

Spontaneous internal biliary fistulas

Bode Rovena, Dracini Xheladin, Celiku Etmond

Department of General Surgery, "Mother Teresa" University Hospital Center,
Tirana, Albania

SUMMARY

Introduction: Internal spontaneous biliary fistulas, or abnormal communications between the gallbladder or biliary ductal system and internal organs of the body, occur secondary to pathologic processes that involve the gallbladder, bile ducts, stomach, duodenum, small intestine, and colon. Their clinical manifestations are produced by local effects of abnormal communication with the adjacent bowel. The chronically diseased and obstructed biliary tract is exposed to contamination by intestinal content and the resulting symptoms are biliary in character. When a calculus has sufficient size to occlude the lumen of the bowel, it passes into the intestinal tract, and findings are primarily those of bowel obstruction.

Aim of the study: The purpose of this study is to evaluate the clinical features and surgical management of spontaneous internal biliary fistulas, found in patients who had not previously had surgical procedures on or near the biliary tract. We reviewed a number of consecutive patients who had been treated for biliary tract diseases during a 4-year period (June 2013-July 2017).

Results: Jaundice was a usual sign of spontaneous internal biliary fistulas, found in all patients. Severe inflammation of the extrahepatic biliary system was present in most cases, resulted from contamination by intestinal content. All of cases were discovered during operation, no one diagnosed preoperatively.

Conclusion: Surgeons must be aware of the possible presence of an internal biliary fistula before surgical intervention, for adopting appropriate imaging modalities before surgery. This might be particularly true when considering laparoscopic cholecystectomy. Since the preoperative diagnostic rate for internal biliary fistula involving the gallbladder is still low, care is necessary before and at the time of surgery especially for elderly patients with cholelithiasis.

Key words: internal biliary fistula, cholelithiasis, peptic ulcer disease, gall bladder carcinoma

Abbreviations: CVD: Cardiovascular Disease; DM: Diabetes Mellitus; PLMD: Pulmonary Disease; HBP: High Blood Pressure; Years: Years Old; CBD: Common Bile Duct Disease

Address for correspondence:

Bode Rovena, Surgery Department, University of Medicine,
Tirana, "Mother Tereza" University Hospital Center, Rruga e
Dibres, 1025 Tirana, Albania, email: rovenabode@gmail.com

Word count: 2964 **Tables:** 05 **Figures:** 01 **References:** 15

Received: - 06 September, 2019

Accepted: - 20 September, 2019

Published: - 27 September, 2019

INTRODUCTION

Internal biliary fistulas are abnormal communications between the gallbladder or biliary ductal system and internal organs of the body, occurring secondary to pathologic processes that involve the gallbladder, bile ducts, stomach, duodenum, small intestine, and colon. The clinical manifestations of internal biliary fistulas are produced by remote or local effects of abnormal communication with the adjacent bowel [1]. The chronically diseased and obstructed biliary tract is exposed to contamination by intestinal content and the resulting symptoms are biliary in character. When a calculus of sufficient size to occlude the lumen of the bowel passes into the intestinal tract, findings are primarily those of bowel obstruction [2]. Two are the most important causes of spontaneously forming internal biliary fistula: (1) chronic cholecystitis and (2) penetration of a peptic ulcer [3]. In fact Aird writes: 'Internal fistula of the gall bladder or cystic duct with duodenum, small intestine, or some other hollow viscus is usually due to the erosion of stone during an attack of acute cholecystitis'. Marshall and Polk in their observations found that 28 of 30 cases of cholecystoduodenal fistula could be attributed directly to chronic gall bladder disease, whereas only two were the results of penetrating peptic ulcers [4]. Ulcerative colitis is another cause of internal biliary fistulae between the colon and the gall bladder, and malignant disease of the biliary tract, stomach, colon, and duodenum have been proved to produce internal biliary fistulae [5]. Hunt et al. have reported that choledochoduodenal fistulas in the periampullary region were iatrogenic, caused by the passage of a rigid choledochal bougie during common duct exploration [6].

The first mechanism involved in the formation of the spontaneous internal biliary fistula is due to adhesions between the gall bladder and adjacent viscera and necrosis of the wall of the gall bladder at the sites of adhesion, which leads to a fistula being formed by a direct tract. The second mechanism is due to an abscess may form around the gall bladder, bursting into contiguous hollow viscera, which then communicate indirectly through the abscess cavity [4, 5].

Although it is sometimes difficult to diagnose correctly the type of internal biliary fistula preoperatively, surgeons must be aware of the possible presence of an internal biliary fistula before surgical intervention. This might be particularly true when considering laparoscopic cholecystectomy and especially in elderly patients where there is a high rate of morbidity and lethality. The recent advances in hepatobiliary imaging techniques have allowed us to reevaluate hepatobiliary diseases. With the aid of endoscopic retrograde cholangiography,

choledochoduodenal fistulas in the periampullary region have been found more frequently than ever.

PATIENTS AND METHOD

We reviewed a number of 871 consecutive patients who had been treated for biliary tract diseases during a 4-year period, from June 2013 through July 2017.

All patients operated by traditional open cholecystectomy, laparoscopic cholecystectomy (as emergency cases and elective cases), other biliary tract diseases especially malignancy are part of the study. All patients were treated for diseases of the biliary tract at the first clinic of the General Surgery Department, "Mother Teresa" University Hospital Centre, Tirana.

Despite the improvement in imaging modalities, have allowed a preoperative diagnosis of biliary fistula, all our cases were discovered during surgery, in the operating room. Ultrasound, Computed Tomography Scan (CT) and Magnetic resonance were employed in most of the patients. Retrograde cholangiography, endoscopic retrograde cholangiography, percutaneous transhepatic cholangiography are imaging modalities that cannot be used in our hospital. And the diagnosis of internal biliary fistula and the type of the fistula was determined by operative findings only. Spontaneous internal biliary fistulas were found in 8 patients of the 871 consecutive cases, presented as two cases with cholecystocolic fistulas, two cholecystogastric fistulas, two cholecystocholedochal fistula (Mirrizi syndrome) and one case presented with gallstone ileus.

Of the 8 cases found and studied, 5 were females and 3 were males (Table 1). Fistulas occur approximately twice as frequently in females as in males. This closely parallels the sex incidence in chronic cholecystitis and cholelithiasis [7].

Age and sex

Of the 8 patients, 5 were females and 3 were males, with a female/male ratio 1:6:1. The mean age was 66 years in the nonobstructive type, in the obstructive type it was 72 years, ranging from 57 to 81. More than 85% of patients were over 60 years old, and range 61-70 in 57% of them (Figure 1).

Past history

Of all the patients, 71% had a history of previous gallbladder disease. All of them gave report of a biliary colic history and jaundice in their past. The obstructive type gave a history of biliary colic many years ago replaced by spastic colic pain time by time.

Internal biliary fistulas were caused by biliary calculi in 7 patients (85.5%) and by malignant tumors (gallbladder carcinoma) in one patient. The clinical features of patients with biliary fistulas are shown in Table 2. Eight fistulas were found in 8 patients. The most common of them were cholecystogastric fistulas in 2 patients (all lithiasis in origin), cholecystocolic fistulas in 2 patients (all lithiasic in etiology), cholecystocholedochal fistulas in 2 patients (one lithiasic and one from gallbladder carcinoma) and a cholecystoduodenal fistula caused by biliary calculi. The duration of illness ranged from 12 years to 3 months (Table 2). Upper abdominal pain, jaundice, and fever were the main symptoms. Diarrhea was present in a patient with cholecystocolic fistula. None of the patients has undergone previous biliary/gastric operations (Table 2).

Preoperative diagnosis

None of the fistulas was diagnosed preoperatively, in both types.

Associated diseases

Associated diseases were found in all of them Hypertension, cardiovascular disease, diabetes mellitus, peptic ulcer disease, chronic pancreatitis, bronchial asthma, hiatal hernia. No multiple fistulas were found. No choledocoduodenal fistulas were found (Tables 3-5).

Classification and distribution of stones

It should be noted that

- It was not taken bile culture in any of patients
- There is no evidence of stone nature in any of patients
- Data on the distribution of stones reflect pre-intraoperative findings
- And define the behavior with CBD exploration

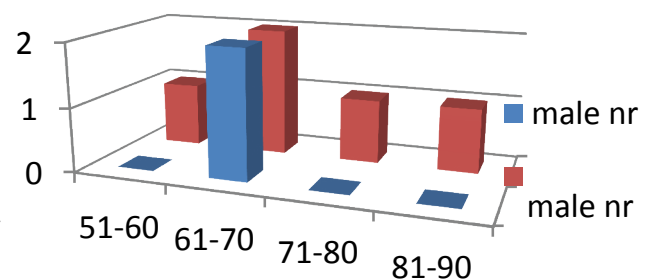


Fig. 1. Age and sex distribution of SBF (spontaneous internal biliary fistulas)

Tab. 1. Patients characteristics preview	Type of fistula	Gender	Age	Cause	History and other diseases
	Patient 1 (cholecystogastric)	female	65 years	lithiasis	biliary colic+, CVD
	Patient 2 (cholecystogastric)	female	63 years	lithiasis	biliary colic+, jaundice, CVD
	Patient 3 (cholecystocholedochal)	female	57 years	gallbladder cancer	biliary colic-, DM,
	Patient 4 (cholecystocholedochal), (Mirrizi)	female	81 years	Lithiasis	biliary colic+, jaundice, PLMD, CVD,
	Patient 5 (cholecystocolic)	male	67 years	lithiasis +chronic pancreatitis	biliary colic+
	Patient 6 (cholecystocolic)	male	64 years	lithiasis	biliary colic+, jaundice, DM
	Patient 7, biliary ileus (Bouveret syndrome)	female	72 years	terminal ileum	Small bowel occlusion clinic+, HBP,
	Patient 8 (cholecystoduodenal)	male	65 years	Lithiasis+peptic ulcer disease	Epigastric periodic, recurrent pain

Tab. 2. Clinical features

Type of fistula	Age/sex	Symptom/signs	Duration of illness
Cholecysto-gastric (2)	63/f	8 months earlier attack of biliary colic, cholecystitis, with the onset of jaundice, fever for some days, actually no jaundice, palpable gb	12 years
	65/f	4 months ago attack of biliary colic, cholecystitis, fever some days, actually no jaundiced	8 years
Cholecysto-duodenal (1)	65	Biliary pain, fever, diarrhea, more 2 years ago, actually no jaundiced	7 years
Cholecysto-choledocal (2)	81/f	7 weeks ago biliary pain, fever, jaundice for 2 weeks, actually jaundiced	4 years
	57/f	One month of biliary pain, fever,	
Cholecysto-colic (2)	67/m	Biliary pain months ago, cholecystitis, jaundice, and fever for some days, diarrhoea time by time	6 years
	64/m	Biliary pain months ago, jaundice and fever for some weeks, diarrhoea time by time	7-8 years
Biliary ileus (1)	72	abdomen distended, intestinal colic, vomiting, stop in feces and gases, more than 6 years ago dg with gall stones, now absent	12 years

Tab. 3. Classification and distribution of stones

Type of fistula	No (patients)	Distribution of gall stones		
		GB	CBD	GB and CBD
Cholecysto-gastric	2	2	0	0
Cholecysto-duodenal	1	1	0	0
Cholecysto-colic	2	1	0	1
Cholecysto-choledocal	2	1	0	1

GB: Gall Bladder, CBD: Common Bile Ducts

Tab. 4. Radiological findings

	Obstructed group (1 patient)	Non obstructed group (7 patients)
Abdominal X-ray dg (no contrast)	No evidence of gallbladder stones, No evidence of air in biliary tract, no evidence of enteric stone Intestinal obstruction	None
Abdominal X-ray dg Barium meal/enema	-None	-None
Abdominal Ultrasound (done in 8 patients)	No evidence of gallbladder stones, dilated small bowel loops, free intra-peritoneal liquid, No evidence of air in biliary tract	Gallbladder stones, contracted-atrophic gallbladder, Normal/dilated extrahepatic biliary ducts, suspect of pancreatic Mass, no aerobilia Hydatid cyst of liver, dilated common hepatic duct,
Computed tomography scans (done in 5 patients)	-None	Gallbladder stones, choledocal stone, contracted- atrophic gallbladder, dilated extrahepatic biliary ducts, suspect of caput pancreatic mass, no aerobilia, no fistula tract Hydatid cyst liver with fistulisation in the CBD, Dilated intra/extrahepatic ducts
Magnetic resonance (done in 2 patients)	-None	-Gallbladder stones, dilated extra/intra hepatic biliary tract, distal choledocal stone, pancreatic mass. -gallbladder stone, Suspect of gallbladder carcinoma or cholangiocarcinoma
Gastroscopy (done in 3 patients)	-None	(1) Gastric ulcer (1) Hiatal hernia, suspicion of bulb infiltration, (1) Erosive gastritis

Tab. 5. Operative findings and procedures	Type of fistula	Intraoperative findings	Surgery
	Cholecysto-gastric (2)	Shrunken gb full of stones, normal common bile duct, traject of fistula isolated	Cholecystectomy+repair of fistula (2)
	Cholecysto-colic (2)	1. Shrunken GB with evidence of stones, 2. GB and choledocal stones, dilated choledoc, shranked gallbladder	1. Cholecystectomy+repair of fistula 2. Cholecystectomy+biliodigestive anastom+repair of fistula
	Cholecysto-choledocal (2)	1. Acute inflamed litiasic GB, stone impacted and eroding through Hartman's pouch into common duct, choledocal decubitus, common bile duct delated exploration revealed no stones, (Mirrizi) 2. GB carcinoma (stone), choledocal infiltration,	1. Cholecystectomy/choledocotomy, choledocal amputation, choledoco-duodenal anastomosis 2. Cholecystectomy/choledocal amputation and hepatico-jejunoanastomosis, Jabulay pyloroplasty
	Cholecysto-duodenal (1)	Shrunken GB, a lot of stones, normal common bile duct	Cholecystectomy+repair of fistula
	Gall stone ileus (1)	Stone impacted in the ileum, 30 cm from the ileocaecal valve	Enterotomy, stone removal

DISCUSSION

Spontaneous internal biliary fistulas are formed between the extrahepatic biliary tract and a variety of adjacent organs. The Incidence is reported to be 0.9%-3.2% of patients with biliary diseases and the most common type is cholecystoduodenal (77%), cholecystocolonic (15%) and cholecystogastric (2%) [8-11]. In our series, the most common type were cholecystocolonic, cholecystogastric followed by cholecystocholedocal and cholecystoduodenal fistulas. Actually, a difference in the incidence may be explained by the fact that choledochoduodenal fistulas have been found more frequently because of the recent development of endoscopic retrograde cholangiography [8]. The clinical features in our study were elderly female, with a long duration of illness and past history of a rapid relief from jaundice, which agrees with previous reports. The symptoms were similar to those of cholelithiasis [9]. Internal biliary fistulas usually occur as a result of pressure necrosis caused by calculi in the inflamed gallbladder or biliary tract. The fistulas were also reported to be formed by peptic ulcer, malignant tumor, trauma and Crohn's disease. Choledochoduodenal fistulas were reported to be caused mainly by peptic ulcer [10-15]. Hunt et al. reported that choledochoduodenal fistulas in the periampullary region were iatrogenic caused by passage of a rigid choledochal bougie during common duct exploration [6]. In our series this possibility should be excluded because no patients had a history of common bile duct exploration [13].

In our series, there was no patient with a choledochoduodenal fistula caused by a peptic ulcer. This can presumably be attributed to the recent therapeutic improvement for peptic ulcers with H2 receptor antagonist.

In the present study, there was no evidence of pneumobilia in none of cases. Although ultrasound and CT examinations were not diagnostic, the presence of the internal biliary fistula should be suggested in some cases by the findings of pneumobilia, an atrophic gallbladder, and biliary stones.

Management may depend on the type and etiology of the internal biliary fistula. Cholecystectomy, removal of common bile duct stones, if present, and repair of the fistula are the best choice in patients with cholecystoenteric fistulas. Dissection of the cystic duct and its junction with the common hepatic and common bile ducts is essential, and because these structures will be surrounded with dense fibrous tissues, their identification is

difficult. If the cystic duct is wide and the common bile duct dilated, exploration of the common bile duct is indicated so as to recognize retained stones or foreign material. In 29%-30% of Marshall and Polk's (1958) series stones were found in the common bile duct. In the event of multiple small stones and debris being present, a choledochoduodenostomy may be indicated following the removal of stones. Cholecystectomy should always be carried out. The closure of the fistulous openings in the colon or duodenum presents little difficulty and the lumen of the bowel is rarely narrowed.

In contrast, the preferred therapy for the periampullary choledochoduodenal fistula is endoscopic papillotomy including fistulotomy in combination with bile duct stone extraction. Cholecystectomy may be mandatory if the patients also have gallbladder disease. In general, laparoscopic cholecystectomy should not be used in the patients with cholecystoenteric or cholecystocholedochal fistulas, although Velez et al. have recently reported a case of a cholecystoduodenal fistula which was successfully repaired laparoscopically.

Those patients presenting with gall stone ileus will require resuscitation with intravenous electrolytes to correct their water/salt imbalance, and, as the electrolyte depletion may be gross, hypertonic solutions may be required. Surgery is directed to the relief of obstruction by the removal of the stones and a search throughout the small bowel for any other stones which may be present and subsequently produce intestinal obstruction.

CONCLUSION

Surgeons must be aware of the possible presence of an internal biliary fistula before surgical intervention, for adopting appropriate imaging modalities before surgery. This might be particularly true when considering laparoscopic cholecystectomy.

Although surgery is the treatment of choice, endoscopic sphincterotomy, and CBD stone extraction/stents placements, percutaneous transhepatic drainage have been said to cause spontaneous healing of the fistula by reducing increased biliary pressure. This has been used in the treatment of elderly unfit patients.

Pneumobilia, a small atrophic gallbladder and a history of disappearance of jaundice in elderly patients may suggest the presence of an internal biliary fistula.

REFERENCES

- | | |
|---|--|
| <ol style="list-style-type: none"> 1. Glenn F, Reed C, Grafe W. Biliary enteric fistula. <i>Surg Gynecol Obstet.</i> 1981;153:527-531. 2. Zwemer F, Coffin-Kwart VE, Conway M. Biliary enteric fistulas: management of 47 cases in Native Americans. <i>Am J Surg.</i> 1979;138:301-304. 3. Tanaka M, Ikeda S. Periapillary choledochoduodenal fistula: an analysis of 83 consecutive patients diagnosed at ERCP. <i>Gastroenterol Endosc.</i> 1983;29:89-93. 4. Marshall SF, Polk RC. Spontaneous internal biliary fistulas. <i>Surg Clin N Amer.</i> 1958;38:679-691. 5. Feller ER, Warshaw AL, Schapiro RH. Observations on management of choledochoduodenal fistula due to penetrating peptic ulcer. <i>Gastroenterol.</i> 1980;78:126-131. 6. Hunt DR, Blumgart LH. Iatrogenic choledochoduodenal fistula: and unsuspected cause of post-cholecystectomy symptoms. <i>Br J Surg.</i> 1980;67:10-13. 7. Tanga RM, Ewing JB. Primary malignant tumors of the gallbladder-report of 43 cases. <i>Surgery.</i> 1970;67:418-426. 8. Puestow CB. Spontaneous internal biliary fistulae. <i>Ann Surg.</i> 1942;115:1043-1054. | <ol style="list-style-type: none"> 9. Griffith CDM, Saunders JH. Cholecysto duodenocolic fistula following abdominal trauma. <i>Br J Surg.</i> 1982;69:99-100. 10. Porter JM, Mullen DC, Silver D. Spontaneous biliary-enteric fistulae. <i>Surgery.</i> 1970;68:597-601. 11. Velez M, Mule J, Brandon J, Kannegieter. Laparoscopic repair of a cholecystoduodenal fistula. <i>Surg Endosc.</i> 1991;5:221-223. 12. Marshall SF, Polk RC. Spontaneous internal biliary fistulas. <i>Surg Clin N Amer.</i> 1958;38:679-691. 13. Ford JA, Soop M, Du J, Loveday BPT, Rodgers M. Systematic review of intraoperative cholangiography in cholecystectomy. <i>Br J Surg.</i> 2012;99:160-167. 14. Sajid MS, Leaver C, Haider Z, Worthington T, Karanjia N, et al. Routine on-table cholangiography during cholecystectomy: a systematic review. <i>Ann R Coll Surg Engl.</i> 2012;94:375-380. 15. Topal U, Savci G, Sadikoglu MY, Tuncel E. Choledochoduodenal fistula secondary to duodenal peptic ulcer. A case report. <i>Acta Radiol.</i> 1997;38:1007-1009. |
|---|--|