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Case Report

Pancreatic actinomycosis: Possible complication of long-term pancreatic stenting, diagnosed on endoscopic retrograde cholangiopancreatography guided pancreatic duct brushings

Varsha Manucha, Oleh Haluszka

Department of Pathology and Laboratory Medicine, School of Medicine, Temple University, Philadelphia, PA, USA

Abstract

Endoscopic ultrasound guided fine needle aspiration and endoscopic retrograde cholangiopancreatography guided biliary and pancreatic ductal brushings are being used as routine procedures to diagnose mass lesions of the pancreas and stenotic lesions of the biliary ducts and pancreas. We report a rare case of pancreatic actinomycosis in a patient with alcohol-induced chronic pancreatitis with biliary and pancreatic duct strictures and occluded pancreatic stents, diagnosed on pancreatic duct brushings.

Key words

Actinomycosis, brushings, endoscopic retrograde cholangiopancreatography, endoscopic ultrasound, fine needle aspiration, pancreas

Introduction

Pancreatic actinomycosis is a rare disease and can masquerade as a malignant process. A tissue biopsy is often necessary for diagnosis. We report a case where the diagnosis was made on pancreatic duct brushings.

Case Report

A 54-year-old African American male was first referred to our hospital 7 years back for endoscopic ultrasonography (EUS) and endoscopic retrograde cholangiopancreatography (ERCP). He was a presumed case of alcohol-induced chronic pancreatitis. At that time, EUS demonstrated moderate chronic pancreatitis and ERCP revealed a biliary stricture, a stricture in the dorsal duct in the head with upstream dilated pancreatic duct in the pancreatic tail. He underwent minor papilla sphincterotomy and bile duct and pancreatic duct stenting. The patient's biliary stricture resolved with biliary stenting, but he had a persistent nonmalignant stricture in the dorsal pancreatic duct. He refused surgery and elected to be treated with intermittent stenting and very intermittent follow-up. Eleven months following replacement of his minor papilla stent he presented to a local hospital with fever, pain, and jaundice and was referred back for an urgent ERCP. On ERCP, he was noted to have a completely occluded minor papilla pancreatic duct stent [Figure 1] and recurrence of his biliary stricture. The pancreatic duct stricture was persistent from prior studies. Cells for cytology were obtained by brushings. The pancreatic stent was replaced, and new bile duct stent was placed.

Two alcohol fixed smears of the pancreatic duct brushings and the brush tip were received at the cytology section. The Papanicolou stained alcohol fixed smears showed cohesive clusters of ductal epithelial cells with mild focal nuclear enlargement and several neutrophils in the background [Figure 2]. The brush tip was received in a separate tube with 50% alcohol. The cells were scraped off the brush tip into the alcohol, which was then centrifuged at 1000 rpm for 10 min. The button obtained was then transferred to another 50 ml centrifuge tube with 1:2 parts

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Address for correspondence:
Dr. Varsha Manucha, Department of Pathology and Laboratory Medicine, Temple University School of Medicine, Philadelphia, PA, USA.
E-mail: varshamanucha@yahoo.com

specimen and Bouin's solution. The tube was centrifuged at 1000 rpm for 10 min. The supernatant was decanted, and the button obtained was submitted for routine tissue processing to prepare a paraffin-embedded cell block. Hematoxylin and eosin stained paraffin embedded sections of the cell block demonstrated fragments of the ductal epithelium, abundant neutrophils and filamentous bacterial colonies (resembling cotton balls) morphologically consistent with actinomyces species [Figure 3].

He received no treatment other that changing the stents with complete resolution of his symptoms. At 1 month follow-up he was doing well clinically with no pain or fevers. No microbiology studies were performed.

Discussion

Actinomycetes are Gram-positive filamentous bacteria, present as a normal commensal of the upper aerodigestive tract, colon, and female reproductive tract. Infection is usually chronic, slowly progressive suppurative disease resulting in a dense desmoplastic reaction and presenting very often as a mass lesion.[1] If left untreated, sinus tracts may form to the skin or surrounding viscera due to its infiltrative nature.[2] Manifestations of actinomycosis within the cervicofacial and thoracic region are well-known.[2] Abdominal actinomycosis accounts for 20% of reported cases and yet remains largely unknown to most clinicians. Its varied presentations are usually considered to represent malignancy rather than an infective process and were once described as "the most misdiagnosed disease."[1,2] The organs most commonly reported to be involved are intestinal wall (ileocecal region)[1,2] and female pelvic organs including ovaries and vagina.[2,3] Pancreatic involvement is extremely uncommon with very few case reports in the literature, a few diagnosed after surgery and some preoperatively on cytology.[1,4-10] Association of pancreatic actinomycosis with long-standing chronic pancreatitis, treated with pancreatic stents has been reported.[1] Underlying immunosuppression, continuing alcohol consumption and diabetes may also have a role in the pathogenesis.[1,10] Attempts at analysis of occluded stents revealed variable number of bacteria of mixed species, calcium carbonate or calcium oxalate crystal, leukocytes, yeast and plant material scattered in the protein matrix.[1] There is no published data about the degree to which actinomycetes can be isolated from occluded stents. Based on the fact that actinomyces cause pathological infection after preceding mucosal breakdown it can be hypothesized that possibly pancreatic stents facilitates the invasion of actinomycetes into the parenchyma of the pancreas already vulnerable by chronic inflammation.[1] There is no pathognomic radiographic sign of actinomycosis, and ultimately tissue biopsy is necessary for diagnosis. The increased likelihood of pancreatic actinomycosis to present with pain abdomen, fever and a pancreatic mass on computed tomography abdomen and/or biliary duct stricture also increases the chances of lesion to undergo EUS and/or ERCP. Fine needle aspiration (FNA)

Figure 1: Pancreatic duct stricture with stent (endoscopic retrograde cholangiopancreatography image)

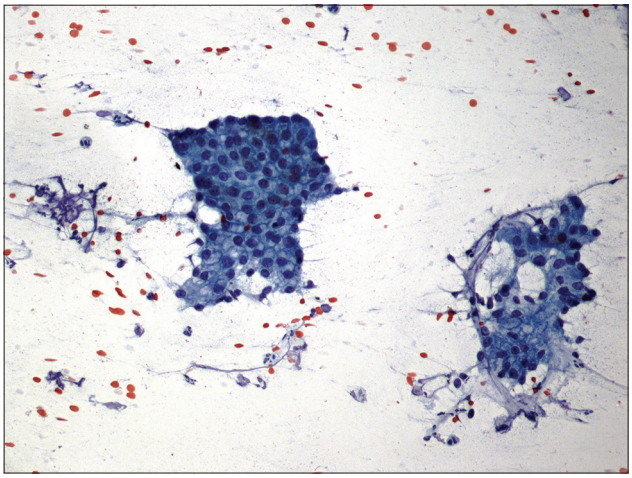

Figure 2: Cohesive clusters of ductal epithelial cells (pancreatic duct brushings, Papanicolou stain, ×200)

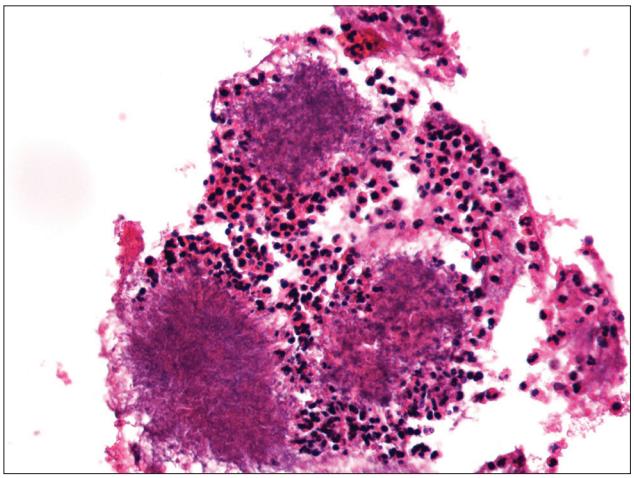

Figure 3: Fragments of benign duct, acute inflammation and filamentous bacterial colonies, consistent with actinomycetes (H and E, ×200)

Table 1: Reported cases of pancreato-biliary actinomycosis diagnosed by EUS-FNA and/or biliary brushings

Author	Clinical history	Presentation	Preliminary diagnosis	Procedure	Follow-up
Harsch *et al.* (2nd patient)[1]	Alcoholism, smoking	Solid mass in pancreatic head, biliary and pancreatic duct strictures (status postbiliary and pancreatic endotherapy)	Neoplastic process	Biliary brushings	Antibiotics
Maestro *et al.*[4]	No significant history	Abdominal pain, occasional fever, solid mass in body of pancreas	Neoplastic process	EUS guided FNA	Antibiotics
Thadani *et al.*[5]	Intrauterine device, no significant clinical history	Jaundice, weight loss, itching, right upper quadrant discomfort, soft tissue mass in gall bladder fossa and pancreatic head	Neoplastic process	EUS-FNA of pancreatic head mass	Antibiotics
Somsouk *et al.*[6]	Alcohol induced chronic pancreatitis	Abdominal pain, mass in pancreatic head	Neoplastic process	EUS-FNA	Antibiotics

EUS-FNA=Endoscopic ultrasound guided fine needle aspiration

has been advocated as a safe tool for a preoperative diagnosis of this pathology in several other sites.[3] No immediate complications following the biopsy are so far reported. After extensive literature search we came across four cases of pancreatic actinomycosis that were diagnosed on cytology, of which three were diagnosed on EUS guided FNA of pancreatic head mass[4-6] and one case was diagnosed on biliary brushings[1] [Table 1]. Preoperative diagnosis in each of the cited cases enabled timely initiation of antibiotics and prevented surgery. If not diagnosed in time, the disease can spread resulting in dissemination infection, abscess and sinus tract formation. The diagnosis can be confirmed by culturing a part of the material obtained through FNA if suspected at the time of onsite adequacy evaluation or by sending the stent for culture analysis. However, special media and transport requirements complicated by overgrowth of concomitant flora make culture of actinomycetes challenging.[1] In contrast, cytological examination (smears/cell block) in an adequately sampled specimen can easily reveal the presence of filamentous bacterial colonies (grains). Presence of associated acute inflammation favors it to be invasive pancreatic actinomycosis and not just a contaminant.

Pancreatic actinomycosis, even though rare, should be suspected in the differential diagnosis of a mass lesion/biliary or pancreatic stricture in a patient with chronic pancreatitis and long-term stent treatment. A EUS guided FNA, or an ERCP guided biliary/pancreatic brushings can aid in early preoperative diagnosis enabling early institution of antibiotics and thereby avoiding surgery besides avoiding the adverse advanced complications of the disease itself.

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