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CASE REPORT | PANCREAS

Spontaneous Infection of a Nonmalignant Intraductal Papillary Mucinous Neoplasm

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ABSTRACT

Intraductal papillary mucinous neoplasm (IPMN) is a pancreatic tumor that originates from the epithelium of the pancreatic duct. Although IPMN cysts can be complicated by infection, this has been reported to involve cysts that have ruptured, fistulized into surrounding organs, undergone malignant transformation, or were recently sampled. We present a 76-year-old man with a history of an IPMN who developed spontaneous cyst infection which was managed with fine-needle aspiration and antibiotics. To the best of our knowledge, this is the first reported case of spontaneous infection of a nonmalignant IPMN. Cyst infection should be considered as a very rare cause of unexplained fevers in patients with history of IPMN.

INTRODUCTION

Intraductal papillary mucinous neoplasm (IPMN) is a pancreatic tumor that originates from the epithelium of the pancreatic duct.[1] Infections arising as a complication of these lesions have been reported; however, infections are uncommon and have been reported to involve peripancreatic spaces or organs as a consequence of fistulizing or ruptured IPMNs.[2] In addition, iatrogenic cases of cyst infection have been reported after endoscopic ultrasound (EUS)-guided fine-needle aspiration, although incidences are rare, occurring in nearly 1% of cases.[3]

CASE REPORT

A 76-year-old man was seen in our clinic for evaluation of a pancreatic cyst. This had been incidentally found on cross-sectional imaging at an outside facility 4 years earlier and was being followed with sequential imaging. He had never undergone endoscopic ultrasonography or sampling of the cyst. His medical history was otherwise notable for coronary artery disease and diabetes mellitus (DM). He had no history of acute pancreatitis, chronic pancreatitis, or recent infections. His family history was unremarkable. He was a former smoker and drank alcohol regularly.

On evaluation, the patient was asymptomatic. He had no abdominal pain or fevers. His white blood cell count, metabolic panel, liver enzymes, lipase, amylase, and carbohydrate antigen 19-9 were all within normal limits. Magnetic resonance cholangiopancreatography was obtained, showing a cyst in the body of the pancreas measuring 2.6 cm in its maximal dimension (Figure 1). Layering was also noted within the cyst. This represented an interval change compared with several previous magnetic resonance cholangiopancreatographies, which had shown a stable simple cyst with no additional concerning features, the last of which was 1 year before the current presentation. An EUS was subsequently performed, redemonstrating a cyst measuring 2.8 $\times$ 1.2 cm in size within the body of the pancreas. Connection to the main pancreatic duct was also noted. Layering debris with increased heterogeneous echogenicity seemed to occupy half of the cyst's content, but no surface nodularity of the cyst was seen. The pancreatic parenchyma seemed normal with no features of acute or chronic pancreatitis. The pancreatic duct and the common bile duct were normal in diameter (Figure 2).

Cyst aspiration was performed, yielding 20 mL of turbid fluid with a pus-like appearance. On analysis, the fluid showed marked acute inflammation, with copious amounts of neutrophils, consistent with pus. The fluid grew *Escherichia coli* (+3) on cultures, had an

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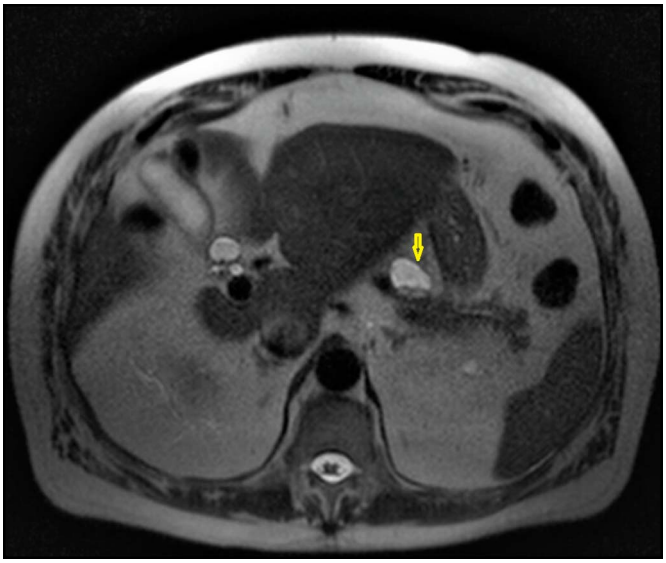

Figure 1. Magnetic resonance cholangiopancreatography showing a 2.6 × 1.2-cm cyst in the body of the pancreas (yellow arrow) with layer noted within the cyst.

amylase level of 2766 U/L, and had a carcinoembryonic antigen (CEA) level of 280 ng/mL. Cytology showed bland columnar epithelial cells without high-grade dysplasia. Mucin immunephenotyping (MUC) was not performed. The patient was believed to have an infected side-branch IPMN and was then treated with a week of oral levofloxacin, at a dose of 750 mg daily. Subsequently, the patient continued to be followed with sequential imaging. An abdominal magnetic resonance imaging obtained 6 months later no longer demonstrated layering within the cyst (Figure 3). The patient remained asymptomatic, with normal lipase, amylase, and inflammatory markers for a year from the initial presentation.

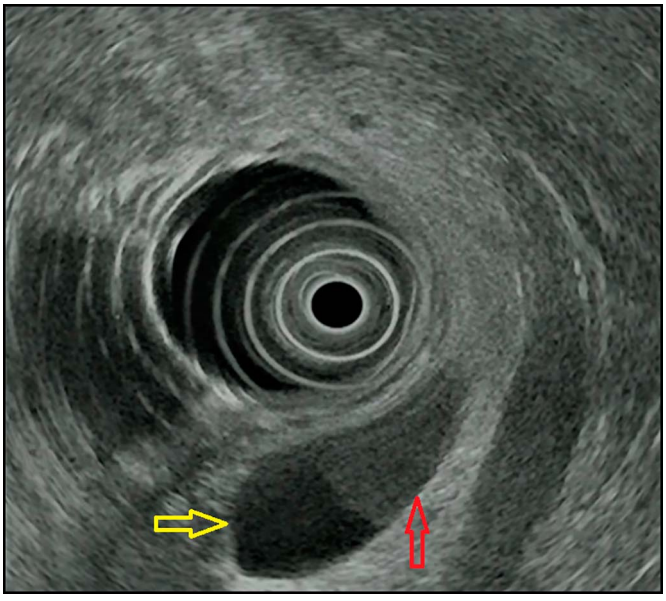

Figure 2. Endoscopic ultrasound showing a 2.8 × 1.25-cm cyst in the body of the pancreas (yellow arrow) with layering noted within the cyst (red arrow).

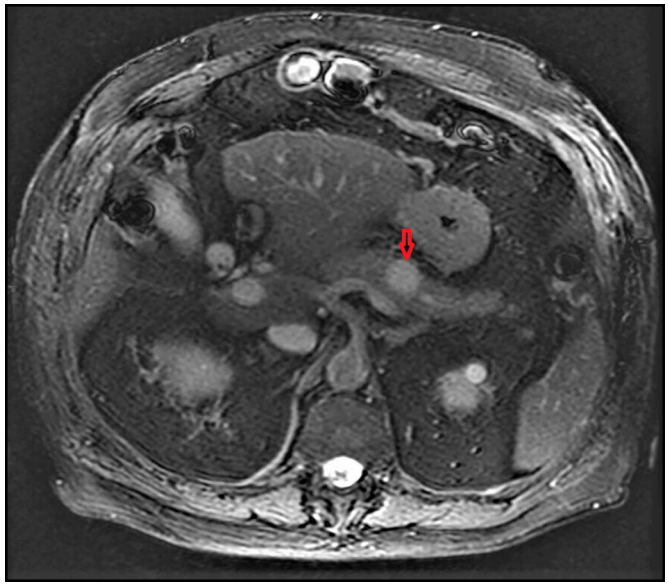

Figure 3. Magnetic resonance imaging of the abdomen showing a 2.8-cm cyst in the body of the pancreas (red arrow) with resolution of the previously noted layering within the cyst.

DISCUSSION

To the best of our knowledge, this is the third case of a spontaneously infected IPMN to be reported, and the first to occur in an asymptomatic patient, in a nondysplastic and nonmalignant lesion.

Two cases of spontaneous IPMN infection have been previously reported.[4] In both cases, patients presented with a sepsis-like picture and were found to have multilocular cystic tumors on imaging. They were managed with cyst drainage of infected fluid that grew enteric organisms, followed by antibiotic therapy.

The mechanism of IPMN infection is unclear. Retrograde bacterial translocation, especially in cases where there is dilation of the papillary orifice because of copious mucin, is a possibility. Other potential mechanisms include the occurrence of acute pancreatitis, a known complication of IPMN,[4] which is associated with bacterial translocation and infection of the pancreatic parenchyma.[5] The patients in both previous cases,[4] as well as our patient, had a history of regular alcohol use or alcohol abuse, both of which are associated with immunosuppression and bacterial translocation.[6] Finally, we note that our patient and one of the previous patients[4] have a history of DM, which has been associated with immunosuppression and spontaneous pancreatic infections.[5]

For the diagnosis of infection, imaging provided an important clue. Fluid layering is an unusual finding in IPMN cysts, and although mucin is commonly observed within these cysts on EUS, it is typically clumped together as a round ball or irregularly shaped cloud-like material within the cyst cavity.

Both previous cases[4] were managed by transpapillary drainage and antibiotics. Although this approach is associated with high rates of pancreatitis and fistulization, approaching 15%,[7]

transpapillary drainage was readily feasible in their cases because the papillary orifices were widely dilated secondary to copious mucin production by the IPMNs. In our case, transmural fluid aspiration was preferred, given its higher safety profile, with rates of complications (most prominently bleeding and infection) being around 1%–2%.[3] Resection was performed in both previous cases[4] because the cysts had undergone dysplastic or malignant transformation. In our case, we did not believe the infection provided a separate indication for resection because the cyst had been stable in size, the patient was asymptomatic, and no dysplastic transformation was noted on cytology.

Spontaneous IPMN infection is an extremely rare occurrence, and presentation may range from absence of symptoms to full blown sepsis, especially in patients with complex cysts, or those whose IPMNs have progressed to dysplasia or carcinoma. Layering within a cyst on imaging secondary to pus formation may provide a clue to the diagnosis, and EUS-guided fine-needle aspiration can provide further diagnostic and therapeutic yields.

DISCLOSURES

Author contributions: A. Ashhab wrote the manuscript and is the article guarantor. A. Podboy wrote the manuscript. S. Lo reviewed and edited the manuscript.

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Informed consent was obtained for this case report.

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