A Rare Cause of Abdominal Pain in a Patient with Acute Necrotizing Pancreatitis

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Abstract

\textbf{Introduction:} Walled-off necrosis (WON) is a potentially lethal late complication of acute pancreatitis (AP) and occurs in less than 10\% of AP cases. It can be located in or outside the pancreas. When infected, the mortality rate increases and can reach 100\% if the collection is not drained. Its treatment is complex and includes, at the beginning, intravenous antibiotics, which permit sepsis control and a delay in the therapeutic intervention, like drainage. Nowadays, a minimally invasive approach is advised. Depending on the location of the collection, computed tomography (CT)-guided drainage or endoscopic necrosectomy are the primary options, then complemented by surgical necrosectomy if needed. Infected WON of the abdominal wall has been rarely described in the literature and there is no report of any infection with \textit{Citrobacter freundii}. \textbf{Case:} We present the case of a 61-year-old man with necrotizing AP complicated by WON of the left abdominal wall, infected with \textit{Citrobacter freundii} that was successfully treated with CT-guided percutaneous drainage and intravenous antibiotics. \textbf{Conclusion:} Infected WON accounts for considerable mortality and its location in the abdominal wall is rare; it can be treated with antibiotics and CT-guided drainage with no need for further intervention.
choledocholithiasis. Cholangiopancreatography (ERCP), performed due to suspected hypertension and no other relevant medical history, who was admitted with only a few cases being reported in the literature. surgical necrosectomy. Extrapancreatic WON is even rarer, involving percutaneous drainage and endoscopic or surgical necrosectomy. A NPC infetada por Citrobacter freundii. Caso: Aprezentamos o caso de um doente do sexo masculino de 61 anos com uma PA necrotizante complicada por NPC da parede abdominal esquerda, infetada por Citrobacter freundii, que foi tratada com sucesso através de drenagem percutânea guiada por TC e antibioterapia intravenosa. Conclusão: A NPC infetada condiciona mortalidade considerável e a sua localização na parede abdominal é rara; pode ser tratada através de drenagem e drenagem percutânea guiada por TC, sem necessidade de intervenção posterior. © 2017 Sociedade Portuguesa de Gastroenterologia Publicado por S. Karger AG, Basel

Introduction

Walled-off necrosis (WON) is a rare and potentially lethal complication of acute pancreatitis (AP) which can occur in or outside the pancreatic tissue. WON is present in 1–9% of all AP cases [1] and it corresponds to an encapsulated collection of pancreatic and/or peripancreatic necrosis that occurs 4–6 weeks after the onset of necrotizing pancreatitis [2]. If left untreated, the mortality can reach 100% [1], mainly because of secondary infection. Currently, a minimally invasive and step-up approach is advised [3, 4], involving percutaneous drainage and endoscopic or surgical necrosectomy. Extrapancreatic WON is even rarer [1] with only a few cases being reported in the literature.

Case Report

We present the case of a 61-year-old black male, with arterial hypertension and no other relevant medical history, who was admitted to our unit for surveillance after an endoscopic retrograde cholangiopancreatography (ERCP), performed due to suspected choledocholithiasis.

The patient had been submitted to an open cholecystectomy 1 month earlier due to acute gangrenous cholecystitis, with intraoperative cholangiogram revealing a stone in the distal common bile duct (CBD). The stone was extracted, but due to maintained cholestasis, a magnetic resonance cholangiopancreatography was performed, showing a dilated CBD and two foci with 6–7 mm suspected to correspond to choledocholithiasis.

The patient underwent an ERCP with difficult cannulation of an intraduodenal papilla. Following the placement of a guide-wire in Wirsung’s duct, cannulation of the CBD was attempted with the double-wire technique, without success. Infundibulotomy was necessary and the cholangiogram revealed a dilated CBD but no evidence of choledocholithiasis. Biliary sphincterotomy was performed, followed by further exploration of the biliary ducts with a Dormia’s basket and no material came out. These findings were presumed to be in relation with biliary stone migration. At the end of the procedure, a 5-Fr, 7-cm-long pancreatic plastic stent was placed as well as administration of 100 mg rectal indomethacin, for postprocedure pancreatitis prophylaxis.

On the next day, the patient developed complaints of mild epigastric pain and intense pain in the left quadrants of the abdomen, poorly controlled by analgesics, and vomiting. At physical examination, he was conscious, febrile (38 °C), normotensive but poorly controlled by analgesics, and vomiting. At physical examination, he was conscious, febrile (38 °C), normotensive but poorly controlled by analgesics, and vomiting. At this time, a mass in the left flank was noticed (Fig. 1). Abdominal ultrasonography and blood cultures were again both negative. Cholestatic enzymes normalized and this alteration was assumed to have occurred in the context of biliary stone migration. However, due to maintained abdominal pain, fever, and a CRP rise to 32.7 mg/dL 72 h later, an abdominal computed tomography (CT) was asked, showing necrosis of the pancreatic tail, acute necrotic collections of both parietocolic gutters, perisplenic and perihepatic spaces and anterior pararenal fascia, and subcutaneous edema of the left abdominal wall. Urine and blood cultures were both negative.

Towards suspicion of infected necrosis, meropenem 1 g q8h was started. Despite initial improvement, 2 weeks later, the left flank pain worsened, fever came back (with temperatures ranging between 38.3 and 38.5 °C), and inflammatory parameters (IP) rose again (with a maximum CRP of 8 mg/dL). At this time, a mass in the left flank was noticed (Fig. 1). Abdominal ultrasonography and CT were performed and identified a well-delineated heterogeneous subcutaneous collection in the left abdominal wall (9 × 6 cm). Vancomycin was added to meropenem empirically, as urine and blood cultures were again both negative.

Even though there was a favorable response in the beginning, 10 days later, there was a new fever peak, of 38.6 °C, and IP rose again (with a maximum CRP of 7 mg/dL). Abdominal CT showed increase in the left abdominal wall collection size (13 × 7 × 8 cm), corresponding to an extrapancreatic WON (Fig. 2).

The patient was submitted to CT-guided percutaneous drainage of the collection with placement of an 8.5-Fr pig-tail catheter.
Citrobacter freundii was isolated in the purulent fluid, susceptible to piperacillin/tazobactam, which was administered for 14 days. Three-times a day irrigation and washing of the WON with a 0.9% saline solution was performed. The patient reported symptomatic improvement, IP normalized, and the collection regressed (Fig. 3), so the drain was removed 15 days after its insertion and the patient was discharged.

A month later, radiologic imaging confirmed persistent resolution of the collection (Fig. 4).

**Discussion**

Acute necrotizing pancreatitis (ANP) is reported to occur in 5–20% of all episodes of AP [1, 2]. The natural history of intra-/peripancreatic necrosis is variable: it may remain solid or liquefy, remain sterile or become infected, persist or disappear [2].
Local complications of AP should be suspected when there is persistence or recurrence of abdominal pain, increases in organ dysfunction, and/or the development of clinical signs of sepsis (fever or leukocytosis) [2], as was the case in our patient. In the context of ANP, local complications are acute necrotic collections and WON. Both can be intra- and/or extrapancreatic, be located in single or multiple sites, and vary in size; they differ according to the time of development after the onset of ANP – WON being the late complication, after 4–6 weeks of the initial episode – and to morphologic characteristics, related to that time. The term “pancreatic abscess” should be abandoned, according to the 2012 Revised Atlanta Classification [2].

WON occurs in 1–9% of all AP cases [1] and consists of necrotic tissue contained within an enhancing wall of reactive tissue. It is a mature, encapsulated collection of pancreatic and/or peripancreatic necrosis and has a well-defined inflammatory wall [2]. The probability of developing WON can be predicted according to Balthazar and Ranson’s radiographic staging criteria; grade E, the most severe, is defined by the presence of two or more fluid collections or gas within the pancreas or peripancreatic inflammation, and the probability of developing WON is 57% [5], as happened in this case report.

The mortality rate from sterile ANP is 10% and rises to 30% when infection is present [6], and WON, especially when infected, is a life-threatening adverse event of AP.

Another interesting peculiarity of this case was the infectious agent of the WON. Colonic translocation of bacterial flora seems to be the responsible for necrotic pancreatic infection [7], and the most typical organisms are gut flora associated and Candida spp. [8]. Citrobacter freundii are gram-negative bacilli and constitute an infrequent etiology of infection in general [9], responsible for only 0.8% of gram-negative infections and more commonly cause urinary tract infections [10]. The antimicrobial susceptibility varies and normally it can be treated with a single antibiotic [9] as in this case. The mortality rate of Citrobacter spp. infections is 6.8% [11].

Ideally, therapeutic intervention is delayed to allow maturation of WON [12] and antibiotics may facilitate this delay [13]. The PANTER study demonstrated that a step-up approach, starting with CT-guided percutaneous drain placement, was superior to up-front open surgical necrosectomy, demonstrating the value of a minimally invasive approach [4]. It is of note that 35% of patients who established necrotic collections did not require any further intervention over simple small-diameter percutaneous catheter drainage [4]. However, they often fail drainage and ultimately require further intervention, either endoscopic or surgical. A “gold-standard” minimally invasive management algorithm would take into account the clinical condition of the patient, anatomical location of the collection, and expertise in the techniques. Many patients may benefit from the use of a multimodal approach with more than one technique [13]. In general, lateral collections and those extending behind the colon are usually better approached from the left or right flank whereas those medial collections where a percutaneous route is compromised by overlying bowel, spleen, or liver, may be better approached endoscopically [13].

In the reported case, an antibiotic known to penetrate the necrotic pancreatic parenchyma (meropenem, a carbapenem) was started as there was no clinical improvement within the first 2 weeks and as soon as the patient showed signs of sepsis, in order to allow necrosis organization. We pursued the step-up approach and used the percutaneous route as he had a lateral WON, in the left flank, and in this case, it was sufficient to reach therapeutic success.

**Statement of Ethics**

Informed consent of the patient was obtained.

**Disclosure Statement**

The authors have no conflicts of interest to declare.
References