



## Rare Association between Gallbladder Perforation, Distant Hepatic Abscess Formation and Choledocolithiasis: Case Report and Literature Review

*Natacha Helena Uryu, Giovana de Figueiredo Godoy, Wellington Forte Alves Junior, Isabel Butter Amim, Emilia Virginia Lima Curvelo Fontes, Adriano Corona Branco and José Francisco de Mattos Farah\**

*Department of Surgery, Hospital do Servidor Público Estadual, Brazil*

### Abstract

The development of a liver abscess linked to gall bladder perforation represents a rare complication of acute cholecystitis. Mortality rates of patients with gall bladder perforation vary from 12% to 16%, of which 86.6% gall bladder perforations are associated to cholelithiasis. Treatment for the rare association between complications of cholecystitis are divergent in the literature, however recent publications reveal a tendency towards using minimally invasive techniques during surgical treatment. A 72-year-old male with history of hypertension and diabetes, presenting abdominal pain, anorexia, and jaundice. Image exams indicated cholecystitis with surrounding abscess formation and a distant abscess, also identifying choledocholithiasis. Topography of the etiology had a significant role when choosing the order of the treatments of the different complications. Several non invasive procedures were applied prior to performing laparoscopic surgery. The patient recovered well and was discharged after 22 days. We believe an individualized approach is at the core of quality patient care. There is a consensus among several authors that the initial control of the septic state in which most patients are found is essential, however, it is important to emphasize that the identification of the correct primary infectious site can help determine the succession of treatments which can also positively influence the outcome of the case.

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#### \*Correspondence:

José Francisco de Mattos Farah,  
Department of Surgery, Hospital do  
Servidor Público Estadual, Brazil,  
E-mail: josefarah@einstein.br

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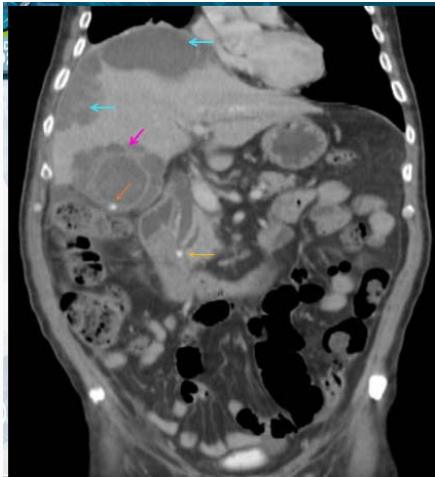
**Keywords:** Acute abdomen; Cholecystectomy; Cholecystostomy; Choledocholithiasis; Laparoscopic; Gall bladder; Liver abscess; Sepsis; Surgery

### Introduction

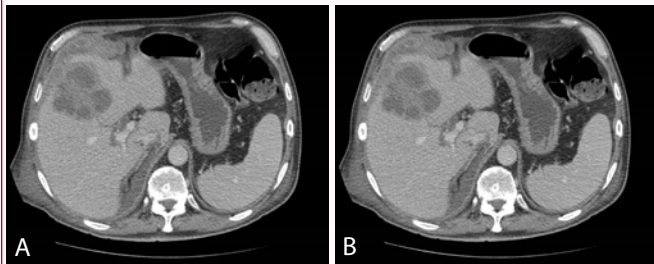
Currently, cholelithiasis is present in more than 10% of the occidental population and represents etiology of 90% of the cases of acute cholecystitis [1]. The complication rate of acute cholecystitis is 15% and mortality rate varies from 3% to 10% [1-3]. Among the main complications that stand out are choledocholithiasis (15%), gall bladder perforation (0.8% to 3.2%), gallstone ileus (0.3% to 0.5%) and intra-hepatic abscess (<1%) [4-7]. Mortality rate of patients with gallstone perforation varies from 12% to 42% [8], and 86.6% of all gallstone perforation are correlated to cholelithiasis. The development of an intra-hepatic abscess due to gallstone perforation represents a rare complication of acute cholecystitis. In 1984, Neimeier classified the gall bladder perforation into three categories according to time: Acute, sub-acute and chronic (Table 1) [8]. Nonetheless, Neimeier's classification does not include the entire pathological spectrum of the disease such as concomitant development of abscesses or cholecystohepatic communications. There is no standardization in the treatment of these complications, literature provides divergent order of care for each complication and reveals a tendency to use the laparoscopic approach for surgical intervention. The case described below demonstrates a great positive impact on the treatment progression of the patient through the use of minimally invasive techniques, prioritizing a decrease in the inflammatory state before submitting him to surgery.

### Case Presentation

A 72-year-old male is transferred to our service; his medical history indicates hypertension and diabetes, without previous surgery, presenting abdominal pain, anorexia, and weight loss of 18% in 3 months. In March of 2021 patient started with complaints of abdominal pain, vomits and hyporexia, went to a local health service which prescribed antibiotics for him, the following months he persisted with low ingestion of nutrients, presenting fever (37.8°C to 38°C) in July of



**Figure 1:** Abdomen CT before procedures showing choledocholithiasis (yellow arrow); Acute gallstone cholelithiasis (orange arrow); Intra-hepatic abscesses (blue arrows); and gallbladder perforation with surrounding collection (pink arrow).



**Figure 2:** Abdominal CT-scan showing the evolution of the hepatic abscess. Figure A - Admission day; Figure B - After drainage of the abscess (7 days later).

2021, by the end of July he was admitted in the local hospital due to the septicemic state for 3 days. He was admitted on this service august 7<sup>th</sup> with physical examination compatible with discrete jaundice with no significant abdominal findings. Laboratory test revealed 34000/mm<sup>3</sup> white blood count, increased serum levels alkaline phosphatase, C-reactive protein (28.8 mg/dL) and direct hyperbilirubinemia, hypoalbuminemia. Abdominal CT findings were compatible with gallstone acute cholecystitis, choledocholithiasis, and two intra-hepatic abscesses in different topography, the largest measuring 10.6 cm (Figure 1). It was established the diagnosis of acute cholecystitis Tokyo 2. Thorax CT showed pleural effusion which was drained (exit of 1,240 ml of liquid).

Initially we opted to realize, concomitant to the use of ceftriaxone and metronidazole, a cholecystostomy simultaneous to the drainage of both liver abscesses (exit of 600 ml of bilio-purulent liquid). It was not noticed contrast extravasation during cholangiopancreatography. After the procedure a decline of C-Reactive Protein (CRP) serum level (27.7 mg/dL) and white blood count (33,500/mm<sup>3</sup>) was noticed.

On the second day hospitalized patient presented without complaints and a decreased on inflammatory markers such as white blood count (26,330/mm<sup>3</sup>) and c-reactive-protein (19.73 mg/dL). It was performed Endoscopic Retrograde Cholangiopancreatography (ERCP) which showed choledocholithiasis with intra and extra-hepatic biliary dilation. Laparoscopic cholecystectomy was

performed on the next day finding a small amount of purulent liquid in the abdominal cavity, abundant adhesions, liver elevated due to inflammatory process, thicker, necrotic, and perforated gallbladder, with purulent liquid output, by the end of the procedure it was removed both drains allocated previously. The first postoperative test results revealed 22,720/mm<sup>3</sup> white blood count and CRP 11.16 mg/dL; and histological analysis were compatible with chronic xanthogranulomatous cholecystitis with acute outbreak and fibrin leukocyte peritonitis. The patient progressed favorably even though he had surgical complication (supra-hepatic and perirenal abscess drained), on the tenth day admission his white blood count 9,200/mm<sup>3</sup> and CRP 7.75 mg/dL.

He stayed hospitalized throughout more twelve days to watch his evolution and to finish use of intravenous antibiotics, during this period a pararenal collection with serohematic content appeared on control CT and it was percutaneously drained. On the day of discharge, he presented without complaints, no abdominal findings on CT images (Figure 2), and laboratory exams showing CRP 0.8 mg/dL, leukocytes 6740/mm<sup>3</sup> and no bacterial growth in blood cultures. After three months follow-up patient presents in good general condition, no complaints since this hospitalization.

## Discussion

Gall bladder perforation associated with intra-hepatic abscess and cholangitis is a rare complication of cholecystitis, entering on the classification proposed by Neimeir as type II (subacute). According to Date RS et al. [8], mortality rate of gall bladder presentation is 10.8% and the most common type of presentation is type 2 (46.2%). Different mechanisms are involved in the etiology process of gall bladder perforation, the most common is cystic duct obstruction, gall bladder distension with consequential increase in the intravascular pressure, altered vascularization, ischemia and necrosis followed by perforation [9]; however, there is other pathogenesis involved in the case of gall bladder perforation with intrahepatic liver abscess, started with the infection of Rokitansky-Aschoff sinuses followed by necrosis, rupture and perforation [9,10].

Since 1984 when Neimeir first classified the gall bladder perforations most treatments reports focused on the gall bladder's removal, associated initially with the abscess drainage (Table 2). On the last five years it was observed an increase on the use of minimally invasive techniques as ERCP and laparoscopic surgery. One case of type II perforation was treated only with ERCP; it had a favorable disclosure [11]. Nikumbh et al. [12] advocates that although radiological evaluation with CT and ERCP is essential prior to surgery in some cases, an early laparoscopic intervention to perform a sub-total cholecystectomy with drain placement is enough to treat both cholecystitis and liver abscess in a definitive manner. Mean while Alshammari et al. [9] affirm that percutaneous drainage and antibiotic therapy could well treat the hepatic abscess followed by a distant laparoscopic cholecystectomy avoiding more invasive procedures.

Although ultrasound remains the main imaging exam for investigation of biliary diseases, the only reliable sonographic sign for gall bladder perforation in "the whole sign" which is a defect in the GB wall. CT-scan is considered the gold standard for the diagnosis of complicated cholecystitis, interruption of the GB wall and the existence of extraluminal gallstones better than sonography. Perforation of the gall bladder can be suspected in case of presence of

**Table 1:** Niemeier classification of gallbladder perforation [8].

I	Acute: Associated with generalized biliary peritonitis
II	Subacute: Fluid localization at sites of perforation, pericholecystic abscess and localized peritonitis
III	Chronic: Formation of interna/external fistulae

**Table 2:** Topography of the etiology had a significant role on choosing the order of the treatments of the different complications.

Author	Year of publication	Characteristics			Treatment
		age	sex	presentation	
Singla et al. [6]	1998	65	M	acute	Open cholecystectomy and abscess drainage
Bhatwal et al. [13]	2012	60	M	acute	Open cholecystectomy and peritoneal lavage
Kamalesh et al. [14]	2012	70	M	acute	Open cholecystectomy then ultrasound-guided percutaneous drainage
Singh et al. [3]	2013	50	M	chronic	Open cholecystectomy then abscess drainage
de Hollanda et al. [15]	2013	50	M	acute	Open cholecystectomy then abscess drainage
Donati et al. [16]	2014	58	M	subacute	ERCP followed by open cholecystectomy then abscess drainage
Alessiani et al. [11]	2014	77	M	chronic	Cholecystostomia
Alshammari et al. [9]	2016	70	F	acute	Abscess drainage then after 3 months open cholecystectomy
Hussain et al. [5]	2016	72	M	acute	Abscess drainage then laparoscopic cholecystectomy
	2016	62	F	subacute	Open cholecystectomy then abscess drainage
Williams et al. [17]	2017	61	M	subacute	ERCP followed by laparoscopic cholecystectomy
Nikumbh et al. [12]	2020	55	M	subacute	ERCP followed by laparoscopic cholecystectomy
Uryu et al.	2021	72	M	subacute	Abscesses drainage then ERCP followed by laparoscopic cholecystectomy

abscess or complex fluid adjoining, and as an intraparenchymal liver abscess, CT image may appear as a hepatic mass uni or multilocular with peripheral enhancement and hyperemia of the adjacent parenchyma [13-15].

Topography of the etiology had a significant role on choosing the order of the treatments of the different complications. The present report has chosen initially to drain the abscesses expecting to prevent a septic shock and reduce the cholecystohepatic inflammation process, once we believe the etiology of the liver abscess being the cholecystitis, differential etiology diagnosis with cholangitis was considered less probable due to the absence of jaundice and total bilirubin of 1.8 mg/dL. Following treatment (ERCP) aimed to remove the biliary obstruction, reducing the inflammatory process in the choledochal duct, and bettering the patient's condition to perform a more secure minimally invasive surgery (laparoscopic cholecystectomy). Because of the complexity of the case, there is a range of variations in the order of therapeutic approach involving the patient's biliary tree, there was no assurance the etiology was in fact from the gall bladder or the cystic duct obstruction and therefore no certainty to direct the sequence of the treatment [16,17].

## Conclusion

We believe individualized approach is the core of the best conduct, there is a consensus among several that initially the control of the septic state in which most patients are found is essential, however, it is important to emphasize that the search to identify the correct primary infectious site to organize the succession of treatments can also influence the outcome of the case. Noninvasive therapeutics was applied even in a complex case of cholecystitis with three associated complications, which may have contributed to the successful treatment of this rare association.

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