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CASE REPORT

Case of Chronic Pancreatic Brucellosis Presenting as *Hemosuccus Pancreaticus*

Angelica C Belo[1], Eugene W Grabowski[2], Chi Zhang[3], David L Longworth[1], David J Desilets[1]

[1]Baystate Medical Center, Tufts University School of Medicine. Springfield, MA, USA.
[2]General Surgery, South Western Vermont Medical Center. Bennington, VT, USA.
[3]North Adams Regional Hospital. North Adams, MA, USA

ABSTRACT

Context *Hemosuccus pancreaticus* is an uncommon cause of upper gastrointestinal bleeding. Chronic infection of the pancreas with Brucellosis causing *hemosuccus pancreaticus* has not been previously reported.

Case report We describe a case of a 75-year-old man presenting with a pancreatic mass and *hemosuccus pancreaticus* secondary to chronic pancreatic brucellosis. Polymerase chain reaction analysis of the pancreatic tissue was positive for brucella after an initial positive serology. ERCP revealed bleeding from the pancreatic duct. Computed tomography scans of the abdomen demonstrated an enlarging pancreatic mass. Endoscopic ultrasound showed a cystic mass in the body of the pancreas. Fine needle aspiration revealed granulomata. Selective mesenteric angiogram failed to reveal the source of bleeding. The patient eventually underwent pancreatic resection with resolution of symptoms.

Conclusion This is the first case of *hemosuccus pancreaticus* due to chronic pancreatic brucellosis reported in medical literature.

INTRODUCTION

Hemorrhage from the pancreatic duct, termed *hemosuccus pancreaticus*, is a rare cause of gastrointestinal bleeding. It may present as massive or obscure gastrointestinal bleeding. In this report, we describe a case of pancreatic mass secondary to chronic brucellosis causing *hemosuccus pancreaticus* in a 75-year-old man.

CASE REPORT

A 75-year-old male dairy farmer presented with recurrent hematemesis. He was born and raised in southern Vermont, where he was a dairy farmer from childhood on a family farm. He recalls having brucella-infected herds in the 1940s and 1950s and drank unpasteurized milk for many years. He was referred in June 2005 with a 7-month history of recurrent idiopathic pancreatitis, anemia, abdominal pain, chills, sweats and weight loss. He never smoked tobacco nor drank alcohol and had no family history of pancreatitis. He had several upper gastrointestinal endoscopies at another hospital which failed to identify a source of bleeding. An ERCP was performed which showed active bleeding from the pancreatic duct consistent with *hemosuccus pancreaticus* (Figure 1). There was also a stricture of the main pancreatic duct at its genu. The biliary ducts

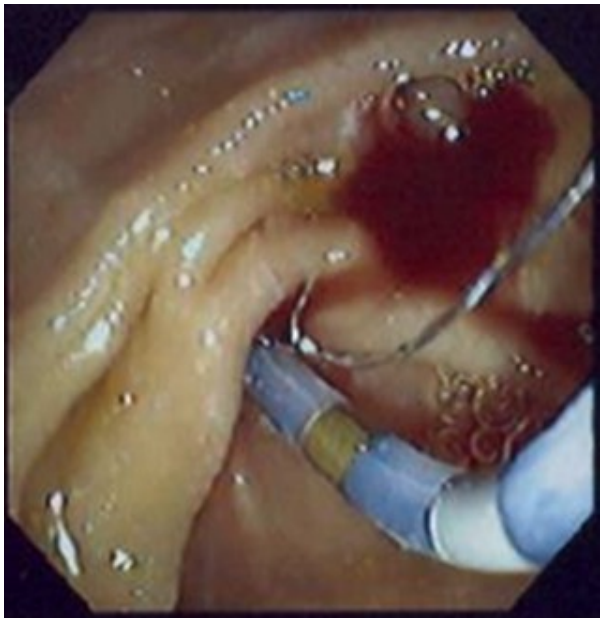

Figure 1. Injection of contrast into the major papilla shows blood gushing out of the minor papilla.

were normal. CT scan of the abdomen demonstrated calcification at the head and tail of the pancreas, an ill-defined hypodense lesion extending from the superior mesenteric artery to the pancreatic calcification at the tail, and two hypodense lesions in the spleen (Figure 2). Subsequent EUS-guided biopsy of this pancreatic lesion revealed granulomatous inflammation. A 5 TU (tuberculin unit) purified protein derivative (PPD) was negative as were serologies for *Histoplasma capsulatum*, *Coccidiodes immitis* and *Blastomyces dermatitidis*. Fungal cultures were obtained because of the granulomatous histopathology on the initial biopsy. We cultured for those classes of organisms that typically produce granulomas. Viral infections do not typically produce this histopathology and thus were not sought. An enzyme immunoassay for *Brucella abortus* on serum was positive, but a follow up indirect fluorescent antibody test against *Brucella abortus* was negative. Because of this discrepancy in serologic findings and the clinical concern about chronic brucellosis, a repeat EUS-guided biopsy of the pancreatic mass was performed. Fresh tissue was transported same day to the Massachusetts State Laboratory (Jamaica Plain, MA, USA), where PCR was positive for *Brucella* species. Subsequent cultures of pancreas, blood and bone marrow failed to grow the organism, however, and histopathology disclosed no visible organisms. On the basis of the history, compatible histopathology and positive PCR, a diagnosis of pancreatic brucellosis was made. Doxycycline and rifampin were initially commenced, but the regimen was changed to doxycycline plus streptomycin upon acquisition of the latter drug. After two weeks of this regimen, he was switched back to doxycycline plus rifampin. His symptoms initially improved, but several weeks later the patient had a further episode of massive hematemesis associated with abdominal pain. Examination revealed marked pallor, tachycardia, and hypotension. His hematocrit was 27% (reference range: 42-52%) and serum lipase was 490 IU/L (reference range: 13-60 IU/L). Upper gastrointestinal endoscopy revealed fresh blood coming from the ampulla of Vater. Selective arteriography of the superior mesenteric artery, celiac axis, splenic artery, and gastroduodenal artery did not reveal a source of bleeding. Repeat CT scan of the abdomen showed an increase in the size of the previously known pancreatic mass. Because of persistent intermittent massive bleeding episodes, the patient underwent subtotal pancreatectomy and splenectomy with complete resolution of his symptoms. At operation, a fluctuant 3.0-3.5

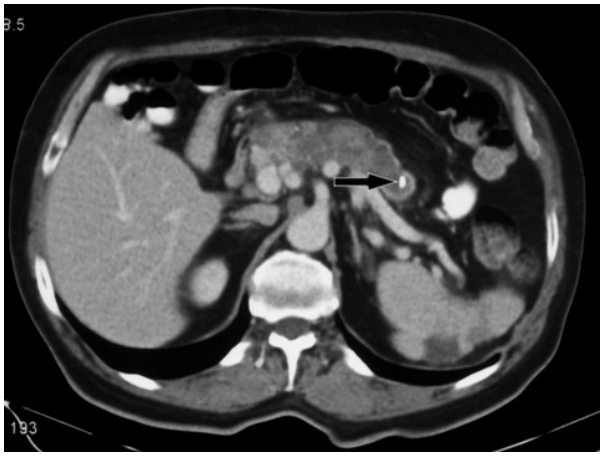

Figure 2. CT scan of the abdomen showing ill-defined hypodense area in the body and tail of pancreas, also two hypodense areas inferiorly in the spleen, pancreatic calcification is present at the tail (arrow).

cm diameter mass was found in the head of the pancreas. Upon opening the specimen a large amount of clotted blood extruded from a dilated pancreatic duct. The mass was an aneurysmally expanded splenic artery with internal clot which had a ruptured wall and blood clot directly communicating with the pancreatic duct (Figure 3). Histology showed chronic pancreatitis and aggregates of lymphocytes. Doxycycline and rifampin were continued postoperatively for 2 weeks. The patient is now an insulin-dependent diabetic and is doing well after one year of follow up.

DISCUSSION

Hemosuccus pancreaticus may pose a significant diagnostic and therapeutic dilemma. The bleeding may range from obscure to massive and life-threatening [1]. Bleeding from the pancreas can occur in the setting of pseudoaneurysm associated with acute or chronic pancreatitis [2], pancreatic pseudocyst, and pancreatic tumor [3]. A bleeding pseudoaneurysm is a rare but serious complication with mortality reaching as high as 40% [4]. The splenic artery is most commonly involved, followed by the gastroduodenal, pancreaticoduodenal, and hepatic arteries [5]. Controversy persists about the optimal therapy for bleeding pancreatic pseudoaneurysm associated with chronic pancreatitis (arteriographic intervention or surgical intervention). Other uncommon reported causes include primary splenic artery aneurysms [6] and hepatic artery aneurysm due to carvenous hemangioma of the liver [7]. Our patient had a history of recurrent hematemesis, anemia, idiopathic pancreatitis, and pancreatic mass due to brucellosis.

His final episode of bleeding was due to erosion of a mycotic pseudoaneurysm of the splenic artery into the pancreatic duct in a setting of chronic brucella infection and inflammation.

Chronic pancreatitis secondary to brucellosis has not been previously described. The pancreas is a rare target organ for brucellosis. We identified two cases of acute pancreatitis secondary to brucellosis in the world's literature [8, 9], and no cases of chronic pancreatitis attributable to this organism. Although we failed to isolate the organism, presumably because of antibiotic therapy, the prior exposure history, positive ELISA, compatible histopathology of the pancreas, and positive PCR on pancreatic tissue for the organism support the diagnosis of brucellosis as the cause of our patient's illness.

Brucellosis is an occupational disease affecting individuals with exposure to the organism through work with livestock or animal products. It is characterized by a variety of often nonspecific clinical symptoms of fever, myalgia, and general malaise, and can manifest with localized disorders of specific organs or tissues [10]. The diagnosis

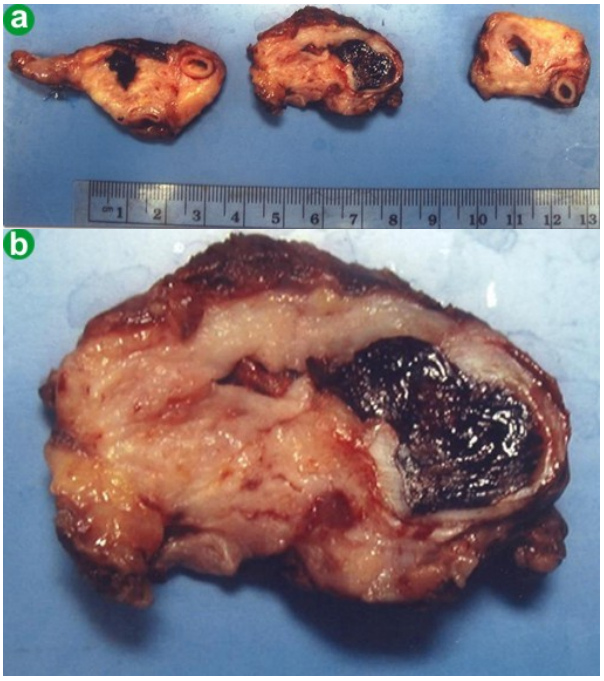

Figure 3. The pancreas cross sections are of the neck, body and tail, proceeding from left to right (**a.**), with the larger image above showing the body just to the right of the midline in the patient showing the mycotic aneurysm of the splenic artery and the fistula into the pancreatic duct. The neck cross section on the left shows the "blood-filled" pancreatic duct in the upper left of the section and the splenic artery to the right.
The body cross section in the middle (also magnified: **b.**) shows the actual pathology, with, again the mycotic aneurysm of the right and the fistula on the left. The final cross section on the right shows the tail of the pancreas with the normal splenic artery lower right and the distal pancreatic duct on the left of the specimen, empty of blood, on the left.

can be established by detection of brucella species in blood cultures or tissue aspirates. Like tuberculosis, brucella species can be notoriously difficult to recover from infected tissues but available serological tests are highly specific and sensitive for detection of brucellosis. The infection is treated with long-term administration of a combination of appropriate antibiotics. Therapeutic failure and relapses, chronic courses and complications such as osteoarticular disease, epididymoorchitis, hepatitis, endocarditis, and neurobrucellosis are characteristic of the disease [11].

Worldwide, brucellosis remains an important disease in people and domesticated animals. This case report further confirms the protean manifestations of brucellosis and reveals previously unreported cause of *hemosuccus pancreaticus* due to chronic pancreatitis associated with chronic brucellosis.

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Correspondence
David J Desilets
Department of Gastroenterology
Baystate Medical Center
Tufts University School of Medicine
Springfield, MA 01199
USA
Phone: +1-413.794.3570
Fax: +1-413.794.8828
E-mail: david.desilets@bhs.org

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