



Pseudoaneurysm of the cystic artery after acute necrotizing pancreatitis



Ann Ital Chir, 2022; 11 - May 30
pii: S2239253X22038026
Online Epub

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A 35-year-old woman, chronic alcoholic, was admitted for an attack of acute, necrotizing pancreatitis. Antibiotics and percutaneous drainage failed to control the septic status and the pancreatic collection. Open surgery allowed a successful necrosectomy and drainage. However, a control CT scan before removal of drains showed a 1 cm diameter pseudoaneurysm of the cystic artery, not present at previous abdominal imaging. A redo laparotomy was performed followed by cholecystectomy with en bloc resection of the pseudoaneurysm and a second look of the peripancreatic area. The patient made an uneventful recovery and was discharged on postoperative day 5. Pseudoaneurysms of the cystic artery after acute necrotizing pancreatitis are very rare. Percutaneous embolization is effective in controlling the pseudoaneurysm, but requires subsequent cholecystectomy within a short delay, due to the risk of gangrene of the gallbladder requiring a further, emergency surgical treatment. Open resection of the pseudoaneurysm en bloc with cholecystectomy appears, therefore, an appropriate treatment of this rare condition.

KEY WORDS: Cystic artery, Pancreatitis, Pseudoaneurysm

Introduction

Pseudoaneurysm of the cystic artery is a very rare condition. As for any pseudoaneurysm its pathophysiology consists of a weakening of adventitial layer either for a mechanical/traumatic, chemical or inflammatory injury. Cholecystitis and iatrogenic injuries are among its most frequent causes¹⁻⁸. During cholecystitis, inflammatory response around the gallbladder wall usually leads to thrombosis of the cystic vessels⁹, nonetheless in rare cases the inflammatory response around the gallbladder wall may lead to adventitial erosion and pseudoaneurysmal dilatation¹⁰⁻¹². Pseudoaneurysms following cholecystitis

may rupture and bleed in the gallbladder lumen with consequent hemobilia clinically resulting in the Quinke's triad (jaundice, melena and upper abdominal pain)^{1,8,13}, upper gastrointestinal bleeding⁴ or even in rectal bleeding, as a consequence of cholecystocolic fistula^{5,14}. Free bleeding in the peritoneal cavity leads to severe anemia, hemoperitoneum and hemorrhagic shock^{1,8}.

Iatrogenic causes include either direct injuries during laparoscopic cholecystectomy or delayed mechanical injuries related to erosion by a surgical clip⁸, mechanical injuries during endoscopic retrograde cholangiopancreatography (ERCP)¹⁵, liver biopsy and liver transplant⁷, arterial erosion by carcinoma of the gallbladder¹⁶, arteriovenous malformations⁷.

Although massive bleeding as a major complication in the setting of acute, necrotic pancreatitis, due to arterial erosion by pancreatic lytic enzymes and intense inflammatory response is well known, cystic artery pseudoaneurysm in the background of this condition is extremely rare and, to our knowledge, has been reported in two cases so far^{17,18}. Due to its rarity and in the underlying setting of necrotic pancreatitis, the optimal treatment of cystic artery pseudoaneurysm is not standardized and may be extremely challenging¹⁸. We report

Pervenuto in Redazione Marzo 2022. Accettato per la pubblicazione Aprile 2022

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one additional case of cystic artery pseudoaneurysm following acute, necrotizing pancreatitis, treated by artery ligation and resection en-bloc with cholecystectomy through an open surgical access, in order to contribute to the definition of an optimal treatment of this rare condition.

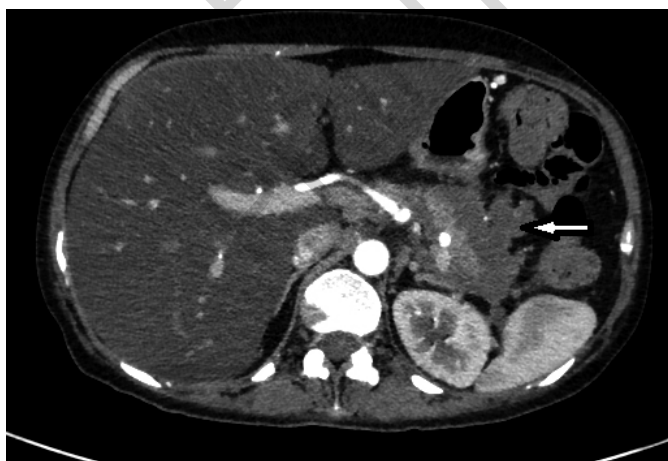
Case Report

A 35-year-old, chronic alcoholic woman, was admitted for an acute necrotizing pancreatitis with a normal gallbladder and absent biliary stones. The condition was associated with sepsis sustained by acinetobacter, hardly controlled with intravenous Tigecycline 100 mg/12 hours, ampicillin +surbactam 3 gr/8 hours and caspofungin 50 mg/24 hours: treatment lasted over two months. Percutaneous drainage by interventional radiologists failed to completely evacuate the peripancreatic collection with persistent septic status (Fig. 1). For this reason a surgical approach was deemed indicated. Through a bi-subcostal laparotomy, an accurate necrosectomy was performed and two, 28 mm, silicone drains were left in place for irrigation and lavage/drainage of the residual cavity in the postoperative course. The septic status slowly regressed and antibiotic treatment could be stopped within 6 weeks: at this time a control CT-scan showed a satisfactory reduction of the cavity, allow-

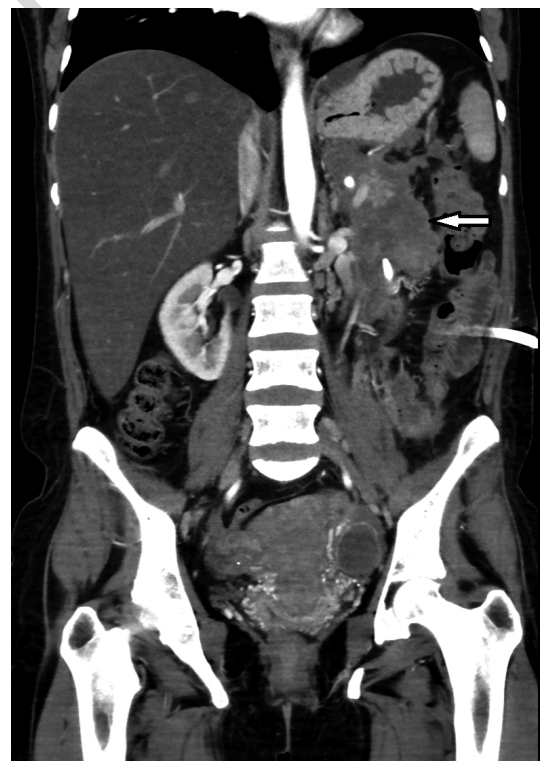
ing retraction of one of the two drains. However, CT imaging at this time, also showed a 1 cm diameter false aneurysm of the cystic artery extending toward its posterior branch, not evident upon initial admission (Fig. 2). A second-look laparotomy was then performed leading to lysis of adhesions, ligation of the cystic artery at its origin after Pringle maneuver and control of the right hepatic artery, aneurysmectomy en-bloc with cholecystectomy and re-exploration of the initial collection which resulted in fibrotic involution. Re-do postoperative course was uneventful: residual pancreatic and subhepatic drains were removed respectively on postoperative day 2 and 3 and the patient was discharged on postoperative day 5, after a satisfactory control CT-scan, showing the absence of peripancreatic fluid collection and other arterial anomaly (Fig. 3).

Discussion

The present report suggests the efficacy of open resection of cystic artery pseudoaneurysm, en bloc with cholecystectomy, following acute, necrotizing pancreatitis. Overall, pseudoaneurysms of the cystic artery are very rare, following most often cholecystitis or iatrogenic injury³⁻⁶. Bleeding or pseudoaneurysmal degeneration of the peripancreatic vessels consequent to arterial erosion during acute necrotizing pancreatitis is well known and



A



B

Fig. 1: (A) CT-scan of the abdomen showing persistent peripancreatic necrotic collection in the transversal slice; (B) incompletely evacuated by percutaneous drain at sagittal view.



A



B

Fig. 2: (A) CT-scan of the abdomen after surgical necrosectomy/drainage of peripancreatic collection: in the transversal view; (B) the peripancreatic collection has disappeared. However a pseudoaneurysm of the cystic artery has developed (sagittal view, slim arrow, B), whereas surgical drains are correctly positioned (large arrow, B).

not infrequent, however the attent of the cystic artery with ensuing pseudoaneurysm during necrotizing pancreatitis is extremely rare, with apparently only two cases reported so far¹⁷⁻¹⁸. In general, as bleeding is a serious complication of such condition, independently from its etiology, its treatment is indicated also at an uncomplicated, asymptomatic stage. However, due to its rarity a standardized treatment protocol is lacking, and in the setting of pseudoaneurysmal degeneration of the cystic artery as a complication of acute necrotizing pancreatitis, the management choice can be subjective and directed by the patient's status¹⁸. Endovascular embolization has become the most frequent approach in the recent period^{1,8}. Isolated pseudoaneurysm embolization may expose to the risk of ischemic gangrene of the gallbladder, within a reported delay of three days¹⁸. For this reason cholecystectomy should be performed with a short delay after pseudoaneurysm embolization^{7,15-16}.

In the reported case, the option of pseudoaneurysm embolization prior to cholecystectomy was considered, based on the assumption that it would reduce the risks of intraoperative pseudoaneurysmal rupture, when gaining access through a redo and scarred surgical field. However, we elected to treat the pseudoaneurysm by open surgery through the previous subcostal incision performed for debridement and evacuation of necrotic tissue, considering that this approach would allow simultaneous resection of the pseudoaneurysm and the gall-

bladder and a second look of any residual necrotic focus with the opportunity of completing its debridement and cleansing. The stable patient's conditions and the elective basis of treatment supported this therapeutical choice.

In conclusion, on an elective basis, open surgical resection of cystic artery pseudoaneurysm following necrotizing pancreatitis en bloc with cholecystectomy, seems a reasonable treatment option for this very rare condition.



Fig. 3: Control CT-scan before patient's discharge showing absence of any other arterial anomaly and of any residual peripancreatic collection.

Riassunto

Una paziente di 35 anni, etilista cronica, veniva ricoverata per un episodio di pancreatite acuta necrotico-emorragica e sottoposta a drenaggio percutaneo della raccolta peripancreatica e terapia antibiotica. Per l'insuccesso di un efficace drenaggio della raccolta per via interventistica e la persistenza di uno stato settico veniva sottoposta ad una necrosectomia/drenaggio per via laparotomia seguita da un riassorbimento della raccolta e risoluzione della sepsi. Tuttavia una TC di controllo prima della rimozione dei drenaggi dimostrava uno pseudo aneurisma dell'arteria cistica, assente al momento del ricovero. Veniva quindi eseguita, attraverso un accesso laparotomico iterativo, una colecistectomia con resezione in blocco dello pseudo aneurisma associata ad un "second look" dell'area peripancreatica. Dopo un decorso post-operatorio regolare, la paziente veniva dimessa in 5^{ta} giornata postoperatoria.

Gli pseudoaneurismi dell'arteria cistica sono molto rari. L'embolizzazione per via percutanea è efficace per nel trattamento dello pseudo aneurisma, ma richiede l'esecuzione di una colecistectomia nell'immediato seguito della procedura a causa del rischio di una perforazione ischemica della colecisti. La resezione chirurgica dello pseudo aneurisma in blocco con la colecistectomia sembra, al momento, la modalità di trattamento più appropriata di questa condizione.

References

- Patil NS, Kumar AH, Pamecha V, Gattu T, Falari S, Sinha PK, et al.: *Cystic artery pseudoaneurysm. A rare complication of acute cholecystitis: Review of the literature.* Surgical Endoscopy, 2022; 36: 871-80.
- Fujimoto Y, Tomimaru Y, Hatano H, Noguchi K, Nagase H, Atsushi H, et al.: *Ruptured cystic artery pseudoaneurysm successfully treated with urgent cholecystectomy: a case report and literature review.* Am J Case Rep, 2018; 19:187-93.
- Kaman L, Kumar S, Behera A, Katariya RN: *Pseudoaneurysm of the cystic artery: A rare cause of hemobilia.* Am J Gastroenterol 1998; 93:1535-37.
- Nakajima M, Hoshino H, Hayashi E, Nagano K, Nishimura D, Katada N, et al.: *Pseudoaneurysm of the cystic artery associated with upper gastrointestinal bleeding.* J Gastroenterol, 1996; 31:750-54.
- Carey F, Rault M, Crawford M, Lewis M, Tan K: *Endovascular. Case report: Cystic artery pseudoaneurysm presenting as massive per rectum bleed treated with percutaneous Coil Embolization,* 2020; 3: 8.
- Nkwam N, Heppenstal K: *Unruptured pseudoaneurysm of the cystic artery associated with acute calculus cholecystitis.* J Surg Case Rep, 2010; 2:4.
- Sunkara PRV, Shah PK, Rakshi K, Choudhary SR, Bohidar NP, Dube Y: *Rupture of cystic artery pseudoaneurysm: A rare complication of acute cholecystitis.* Indian J Surg, 2018; 80: 87-9.
- Taghavi SMJ, KumaR MJ, Prabha RD, Puhalla H, Sommerville C: *Cystic artery pseudoaneurysm: Current review of aetiology, presentation, and management.* Surgery Research And Practice, 2021; 4492206: 1-6.
- England RE, Marsh PJ, Ashleigh R, Martin DF: *Pseudoaneurysm of the cystic artery: A rare cause of hemobilia.* Clin Radiol, 1998; 53: 72-5.
- Saluja S, Ray S, Gulati M, Pal S, Sahni P, Chattopadhyay TK: *Acute cholecystitis with massive upper gastrointestinal bleed: A case report and review of the literature.* BMC Gastroenterol, 2007; 7:12.
- Akatsu T, Tanabe M, Shimizu T, Handa K, Kawachi S, Aiura K, et al.: *Pseudoaneurysm of the cystic artery secondary to cholecystitis as a cause of hemobilia: Report of a case.* Surg Today, 2007; 37: 412-17.
- GLaysher MA, Cruttenden-Wood D, Szentpali K: *A rare cause of upper gastrointestinal haemorrhage: ruptured cystic artery pseudoaneurysm with concurrent cholecystojejunal fistula.* Int J Surg Case Rep, 2014; 5:1-4.
- Berry R, Han J, Kardashian Aa, Larusso NF, Tabibian JH: *Hemobilia: etiology, diagnosis and treatment.* Liver Research, 2018; 2: 200-08.
- Lee JW, Kim MY, Kim YJ, Suh CH: *CT of acute lower GI bleeding in chronic cholecystitis: Concomitant pseudoaneurysm of cystic artery and cholecystocolonic fistula.* 2006; 61:634-36.
- Proenca AL, Gomes FV, Costa N, Billim T, Luz JH, Coimbra E: *Transarterial embolization of iatrogenic cystic artery pseudoaneurysm.* Ge Port J Gastroenterol, 2020; 27:115-18.
- Mahalingam S, Shaikh OH, KumbhaR LS, Mohan A: *Cystic artery pseudoaneurysm due to carcinoma of the gallbladder.* BMJ Case Rep, 2021; 14: e241714
- Delgadillo X, Berney T, De Perrot M, Dider D, Morel P: *Successful treatment of a pseudoaneurysm of the cystic artery with microcoil embolization.* JVIR, 1999; 10:789-92.
- Thillai M, Sethi P, Menon RN, Kader NP: *Cystic artery pseudoaneurysm following acute necrotizing pancreatitis.* BMJ Case Rep, 2017; 2017: bcr2016218891.