

RAPID COMMUNICATION

## Internal biliary fistula due to cholelithiasis: A single-centre experience

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### Abstract

**AIM:** To discuss about the perioperative problems encountered in patients with internal biliary fistula (IBF) caused by cholelithiasis.

**METHODS:** In our hospital, 4130 cholecystectomies were carried out for symptomatic cholelithiasis from January 2000 to March 2004 and only 12 patients were diagnosed with IBF. The perioperative data of these 12 IBF patients were analyzed retrospectively.

**RESULTS:** The incidence of IBF due to cholelithiasis was nearly 0.3%. The mean age was 57 years. Most of the patients presented with non-specific complaints. Only two patients were considered to have IBF when gallstone ileus was observed during the investigations. Nine patients underwent emergency laparotomy with a pre-operative diagnosis of acute abdomen. In the remaining three patients, elective laparoscopic cholecystectomy was converted to open surgery after identification of IBF. Ten patients had cholecystoduodenal fistula and two patients had cholecystocholedochal fistula. The mean hospital stay was 13 d. Two wound infections, three bile leakages and three mortalities were observed.

**CONCLUSION:** Cholecystectomy has to be performed in early stage in the patients who were diagnosed as cholelithiasis to prevent the complications like IBF which is seen rarely. Suspicion of IBF should be kept in mind, especially in the case of difficult dissection during cholecystectomy and attention should be paid in order to prevent iatrogenic injuries.

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**Key words:** Internal biliary fistula; Cholecystoduodenal fistula; Cholelithiasis; Iatrogenic injuries

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### INTRODUCTION

A biliary fistula is an abnormal passage or communication from the biliary system to an organ, cavity, or free surfaces. Fistula are classified as external (biliary-cutaneous) or internal (biliobiliary, bilioenteric, bronchobiliary)<sup>[1]</sup>. Internal biliary fistula (IBF) is associated with chronic cholelithiasis in 90% of the cases.

Preoperative diagnosis of IBF is difficult<sup>[2]</sup>. As the symptoms of IBF include abdominal pain, fever, nausea, vomiting, flatulence, fat intolerance, diarrhoea and weight loss, which are all non-specific and seen in most gastrointestinal pathologies, the diagnosis is often not suspected preoperatively<sup>[3]</sup>. The diagnosis is usually made peroperatively<sup>[4-6]</sup>.

### MATERIALS AND METHODS

A total of 4130 cholecystectomies were carried out for symptomatic cholelithiasis in Ankara Numune Teaching and Research Hospital, Ankara, Turkey from January 2000 to March 2004 and only 12 patients were diagnosed with IBF. The preoperative and peroperative findings as well as postoperative course of the patients with IBF were evaluated from their hospital records.

### RESULTS

#### Pre-operative findings

Of 4130 cholecystectomies, 12 patients (5 women and 7 men) were diagnosed as IBF. The mean age of these 12 patients was 57 years and 8 of these patients were above 60 years. Five of the 12 patients were previously diagnosed cases of cholelithiasis.

Tenderness in the right hypochondrium was the most frequent physical finding presented in 10 of the 12 (83.3%) patients. One case presented with abdominal pain in the right lower quadrant, with nausea, vomiting and abdominal distension, that was preoperatively diagnosed as acute appendicitis.

Preoperative diagnosis of IBF was achieved in only two patients with gallstone ileus and based primarily on plain abdominal X-ray. The consistent findings were air-

Table 1 The characteristics of the patients

No.	Age/sex	Pre-operative diagnosis	Peri-operative findings	Operation	Hospital stay (d)	Morbidity during postoperative course
1	69/F	Gallstone ileus	Double CDF + gallstone ileus + iatrogenic sigmoid perforation	Cholecystectomy + enterolithotomy + T-tube + primary sutures to duodenal wall + Bogota bag + primary colon repair	45	Bile leakage
2	80/M	Acute abdomen, Acute appendicitis? Ileus?	Gallstones, CDF + jejunal mass + gallstone ileus	Cholecystectomy + choledochotomy + T-tube + primary sutures to duodenum + tube duodenostomy + appendectomy + jejunotomy + removal of stone	16	Myocardial infarction, death
3	60/M	Obstructive jaundice	CCDF	Partial cholecystectomy + repair of common bile duct over T-tube	20	Wound infection
4	22/M	Cholecystocholedocholithiasis	Adhesions, conversion to laparotomy, CDF, acute cholecystitis	Cholecystectomy + T tube insertion + primary sutures to duodenal wall	10	NC
5	67/F	Cholelithiasis	Adhesions, conversion to laparotomy, CDF	Cholecystectomy + primary sutures to duodenal wall	8	NC
6	54/F	Acute abdomen	CDF	Cholecystectomy + choledochotomy + T-tube insertion + primary sutures to duodenal wall	8	NC
7	71/M	Choledocholithiasis	Mirizzi's syndrome, CDF	Cholecystectomy + choledochotomy + T tube insertion + primary sutures to duodenal wall	6	NC
8	30/F	Cholelithiasis	CDF	Cholecystectomy + primary sutures to duodenal wall	4	NC
9	66/M	Cholelithiasis	Adhesions, conversion to laparotomy, CDF	Cholecystectomy + T-tube insertion + primary sutures to duodenal wall	8	Wound infection
10	38/M	Acute cholecystitis	Acute cholecystitis, CDF	Cholecystectomy + T-tube insertion + primary sutures to duodenal wall	34	ARDS, death
11	62/M	Acute abdomen, gallbladder perforation	CDF, perforation of gallbladder, pleural effusion	Cholecystectomy + T-tube insertion + primary sutures to duodenal wall + chest tube insertion <sup>1</sup>	28	GI bleeding, bile leakage, sepsis, death
12	70/F	Acute cholecystitis, obstructive jaundice	CCDF, acute cholecystitis	Cholecystectomy + Hepaticojejunostomy	25	Bile leakage

CDF: Cholecystoduodenal fistula; CCDF: Cholecystocholedochal fistula; NC: No complications. <sup>1</sup>This is the primary operation performed. Patient also underwent repeated procedures due to bile leakage.

fluid levels, distended small bowel loops and a radio-lucent stone outside the biliary tree.

Endoscopic retrograde cholangiopancreatography (ERCP) performed in two patients due to obstructive jaundice and choledocholithiasis failed to show any fistulous opening in any of them.

Among four patients diagnosed preoperatively as acute abdomen, two patients had serious coronary failure and other two had history of chronic obstructive pulmonary disease.

Nine of the 12 patients with IBF underwent emergency laparotomy with the pre-operative diagnosis of acute abdomen. In remaining three patients, elective laparoscopic cholecystectomy was converted to open surgery after identification of IBF.

### Peroperative findings

In the 12 patients with IBF, peroperative findings were dense adhesion around gallbladder requiring of sharp dissections, fibrosed and contracted gallbladder, encountering difficulty in identifying cystic duct, cystic artery and common bile duct.

Intra-operative cholangiography (IOC) had to be performed only in 4 of the 12 patients in order to prevent iatrogenic injuries due to the aforementioned findings. Nevertheless, IOC was useful in identifying IBF in only one patient. The fistula tract was identified after sharp dissection in the remaining 8 patients. Regarding the types of fistulas, cholecystoduodenal fistula (CDF) and cholecystocholedochal fistula (CCDF) were found in 10

and 2 patients, respectively.

The fistula orifice in the common bile duct was usually closed by T-tube placement after exploration of the common bile duct for stone retrieval. The fistula orifice in the duodenum was primarily repaired and T-tube was inserted with the aim of biliary decompression in the patients with CDF. Cholecystectomy plus fistula repair, enterotomy and removal of the gallstone (enterolithotomy) were performed in two patients with cholecystoduodenal fistula (CDF) plus gallstone ileus. The operative details and other details are shown in Table 1.

### Postoperative course

The mean (range) hospital stay was 13 (4-45) d. Two wound infections, three bile leakages and three mortalities were observed in postoperative period. Thus, the mortality rate was 25% (3/12) and morbidity rate was 42% (5/12).

One death occurred due to myocardial infarction on the 16<sup>th</sup> postoperative day after cholecystectomy + choledochotomy and T-tube insertion + repair of fistula + tube-duodenostomy + jejunotomy and removal of stone. The second death occurred due to acute respiratory distress syndrome (ARDS) on the 18<sup>th</sup> postoperative day after cholecystectomy + T-tube insertion + primary closure of the duodenum. The third death occurred following repeated procedures (abdominal lavage for intra-abdominal interloop abscess + T-tube reinsertion) due to septic complications caused by bile leakage on the 28<sup>th</sup> d after cholecystectomy + T-tube insertion + primary closure of the duodenum.

One of the two patients had bile leakage after being discharged from the hospital with T-tube and T-tube was extracted after one month later. External biliary fistula was observed in the other patient and the fistula was closed after 20 d.

## DISCUSSION

Internal biliary fistula (IBF) occurs due to acute inflammation with obstruction of the cystic duct resulting in adhesions of the gallbladder to the adjacent viscus, usually to the duodenum; and repeated attacks of inflammation including gangrenous changes of the gallbladder wall and the wall of the adherent viscus, with eventual erosion and fistula formation<sup>[1]</sup>. The common causes of IBF include cholelithiasis, peptic ulceration, malignant neoplasm (gallbladder, bile duct, duodenum, pancreas, or stomach), Crohn's disease of the duodenum and paraduodenal abscess<sup>[1,7,8]</sup>. In this study, IBF due to cholelithiasis was evaluated.

Our data showed the IBF incidence of 0.29% (12/4130) and all fistula were secondary to cholelithiasis, which is less than the previous reports<sup>[9,10]</sup>.

Our patients with IBF usually presented with non-specific signs or symptoms of biliary fistula. Hence, the preoperative accurate diagnosis was made only in 16.6% (2/12) patients in the present study, which is in agreement with the previous study<sup>[3]</sup>. In the present study, gallstone ileus was suspected on plain abdominal X-ray in only one patient, which was confirmed by operative findings and stone was removed by enterotomy. However, some classical findings on plain abdominal radiography include pneumobilia, intestinal obstruction, aberrantly located gallstone and change of location of a previously observed stone<sup>[1,3]</sup>. Although ultrasonography is extremely useful in detecting a fistula, pneumobilia, impacted stones and the presence of residual cholelithiasis and/or choledocholithiasis<sup>[11]</sup>, none of the IBF in our patients was detected preoperatively by ultrasonography.

As the symptoms of IBF include abdominal pain, fever, nausea, vomiting, flatulence, fat intolerance, diarrhoea and weight loss, which are all non-specific and seen in most gastrointestinal pathologies, the diagnosis is often not suspected preoperatively<sup>[3]</sup>. Preoperative diagnosis according to the symptoms was not possible in any of the our cases. Jaundice secondary to common bile duct obstruction was reported to be a common finding in patients with cholecystocholedochal fistula (CCDF)<sup>[9,12]</sup>. Although two patients in our study presented with jaundice and underwent ERCP, we were unable to detect any fistulous opening.

Most studies report that the fistulas are detected incidentally or unsuspectedly during ERCP or other radiologic examinations performed for investigation of biliary or pancreatic diseases<sup>[13]</sup>. Although ERCP failed to detect IBF in one patient in the present study, ERCP is the principal tool for diagnosis of IBF<sup>[4]</sup>. It can easily show the fistula orifice, fistula tract, and communication with the biliary tree, as well as the etiology. ERCP has therapeutic potential by endoscopic sphincterotomy

**Table 2** Data on diagnosis and management of internal biliary fistula in literature

Authors	Case number	Peri-operative findings	Procedure
Silecchia <sup>[18]</sup>	5	CCDF, Type II Mirizzi syndrome	NA
Carlei <sup>[19]</sup>	5	CDF	LC + repair
Angrisani <sup>[6]</sup>	34	CDF, CJF, CCF.	LC + repair in 14 patients, Conversion to laparotomy in 20 patients
Moreno Ruiz <sup>[20]</sup>	17	14 CDF, 3 CCF, 2 CGF	LC + repair

NA: Surgical data not available; LC + repair: Laparoscopic cholecystectomy + fistula repair; CDF: Cholecystoduodenal fistula; CCDF: Cholecystocholedochal fistula; CCF: Cholecysto-colic fistula; CGF: Cholecystogastric fistula; CJF: Cholecystojejunum fistula.

and choledocholithotomy<sup>[14]</sup>. CT is the most appropriate imaging test for further evaluation because sonographic diagnosis is often difficult<sup>[15]</sup>. Pickhardt *et al*<sup>[16]</sup> reported that MRCP might be useful in selected cases to confirm the diagnosis<sup>[16]</sup>.

In our study, all fistulas were diagnosed intra-operatively. Suspicion of IBF should be kept in mind, especially in cases of difficult dissection during cholecystectomy due to small, contracted, chronically inflamed and densely adherent gallbladder, and attention should be paid in order to prevent iatrogenic injuries. Moreover, intra-operative cholangiogram through the gallbladder help to identify any existing fistulous tract.

Cholecystoduodenal fistula (CDF) is the most frequent type (70%-90%) of IBF, followed by cholecystocolonic (10%), choledochoduodenal, choledochogastric, cholecystogastric, and duodeno-left hepatic duct fistulas<sup>[1,17]</sup>. Similarly in this study, cholecystoduodenal fistula (CDF) and cholecystocholedochal fistula (CCDF) were found in 10 (83.3%) and 2 (16.6%) patients, respectively. Two of our patients with cholecystoduodenal fistula had gallstone ileus (20%).

The standard treatment of IBF is cholecystectomy and repair of the fistulous opening; however, it was performed in only two cases in our study<sup>[2]</sup>. Common bile duct exploration, T-tube insertion, choledochoduodenostomy, tube duodenostomy, enterolithotomy and hepaticojejunostomy were also used as an additional procedure when indicated.

It was initially reported that when the IBF was diagnosed during laparoscopic cholecystectomy, it carried a higher rate of conversion to laparotomy<sup>[6,18]</sup> (Table 2). Nowadays, as the surgeon is skilled in advanced laparoscopic procedures, such as duodenal mobilization and intracorporeal suturing and knotting, the rate of conversion is low, CDF is no longer considered a contraindication for laparoscopic treatment<sup>[19-21]</sup>. Although the procedure was started laparoscopically in three cases in the present study, it was converted to the open due to technical difficulties.

The mortality rate of IBF was 25% (3/12) in our study, which is slightly higher than that reported in literature (i.e. 15% to 22%)<sup>[1,9]</sup>; this might be explained as the patients underwent emergency operation for acute abdomen without enough preoperative evaluation.

In conclusion, cholecystectomy has to be performed in the early stage in the patients who were diagnosed as cholelithiasis to prevent the complications like IBF which is seen rarely. Suspicion of IBF should be kept in mind, especially in case of difficult dissection during cholecystectomy and attention should be paid in order to prevent iatrogenic injuries.

## COMMENTS

### Background

Internal biliary fistula (IBF) is seen rarely and it is a complication of chronic cholelithiasis. Preoperative diagnosis of IBF is difficult because of non-specific signs or symptoms. Although ultrasonography and ERCP are extremely useful in detecting a fistula, the diagnosis of IBF is usually made peroperatively.

### Research frontiers

This study showed high morbidity, mortality rate and the peroperative problems encountered in patients with IBF.

### Innovations and breakthroughs

Even though IBF is seen rarely, it must be remembered in the patients with chronic cholelithiasis and IBF should be diagnosed during USG and ERCP. Currently, treatment of IBF may be performed during laparoscopic cholecystectomy, but in this case the surgeon must be very skilled in advanced laparoscopic procedures.

### Applications

Cholelithiasis has to be treated in early stage to prevent IBF. Suspicion of IBF should be kept in mind, especially in cases of difficult dissection during cholecystectomy due to small, contracted, chronically inflamed and densely adherent gallbladder.

### Terminology

Cholecystectomy: Removing of gall bladder *via* operation. Choledocholithiasis: The position of existing one or more stones in bile ducts. Choledochotomy: Incision which is performed on ductus choledochus. Duodenostomy: Surgical fistula which is performed between duodenum and another anatomic area. Enterolithotomy: Removing of stone from intestine. Hepaticojejunostomy: Anatomises which is performed between ductus hepaticus and proximal jejunum.

### Peer review

The authors investigated the occurrence of internal biliary fistula seen among the patients receiving cholecystectomy due to cholelithiasis. They concluded that laparoscopic cholecystectomy has to be performed in early stage in the patients who were diagnosed as cholelithiasis to prevent from complications like IBF which is seen rarely. IBF suspicion should be remembered during difficult dissection encountered in peroperative period and attention should be paid in order to prevent iatrogenic injuries.

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