

Spontaneous Internal Biliary Fistula, Obstructive and Nonobstructive Types:

Twenty-Year Review of 55 Cases

OSCAR H. PIEDAD, M.D., PHILIP B. WELS, M.D.

From the Department of Surgery, State University of New York at Buffalo and Millard Fillmore Hospital, Buffalo, New York 14209

REFINEMENTS in radiologic technics, leading to earlier diagnosis of asymptomatic gallstones and prompt referral of these patients to the surgeon, have resulted in a decreased overall incidence of spontaneous internal biliary fistulas,^{9, 36} even though, as Porter *et al.*²⁶ put it, more are being found preoperatively today. However, diagnostic awareness of the possibility of bilioenteric fistula leaves much to be desired, and its presence is still too often first diagnosed at operation.

No large series of spontaneous internal biliary fistulas have been reported since those of Judd and Burden¹⁹ in 1925 (153 cases), Kehr²¹ (100 cases) and Marshall and Polk²² in 1958 (41 cases).

We therefore decided to review all cases of spontaneous internal biliary fistula encountered at the Millard Fillmore Hospital in the 20-year period from 1950 to 1970.

We arbitrarily divided the fistulas into two types which we termed "nonobstructive" and "obstructive," because, although gallstone ileus usually is a complication of the former type, the two clinical entities are distinct, as are also their treatment and outcome. We further compared the cases in the two decades represented: 1950 through 1959 and 1960 through 1969, particularly as to preoperative diagnosis, sequelae and mortality.

Present Study

Material

Of a total of 55 patients with spontaneous internal biliary fistula in the period studied, 39 were seen in the first 10-year

period and only 16 in the past 10 years. Gallstone ileus comprised the 19 total obstructive cases, 12 occurring in the first and seven in the second decade (Table 1).

Age and Sex. In the nonobstructive type, the average age was 60 years; in the obstructive type, 67 years. Over 85% of the total patients were 65 years of age or over (range 48-83). Typically, females predominated, in the ratio of 4:1.

Past History. Of the total patients 24, or 44% (18 with the nonobstructive and 6 with the obstructive type), had a history of previous gallbladder disease. Interestingly, of 33 patients who had emigrated to this country from Europe, 22 initially denied any history of gallbladder disease, which could be attributed partly to a language barrier, and at first refused surgical treatment.

Preoperative Diagnosis. From Table 1 it is evident that there was improvement in the preoperative diagnosis in both types in the 20-year period studied (from 23% to 56%). In the nonobstructive type the diagnostic accuracy was only 19% as compared to 63% in gallstone ileus, emphasizing the difficulty of diagnosis in the former type. In six of the 36 cases fistula was suspected by the abdominal scout film and confirmed by x-ray study with contrast media. In the gallstone ileus, or obstructive type, 10 of the 12 total diagnoses were made on plain radiographs of the abdomen; the remaining two were made clinically. One was a recurrent gallstone ileus and the other emesis of the gallstone.

Associated Diseases. Forty-four patients (80%) had previously diagnosed associated

TABLE 1. *Spontaneous Internal Biliary Fistula—55 Cases*

Type	Nonobstructive		Total Cases	Obstructive		Total Cases
	1950-1959	1960-1969		1950-1959	1960-1969	
Period of study	1950-1959	1960-1969		1950-1959	1960-1969	
No. of cases	27	9	36	12	7 (incl. 1 recurr.)	19
No. preoperatively diagnosed	3 (11%)	3 (33%)	6	6 (50%)	6 (86%)	12
Deaths	4 (15%)	1 (11%)	5	4 (33%)	1 (14%)	5
Causes of death	*Pancreatitis *Ext. bil. fistula Myocard. infarction Pneumonia	*Common-duct injury		Diabet. acidosis Electrolyte imbal. Pul. embol. Pneumonia	Re-obstruct. due to adhesions	

* Attributed to the surgical procedure.

disease, 25 of which were arteriosclerotic cardiovascular disease; this was followed by pulmonary disease and diabetes.

Morbidity. Wound infection was the most common postoperative complication, followed by partial wound dehiscence, pulmonary infections, atelectasis, and lastly electrolyte imbalance.

Mortality. In 1950-1959, four of the eight deaths were attributed to cardiopulmonary complications. One patient in severe diabetic acidosis died while being prepared for operation. In 1960-1969, the only death in the nonobstructive type was attributed to technical error, and the only one in the obstructive type was caused by re-obstruction due to adhesions.

Discussion

Nonobstructive Type

Spontaneous bilioenteric fistula is produced most frequently by gallstones (approximately 90%¹⁷), followed by peptic ulcer (6%), and the remainder (4%) by malignancy or trauma.¹⁵ Of the 55 cases in the present series, 53 were caused by chronic cholecystitis with cholelithiasis and two by duodenal ulcer. As soon as an internal fistula is formed, the patient is relieved of the distress from common-duct obstruction, a "fortunate accident" says Dean.⁷ Halsted,¹⁶ however, states that nature's methods are "very crude and at-

tended with much suffering and great danger, immediate and remote," his inference being that the fistula produced is a potential source of complications, among them gallstone ileus.

The most common communication (69-70%) is said to be cholecystoduodenal,^{17, 26} followed by cholecystocolonic (14%) and cholecystogastric (6%).²⁶ In our series the type of fistula was ascertained only in the nonobstructive type (Table 2).

Sequelae of the nonobstructive type of biliary fistula are ascending cholangitis, particularly when the colon is involved⁵; gallstone ileus⁶; loss of weight³⁴; malabsorption syndrome,¹¹ gastrointestinal hemorrhage (rarely),^{18, 20} and development of carcinoma.¹ Bossart *et al.*³ found the incidence of malignancy in biliary fistula to be 15% as compared with 0.8% of gallbladder carcinomas noted in cholecystectomies.

Obstructive Type

The incidence of gallstone ileus is generally accepted to be approximately 2% or less of all mechanical bowel obstructions, but is significantly higher (23-24%) in patients over 65 years of age.^{15, 23}

The 19 cases of gallstone ileus in the present series represented 0.38% of all (4,897) cases of mechanical bowel obstruction surgically treated in this hospital

in the 20-year period under study and 34.5% of the spontaneous internal biliary fistulas in our series.

It is commonly stated that the gallstone must be over 2.5 cm. in diameter to produce symptomatic ileus.¹² (Turner³³ described a stone measuring more than 17 cm.) The gallstone passes through the fistula as soon as it is in the lumen of the viscus, and usually proceeds distally. Occasionally it proceeds in a proximal direction and in these instances it may be vomited; or sometimes it becomes impacted at the pylorus, producing duodenal obstruction. In 80–90% of cases the stone will migrate with peristalsis and be passed spontaneously.²⁹ This is supported by the finding that in a series of 104 cases of spontaneous internal biliary fistulas the incidence of gallstone ileus was only 14%.⁶

If the stone becomes impacted it is most often found at the terminal ileum, less often at the jejunum. In our series 63% were impacted at the terminal ileum and 26% at the jejunum (Table 2); in all of the latter there were adhesions between the jejunum and abdominal wall. In these latter patients a wrong preoperative diagnosis was made—the abdominal surgical scar being the culprit. The presence of a surgical scar does not eliminate the possibility of gallstone ileus. On the other hand, it should stimulate awareness to the possibility of this diagnosis, especially in the elderly patient with no history of cholecystectomy. Noskin and Tannenbaum²⁴ described an interesting case of a gallstone palpated through an incisional hernia. A stone occasionally can be impacted at the sigmoid colon, especially if the colon is diseased.²⁷ In our series there was one case of sigmoid colon obstruction (Table 2).

A controversial mechanism is the migration of a small stone through an intact common duct to produce eventual obstruction,^{18,31} a mechanism not accepted by some.³⁴ Warren and Swan³⁵ reported a small-bowel obstruction produced by gallstone in the absence of a gallbladder.

TABLE 2. *Type of Fistula and Location of Obstruction: 55 Cases*

Nonobstructive		Gallstone Ileus	
Type of Fistula No. of Cases		Location of Obstruction No. of Cases	
Cholecysto- duodenal	22 (61%)	Terminal ileum	12 (63%)
Cholecysto- colonic	6 (17%)	Jejunum	5 (26%)
Multiple fistulas			
Cholecysto- duodeno- colonic	4	Pylorus	1
Cholecysto- choleodocho- duodenal*	3	Sigmoid colon*	1
Cholecysto- gastrocolonic	1	—	
Total	36	Total	19

* In the three cholecystocholeodocho duodenal fistulas, two were caused by duodenal ulcer. The sigmoid colon obstruction occurred in a patient with diverticulosis.

Diagnosis

1. Nonobstructive Type

A history of biliary-tract disease is more readily elicited in the nonobstructive type, as the acute symptoms of bowel obstruction often mask those of the biliary tree. In the present series 18 of 36 patients with the nonobstructive type had such a history.

The symptoms of this type are nonspecific and similar to those of any chronic biliary-tract disease. Although the cholecystoduodenal fistula seldom produces ascending cholangitis, it is said to be the most difficult to diagnose and also the one that most frequently produces gallstone ileus.⁸ Jaundice was observed in 11 of 15 of Puestow's²⁸ cases. Physical findings are not remarkable; however, a vague fullness can be palpated in the right upper abdominal quadrant which represents the fistula and adhesions. This was present in 12 of the 36 patients in our series.

In general, preoperative recognition is more difficult in the nonobstructive type. In the series of Judd and Burden¹⁹ in 1925 only two out of 153 cases were diagnosed preoperatively. In the present series only six of 36 cases were diagnosed before surgery; the remaining 30 cases were found

incidentally in a total of 4,720 cholecystectomies performed in this hospital in the past 20 years. Kehr²¹ is often quoted as finding 100 fistulas in 2,000 cholecystectomies.

The presence of air in the biliary tree is suggestive of a diseased biliary system but not diagnostic of fistula. In the 267 cases of biliary fistula reported by Borman and Rigler² 30% revealed air in the biliary tree. The x-rays in the six nonobstructive cases in our series that were preoperatively diagnosed showed air in the biliary tree. This finding is often recognized only in retrospect.

The most important diagnostic roentgen finding is reflux of a contrast media into the biliary tree. In our rare case of cholecystogastrocolonic fistula, the fistula was demonstrated by x-ray examination with barium enema but was not seen in the upper gastrointestinal tract series.

2. Obstructive Type

The clinical picture of gallstone ileus is not unlike that of any mechanical bowel obstruction. During migration of the stone, symptoms are minimal or absent. Thus the onset is insidious, occasionally acute. The stone can produce intermittent obstruction due to the "tumbling" or migrating phenomenon.^{6, 12} Recurrent, incomplete bowel obstruction can be present for weeks or months before complete blockage occurs. Drucker¹⁰ reported a latent period of 9 months. The average duration of complete obstruction in previous reports ranged from 4.5 to 7 days.^{6, 15} In the present series the average was 5 days.

A plain radiograph of the abdomen, as fully described by Rigler, Borman and Noble,³⁰ is the most helpful. Air in the biliary tree is significant and has been reported in up to 93% of cases in a retrospective study,³⁰ which implies lack of awareness preoperatively. In our series the x-rays in eight, or less than half of the 19 cases of gallstone ileus, showed air in the biliary tree; there was no retrospective review of the films.

Another significant x-ray finding is visualization of an ectopic radiopaque stone that varies in location. This was demonstrated in seven of 10 cases in our series which were diagnosed by x-ray study. Drucker¹⁰ showed the importance of barium swallow x-ray in the diagnosis of gallstone ileus. Haffner *et al.*¹⁵ found that when all these x-ray findings were considered, a diagnosis could be achieved in 85% of cases.

Treatment

1. Nonobstructive Type

As the majority of nonobstructive cases are found incidentally in the operating room, if one is not aware of the disturbed anatomic features, operation is likely to be a difficult and dangerous procedure.

The anatomic features of nonobstructive biliary fistula of greatest surgical importance are: (1) the presence of dense adhesions in the right subhepatic area, (2) a small fibrotic gallbladder adherent to a viscera, and (3) a dilated common duct. As the common duct also may be obstructed,^{14, 17} it is mandatory positively to identify and explore the duct before excising the fistula and the gallbladder.

We find intraoperative cholangiograms most useful in identification of the important structures as well as in confirming choledochal obstruction. The gastrointestinal tract should be meticulously explored, especially if the stone is faceted. Bowel preparation is important, particularly when the colon is involved. If the fistula is between the common duct and the duodenum, a duodenal ulcer is "almost always" present according to Porter *et al.*²⁶ In our series subtotal gastrectomy was done for this type of fistula; others have used vagotomy with drainage.

Internal biliary fistulas of the nonobstructive type are nearly always managed by a single-stage surgical procedure, as were the 36 cases in our series. Hicken and Coray,¹⁷ however, perform a preliminary decompression of the colon or common duct.

In the present series the case fatality rate decreased from 14.7% in 1950–1959 to 11.1% in the past 10 years. (Marshall and Polk in 1958 cited a rate of 12.5%.²²) The single fatality in the latter period was due to transection of the common duct, which was followed by rapid progression to liver failure. This technical catastrophe could have been avoided by the use of intraoperative cholangiography. Three of the five total deaths were directly related to surgery. The authors, in a previous publication,²⁵ pointed out safeguards in the surgical approach to spontaneous internal biliary fistula.

2. Obstructive Type

The primary surgical treatment, as in any mechanical bowel obstruction, is laparotomy. Moore and Baker,²³ however, described a nonoperative method by flooding the terminal ileum with barium in poor-risk patients. Correction of the fluid and electrolyte imbalance, restoration of any metabolic derangements, and the use of intestinal intubation are essential preparations for surgery.

When the obstructing stone is located, an attempt should be made at milking it into a healthy bowel, followed by enterolithotomy, but often this is not possible because of the edema, inflammation, and spasm that exist at the site of impaction. The rest of the bowel should be explored for the presence of another asymptomatic stone. Tuell³² found more than one stone large enough to obstruct the bowel in 14% of 500 cholecystectomies. If another stone is detected near the site of the enterotomy, this should be gently milked to the incision. If the stone is in the sigmoid colon, one should attempt carefully to milk it toward the rectal ampulla and then extract it from the rectum.¹²

Some use a single-stage surgical procedure for treatment of gallstone ileus in certain cases.^{1, 6, 12} In this procedure, excision of the fistula and cholecystectomy are performed in addition to enterolithotomy. Fraser¹³ advises only cholecystostomy and enterolithotomy whenever feasible.

The authors prefer the two-stage procedure, as the majority of these patients are elderly, precariously sick, and debilitated, and we believe they are better able to tolerate a shorter operative procedure. Also, removing the gallbladder and fistula could be technically difficult and tedious because of obesity and the presence of severe, dense adhesions in the right upper abdominal quadrant. We believe it is preferable to do the cholecystectomy and excision of the fistula at a later date when conditions are more favorable. However, in the present series only 28% of the patients returned for the second procedure and they are presently alive and well; the remaining 10 either refused operation or were lost to follow-up.

It is generally agreed that the fistula and gallbladder should be removed to prevent recurrent ileus (5–10%^{4, 23}) and other sequelae of the fistula. In some instances the fistula may close spontaneously provided it is small and there is no distal obstruction of the common duct.²⁹ Deckoff⁸ emphasized the importance of further x-ray studies to demonstrate the fistula prior to the patient's discharge.

The mortality rate of gallstone ileus has decreased tremendously in the past half-century (from 75% prior to 1925 to 28% in 1960),²⁹ and to less than 15% in the past decade.⁶ The mortality is, however, still too high and ways must be found further to reduce this.

Summary and Conclusions

A series of 55 cases of spontaneous internal biliary fistula encountered at the Millard Fillmore Hospital, Buffalo, N. Y., in the 20-year period, 1950 to 1970, is presented.

The cases were arbitrarily divided into nonobstructive and obstructive (gallstone ileus) types, and the preoperative diagnoses and mortality rates for both types in the two decades studied were compared. Both showed great improvement in the latter decade, which is in agreement with the recent literature.

We found a high incidence of spontaneous internal biliary fistula among the immigrant patients. This might be explained by their "conservative" attitude toward surgical treatment for asymptomatic gallstones. For this reason we believe gallstone ileus should always be considered in the differential diagnosis of bowel obstruction in these patients.

In an effort further to reduce the death rate and decrease the morbidity in spontaneous internal biliary fistula, the following points are emphasized.

For the nonobstructive type: Better understanding of the anatomic distortions, leading to refinements in operative technic.

For the obstructive type: Earlier diagnosis of gallstone ileus and earlier surgical treatment.

Finally, the ultimate answer to the prevention of internal biliary fistula formation is cholecystectomy, whenever feasible, in all patients in whom gallstones are present.

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