

PERFORATIONS OF THE GALLBLADDER AND CHOLECYSTOBILIARY FISTULAE: A REVIEW OF MANAGEMENT AND A NEW CLASSIFICATION

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Experiences with seven cases of gallbladder perforation managed in Kingston, Jamaica, at the District of Columbia General Hospital, and other Howard University affiliated hospitals are presented. The results of a review of 197 consecutive biliary operations at DC General Hospital for occurrence of this entity are presented.

Gallbladder perforation is a complication of cholecystitis in 1 to 4 percent of cases. Niemeier¹ classified this complication in three types in 1934, and currently these are described as type 1—free perforation, type 2—perforation with abscess, and type 3—chronic perforation with cholecysto-enteric fistula.

The gallbladder may, in extremely unusual occurrences, perforate into the biliary tree itself with significant operative implications, and therefore Niemeier's classification can be modified to include cholecystobiliary fistulae formation as type 4. Seven case reports manifesting all four types of perforation are presented, representing the spectrum of current treatment and diagnostic options.

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CASE REPORTS

Case 1

A 24-year-old woman of the Rastafarian faith had a long history of dyspepsia and fatty food intolerance. She was admitted to Kingston Public Hospital, Kingston, Jamaica, on September 10, 1984, with generalized peritonitis. At exploratory laparotomy she was found to have a gallbladder perforation at Hartmann's pouch. No stones were found, and cholecystostomy and drainage were effected. She recovered with antibiotics and supportive care, and was discharged 15 days later with a discharge diagnosis of gallbladder perforation secondary to acalculous cholecystitis. The patient was readmitted 2½ months later with bile still draining from the cholecystostomy site. At the second operation, dense adhesions were encountered, and a cholecystectomy was performed. A dilated common hepatic duct was inadvertently entered and used for a Roux-en-Y common hepatic duct and jejunostomy reconstruction. A postoperative cholangiogram was taken using a T tube left in the common bile duct, which demonstrated a stone in the terminal common bile duct. The patient required postoperative treatment for septicemia, but otherwise recovered uneventfully.

Case 2

A 64-year-old black woman was operated on at Andrews Memorial Hospital in Kingston, Jamaica,

on March 15, 1982. She experienced right upper quadrant pain for two weeks before admittance and was evaluated with an outpatient sonogram (Figure 1) that demonstrated gallstones and a pericholecystic mass consistent with gallbladder perforation. At surgery the gallbladder was perforated at the fundus and was adjacent to a walled-off abscess between the gallbladder, liver, and colon. Uneventful recovery followed cholecystectomy, drainage, systemic antibiotics, and supportive care.

Case 3

A 62-year-old black woman was admitted to the DC General Hospital medical service on July 16, 1982, with uncontrolled diabetes mellitus, chills, fever, and mental confusion. No abnormal physical findings were present initially in the abdomen. After three weeks in the hospital, her alkaline phosphatase level was noted to be increased to 660 U/L. Serum bilirubin and other liver function tests yielded normal results. A computerized tomogram (CT) of the abdomen (Figure 2) demonstrated multiple liver abscesses. Exploratory laparotomy done almost one month after admission demonstrated a hepatic perforation of the gallbladder body associated with gangrene, gallstones, and a large liver abscess with over 700 ML of pus. Cholecystectomy with extensive drainage of the liver and gallbladder bed, systemic antibiotics, total parenteral nutrition, and supportive care resulted in recovery and discharge from the hospital one month after surgery. She has had no further problems.

Case 4

A 66-year-old black man was operated on at Greater South East Community Hospital, Washington, DC, in January 1986. He was known to have moderately severe obstructive lung disease. He presented with a coincidental constellation of diagnoses, including acute gouty arthritis of the left foot, left ureteric colic with the spontaneous passage of a kidney stone through the urethra, and acute cholecystitis manifested by nausea, vomiting, right upper quadrant tenderness, and a right upper quadrant mass on palpation initially thought to be a dilated gallbladder. A slightly enlarged gallbladder, with gallstones and a thickened wall, was demonstrated on a sonogram. The patient responded poorly to parenteral antibiotics and supportive care. On January 9, 1985, he under-

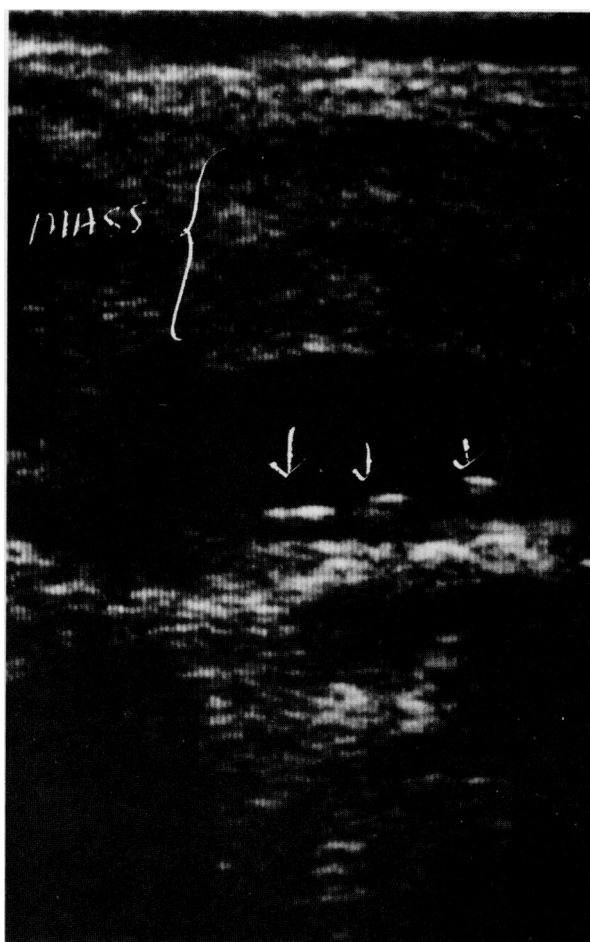


Figure 1. Sonogram of a type 2 perforation demonstrating a gallbladder, stones, pericholecystic hypoechoic areas, and inflammatory mass

went laparotomy and was found to have an inflamed gallbladder that was partly intrahepatic with perforation in its mid-portion. The mass palpated in the right upper quadrant was actually omentum and transverse colon firmly attached to the gallbladder and the under surface of the liver. A cholecystectomy and a cholangiogram taken through the cystic duct were done and the right upper quadrant was drained. The patient made an uncomplicated recovery, and was discharged within one week of surgery.

Case 5

A 77-year-old, white, male, nursing home inmate was admitted with intestinal obstruction secondary to volvulus of the sigmoid colon. The diagnosis of volvulus was confirmed on barium enema. A circular

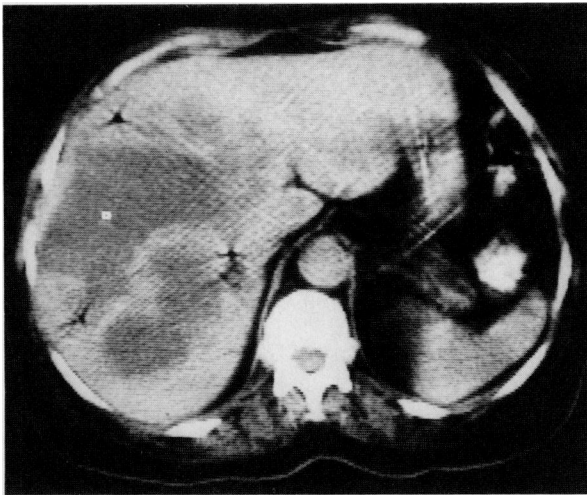


Figure 2. CT of abdomen demonstrating a large multi-locular abscess in a 62-year-old woman with a type 2 hepatic perforation of the gallbladder

radiodense mass was seen on a plain x-ray film of the abdomen in the right lower quadrant (Figure 3). At emergency laparotomy for the volvulus, this mass was found to be a gallstone impacted in the terminal ileum. Dense adhesions were palpated in the area of the gallbladder and the duodenum; they were not disturbed. The twisted sigmoid colon was resected, the rectal end was closed, and the proximal descending colon was converted into an end colostomy. Longitudinal ileotomy was effected in the partially obstructed ileum proximal to the stone, and this was closed in two layers after removal of the stone (Figure 4). The patient had an uneventful recovery and returned to the nursing home.

Case 6

A 68-year-old Chinese-Jamaican woman was operated on at St. Joseph's Hospital, Kingston, Jamaica, in 1981 for obstructive jaundice. She had cholangitis prior to surgery. At laparotomy she was found to have a contracted fibrous gallbladder with a large fistula between the proximal end of the body of the gallbladder and the common hepatic duct. The common duct was dilated and had a single large stone in it. The cystic duct was small and fibrous. Cholecystectomy was completed. The common duct was explored and choledocholithotomy was done through the opening in the hepatic duct. This opening was able to be apposed without tension, and closure by transverse side-side suture without compromise to the lu-

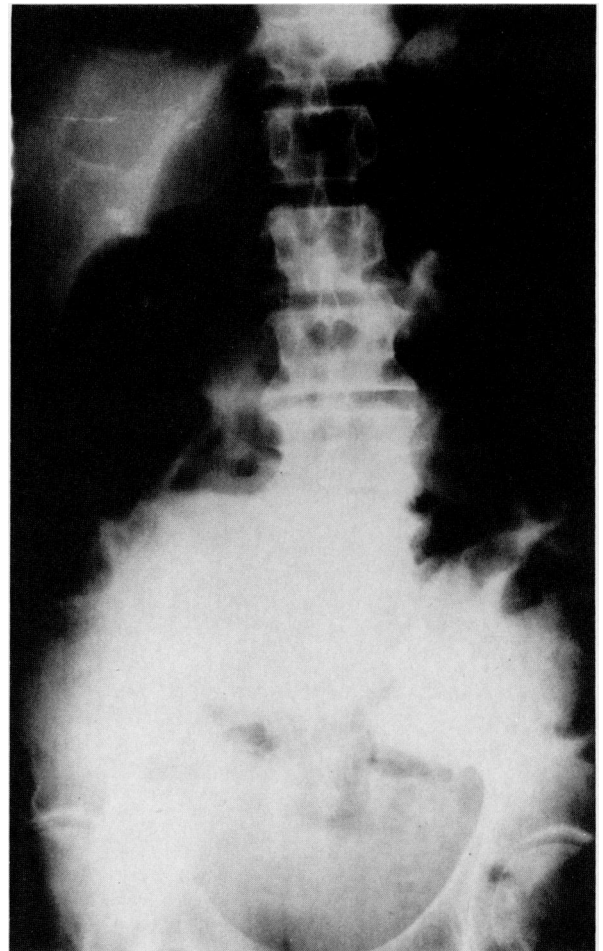


Figure 3. Gallstone in ileum in a 77-year-old man with a cholecystoduodenal fistula (type 3 perforation). He also had a volvulus of the sigmoid colon

men was effected. A T tube was inserted with each of the short limbs inserted in the left and right hepatic ducts, respectively, with the long limb exiting through the common duct below the repair. The patient made an uneventful and full recovery. The T tube was removed three months later after a cholangiogram was taken in the office (Figure 5).

Case 7

A 68-year-old diabetic woman with known glaucoma and schizophrenia was admitted with cholangitis on January 4, 1984. She had a temperature of 103.6 °F and laboratory tests revealed that her bilirubin level was 4.7 mg/dL and she had a white blood cell count of 20,500 mm³. Gallstones were revealed on a sonogram. The patient responded to intravenous



Figure 4. Gallstone of 1½ in was removed from the terminal ileum

antibiotics therapy, and on January 6, 1984, endoscopic retrograde cholangiopancreatography (ERCP) was done (Figure 6). Although not interpreted correctly at first, a cholecystocholedochal fistula and a large stone in the common hepatic duct were demonstrated on ERCP. The operation was done on January 12, 1984, and the findings included a normal size cystic duct and a fibrous, shrunken gallbladder with Hartmann's pouch firmly applied to the common bile duct. In dissecting off the gallbladder, the common duct was inadvertently entered through the fistulous opening. The stone was extracted through this opening, and reconstruction of the biliary tree was completed with a choledochoduodenostomy. The patient's full recovery was uneventful.

DISCUSSION

Perforation of the gallbladder is an infrequent and serious complication of cholecystitis. Duncan² in 1844 is credited with having been the first to report on a case of gallbladder perforation. Niemeier¹ in 1934 reported one case, and first classified this entity



Figure 5. Postoperative cholangiogram three months after repair of a type 4 cholecystobilia fistula in the common hepatic duct. The stenting tube was removed

in three types: type 1, chronic perforation with fistula formation between the gallbladder and another viscus; type 2, subacute perforation in which the gallbladder perforation has been localized as a pericholecystic abscess from the rest of the abdomen by adhesions and adjacent structures; and type 3, acute perforation with generalized peritonitis.

Subsequent reports on this complication by Fletcher and Ravdin³ in 1951 and Roslyn and Busuttil⁴ in 1979 have given credit to Niemeier but have described type 1 perforations as acute perforations of the gallbladder with peritonitis; type 2 as subacute perforation of the gallbladder with abscess; and type 3 perforations as chronic perforations with cholecystoenteric or cutaneous fistula formation.

An even more rare complication of cholecystitis and manifestation of gallbladder perforation is that of cholecystobiliary fistula formation in which a communication between the gallbladder and the common bile duct or its more proximal divisions develops as in cases 6 and 7 in our series. There have been two prior reports in the English literature on this seldom encountered event by Howard et al.⁵ in

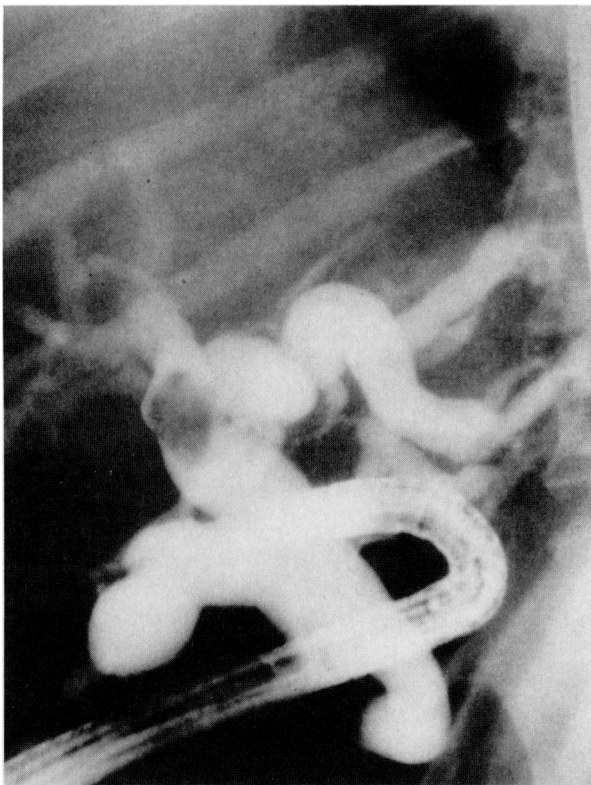


Figure 6. Immediate filling of a shrunken gallbladder through a large cholecystocholedochal fistula (type 4 perforation) is seen on endoscopic retrograde cholangiopancreatogram (ERCP). A large stone is seen in the common hepatic duct. This is the first reported ERCP demonstration of a cholecystobiliary fistula

1951 and Corlette and Bismuth⁶ in 1975. In these two reports there were 23 true cases of cholecystobiliary fistulae formation. The two cases reported here would bring the number of reported cases to 25.

The surgical implications of this unusual occurrence are sufficient, in the authors' opinion, to justify modification for the second time of Niemeier's original classification of gallbladder perforation to include cholecystobiliary fistulae. The following modification of the classifications is proposed (Figure 7): type 1, acute perforation with generalized peritonitis; type 2, subacute perforation with localized pericholecystic abscess and phlegmon; type 3, chronic perforation with cholecysto-enteric fistula; and type 4, chronic perforations with cholecystobiliary fistula formation.

All previously reported cases of type 4 cholecystobiliary fistulae patients including the two cases reported in this paper were jaundiced.

In a remarkable article entitled, "Perforation of the

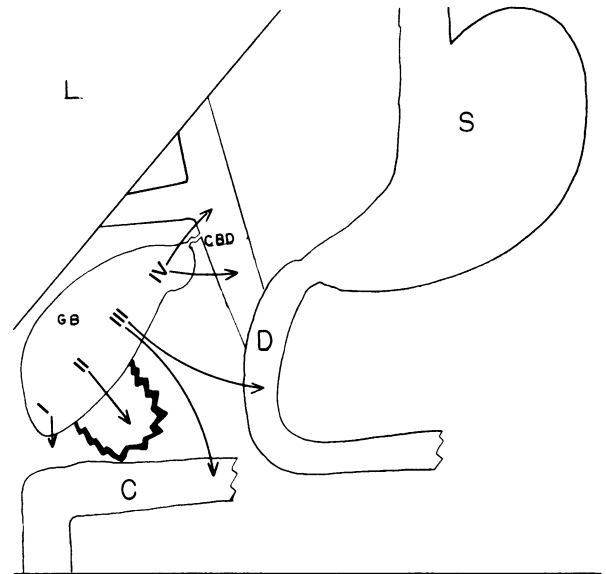


Figure 7. Schematic representation of gallbladder perforations and modified Niemeier's classification: type 1, free perforation; type 2, perforation with abscess; type 3, perforation with cholecysto-enteric fistula; and type 4, perforation with cholecystobiliary fistula formation. C, colon, CBD, common bile duct, D, duodenum, GB, gallbladder, L, liver, and S, stomach

Gallbladder," in the journal in which Niemeier published his classification, Eliason and McLaughlin⁷ reported nine cases of gallbladder perforations in 490 patients admitted for biliary tract disease. His conclusions have stood the test of time and are repeated here:

1. Perforations occur in 1 to 3 percent of biliary cases.
2. Perforations generally occur in patients with a long history of chronic calculous cholecystitis.
3. A correct diagnosis is rarely made prior to laparotomy.
4. Early operation is urged in all cases of acute cholecystitis that do not promptly subside under adequate palliative management.
5. The operative procedure to be done is dependent on the nature of the lesion found at surgery.
6. High mortality is associated with perforation.
7. Unless there is an early contraindication, elective surgery is recommended for calculous cholecystitis.

Gallbladder perforations have been reported to occur with an incidence of 1 to 4 percent of cases with cholecystitis. Fletcher and Ravdin,³ in a study of 2,807 cases of acute and chronic cholecystitis, reported an

incidence of 1.6 percent with the majority of cases being type 2 with associated pericholecystic abscess formation; chronic perforations with cholecystoenteric fistula (type 3) were second in incidence and free perforations (type 1) were the least frequent. This relative frequency of occurrence of each type of perforation seems representative of the reported experiences. The relative mortality associated with each type also seems to be a constant with 40 percent mortality reported for type 1, 4 percent for type 2, and 0 percent for type 3 in the same series. Williams and Scobie⁸ in a review of 496 operated biliary cases out of 4,890 patients with chronic cholecystitis seen at Ottawa Civic Hospital reported a 3.8 percent incidence of gallbladder perforation.

Several authors have postulated as to the pathogenesis of perforation. Williams and Scobie⁸ suggested that mucus secretion and distension followed cystic duct obstruction, and when intraluminal pressure exceeded arterial perfusion pressure, ischemia, edema and subsequent gangrene predisposed to perforation. Fry and colleagues⁹ additionally suggested that tissue invasion by bacteria could result in thrombosis of nutrient vessels and cause gangrene and perforation of the gallbladder wall. Also proposed was the possibility of cystic artery thrombosis as a result of associated infection with consequent gallbladder ischemia, gangrene, and perforation. The gallbladder fundus, with the terminal blood supply of the viscus, is the site most frequently reported to perforate.¹⁰ Roslyn and Busuttill reported several risk factors that included old age, arteriosclerosis, diabetes mellitus, immune suppression, and steroidal drug usage. Lennon and Green¹¹ in their series, found many patients that were elderly had associated cardiorespiratory diseases, but could find no association with other pathology.

A correct preoperative diagnosis is generally not the rule. Roslyn and Busuttill⁴ reported that only 20 percent of cases were suspected before surgery. A high index of suspicion and awareness of the possibility of this complication will likely result in improvement in diagnosis. Several clinico-radiologic features may be relevant:

1. A careful history and physical examination should be undertaken because it is known that these complications occur in greatest frequency in the elderly or in immune-suppressed patients with associated systemic disease and especially in those with a long history of calculous cholecystitis.

2. A palpable right upper quadrant mass associated

with a nondistended gallbladder on sonogram might be an indication as it was in case 4 of our series.

3. Jaundice in a patient with calculous disease should make one conscious of the possibility of a type 4 perforation. This may enhance the interpretation of biliary imaging studies and intraoperative findings and pave the way for optimal reconstruction of the biliary tree.

4. The chest and abdominal x-ray findings of a right pleural effusion and mottled gas or localized ileus in the right upper quadrant may be suggestive of gallbladder perforation.

5. The sonogram, as in case 2, may be helpful. Madrazo et al.¹² described a gallbladder as being distended if it is greater than 3.5 to 4 cm in diameter and with a thickened wall greater than 3 mm. He suggested that a distended gallbladder with edema of the wall may be the earliest sign of impending perforation. A highly suggestive, if not specific, sign of perforation, is the finding of a halo effect around the gallbladder and hypoechoic masses surrounding a poorly defined gallbladder.

6. The CT scan may be the method of choice for evaluating patients suspected of having gallbladder perforation according to Pedroza et al,¹³ who noted that ultrasonography may fail because of increased intestinal gas or pain. He gave the following direct and indirect findings of gallbladder perforation on CT. Direct indicators of perforation by CT scan included the demonstration of either calculi outside the gallbladder or a ruptured segment of the gallbladder wall. Indirect indicators included the demonstration of an abscess outside the gallbladder (as in case 3 with the liver abscess), the presence of gallstones, and thickening of the gallbladder wall.

7. Biliary imaging, either by percutaneous transhepatic cholangiogram or ERCP, may play a significant role in the jaundiced patient with type 4 perforations. The diagnosis in case 6 of our series was clearly made by ERCP, although the correct interpretation was retrospective and postoperative. This reinforces the adage that "you won't see it if you don't think it." This case is the only reported one at the time of writing of cholecystobiliary fistula (type 4) perforation demonstrated on ERCP.

8. Exploratory laparotomy may be necessary if none of the above findings is present in a particular case. Of principal importance, however, is the doctor's recognition of an acute abdomen and prompt laparotomy on those patients who fail to improve on an appropriate nonoperative regimen that includes na-

sogastric decompression, intravenous fluids, and systemic antibiotics. In this vein, I am forced to recall the admonition of O. W. Hoffer, premier surgical teacher and technician, to a second year resident on morning rounds at Norfolk Community Hospital, Virginia after a long night of well intended procrastination on a male patient with vague right lower quadrant tenderness, that "an open abdomen keeps no secrets."

At operation the proper procedure to be done is determined by local findings and the patient's general condition. Proper drainage is practically always necessary and is advised routinely. Cholecystectomy is preferable and desirable when it can be safely conducted. Cholecystostomy and drainage may be the procedure of choice, especially in type 1 cases in which the patients are most often physiologically compromised and would not tolerate well extensions of surgery and anesthesia at the initial operation.

In type 4 cases of cholecystobiliary perforations, cholecystectomy and choledocholithotomy are effected. Reconstruction of the biliary tree may include duct enterostomy as in case 7 or simple closure over a T tube as in case 6. Corlette and Bismuth⁶ have advised leaving a piece of gallbladder wall in place for closure without obstruction.

A review of 197 consecutive cholecystectomies at DC General Hospital over an eight-year period revealed a 1 percent incidence of gallbladder perforations. The average age of these patients was 50.9 years with 70 percent females and 30 percent males.

Among the seven patients reported with gallbladder perforations, the average age was 61 years, 10 years above the general population of our patients undergoing cholecystectomy. The sex ratio in perforated cases was the same as in the nonperforated cases of cholecystitis, 71.5 percent female and 28.5 percent male. The correct diagnosis was made preoperatively in only two of seven cases and should have been made in two additional cases. There was no mortality and no long-term morbidity in this series of gallbladder perforations.

CONCLUSIONS

Gallbladder perforation is an infrequent but important complication of cholecystitis occurring in 1 to 4 percent of cases. Cholecystobiliary fistula formation is occasionally a manifestation of perforation with important therapeutic implications. The authors have classified this complication as type 4 in a second modification of the Niemeier classification.

Gallbladder perforation tends to occur in an older subset of patients with gallbladder disease. These patients often have associated chronic medical illnesses. The plain x-ray film, abdominal sonogram, computerized tomogram of the abdomen, and biliary imaging by endoscopic retrograde cholangiopancreatogram or percutaneous trans-hepatic cholangiogram in the jaundiced patient may, at times, complement the history and physical examination and ultimately facilitate the timing of operative intervention and choice of procedure. A high index of suspicion and awareness of the possibility of this complication could likely result in an improvement in preoperative diagnosis.

With current surgical techniques, antibiotics, and supportive measures, gallbladder perforations can be treated successfully with little greater morbidity and mortality than other patients with cholecystitis.

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