A rare cause of hemosuccus pancreaticus: Primary splenic artery aneurysm ruptured into pancreatic serous cystadenoma

Hemosukkus pankreatikusun nadir bir nedeni: Pankreatik seröz kistadenoma içine primer splenik arter anevrizma rüptürü

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Hemosuccus pancreaticus is a rare clinical condition defined as bleeding into the pancreatic duct from a peripancreatic artery. We present here a 57-year-old woman admitted to our clinic with abdominal pain, tar-colored stool and confusion. Further investigations were done because of severe anemia. Abdominal computerized tomography revealed intraabdominal hematoma. Laparotomy was performed, which confirmed that intraabdominal haemorrhagia had occurred with the rupture of a splenic artery aneurysm into a pancreatic serous cystadenoma, which ruptured into the abdomen because of high pressure. This is an interesting case diagnosed with multidisciplinary approaches.

Key words: Hemosuccus pancreaticus, splenic artery aneurysm, obscure bleeding

Hemosukkus pankreatikus, peripankreatik bir arterden pankreatik kanal içine kanama olarak tanımlanan nadir bir klinik tablodur. Biz bu yazıda kliniğimize karın ağrısı, koyu renkli dışkılama ve şuur bulanıklığı ile başvuran 57 yaşında bir kadın hasta sunduk. Ciddi anemisi olması nedeniyle daha ileri incelemeler yapıldı. Bilgisayarlı batın tomografisinde intraabdominal hematom saptandı. Ardından laparatomi yapıldı ve splenik arter anevrizmasının pankreatik seröz kistadenom içine açıldığı, artan kist içi basınçtan dolayı kistin batın içine rüptürüne bağlı intraabdominal hemoraji meydana geldiği belirlendi. Bu ilginç olguya multidisipliner yaklaşım sonucu tanı konuldu.

Anahtar Kelimeler: Hemosukkus pankreatikus, splenik arter anevrizması, obscure kanama

INTRODUCTION

Hemosuccus pancreaticus (HP) is defined as bleeding into the pancreatic duct from a peripancreatic artery. This condition is also referred to as "wirsungorrhagia" or "pseudo-hemobilia". The cardinal features are episodic gastric pain associated with elevated serum amylase level and passage of melena. HP most commonly follows pseudoaneurysm and pseudocyst formation secondary to acute or chronic pancreatitis (1). Although it has a rare occurrence, HP should be considered in the differential diagnosis of all causes of obscure upper gastrointestinal bleeding in patients with chronic pancreatitis, whether or not it is accompanied by pain. HP is the underlying cause of only 2% of hemobilia patients. It is relatively difficult to diagnose; endovascular embolization and/or pancreatectomy are possible treatment modalities.

We present a rare cause of gastrointestinal bleeding due to rupture of splenic artery aneurysm into pancreatic serous cystadenoma (SCA).

CASE REPORT

A 57-year-old woman presented to our clinic with abdominal pain, tar-colored stool and a decreased level of consciousness. She was admitted to the hospital complaining of hematochezia. She had two episodes of epigastric pain with melena three months before her admission to hospital. Gastroscopic examination revealed grade B reflux esopha-

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gitis and *Helicobacter pylori (H. pylori)* positive antral gastritis. Eradication therapy for *H. pylori* was applied. Colonoscopic examination was normal. The patient complained of increasing malaise and rectal bleeding one week prior to admission to the hospital. She received four units of fresh frozen plasma and four units of packed red blood cell suspension due to low hematocrit (Hct 20%). The patient was admitted to our clinic to investigate the cause of the gastrointestinal bleeding. Her medical history revealed that she had experienced abdominal pain five years ago, and abdominal computerized tomography (CT) scan demonstrated a cystic lesion on the pancreatic tail (Figure 1). During that period, her serum amylase and lipase levels were normal and there were no signs of chronic pancreatitis. Her family history was normal. She did not smoke cigarettes or consume alcohol. Upon physical examination, she was confused and her skin was pale with loss of turgor. Arterial blood pressure was 80/60 mmHg, and pulse was 120 beats/minute and rhythmic. Her respiratory rate was 24 breaths/minute and her breath sound was normal. Abdominal distension was observed with decreased bowel sounds. There was increased sensitivity in all quadrants upon palpation and abdominal guarding was also present. Laboratory results were as follows: Hct 12%, leukocytes 10,300/mm^3, thrombocytes 210,000/mm^3, blood glucose 77 mg/dl, blood urea nitrogen (BUN) 34 mg/dl, creatinine 1.2 mg/dl, sodium 142 mEq/L, potassium 3.9 mEq/L, calcium 7.9 mg/dl, inorganic phosphorus 3.7 mg/dl, aspartate aminotransferase (AST) 13 U/L, alanine aminotransferase (ALT) 12 IU/L, lactate dehydrogenase (LDH) 392 IU/L, alkaline phosphatase (ALP) 66 IU/L, gamma glutamyl transferase (GGT) 8 IU/L, triglyceride 141 mg/dl, cholesterol 160 mg/dl, total bilirubin 0.99 m/dl, direct bilirubin 0.19 mg/dl, total protein 6.1 g/dl, albumin 3.1 g/dl, serum iron levels 27 µg/dl, total iron binding capacity 162 µg/dl, and normal levels of serum and urine amylase. The general condition of the patient was poor. She was in hypovolemic shock with low urine output (50 cc/hour). Central venous pressure catheter was placed to begin replacement of serum physiologic (isotonic NaCl) and blood transfusion. No active bleeding was observed by gastric lavage. Upon rectal lavage, the patient had watery melena. To detect the cause of bleeding, celiac angiography was performed, which revealed two splenic artery aneurysms and medial displacement of the liver (an indirect sign of intraabdominal bleeding) (Figure 2). Abdominal CT scan was performed, and generalized intraabdominal hematoma was detected (Figure 3). The patient underwent laparotomy on the suspicion of splenic artery aneurysm rupture. The laparotomy confirmed that intraabdominal haemorrhagia had occurred due to rupture of splenic artery aneurysm to pancreatic cystic lesion. Distal pancreatectomy and splenectomy were performed (Figure 4). Macroscopic and microscopic examination of the pancreas specimens revealed that the lesion on the pancreatic tail was a SCA. The lining of most parts of the cystic cavity was denuded and single layered cuboidal cell lining was observed in

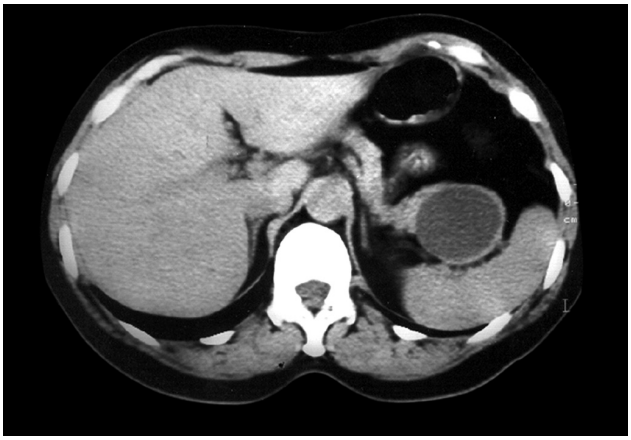

Figure 1. A 4 cm cystic mass arising from the tail of the pancreas. The lesion is well-defined and does not show enhancement on CT after intravenous contrast injection.

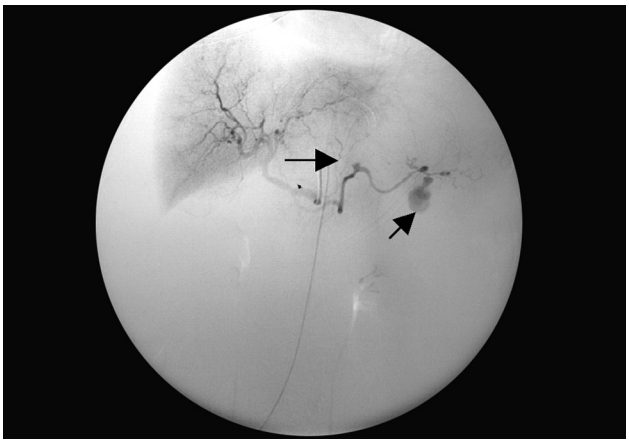

Figure 2. Celiac angiography at the mid-arterial phase showing two aneurysms of the splenic artery. The first is a 1 cm aneurysm arising from the 1/3 proximal segment of the splenic artery superiorly. The second is an irregular fusiform-shaped aneurysm at the level of the splenic hilum approximately 3.5 cm in diameter (bleeding into the pancreatic cystadenoma). Note the displacement of the liver from the right abdominal wall, which is an indirect finding of intraabdominal fluid.

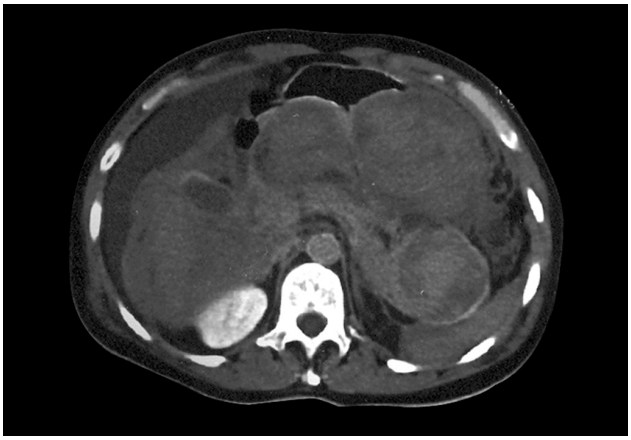

Figure 3. Axial abdominal CT view after intravenous contrast injection. There is free fluid in the perihepatic and peripancreatic areas. The cystic mass arising from the tail of the pancreas is filled with heterogeneous fluid consistent with hematoma. Furthermore, displacement of intestines due to intraabdominal hematoma is seen.

a few areas. Surrounding areas were hemorrhagic (Figure 5a, 5b). Detection of CD34 negative epithelium by multiple cross- section specimens of the cystic lesion confirmed that it was not originated from the aneurysmal vascular lesion. There was no sign of pancreatitis or malignancy. Tumor markers (Ca 19-9, CEA) were in normal range. Postoperative celiac angiography showed no other aneurysmatic structures; postoperative abdominal CT angiography was also normal. She was diagnosed with rupture of a splenic artery aneurysm into pancreatic SCA. After her general health status improved, the patient was discharged from the clinic to be followed on an outpatient basis. During the one-year follow-up period, she experienced only diabetes because of pancreatic resection.

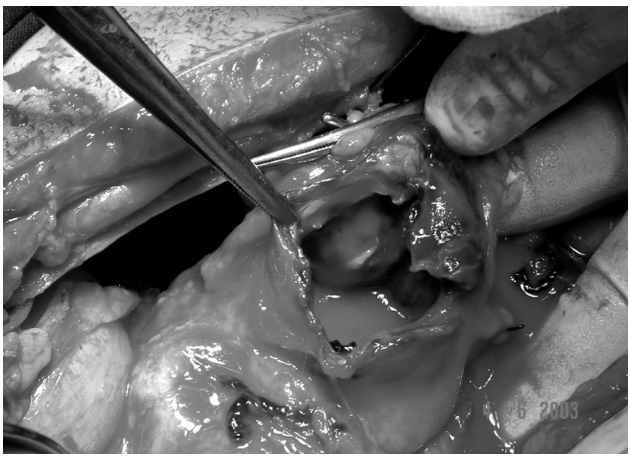

Figure 4. Appearance of active bleeding into the pancreatic cystic lesion during the operation.

DISCUSSION

Hemosuccus pancreaticus is a rare clinical condition with a classical triad of symptoms consisting of recurrent abdominal pain, transient hyperamylasemia and melena. The HP term was first used by Sandblom (2) to define a type of hemobilia in which there is bleeding from the pancreatic duct into the gut via the ampulla of Vater. Sandblom described three cases of pancreatitis in which visceral artery aneurysms ruptured into the pancreatic duct (3). Camishion et al. (4) defined three possible causes of pancreatic duct hemorrhage as follows: (a) rupture of a primary splenic artery pseudoaneurysm to the pancreatic duct; (b) splenic artery pseudoaneurysm rupture into the pancreatic duct because of pancreatitis; and (c) bleeding from a cyst wall in chronic pancreatitis. Here, we report an interesting case admitted to emergency service with hypovolemic shock. She was diagnosed by angiography with intraabdominal bleeding due to rupture of a splenic artery aneurysm into a pancreatic SCA.

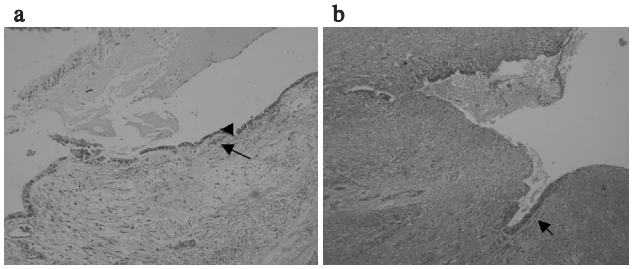

Figure 5a, 5b. Single-layered cuboidal cells lining the cystic cavity and denuded areas (arrow). (Hematoxylin and eosin-H&E, original magnification x100).

Pancreatic SCA is generally asymptomatic and occurs primarily in females. These lesions are well-defined masses radiologically containing multiple septae cysts (<2 mm) with calcifications. Hypervascularization can be seen in these calcifications and septae. However, macrocystic SCA has atypical radiologic appearance and its incidence rate is 10% in all SCAs. These lesions can be unilocular and well limited. Generally, there is no solid component or septae. Peripheral contrast enhancement can be seen after IV contrast agent injection (5-8). However, distinction between SCA, mucinous cystadenoma (MCA) and pseudocyst was not possible in our case five years ago because of low quality CT. Absence of typical calcification (for MCA) and presence of CD 34 negative epithelium

(pseudocyst has no epithelium) suggested that this lesion was a macrocystic SCA, which is rare form of SCA.

Most cases of HP are due to pancreatitis (9). Peripancreatic vessels are affected in 10% of cases of pancreatitis. There are many mechanisms by which the pancreatic and peripancreatic vessels can be involved. Proteolytic enzymes released during the episode of pancreatitis can directly erode the numerous arteries in and around the pancreas, resulting in hemorrhage or formation of a pseudoaneurysm. Pseudoaneurysm may develop within the pancreatic pseudocyst and in some cases rupture into the neighborhood pseudocyst (10). The splenic artery, because of its contiguity with the pancreas, is the vessel most commonly involved in pancreatitis (11). Splenic and superior mesenteric vein thrombosis ensues due to the pressure effect of the inflammatory mass, and bleeding may also occur. The gastroduodenal and pancreaticoduodenal arteries are frequently involved, while the left gastric, hepatic and small intrapancreatic branches are less often implicated. However, there were no signs or symptoms of acute or chronic pancreatitis in our case and there was no pseudocyst.

Another cause of HP is visceral organ vascular malformations. The incidence of splenic artery aneurysm is reported to be 0.98% (in a study containing 195,000 autopsy cases) (12). The etiologic factors of splenic artery aneurysm include atheromycotic emboli, portal hypertension, splenomegaly and trauma. Such primary aneurysms are generally asymptomatic. If rupture occurs, it is usually into the pseudocyst (13, 14). In our case, we detected splenic artery aneurysms, and rupture occurred into the SCA. Distinction between aneurysm and pseudoaneurysm is not possible by angiography. In our case, absence of acute or chronic pancreatitis led us to consider that aneurysms were primary splenic artery aneurysms. However, bleeding from the pancreatic duct has been most commonly attributed to visceral artery pseudoaneurysms (40%) (15). Other unusual causes include pancreatic lithiasis (16, 17), pancreatic tumors (18), and arteriovenous malformations (19), and pancreas divisum with chronic pancreatitis has been reported to be as high as 10% (1). HP may rarely develop with pancreatic cystadenoma and cystadenocarcinoma as precipitating factors (such carcinomas account for only 10% of all pancreatic malignancies) (20). If HP develops with a malignancy, the cause is most probably splenic vein thrombosis or direct invasion of the splenic artery with tumor (18, 21, 22).

Most patients present with episodes of intermittent left upper quadrant or epigastric pain, hematemesis, melena, and a history of pancreatitis. In patients with pancreatitis, a ruptured arterial pseudoaneurysm should be suspected if there is a persistent or rapid increase in abdominal pain, gastrointestinal bleeding with no obvious cause, and associated hemodynamic instability or decreasing Hct. However, some cases are asymptomatic or present with nonspecific symptoms such as abdominal pain and weight loss (23). HP diagnosis in asymptomatic patients is difficult. In our case, there were no symptoms or signs in the patient history compatible with chronic or acute pancreatitis, except epigastric pain upon admission to the hospital. Histological examination also showed no signs of pancreatitis.

Imaging methods are very important in early diagnosis and treatment. Ultrasonography (24), CT, visceral angiography, technetium 99m scintigraphy (25), and endoscopic retrograde cholangiopancreatography (ERCP) are the chief modalities used in the treatment and diagnosis (26). Visceral angiography is the most sensitive investigation for the detection of a visceral artery pseudoaneurysm (17, 27). Visualization of a direct communication with the pancreatic duct has been reported rarely.

The therapy of choice for HP is through invasive methods. Endovascular embolization is the most effective and the safest method of treatment. However, surgery is often the procedure of choice, especially when there is a pancreatitis-related indication for surgery or severe acute intraabdominal bleeding, as in our case (28).

To our knowledge, this is the first reported case of acute bleeding from a visceral vascular malformation (splenic artery aneurysm) into a benign pancreatic cyst (pancreatic SCA) that then caused intraabdominal bleeding.

In conclusion, HP can be seen even in the absence of chronic pancreatitis. Therefore, HP must be considered as a cause of bleeding, and in the case of active bleeding in which the etiology is unconfirmed, celiac angiography must be performed as soon as possible. The diagnosis and treatment of HP can be achieved through cooperation between the medical, surgery, and radiology departments.

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Note from the Editor:

The case described in this report created an interesting controversy between one reviewer and the authors, parts of which we decided as the Editorial Board, to bring to the attention of our valuable readers who can make their own mind on the diagnostic controversy of this case.

September 28, 2006: Reviewer's critique

The subject of this article is a case brought to the hospital in a very severe clinical condition. There was bleeding into the abdomen and the gastrointestinal system (GIS), and it was found that the bleeding was originating from splenic artery aneurysms. A timely diagnosis was made and the patient was referred for a life-saving successful surgical intervention. This is a very important achievement.

However, in pathologies causing such conditions, I am of the opinion that cystic lesions in the pancreatic tail should not be interpreted as serous cystadenoma and that the aneurysm in the splenic artery should not be considered as primary splenic artery aneurysm.

1. Serous cystadenoma of the pancreas is a hypervascular lesion involving solid and cystic structures. Both angiography and contrast computerized tomography (CT) provide special images because this lesion is hypervascular. There is no finding of a vascular lesion in the pancreatic tail in celiac angiography of this case. Pre-ope-

rative CT image (Figure 2) was not sufficient to make an interpretation. In CT section in Figure 5, the lesion is similar to pseudocyst. There are no characteristics of a serous cyst. The only possibility other than pseudocyst is mucinous cystadenoma.

2. It was reported that the patient had abdominal pain five years ago and that a cystic lesion was found in the pancreatic tail according to the CT of the patient at that time. When a lesion is present in the pancreatic tail, neighboring the splenic hilus and therefore associated with the splenic veins for a long period, it is not rational to interpret the aneurysm in the splenic artery as a primary splenic artery aneurysm. Moreover, image of the aneurysm is compatible with pseudoaneurysm.
3. In addition, it is not possible for a mass like serous cystadenoma involving solid structures to cause recurrent wirsungorrhagia into the pancreatic channel from the splenic artery (It was reported that the patient had wirsungorrhagia twice three months ago).
4. Wirsungorrhagia and bleeding into the abdomen most often occur as a complication of pancreatic pseudocysts. It is possible that a pseudocyst associated with the pancreatic channel could cause damage to the neighboring splenic artery wall and lead to a pseudoaneurysm, and hence result in bleeding.
5. It was reported that there was no acute or chronic pancreatitis based on the laboratory and laparotomy analyses of this case. There was also no finding of chronic pancreatitis in the tomographies. However, if the patient had had an acute pancreatitis long ago, it is possible that no findings would be seen in the laparotomy. It was reported in the patient's history that the patient experienced abdominal pain for a while and that a cyst was found in pancreatic tail in the CT made at that time.

For these reasons, this article cannot be published as it is. However, it can be compiled after pathology preparations are revised and necessary arrangements are made.

November 25, 2006: Response from the authors

1. Serous cystadenoma of the pancreas is mostly asymptomatic and more frequent in women. Serous cystadenoma is localized 50% in the pancreatic head. The lesions typically contain well-circumscribed multiple cysts and septa less than 2 mm of diameter and can be calcified in these septa. Both these septa and central/septal calcifications can be hypervascular. When the lesion is small, it can show homogeneous contrasting in CT, but in larger lesions the contrasting may only be seen in septa and solid parts. However this classical information is partially not valid for "macrocystic serous cystadenoma" which constitutes a rare form of serous cystadenoma and responsible for 10% of all pancreatic serous cystadenomas. In macrocystic forms, lesion may be unilocular and well-circumscribed. Mostly they do not contain septation and solid component. After the administration of contrast, like pseudocyst or mucinous cystadenoma, they may either not enhance contrast or show peripheral contrasting. Because they are unilocular, have a formed wall structure, are mainly localized in the pancreatic corpus and tail and have contrasting properties, lesions can take part in dfferential diagnosis together with pseudocyst and mucinous cystadenoma. The differentiation of these pathologies can only be made by fine needle biopsy, examination of aspirate liquid in terms of biochemical and tumor markers and perioperative frozen sampling. Glycogen rich cells and low CEA levels can be observed, but these aspirate samples are insufficient in nearly 50% of the patients, so the diagnosis can mostly be made pathologically. The CT of the case which was performed 5 years ago was not qualified enough in terms of radiology, so it can not make a differentiation between serous/mucinous cystadenoma and pseudocysts. As a result the lesion is not a standard serous cystadenoma or pseudocyst with a calcified wall or mucinous cystadenoma. On the other hand, peripheral calcification which is typical of MVA does not exist. Hence, it is considered as a macrocystic variant of serous cystadenoma.
2. Aneurysm or pseudoaneurysms are terms that can only be identified as pathologic. There is no significant difference between them in terms of angiographic imaging. As we stated, the possibility of the aneurysm in the patient with a cystic lesion detected in the tail of pancreas 5 years ago to become secondary to the pseudocyst (although there is no history of pancreatitis) is high due to the scarcity of primary splenic ar-

tery aneurysm. However, it is unusual for the pseudocyst detected 5 years ago not to cause complications and stay nearly the same size within this period. However, our first diagnosis was the formation of secondary pseudoaneurysm to the pseudocyst until the postoperative pathologic diagnosis.

3. Based on the existing literature, we agree with your opinion which states that the serous cystadenoma may not cause recurrent wirsung hemorrhage. We wrote this case due to its rare nature.
4. One of the most common causes of wirsung hemorrhage and intraabdominal hemorrhage as stated in your paper is the damaging of the near peripancreatic artery by way of enzymatic destruction by a pseudocyst adhering to wirsung duct and rupturing of the developed pseudoaneurysm. However a pancreatic situation that can cause enzymatic destruction is not documented in this case. One of the aneurysms in the splenic artery (larger one) was located in the tail of pancreas and was the bleeding aneurysm ruptured inside the pancreatic microcystic serous adenoma. The other aneurysm was a smaller one at the level of pancreatic corpus and not associated with bleeding. The presence of two aneurysms supports the diagnosis of primary aneurysm. Accordingly, we taught that the pancreatitis to cause the formation of a pseudoaneurysm both in corpus and tail (the distance between them is nearly 10 cm) should be very extensive and show sequel findings (such as calcification, atrophy, etc.).
5. We have some doubts about whether the case had a pancreatic attack or not 5 years ago. However, according to the clinical data and anamnesis of the patient, there was no documented previous pancreatitis attack. Although there is a history of acute pancreatitis, detection of epithelium in the existing cystic lesion in the histopathological examination clearly differentiates it from pseudocyst. In the histopathological examination and biochemical profile in all recent abdominal CT and pancreatectomy materials, there are no findings in favor of acute or chronical pancreatitis.

January 19, 2007: The reviewer asks to see the pathological specimens of the case

April 3, 2007: Critique of the reviewer after examining pathologial specimen

The pathologial preparations requested from the author were examined and they were found to be compatible with serous cyst adenoma. However, the clinical condition of the patient and findings of radiological imaging were found compatible neither with serous cystadenoma nor with primary aneurysm of the splenic artery. In other words, there is incompatibility between the clinical condition of the patient and radiological findings with the pathological findings sent.