEUOBIČAJENI UZROK PERFORACIJE DVANAESNIKA- PRIKAZ SLUČAJA

SAŽETAK

Ektopično ili ostatno tkivo gušterače, je rijetka kongenitalna anomalija kod koje se tkivo gušterače nalazi smješteno izvan anatomskih granica gušterače bez vaskularnih ili drugih veza sa tijelom žlijezde. Anomalija se pojavljuje duž cijelog GI trakta, ali se najčešće nalazi u želucu i proksimalnom dijelu tankog crijeva. Najveći broj bolesnika sa ektopijom gušterače je bez simptoma, no u onih bolesnika kod kojih se jave, simptomi su raznoliki. Prikazujemo bolesnika sa akutnim kirurškim abdomenom uslijed perforacije dvanaesnika uzrokovane upalom ektopičnog tkiva gušterače. Histološki pregled reseciranog duodenuma i želuca je pokazao upalu heterotopičnog tkiva gušterače sugerirajući »pankreatitis u želucu i dvanaesniku«. Prema literaturi, do sada je opisano tek nekoliko slučajeva perforacije želuca uslijed postojanja ektopičnog tkiva pankreasa. Čini se da je naš slučaj rijedak, gotovo jedinstven. Kod bolesnika sa akutnim abdomenom potrebno je diferencijalno dijagnostički razmotiti mogućnost postojanja ektopičnog tkiva gušterače.