External Postoperative Biliary Lithiasic Fistulae Associated with Choleperitoneum

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ABSTRACT

Introduction: Laparoscopic cholecystectomy is now a standardized technique, unanimously accepted and considered in mini-invasive techniques. On the other hand, surgical indication for cholecystectomy is based on a series of clinical, biological and imaging data, extremely well coded.

Material and method: The authors bring to discussion 2 cases of patients who developed, after laparoscopic colecis-tectomy, in an interval of 9 months in one case, and 6 weeks the other case, a rebound of their initial biliary disease, this

REZUMAT

Introducere: Colecistectomia laparoscopicã reprezintã astãzi o tehnicã standardizatã, devenitã deja clasicã în cadrul tehnicilor mini-invazive cu o indicaåia operatorie care se bazeazã ãi pe o serie de date clinice, biologice æi imagistice, extreme de bine codificate.

Material æi metodã: Autorii aduc în discuåie douã cazuri la care, dupã colecistectomia laparoscopicã, la un interval de 9 luni respectiv 6 sãptãmãni, suferinåa biliarã a recidivat printr-o severã complicaåie peritonealã, ca urmare a unei fistule de bile biliarã principãlã, datorate unei litiaze biliare necunoscute.

Discuåii: La ambele cazuri, iniåial s-a intervenit chirurgical pe cale laparoscopicã pentru asemãnãtoare forme de colecis-titã acutã litiazicã, farã ca datele biologice, simptomatologia, imagistica sau constãrile intraoperatorii sã semnaleze prezenåa unei forme de litiazã a cãii biliare principale. La ambele cazuri colecistectomia laparoscopicã s-a desfãæurat în bune condiåiuni, iar evoluåia postoperatorie a fost favorabilã o lungã perioadã de timp, pânã în momentul noului episod al suferinåei biliare. Acest nou episod a debutat în ambele cazuri prin instalarea unui icter frust, urmat rapid de declanåauia unei peritonite biliare prin fistulã biliarã litiazicã. Doar la unul din pacienåi reintervenåia pe cale clasicã a permis rezolvarea leziunii, la celalalt caz reintervenåia clasãcã fiind urmatã de multiple recidive æi intervenåii endoscopice, pânã la un rezultat stabil.

Concluzii: Lucrarea supune discuåiei o serie de supoziåii privind mecanismul de apariåie al fistulei biliare, corectitudinea conduitei terapeutice æi a explorãrilor paraclinice respectiv aspectul restant, recidivant, migrat sau independent al litiazei biliare.

Cuvinte cheie: fistule litiazice, extern, postoperator, coleperitoneu

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Case Report

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time with severe manifestations of peritoneal complications as a result of a main biliary duct fistulae, due to an unknown lithiasic process.

**Discussion:** In both cases the initial surgical intervention was a laparoscopical procedure for acute lithiasic cholecystitis, both with the same clinical behaviour. In both cases there was no biological, symptomatical, imagistic or intraoperative reason to suspect a form of main biliary duct lithiasis process. In both cases the procedure was undertaken in good conditions and the postoperative evolution was favorable for a long period of time, up to the moment of the new bile-related symptoms. This new event’s first symptoms were a frust icter, followed shortly by a biliary peritonitis as a result to the biliary fistulae. In only one of these 2 cases the classical second approach was able to lead to the end of the symptoms, in the other case multiple endoscopic procedures being needed for a stable and favorable outcome.

**Conclusions:** Our report is now facing a series of suppositions regarding the mechanism that made possible the biliary fistulae, the accuracy of the therapeutic decisions and the paraclinical explorations as well as the real origin (migrating, independent, recidivant?) of the biliary lithiasis.

**Key words:** external, lithiasic fistulae, postoperative, choleperitoneum

**INTRODUCTION**

Laparoscopic cholecystectomy is regarded as a safe procedure, which could allow only with great difficulty the possibility of unpleasant progression or intraoperative surprises [2]. Both findings are now able to allow in most cases an optimal therapeutic conduct and a most simple postoperative evolution, devoid of consequences.

But, however, even with diagnosis establishment and surgical indication with maximum precision, such as a very advanced surgical technique, surgery further reserves the right to not always provide nor pleasant, nor easy to solve surprises.

By external biliary fistula one understands a communication between the biliary tree and the peritoneal cavity, due to a breach in the intra- and extrahepatic bile ducts, which allows the discharge of bile in the free peritoneum. This in turn can be controlled or uncontrolled [5].

External biliary fistulas are labeled as controlled when the bile is exteriorized via an intraperitoneal trajectory, formation of a biliary collection being absent or uncontrolled, when manifesting as a coleperitoneum. In turn, the coleperitoneum is represented by an accumulation of bile in the peritoneal cavity, initially sterile, and then the associated inflammatory factor triggering bacterial translocation from the intestinal lumen, to turn into an infected coleperitoneum, which is in reality a biliary peritonitis [4, 5].

But perhaps bacterial translocation is not the only possibility of contamination, as biliary lithiasis fistulas stimulate the choledochal bile discharge through the biliary lumen, rich in germ by associating angiocholitis, which becomes another cause of biliary peritonitis.

External biliary fistula was considered in the nineteenth century, a common complication of diseases of the biliary tree. The first description is attributed to Thiselius in 1670, then in 1890 COURVISIER described 169 cases, Naunyn 184 cases in 1896, and in 1897 BONNET, 122 cases. However, in the last 50 years, due to the evolution of medical science and surgical techniques, less than 20 cases have been mentioned in the literature. Of these latter studies, two cases mention coexistence of spontaneous external biliary fistula with an internal one, the last of which, by far, seems to be the most common [1].

**MATERIAL AND METHOD**

We describe the cases of 2 patients, operated on in our service, in which laparoscopic cholecystectomy was performed for acute lithiasic cholecystitis, with favorable postoperative evolution. Both cases developed a clinical picture of biliary peritonitis through external lithiasic biliary fistula, after a uneventful postoperative evolution and after a relatively long period of suffering, due to a unknown and unsanctioned main biliary duct lithiasic.

CASE I. The first story concerns a patient, M.A. aged 45, patient chart no. 275 of January 17 2002, who is admitted under emergency status with a diagnosis of acute lithiasic cholecystitis, confirmed by clinical, biological and imaging investigations.

Ultrasound detected a distended gallbladder with longitudinal diameter of approx. 80 mm, with wall thickening and edema, occupied by numerous calculi, the greatest one being 7 mm, mostly mobile, with others fixed in the infundibular region. The main bile duct was free, and of normal size.

Laparoscopic colecistectomy was performed, revealing a vesicular hydrops with significant edema in the infundibulo-cystic and hepatic pedicle regions, unable to view the main bile duct. Cholecystectomy proceeded under good circumstances, the only notable element being a short cystic duct, with a narrow lumen, unoccupied and extremely edematous, lacking sufficient space for clipping, so that it was ligated with an extracorporeal knot. Postoperative evolution was favorable, the patient being discharged on postoperative day 4, under surgically cured status.
On October 18 of the same year, after 9 months, the patient is readmitted under emergency status with the clinical picture of an infected choleperitoneum in generalized biliary peritonitis stage, presenting sclerotegumentary jaundice, fever and generalized muscular defense in the abdominal area. From her medical history we learn that the current suffering dates back approximately a week, when she noticed the jaundice development, and requested admission in a regional hospital. As an inpatient, she presented an episode of right upper quadrant pain after about two days, a vivid pain that later decreased in intensity without completely receding, but then spread throughout the peritoneal cavity.

In our service, upon admittance, presence of jaundice and of peritoneal irritation signs are observed, in absence of bowel movement for feces and gas, septic fever, and an added hepatorenal failure, in the oligoanuric stage. Laboratory data reveal a total bilirubin level of 12.36 mg.%, with direct bilirubin 7.00 mg.%, AST 53 u/l, ALT 81 u/l, alkaline phosphatase 324 u/l, creatinine 1.6 mg/dL, serum urea 7.2 mg/dL, leukocytosis with 15,000/mmc, and ESR of 115 mm per hour. Abdominal ultrasound revealed presence of liquid in the peritoneal pouches with predominance in the subhepatic and right subphrenic region, without intrahepatic biliary duct dilation and without offering any information on the pancreatic morphology or the main bile duct, due to intense aerocolia.

The CT exam revealed multiple right subdiaphragmatic liquid collections, peripherally, anterior to the left lobe and to the IVth segment. (Fig. 1)

Thus, a diagnosis of obstruction jaundice, angiocholitis, external biliary fistula, generalized biliary peritonitis is set, with no available element suggesting the possibility of lithiasic jaundice.

Reintervention. After a short period of diuresis stimulation and hydro-electrolyte, volemic and acid-base rebalancing, surgery is performed on the 19th of October 2002, by wide right subcostal incision, detecting a seclud-
mental control of the main bile duct failing to identify any stenotic or lithiasic obstacle. We thus decided to limit the intervention to axial external biliary drainage, placed through the same perforation orifice with right trans-hepatoparietal exteriorization, leading the distal end of the drain tube to the point of the terminal choledochus, next to the papilla. (Fig. 4)

The choice of an axial drainage was based on the possibility of a ductal stenosis which passed unobserved, thus allowing ductal reinforcement and recalibration, parallel to biliary duct decompression.

Drainage method was influenced as well by the extreme friability of the biliary wall during several attempts of suture perforation, when the suture wires cut through the tissue while knot tying. Thus, we considered that a KEHR type drain could lead to a permanence of the biliary fistula, especially after its removal, choosing instead an axial drain. Unfortunately, the fistulous orifice remained open, communicating freely with the peritoneal cavity, possibly even wider than the initial perforation, due to the extreme friability of the choledochal wall, to the instrumental exploration and the impossibility of a tight suture.

Left and right subphrenic, subhepatic drainage, of the parietocolic and pelvic recesses, after abundant lavage of the peritoneal cavity with betadine serum, were the last gestures of this intervention, which preceded the closure of the abdominal wall, leaving room for maybe the only possibility of clearing our uncertainties, retrograde endoscopic cholangiography.

Postoperative evolution was indeed simple, the only notable events being labeled as part of an evolution favorable in all ways.

We refrained from any investigation with contrast material throughout these two weeks, in order to prevent increasing the pressure in the ducts and to allow closure of the fistulous opening, which was confirmed by complete reduction of the subhepatic drainage. These are once again some of the valences of axial drainage, which leaves the main bile duct intact, allowing even a spontaneous healing of a fistulous opening under the conditions of continuous decompression of the biliary tree.

An endoscopic Oddi papillosphincterotomy followed, the surprise being the identification of a main bile duct remnant lithiasis, consisting of multiple calculi located in the right and left hepatic ducts, which could not be fully extracted using the Dormia probe due to the endoluminal presence of the axial drainage tube. Further control examinations revealed spontaneous evacuation of the bile stones from the main bile duct via the large papillosphincterotomy previously performed and complete closure of the biliary fistula. After approximately two weeks, the patient is retransferred to our service, axial drainage is suppressed, and she is discharged, surgically cured. (Fig. 5)

In February 2004, the patient is readmitted for a new episode of jaundice, endoscopic exploration revealing the restenosis of the Oddi papillosphincterotomy, and a new lithiasis recurrence. A new endoscopic Oddi papillosphincterotomy is performed, multiple choledochal calculi being evacuated with the aid of a Dormia probe, intervention resulted in the remission of clinical phenomena and favorable evolution until present. (Fig. 6)

CASE II. The patient P.M., 74-year-old, patient chart 1149 / 19.11.2002, is admitted for emergency treatment with a diagnosis of acute lithiasic cholecystitis. Ultrasound detects a distended gallbladder, elongated, with wall thickening, and the image of a calculus of 5/10 mm, located in the infundibulum, immobile. Free main bile duct, with normal size.

Laparoscopic surgery is performed on 22.11.2002, laparoscopically, describing the appearance of acute cholecystitis with important parietal and infundibular edema, showing a short cystic duct with normal diameter, but edematous. Laparoscopic cholecystectomy is
concluded, the operative piece extracted having a lithiasic content represented by two oval calculi, blackish, with olive appearance, and diameter of approx. 1 cm.

Postoperative evolution was favorable, the patient was discharged on postoperative day 7, surgically cured. The same simple and uneventful evolution continued the following 6 weeks, until the 15.01.2003, when the patient is readmitted with symptoms of angiocholitis, marked by colicky pain, jaundice and fever, which culminated with paroxysmal pain in right upper quadrant, followed by phenomena of acute peritonitis.

Reintervention. Emergency surgery is performed via a median xipho-subumbilical incision, revealing generalized infected choleperitoneum, accompanied by adherences and false membranes. At the level of the dilated bile duct, approximately 15 mm in diameter, a longitudinal perforation with necrotic margins is observed, about 2.5 cm long, located on the anterior aspect, starting under the biliary convergence and up to the superior margin of the duodenum, through which a purulent bile was oozing. At distal compression of the intrapancreatic common bile duct, a dark oval calculus is evacuated through the choledochal wound, resembling an olive, approx. 1 cm in diameter, similar to the two vesicular calculi extracted during laparoscopic cholecystectomy.

This enormous breach of the main bile duct allowed a comfortable endoluminal exploration, both instrumental, as well as visual, without noticing any other calculus. Also, passage of the metal instrument in the papilla or liver ducts confirmed their perfect permeability. (Fig. 7)

In this case, placing an external biliary drainage in axial manner is decided for and performed, with transhepatoparietal externalization, followed by an attempt of suturing the perforation. An appropriate suture was not
managed this time either, due to extreme friability of the ductal wall. Without having even by far the certainty of a tight suture, we had to abandon a biliary fistula in contact with the peritoneal cavity, hoping for a minimal chance of improvement, as a result of continuous decompression of the biliary tree by keeping the axial drainage in continuous declivous position and of presence of a permeable subhepatic drainage.

Abundant saline lavage of the peritoneal cavity with iodine solution and polivalent drainage, with multiple drain tubes, completed this procedure, the patient’s condition remaining stable. (Fig. 8)

Postoperative evolution was tedious due to large losses of fluid and electrolytes, external biliary drainage was approximately 700 ml daily, plus peritoneal and especially subhepatic drainage, which would take over the fistula flow, oscillating between 200-300 ml of bile for a period of more than two weeks, in order to recede completely after about a month. Therefore, this time as well, we managed to close the biliary fistula by continuous biliary duct decompression, fluid losses externalized through the external biliary drainage requiring a highly balanced intensive compensation therapy.

We expected axial drainage maintenance for a further period of one month, gradually lifting the drainage container up to the level of the body, when the favorable evolution was interrupted by a bronchopneumonia, in the remission period of which a massive fatal pulmonary embolism occurred.

Both cases raise through their particular evolution a series of questions that we will try to answer more from experience rather than from the literature, where we found only a few brief references supporting our claims.

In both cases, the onset of the secondary clinical suffering was represented by jaundice development, followed by perforation and a clinical picture of coleperitoneum, which evolved into a biliary peritonitis. Since then, in both cases, either an accidental main bile duct injury, occurred during laparoscopic cholecystectomy and with late manifestation, or a biliary obstruction, lithiasis likely, was suspected, considering the history of lithiasis.

DISCUSSION

Both cases were concerning a lithiasis of the main bile duct, unreported and previously unknown, remnant or recurrent, discovered due to biliary lithiasis perforation lesions followed by coleperitoneum, occurring at a distance from the laparoscopic cholecystectomy.

Can we ascribe an iatrogenic attribute to these fistulas or must we consider them as independent and without any connection to the technical defects of the previous laparoscopic surgery [6]?

In case of iatrogenic lesions, external biliary fistula is usually the negative response to surgery, early or late, the result of an unnoticed lesion either by electrical burn, or by clipping, or by unexpected devascularisation of a segment of the main bile duct. The consequences of these gestures are however manifested much earlier, within a few days or up to a maximum of one week after surgery [8].

There is of course the possibility of a main bile duct stenosis, with validation in a much longer period of months or years, isolated or associated with a persistent biliary fistula with low flow; occurrence of stenosis is usually a consequence of biliary fistula, which causes inflammatory lesions which progress towards parietal fibrosis. Remnant or recurrent biliary lithiasis is an aggravating factor, able to complete the etiopathogenesis of these external biliary fistulas, either by direct mechanical abrasion aggression and parietal decubitus, or by its redoubtable consequence - angiocholitis, or by both [11].

Both cases involve external biliary fistulas located in the main bile duct, probably occurred amid necrotic lesions that led to the development of scars, whose detachment primed perforation. In both cases, the intraoperative findings clearly led to the conclusion of ductal wall necrosis, judging by the appearance of the perforation margins, by its extreme friability upon attempt to suture and by after the purulent appearance of the exteriorized bile [7, 9].
Also in both cases we are dealing with the indubitable aggression of bile duct calculi and their perforating consequences, situations in which both the mechanical as well as the infectious element lead to the onset of decubitus lesions and local vascular thrombosis, followed by ischemia, necrosis and perforation. In addition, calculus presence involves a series of pressure balances and imbalances in the biliary tree, hyperpressure contributing to the onset of perforation and maintenance of the fistulous tract.

An additional thought is that the biliary fistula occurred every time at a distance from the laparoscopic cholecystectomy, which allows in the case of very few exceptions the consideration of possible complications due to laparoscopic technique, the period of time passed being too long to invoke late validation of these injuries.

Thirdly, the question of appropriateness and effectiveness of the axial drainage technique approached at reintervention is raised, which ended each time with favorable postoperative evolution.

In neither case was an appropriate suture of the fistula succeeded, due to the extremely brittle biliary wall, both patients developing spontaneous healing of the fistulous orifice thanks to decompression of the biliary tree and to angiocholitis remission due to the benefits of continuous and declivous external biliary drainage. Axial drainage was able to lead to the closure of a main bile duct fistula, even in the presence of remnant calculi, thanks to the ability to decompress the biliary tree, the remission of angiocholitis and relaxation of the fistulous tract through a permanent and effective external biliary drainage [3].

In both cases surgery was performed in such a manner that it would allow the transformation of a generalized peritonitis in a localized peritonitis, or more precisely, in a collection of biliary fluid fueled by the fistula, but permanently drained out. This quite common tactic in the face of an external biliary fistula has a good chance of determining the closure of the fistulous tract only in case of an unoccupied main bile duct and, especially, through continuous decompression of the biliary tree, preferably via an external axial type drainage. Surprisingly, in the first case this technique allowed the closure of the fistulous tract even in the presence of some remnant calculi, in a young person [10, 12].

In fact, the essence of surgical treatment efficiency applied to these two cases lies in maintaining in declivous position the axial decompression drainage of the biliary tree for as long as possible, accompanied by limiting of the peritoneal source, transforming a generalized biliary peritonitis in localized peritonitis, as a subhepatic coloenteroneum, permanently drained externally [4].

Even in these extreme situations, endoscopic retrograde cholangiopancreatography and Oddi endoscopic sphincterotomy offer the chance of diagnosing biliary lithiasis and of extraction or evacuation of the calculi.

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CONCLUSIONS

These two cases allow a series of considerations which can be briefly formulated:

1. Remnant or recurrent lithiasis, asymptomatic and unknown, may occur at a distance from laparoscopic cholecystectomy, by perforation of the main bile duct, resulting in external biliary fistulas and biliary peritonitis.
2. Participation of laparoscopic cholecystectomy in the mechanism of occurrence of these fistulas may be a consequence of small areas of main bile duct devascularization, left unnoticed.
3. These perforations of the main bile duct present extremely friable necrotic margins, preventing any attempt to suture the fistulous opening.
4. The operative conduct in these situations is continuous decompression of the biliary tree through an external biliary drainage, preferably in axial manner, accompanied by appropriate subhepatic drainage to decrease fistula flow and transform the generalized biliary peritonitis into a localized bile lake, easy to control.
5. Next, endoscopic retrograde cholangiopancreatography and Oddi endoscopic sphincterotomy offer the chance of diagnosing biliary lithiasis and of extraction or evacuation of the calculi.
6. Any pre- or intraoperative suspicion of associated biliary lithiasis imposes widened exploration through peroperative laparoscopic cholangiography, during laparoscopic cholecystectomy.

REFERENCES