CASE REPORT

Multiple Pancreaticocolonic Fistulas Involving the Transverse and Descending Colon with Multiple Walled-Off Pancreatic Necroses: Reporting of a Complex Case Treated Surgically

Khaled E Elshaar¹, Hamada A Alwakeal¹, Alaa A Hakami¹, Ahmed M Osman², Laila H AbuAleid¹, Mohammed A Abdulmughni³, Shaima A Maghadi¹

¹Department of General Surgery, ²Intensive Care Unit, and ³Department of Radiology, King Fahd Central Hospital, Jazan, Saudi Arabia

ABSTRACT

Context Pancreaticocolonic fistula is a rare and potentially critical complication of necrotizing pancreatitis. We report a complex case of multiple Pancreaticocolonic fistulas that was successfully treated with extended left hemicolectomy. **Case report** A forty-three-years-old male patient, presented in our emergency department with epigastric pain, vomiting, diarrhea and weight loss for 4 weeks duration, with past history of acute gall stone pancreatitis 10 weeks earlier. Contrast enhanced computed tomography abdomen showed multiple Walled-off Pancreatic Necrosis in the peripancreatic, right paracolic and left paracolic regions up to left inguinal region with extensive Pneumoretroperitoneum. The periduodenal collection caused duodenal compression. Laparotomy done for pancreatic necrosectomy, relieving the duodenal compression, and drainage of all collections. We noticed multiple Pancreaticocolonic fistulas, 5 in numbers, between the transverse and descending colon and their neighboring collections, extended left hemicolectomy done. Multiple abdominal drains had been put for continuous postoperative irrigation. The patient discharged home but after 3 months of a hectic post-operative course. **Discussion** To the best of our knowledge this is the first case to be reported in the literature with multiple walled-off pancreatic necrosis associated with multiple Pancreaticocolonic fistulas (5 in Number) in both transverse and descending colon. **Conclusion** Necrotizing pancreatitis is a devastating disease, the presence of Pneumoretroperitoneum does not essentially only point to infected necrosis, but the possibility of Pancreaticocolonic fistula should always be kept in mind and searched for the diagnosis of pancreaticocolonic fistulas.

INTRODUCTION

Colonic fistulas occur in 3%–10% of patients with severe acute pancreatitis [1]. An air pocket in a necrotic area of the pancreas usually indicates that infected necrosis is present and/ or there is a fistula to the gastrointestinal tract. The root of the mesocolon serves as a potential route for spread of inflammatory mediators to the colonic wall. This inflammation may lead to thrombosis of mesenteric vessels and subsequently to necrosis of the colonic wall [2]. The consequences of a colonic fistula may be more severe than those of fistulas at other sites of gastrointestinal tract because of the heavy load of multiple organisms [3]. We report a complex case of multiple pancreaticocolonic fistulas that was successfully treated with extended left hemicolectomy along with pancreatic necrosectomy, drainage of all Walled-off Pancreatic Necrosis collections.

Received April 21th, 2018 - Accepted June 05th, 2018 **Keywords** Pancreatic Fistula; Pancreatitis, Acute Necrotizing; Retropneumoperitoneum **Abbreviation** PCFs pancreaticocolonic fistulas; WOPN walled-off pancreatic necrosis **Correspondence** Khaled E Elshaar King Fahd Central Hospital Jazan 45196, B.O. 8123 Saudi Arabia **Tel** +966501479918 **E-mail** khaledshaar2001@yahoo.com

CASE REPORT

A forty-three-years-old male patient presented to an outside hospital, 10 weeks before he presented to us, with biliary pancreatitis, for which he was admitted, at that time his laboratory findings showed, leukocytosis $(12.3 \times 10^9/L)$, Hemoglobin 8 g/dL, HCT 25%, ESR 60 mm/H, CRP >20 mg/dL (N: 0-0.3), S. Amylase 475 U/L (Figure 1).



Figure 1. Acute Pancreatitis with Peripancreatic Collection.

The treating physician referred the patient to another institution where he had been admitted in ICU as the patient developed Adult Respiratory Distress Syndrome (ARDS) secondary to sepsis, and kept on ventilator for 12 days, after which he was discharged to general ward. Gradually, he tolerated fat free diet. Before the patient discharge (after 6 weeks of admission), CECT done for him, **Figure 2** showed WOPN along with paracolic collection and there was no Pneumoretroperitoneum. As the patient was asymptomatic at that time, no any form of interventions had been done for the collections by his treating physician.

After discharge by 4 weeks, the patient presented in our emergency department with dull aching epigastric pain, radiated to the back associated with anorexia and repeated vomiting and intolerance to food, with recent loss of about 25 kg of his weight.

His most distressing complaint was frequent diarrhea which was purulent, yellowish in color with offensive odor, about 5 times per day. No history of fever or night sweating.

No past history of chronic illness but he had had gall stones which had not been treated yet.

No past history of drugs or alcohol intake, and no history of surgical intervention before.

On examination; the patient looked ill, tachypnic, dehydrated and pale, but was not jaundiced

Temp 37.2, P 125 b/m, RR 28/m, BP 127/75 mmHg, Weight 79 kg (it was 125 kg before), Height: 175 cm. The abdomen was distended, there was an ill-defined tender epigastric mass about 20×20 cm which represented the pancreatic phlegmon. The rest of the abdomen was soft and lax. Hernia orifices were intact. Digital rectal examination revealed yellowish colored loose and offensive stool without blood. Lower limbs were edematous, more in the left side, but not tender. **Table 1** showed the laboratory data on admission.

CT chest, showed evidence of moderate left pleural effusion. CT abdomen and pelvis with IV and oral contrast showed WOPN (Figure 3). The peripancreatic collection caused compression and narrowing of the second part of duodenum. The midpart of transverse colon was collapsed and closely related to the large preipancreatic fluid collection. There was extensive Pneumoretroperitoneum within a huge retroperitoneal septated and multiloculated collection in the lesser sac, which extended to left and right subhepatic and left paracolic gutter up to left inguinal region (Figure 4).



Figure 2. CECT Abdomen Sagittal view; Walled-off Pancreatic Necrosis (WOPN), with Paracolic Collection.

 Table 1. Laboratory Investigations on Admission.

Lab. Investigation	Value	Normal Range
HGB	7.3 g/dL	13-18
WBC	8×10^9/L	4.5-10
НСТ	23%	40-45
PLATELET COUNT	181×10^9/L	150-400
GLUCOSE RANDOM	7.7 MMOL/L	3.9-9.4
РТТ	33.2 Sec.	25-35
INR	1.3	0.8-1.2
ESR	120 mm/h	Feb-20
BUN	4 mmol/L	2.6-6.4
SODIUM	135 mmol/L	135-153
CREATINNE	50 mmol/L	50-115
POTASSIUM	4.7 mmol/L	3.5-5.3
MAGNESIUM	0.81 mmol/L	0.74-1
PHOSPHORUS	0.71 mmol/L	0.81-1.58
CALCIUM	1.7 mmol/L	2.2-2.62
T.BILIRUBIN	10.4 UMOL/L	0-17
CONJUGATED BILIRUBIN	0.3 UMOL/L	0.6
ALKALINE PHOSPHATASE	288 U/L	50-137
AST	23 U/L	15-37
ALT	15.9 U/L	Oct-50
AMYLASE	28 U/L	25-115
LIPASE	12 U/L	13-60
PCT (Procalcitonin)	Not Available	



Figure 3. Axial CECT Abdomen shows Necrotizing Pancreatitis with Extensive Pneumoretroperitoneum.

These extensive air foci were supposed to be due to either superadded infection or the presence of PCF.

Lower limbs Duplex US showed left superficial femoral and popliteal veins thrombosis so, IVC catheter was inserted. The patient was admitted to the ICU and parenteral nutrition started.

As the patient was in SIRS, and he had element of gastric outlet obstruction along with extensive pneumoretroperitoneum, within non amenable multiseptated collections for percutaneous drainage, we had prepared him for exploratory laparotomy aiming for pancreatic necrosectomy, drainage of all collections, relieving the gastric outlet compression and tackling the possible colonic fistulas.

Operative Notes

Upon entry of the peritoneal cavity, there was a big phlegmon in the region of the lesser sac, entry of the lesser sac was quite difficult, a gush of foul smelled thick pus came out, the pancreatic necrosis was already well demarcated, necrosectomy done **(Figure 5)**. Opening of all sub hepatic and paracolic collections with removal of all necrotic tissues from within the collections done. Total amount of pus aspirated about 3 liters.

Upon inspection of the transverse and descending colon which looked dark in color and stenosed, there were 5 fistulas, 2 in numbers between the transverse colon and the lesser sac collection (Figure 6), and 3 fistulas between the descending colon and its corresponding left paracolic collection (Figure 7). So extended left hemicolectomy done with side to side colonic anastomosis, along with protecting loop ileostomy. The sizes of the fistulas ranged between 1 to 2.5 cm in diameter.

Subtotal cholecystectomy done as there was extensive adhesions, in the triangle of Calot. Four drains were inserted, 2 in the region of pancreatic remnant, for planned post operative saline irrigation, closure done but the skin left open.

The patient shifted back to ICU, kept on total parenteral nutrition. Cultures of pus and pancreatic necrosis showed E.Coli and Enterococcus faecium, so appropriate antibiotics started along with antifungal agent.

Continuous saline irrigation was started as the upper drains brought remnant of necrotic tissues. Irrigation stopped after couple of weeks.

Postoperative course was hectic, as the patient developed the following complications:-

- 1. Wound infection, treated with frequent dressing.
- 2. Upper limb DVT involved left internal jugular, subclavian and cephalic veins, although he was on heparin.
- 3. HIT (Heparin induced thrombocytopenia), we stopped heparin.
- 4. Right lower lobar pneumonia, resolved later on.
- 5. Biliary leak **(Figure 8)** as the gall bladder stump gave way due to slipped CBD stone, for which ERCP and sphinterotomy done. The fistula closed later on.
- 6. Multiple splenic infarctions (Figure 9), resolved later on, evidenced by MRI abdomen.
- 7. Prolonged intubation, invited for percutaneous tracheostomy.

The patient discharged from the ICU after 8 weeks to general ward.

Follow up CT abdomen done after 8 weeks of admission showed complete resolution of the inflammatory process of the pancreas, as well as all the loculated collection.



Figure 4. Abdominal CECT Coronal View shows Walled-off Paracolic Collections with Extensive Air Foci.



Figure 5. Necrosed Pancreatic and Parapancreatic Tissues.



Figure 6. The 5 Instruments point to The 5 fistulas; 2 in The Transverse Colon and 3 in The Descending colon.

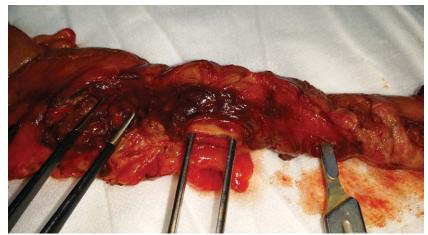


Figure 7. Close-up View of the 3 Fistulas in Left Colon.



Figure 8. MRCP shows Biliary Leak.

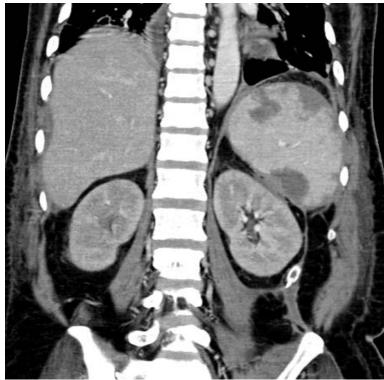


Figure 9. CECT Abdomen shows Multiple Splenic Infarctions

3 weeks later the patient discharged home on warfarin.

The loop ileostomy had been taken down later along with excision of the remnant of the gall bladder to avoid stone formation in the future.

DISCUSSION

Acute necrotizing pancreatitis is diagnosed when more than 30% of the gland is affected by necrosis and accounts for 5% to 10% of pancreatitis cases [4]

PCFs occur in 3%–10% of patients with severe acute pancreatitis [1]. An air pocket in a necrotic area of the pancreas usually indicates that infected necrosis is present and/or there is a fistula to the gastrointestinal tract [2].

In the current literature, there are several theories concerning colonic involvement in the course of acute necrotizing pancreatitis the most important mechanism is the spread of pancreatic enzymes and arising necrosis of surrounding tissues [5, 6].

The root of the mesocolon serves as a potential route for spread of inflammatory mediators to the colonic wall. This inflammation may lead to thrombosis of mesenteric vessels and subsequently to necrosis of the colonic wall with subsequent fistula formation [2].

PCFs can also be an adverse complication of interventional treatment of acute necrotizing pancreatitis, such as percutaneous drainage of pancreatic fluid collections [7, 8].

Typically, the clinical manifestations of PCF include diarrhea, hematochezia, and fever. Less frequently, however, PCF may result in a large bowel obstruction, which accounts for less than 1% of patients with PCF [1, 9,

10]. Mateusz *et al.* [11] reported symptoms likely related to the presence of PCF only in 12 out of 21 patients (57.14%). Melena was noted in seven patients, while hematochezia with diarrhea was observed only in three patients.

In our patient, however, the most distressing complaint was the frequent passage of foul smelled diarrhea but there was no hematochezia or fever.

Unless kept in mind, the diagnosis of PCF is considered to be difficult due to unspecified symptoms [10, 12, 13, 14].

Conventional CT is not considered a successful method in demonstrating pancreatico-colonic fistulas and its sensitivity is low as compared to ERCP [15]. The most probable reason for its low sensitivity is the failure of the oral contrast material to induce luminal distension and pressure to fill the fistulous tracts. The sensitivity of the CT in detecting fistulas can be increased by using rectally administered contrast material [16].

Although in our case we had done preoperative CT using rectal water-soluble contrast media we were not able to directly detect the fistulous tracts, may be due to the high pressure in the collection interfering with backward flow of the contrast from the lumen of the colon back to the collections. The only the indirect evidence for the presence of PCF was the presence of extensive pneumoretroperitoneum.

ERCP is considered the best modality for the detection of pancreatico-colonic fistulas [15].

In one study, ERCP was able to discover the presence of PCF in all studied patients with WOPN and PCF [11]. The advantage of ERCP over other imaging modalities is the added benefit of pancreatic stent placement in order to bridge the disruption site [17, 18]. In our case, however we did not ask for ERCP as a diagnostic tool, first as the patient was not jaundiced, second the patient was just started on heparin drip for the acute lower limb DVT, thirdly we concerned about the possible added complications of ERCP to the already complex presentation of the patient.

The diagnosis on infected WOPN is crucial for early institution of antibiotics and further interventions as needed. CT findings suggestive of an infection include the presence of extra-luminal gas, but still some authorities believe that a definitive diagnosis can only be confirmed when a percutaneous image guided or endoscopic ultrasound (EUS)-guided fine needle aspiration (FNA) is positive for bacterial/fungal stain or culture [19].

In our case, infected WOPN was suspected indirectly by the presence of extensive air foci within all collections, and confirmed by the positive cultures which showed E.Coli and Enterococcus faecium.

While sterile necrosis is associated with 5 % to 10 % mortality rate, the mortality rate increases to 20%–30% when necrosis becomes infected [20, 21, 22]. Thus, early recognition and institution of appropriate therapy is necessary.

Two thirds of necrotic pancreatic collections are sterile and will resolve with conservative management, while the remainder will become infected and will require further intervention [23].

An infected pancreatic fluid collection dictates drainage. Endoscopic drainage should be preferred over radiological one whenever possible due to better drainage and decreased risk of percutaneous fistula, and over surgery because of high morbidity and mortality of the latter [24].

Accepted indications for surgical intervention of WOPN include proven infected necrosis, clinical deterioration, or persistent symptoms due to complications of pancreatic infection.

Our patient, presented to us in sepsis manifested by features of Systemic Inflammatory Response Syndrome (SIRS), with Pulse 125 b/m and Respiratory Rate of 28 /m, beside the proved positive culture from the collections which grown *E. Coli* and *E. Faecium*, the source of sepsis.

There is a general consensus to delay intervention to at least 3-4 weeks after onset of disease and preferably as late as is feasible [25, 26], to allow better demarcation of the necrosis and to avoid unnecessary removal of normal pancreatic tissues which can cause post operative endocrine and exocrine insufficiency.

Open necrosectomy used to be the standard approach for treating infected pancreatic necrosis. This approach is associated with high morbidity and mortality rates (up to 95% and 25%, respectively), as well as debilitating complications such as entero-cutaneous and pancreaticocutaneous fistula formation [27, 28].

However, over the last decade, a variety of minimally invasive interventions for the treatment of acute necrotizing pancreatitis have been introduced as alternatives to the traditional open necrosectomy [29]. Laparoscopic approaches for pancreatic necrosectomy have proved efficacy with a survival rate as high as 85% [30, 31], and is associated with a lower incidence of new onset organ failure post-operatively when compared to the open approach [32, 33]. The current technique for laparoscopic necrosectomy is using a retrogastric transmesocolic or retroperitoneal approach to the lesser sac [34]. Another alternative after the placement of the percutaneous drain would be a videoscopic-assisted retroperitoneal debridement (VARD), where a small 5 cm subcostal incision is made to allow larger pieces of solid debris to be removed.

The PANTER trial (PAncreatitis, Necrosectomy versus sTEp up appRoach) assigned patients with pancreatic necrosis and infected necrosis to either primary open necrosectomy or a step up approach, where a percutaneous drain was initially placed followed by minimally invasive retroperitoneal necrosectomy when needed. It showed that a minimally invasive step up approach was associated with a lower rate of major complications and death when compared to open necrosectomy [35]. The step up approach has proven its efficacy since most necrotic collections are percutaneously accessible and large pieces of necrosis can be removed; however, there is risk of developing a chronic pancreatico-cutaneous fistula with this approach [36].

However, till date, there are no established guidelines for the treatment of WOPN with colonic fistula.

There are recent reports of several cases of PCF have been successfully treated by endoscopic interventions, such as pancreatic stent, [37] endoscopic clip with fibrin glue, [38, 39] and over-the-scope clip system. While Renteln *et al.* [40] reported upon the efficacy of conventional endoscopic clips for the closure of small colonic perforations, endoscopic clips may not be sufficient for the closure of larger perforations [41].

Mateusz *et al.* [11], studied the efficiency and safety of endoscopic treatment in 21 patients with WOPN complicated with PCF, where, transmural, necrogastrostomy or necroduodenostomy, drainage was attempted in all patients with symptomatic WOPN, but drainage was not performed if the distance between the wall of the fluid collection and the gastrointestinal wall exceeded 15 mm.

The place of fistulotomy was chosen under EUS guidance. Fistulotomy was performed on the top of the largest protuberance of the necrotic collection into the gastrointestinal wall. The opening between the lumen of the gastrointestinal tract and the lumen of the necrotic collection was widened with the use of a bougie dilator. A nasocystic drain and several double-pigtail or 10 French

stents were inserted into the cavity lumen of the collection, through which postproceural saline irrigation of the necrotic collection was done.

Complete therapeutic success of WOPN complicated with PCF was achieved in 16 of the 21 patients (76.19%), while the closure of PCF was confirmed by CECT imaging in 17 of the 21 patients (80.95%), The recurrence of WOPN was observed in six of the 21 (28.57%) patients during follow-up, but no PCFs were found in any of the patients with recurrence of pancreatic fluid collection.

In our case, however, we have taken the decision for surgical intervention. First, as the patient had multiseptated multiple collections in the lesser sac, right and left paracolic gutter up to the left inguinal region, which were not amenable to percutaneous drainage, beside, we don't have the facility of endoscopic drainage. Second, we couldn't exclude the existence of the multiple PCFs preoperatively. Thirdly, the patient could not tolerate orally with repeated vomiting, as he had an element of gastric outlet obstruction as the duodenum was compressed by the peripancreatic collection.

Till date, we don't know whether the endoscopic drainage with or without over the scope clipping of the multiple large fistulas (more than 10 mm) could succeed with our patient or not.

Shuhei *et al.* [42] treated one patient with solitary PCF at the splenic flexure of the transverse colon after Endoscopic Ultrasound-Guided Cyst Drainage for Pancreatic Pseudocyst, surgically with a diverting colostomy and abscess drainage. After the surgery, the patient's condition improved and was discharged on the 37th day.

Although, In our patient the option of extended left hemicolectomy with diverting ileostomy was an aggressive option, we have chosen it as we doubted the healing of these multiple wide fistulas if we did only drainage with diverting ileostomy or colostomy and to the best of our knowledge we couldn't find a similar case in the literature with multiple large PCFs treated conservatively or with other minimally invasive or surgical approaches.

CONCLUSION

Despite the aggressive surgical strategy and number of following complications, our patient discharged home safely, and on follow up, he had no recurrence of the WOPN or the PCFs.

Although PCF could be diagnosed during ERCP, in our humble opinion, the presence of PCF should be suspected in cases of WOPN with recent history of persistent diarrhea, the presence of air foci within the collection, the isolation of Enterococcus faecium or E- faecalis from WOPN aspirate, and if open drainage and necrosectomy was decided it would be wise to check the nearby colon for the presence of PCF.

Conflicts of Interest

There are no conflicts of interest.

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