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An unusual cause of acute abdomen: Mesenteric heterotopic pancreatitis causing confusion in clinical diagnosis

Akut karının alışılmadık bir nedeni: Klinik tanıda karışıklığa neden olan mezenterik heterotopik pankreatit

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Heterotopic pancreas is defined as the presence of pancreatic tissue that lacks anatomic and vascular continuity with the main body of the pancreas. Frequent symptoms and signs are epigastric pain, abdominal fullness and tarry stools. The most frequent locations of heterotopic pancreas tissue are the stomach and jejunum; however, there are a few reported cases of heterotopic pancreas in the mesentery of the small intestine. Heterotopic pancreas may or may not cause complications related to the pathologic conditions of the pancreas itself. Here we present a case showing an unusual cause of acute abdomen, which caused confusion in the clinical diagnosis preoperatively. The definitive diagnosis was achieved only after histopathologic examination in the postoperative period. Final diagnosis of the patient was mesenteric heterotopic pancreatitis, which was a complication of heterotopic pancreas itself with a rarely seen location. In conclusion, mesenteric heterotopic pancreatitis is seen very rarely and may be an unusual cause of acute abdomen. If the pathologic condition develops in the heterotopic tissue, as in the case of heterotopic pancreas, signs and symptoms of the disease may cause confusion in the clinical diagnosis. We agree that preoperative diagnosis of heterotopic pancreas is still difficult, even in a symptomatic patient.

Key words: Acute abdomen, mesentery, heterotopic pancreatitis

Heterotopik pankreas, pankreasın ana gövdesi ile anatomik ve vasküler bağlantısı olmayan pankreas dokusunun bulunması olarak tanımlanmaktadır. Sık belirti ve bulguları epigastrik ağrı, karında dolgunluk ve katran gibi dışkıdır. Dokusunun en sık lokalizasyonu mide ve jejunumdur, ancak ince barsak mezenterinde yerleşmiş bir kaç heterotopik pankreas vakası bildirilmiştir. Heterotopik pankreas pankreasın kendisine ait patolojik durumla ilgili olan ve olmayan komplikasyonlara neden olabilir. Biz burada preoperatif olarak klinik tanıda karışıklığa neden olan akut abdomenin alışılmadık bir nedenini bulunduran bir vaka sunduk. Kesin tanı ancak postoperatif dönemde sonuçlanan histopatolojik inceleme sonrasında başarıldı. Son tanı çok nadir bir lokalizasyonda görülen heterotopik pankreasın kendisine ait bir komplikasyonu olan mezenterik heterotopik pankreatitti. Sonuç olarak mezenterik heterotopik pankreatit çok nadir görülmektedir ve akut karının alışılmadık bir nedeni olabilir. Eğer bir dokunun patolojik durumu heterotopik olanında oluşursa, heterotopik pankreas vakasında olduğu gibi hastalığın belirti ve bulguları klinik tanıda karışıklığa neden olabilir. Biz, semptomatik hastalarda dahi preoperatif olarak heterotopik pankreas tanısının halen zor olduğu gerçeğine katılmaktayız.

Anahtar kelimeler: Akut karın, mezenter, heterotopik pankreatit

INTRODUCTION

Heterotopic pancreas (HP), which is also referred to as aberrant pancreas, choristoma and adenomyoma, is defined as the presence of pancreatic tissue that lacks anatomic and vascular continuity with the main body of the pancreas (1, 2). The most frequent locations of heterotopic pancreas tissue are the stomach and jejunum (3); however, there are a few reported cases (4-6) of heterotopic pancreas in the mesentery of the small intestine. The majority of the cases are found coincidentally at the time of surgery for other abdominal conditions (7). As the preoperative diagnosis of heterotopic pancreas is difficult, frozen section examination is mandatory to establish the diagnosis (8). In a series of 15 patients, the diagnoses in 40% of the patients were made only after postoperative histopathologic examination (7).

Here we present a case with an unusual cause of acute abdomen, which caused confusion in the clinical diagnosis preoperatively. Final diagnosis of the patient was mesenteric heterotopic pancreatitis, which was a complication of the heterotopic pancreas itself with a rarely seen location.

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CASE REPORT

A 75-year-old female admitted to the hospital with acute periumbilical pain, nausea and vomiting. Her physical examination was suggestive of acute abdomen with peritoneal irritation findings. Abdominal ultrasonography (US) revealed cholelithiasis and gallbladder wall thickening. Blood chemistry results were: alanine aminotransferase (ALT): 38 U/L, aspartate aminotransferase (AST): 48 U/L, amylase: 1311 U/L, lipase: 1258 U/L, total/direct bilirubin: 2.4/0.5 mg/dl, creatinine: 1.4 mg/dl, blood urea nitrogen (BUN): 65 mg/dl, C-reactive protein (CRP): 127 mg/L, and white blood cells (WBC): 9650/μL. The clinical diagnosis of mild biliary pancreatitis was made according to the Ranson criteria and supportive medical treatment was started. Although the complaints of the patient and biochemistry findings improved with the medical treatment, abdominal examination findings persisted. Laparoscopic cholecystectomy was planned on the fifth day of her admission. However, a mass of 10x12 cm was discovered beneath the umbilicus on palpation of the abdomen under general anesthesia. US repeated on the operating table before the surgical intervention revealed a pseudokidney image suggesting an abdominal tumoral mass originating from the intestine or mesentery.

Because of the mass, the patient underwent laparotomy, which revealed normal pancreas, cholelithiasis, and an inflammatory mass located in the mesentery of the proximal jejunum, 15x8x5 cm in size and containing small abscess foci and necrotic tissue. Frozen section of the mass showed a benign histology. After microbiological sampling, the abscess foci were drained, a great portion of the inflammatory mass was excised and debridement of the necrotic tissue and cholecystectomy were performed. Except for amylase and lipase values at two-fold of upper limits, postoperative recovery was uneventful and the patient was discharged on the seventh postoperative day. After histopathological examination (Figure 1a and 1b), definitive diagnosis of the patient was mesenteric heterotopic pancreatitis.

Thereafter, the patient was invited to the hospital for 1-, 6- and 12-month follow-ups. In the first month examination, abdominal US of the patient was normal, and amylase was two-fold the upper limit. At the 6th and 12th postoperative month examinations, the patient had no complaints, normal blood biochemistry and normal abdominal US findings.

DISCUSSION

Heterotopic pancreas is seen rarely, with an incidence of 0.25% of the findings of all abdominal operations performed during the same period (7). It is encountered most commonly in the fourth, fifth and sixth decades, with a slight male predominance (7, 8). Only 33-47% of the patients are symptomatic, and 41-66% of HP cases are found incidentally at operation for other surgical conditions (3, 7, 8). Frequent symptoms and signs are epigastric pain (77%), abdominal fullness (30%), tarry stools (24%), vomiting (18%), and diarrhea (18%) (8). Complications of HP may or may not be related to pathologic conditions of the pancreas itself (7, 8). Those related to the pancreas may be inflammation, cyst or pseudocyst formation, abnormal hormone secretion and malignant degeneration (7, 9, 10). Those unrelated to the pancreas may be gastric outlet, intestinal or common bile duct obstruction, intussusception or bleeding (7, 8, 10). Mesenteric heterotopic pancreatitis is a rare cause of acute abdominal pain (4). The inflammatory mass in our case was one of the signs of the disease but was overlooked because it masked voluntary guarding and was recognized only after muscular relaxation under general anesthesia. Therefore, the clinical presentation of the patient with acute abdomen was initially decided to be related to biliary pancreatitis; however, after histopathological examination, the cause of acute abdo-

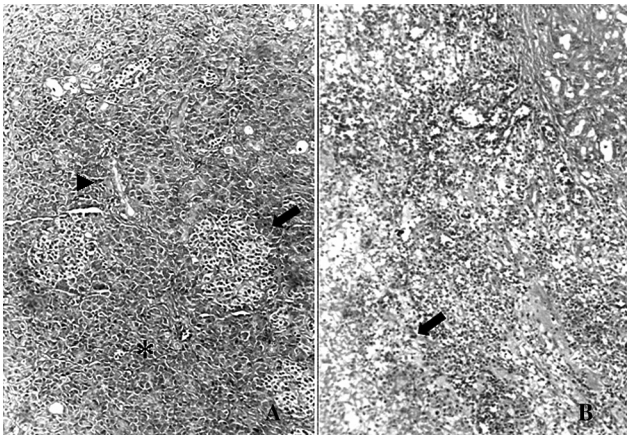

Figure 1. The histopathological examination of the surgical specimen revealed pancreas tissue having both exocrine and endocrine components (acini [asterisk], ducts [arrow head] and Langerhans' islets [arrow]) (Figure **1A;** hematoxylin and eosin [H&E], x100) among the intensive inflammatory cell infiltration and necrosis (arrow) (Figure **1B;** H&E, x200). The inflammatory cell infiltration consisted of mixed inflammatory cells rich in eosinophils. The definitive diagnosis was mesenteric heterotopic pancreatitis.

men was determined as mesenteric heterotopic pancreatitis, which was a complication of the HP itself.

The most frequent location of HP tissue is the stomach (47%), followed by jejunum (35%), duodenum (11.7%) and ileum (5.8%) (3). Although mesenteric location of HP is rare, there are a few reported cases of HP (4-6) in the mesentery of the small intestine; thus, our patient is in the very rarely seen group.

Gastroduodenoscopy, upper gastrointestinal series and abdominal US are the three frequently used diagnostic tools among the 12 different kinds of imaging studies, some of which are endoscopic US, computerized tomography (CT), endoscopic retrograde cholangiopancreatography, and magnetic resonance (3). Abdominal US may be normal or, like CT, show a cystic or heterogeneous mass in different parts of the abdomen (3, 4, 11, 12). Preoperative biopsy may not help in the diagnosis of HP, and except for a few cases (3), definitive diagnosis of HP was not established preoperatively even in the symptomatic patients (3, 7, 8). Preoperative diagnosis of HP is still difficult regardless of the recent advances in diagnostic tools and techniques (3, 11). Physical examination findings and laboratory findings directed us toward the diagnosis of biliary pancreatitis as the possible cause of acute abdomen. The mesenteric mass, which can be revealed by US, was overlooked because of the inadequate initial US examination. It was not possible for us to make an accurate diagnosis preoperatively because an unusual cause of acute abdomen like mesenteric heterotopic pancreatitis was not suspected. Only after the nontender abdominal palpation and repeated US under general anesthesia before the laparotomy was the initially overlooked tumoral mass recognized.

Intraoperative frozen section may help in the diagnosis of HP in many cases (8) and prevent unnecessary, more extensive surgery (10); however, it will not always lead to a correct diagnosis (1). Thus, the diagnosis of HP after postoperative histological examinations is not infrequent (7). In the present case, although frozen section excluded malignancy, it did not help in making an exact diagnosis. As the inflammatory tumoral mass contained small abscess foci and necrotic tissue, infected mesenteric lymph nodes was a probable diagnosis during the operation.

Conservative treatment and follow-up or removal of HP to prevent future complications are the recommended treatment options (1, 8). Resection of the tissue-bearing area is advisable when the condition is encountered coincidentally during the operation (7). Although the clinical symptoms of patients disappear completely after surgical removal of the aberrant tissue (8), hyperamylasemia may persist for five weeks postoperatively (1).

We performed a minimal surgical procedure for the safe treatment of this patient, since the frozen section was benign and it was not possible to resect the inflammatory mass totally due to its deep location in the mesentery surrounding the whole vasculature of the intestine. The definitive diagnosis was not achieved until results of the histopathologic examination in the postoperative period were obtained. Like in our patient, an inflammatory abdominal mass with abscess formation may histologically present as an ectopic pancreas composed of the cell types found in normal pancreatic tissue with both endocrine and exocrine functions (8, 13).

In conclusion, mesenteric heterotopic pancreatitis is seen very rarely and may be an unusual cause of acute abdomen. If the pathologic condition develops in the heterotopic tissue, as in the case of HP, signs and symptoms of the disease may cause confusion in the clinical diagnosis. We agree that preoperative diagnosis of HP is still difficult (3), even in a symptomatic patient.

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