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CASE REPORT

Case Report: Pancreatic adenocarcinoma presenting as acute obstructive suppurative pancreatic ductitis [version 1; peer review: 1 approved, 1 approved with reservations]

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Abstract

Acute obstructive suppurative pancreatic ductitis (AOSPD) is a rare form of infection primarily arising within the pancreatic duct in the setting of chronic pancreatitis. We present a case of AOSPD precipitated by obstructive adenocarcinoma of pancreatic head in an elderly woman with a past medical history of chronic pancreatitis, alcohol use disorder and, advanced dementia, who developed progressive abdominal pain during her hospital admission for urinary tract infection. Endoscopic retrograde cholangiopancreatography (ERCP) with balloon sweep of pus and stent placement resulted in prompt abdominal pain resolution. Our case highlights a rare presentation of AOSPD as a harbinger of pancreatic malignancy.

Keywords

Pancreatic adenocarcinoma, AOSPD, acute suppurative pancreatic ductitis, malignancy, pancreas, chronic pancreatitis, obstruction, sphincterotomy

This article is included in the **Oncology** gateway.

Open Peer Review

Approval Status

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2. **Andreas Minh Luu** ,

Any reports and responses or comments on the article can be found at the end of the article.

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Introduction

AOSPD, first described in 1995, is a rare consequence of chronic pancreatitis characterized by an acute infection of the pancreatic ducts while sparing pancreatic parenchyma. The distinguishing feature is a lack of pseudocyst, abscess, or necrosis formation. Literature remains sparse in the domain of AOSPD, with a recent literature search only mentioning this entity in over 29 cases to date¹⁻¹². Pancreatic malignancy is also known to promote AOSPD by obstructing pancreaticobiliary secretion outflow ensuing in subsequent infection^{2,9}. We describe a case of AOSPD precipitated by underlying pancreatic adenocarcinoma.

Case presentation

A 71-year-old female with a past medical history of chronic alcohol use, chronic pancreatitis and advanced dementia, presented to our tertiary care setup after a fall with confusion, a day after being discharged from an outpatient facility where she had undergone a colonoscopy. Vitals were remarkable for tachycardia (105 beats per minute), blood pressure 90/70 mmHg, temperature of 100.4F and respiratory rate of 18 per minute. On admission, her blood work-up was significant for an elevated white blood count of 12.4k/ μ L with urinalysis, positive for nitrites, elevated white blood cells, and bacteria. Blood and urine cultures were sent as part of workup for suspected sepsis, and the patient was started on ceftriaxone (2 grams) to cover for suspected sepsis secondary to urinary tract infection, which was considered as a cause of fall and altered mental status at presentation. Ceftriaxone was continued as urine cultures returned positive with the growth of *Klebsiella pneumoniae* and *Escherichia coli* with improvement in mentation 24 hours after admission.

However, two days later, the patient developed new onset of progressively worsening, dull epigastric pain with no alleviating or aggravating factors. The pain did not correlate with breathing, position, or food intake. The patient reported that she had for had previous similar abdominal pain episodes six months prior. Magnetic resonance cholangiopancreatography (MRCP) was performed, which showed chronic pancreatitis and a 1 cm calculus in the main pancreatic duct; however, we pursued no intervention. She again had an episode two months prior with elevated white cell count, which responded to a short antibiotic course, but no infection source was found. The patient had no fever or hemodynamic instability. Her labs were significant for AST 156, ALT 182, ALP 949, GGT 2859, Bilirubin 1.1, lipase 68. It was noted that AST, ALT, and ALP were elevated at the previous encounter where she had the first episode of abdominal pain, six months before current admission. Blood cultures at the time of presentation had been negative to date.

Due to epigastric pain, an abdominal ultrasound performed showed a 14mm stone in the pancreatic head with a dilated pancreatic, common hepatic and common bile duct (CBD), measuring 11mm, 9mm, and 11mm respectively (Figure 1 and Figure 2). On day 5, the patient's hemodynamic parameters worsened with a surge in leukocytosis, and thus, transferal to intensive care with a change in antibiotic regimen to vancomycin

Figure 1. 14 mm stone in the pancreatic head with dilated duct.

Figure 2. Dilated pancreatic duct of 11 mm within the head of pancreas.

and piperacillin-tazobactam was made. Abdominal computed tomography (CT) performed to look for a possible abscess revealed a 3cm obstructive mass in the pancreatic head, suspicious for malignancy with dilation of CBD and pancreatic duct (PD) measuring 16mm and 11mm respectively. CT also demonstrated extensive pancreatic desiccation consistent with the diagnosis of chronic calcific pancreatitis.

Endoscopic retrograde cholangiopancreatography (ERCP) was performed for suspected acute cholangitis secondary to obstruction from pancreatic mass, showing high-grade distal CBD and PD stricture. Sphincterotomy was performed first for the pancreatic duct, then the distal CBD and the stricture in CBD and PD, dilated by using a 4 mm $\times$ 4 cm dilating balloon catheter. Upon 4 mm dilation of the PD, a significant

amount of pus with debris and stone fragments were extruded. The dilating balloon catheter was removed and a 9–12 mm injecting below retrieval balloon catheter was used to sweep the PD, yielding more pus, debris, and stone fragments. Brushings were obtained from the PD stricture followed by pediatric forceps biopsy of the CBD stricture to rule out malignancy. Subsequently, a 10 Fr × 9 cm biliary stent was placed into the CBD and a 7 Fr × 7 cm stent in the PD with prompt drainage from both stents consistent with the diagnosis of AOSPD (Figure 3 and Figure 4).

The patient's sepsis resolved after the pancreatobiliary drainage with prompt resolution of leukocytosis, transaminitis, and total

Figure 3. Balloon sweep of pancreatic duct with removal of pus and sludge.

Figure 4. Common bile duct and pancreatic duct stents at the end of endoscopic retrograde cholangiopancreatography.

bilirubin levels. Piperacillin-tazobactam was continued for ten days. Biopsy and brushings confirmed hepatobiliary primary invasive adenocarcinoma. Given the grave prognosis, palliative care was proposed. The patient opted for home hospice care.

Discussion

AOSPD is a very rare entity and its mention in literature is sparse. AOSPD presentation overlaps with cholangitis and this mimicry makes misdiagnosis or non-recognition as a separate clinical entity difficult. AOSPD presentation varies from acute to chronic, the most common symptom being abdominal pain^{1–5,8–12}. Less commonly, it may also present as sepsis or septic shock^{1–4}. However, the presence of infection does not always correlate with clinical manifestation. This was evident from cases of asymptomatic presentation, where lab work uncovered the underlying infection incidentally^{6,7}. Pancreatic imaging plays a pivotal role. The main pancreatic duct diameter was significantly higher in AOSPD with Chronic Pancreatitis (CP) than Acute-on-Chronic Pancreatitis patients¹⁰. AOSPD as the initial presentation of pancreatic malignancy is reported only once before, besides our case¹⁰.

The presence of pancreatic ductal stones precipitated due to pancreatic secretion stasis also serves as a nidus of infection generation leading to septicemia^{1–5}. Besides enteric pathogens, respiratory pathogens have been increasingly reported in association with AOSPD, especially *Klebsiella*^{3,5}. In our case, the pancreatic fluid and urinary cultures grew *Klebsiella* of the same species; however, no bacteremia was present.

The mainstay of AOSPD diagnosis is through ERCP detection of purulent discharge from the main pancreatic duct, seen in the absence of associated lesions like an abscess or pancreatic pseudocyst. MRCP presumptively diagnosed one case by demonstrating pancreatic duct dilation. However, the diagnosis was still confirmed after ERCP lead to symptom resolution².

Treatment is through source control via ERCP drainage and appropriate antibiotic administration. Notably, endoscopic naso-pancreatic drainage (ENPD) was additionally performed following ERCP in one reported case¹¹. At this time, there are no established guidelines for the duration of therapy. Broad-spectrum antibiotic with enteric anaerobic coverage (vancomycin with piperacillin-tazobactam, ceftazidime with gentamicin) given for 7–14 days, is the mainstay of therapy^{1–6}.

AOSPD is a rare and potentially fatal diagnosis that requires expeditious recognition and treatment with referral to a facility with ERCP capabilities and expert infectious disease management. Future reviews and studies should work toward universal guidance in management.

Consent

Written informed consent for publication of clinical details and clinical images was obtained from the patient.

Data availability

All data underlying the results are available as part of the article.

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Andreas Minh Luu

Dear authors,

Thank you very much for the opportunity to review this nicely written manuscript. I would suggest minor revisions prior to acceptance of the manuscript.

First of all, what was the primary underlying cause of the main pancreatic duct dilation? Was it the pancreatic stone, the tumor or even both?

Please add a CT-scan sequence showing the pancreatic head cancer.

Was the stone removed completely?

I am looking forward for your revision.

Is the background of the case's history and progression described in sufficient detail?

Partly

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners?

Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Pancreatic surgery

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Reviewer Report 19 July 2021

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Shehzeen Fatima Memon

The report is well-written and well presented with a firm approach towards the diagnosis from the beginning to the end. Being a rare case, authors have thoroughly covered the appropriate investigations, diagnostic measures and treatments that need to be carried out for pancreatic ductitis presenting in the head of pancreas carcinoma. There are, however, a few grammar mistakes that need to be addressed by the authors.

The case given is of an elderly woman with existing co-morbidities of chronic pancreatitis and alcohol use, presenting to the setup with a fall and the diagnosis of UTI was established. Later on, her new abdominal pain episode and a previous history of a similar episode led to a MRCP that showed a stone in main pancreatic duct. Her liver function tests were elevated after the second episode as well as after the first one six months back. Abdominal U/S now showed a stone at the pancreatic head with dilation of surrounding CBD and pancreatic duct. Leukocytosis helped establish the diagnosis of duct inflammation while ERCP revealed a stricture due to obstruction with the balloon sweep showed pus and debris further strengthening the grounds for the presumptive diagnosis. An appropriate therapy of Piperacillin-tazobactam was started as *Klebsiella* was found in the fluid meanwhile pus drainage was done via balloon catheter and a stent was placed to relieve obstruction. The case has been extensively covered for physicians to benefit from.

Is the background of the case's history and progression described in sufficient detail?

Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Yes

Is sufficient discussion included of the importance of the findings and their relevance to

future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners?

Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Internal Medicine and Neurology

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

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