

“Spontaneous biliary-enteric fistulas (SBEF) : an update

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Abstract

Spontaneous biliary-enteric fistulas (SBEF) can be defined an abnormal communication between the biliary tree and the gastrointestinal tract which develops as complication of a biliary or gastrointestinal disease.

In almost 90% of cases the cause of SBEF is chronic recurrent cholelithiasis. Less common causes are penetrating peptic ulcers and neoplastic infiltration from the biliary or gastrointestinal tract

The most common type of SBEF is cholecystoduodenal fistula and the least common is choledochoduodenal fistula. There are various complications associated with SBEF but often these are not early or timely identified and diagnosis and treatment may be delayed for years.

The most known complication, which can be considered pathognomonic for many SBEF, is gallstone ileus, either case of acute or chronic intestinal obstruction.

Gallstone ileus features a rather rare complication of a rather common pathology, that means biliary lithiasis, which is mainly found in women over the age of 65, with a male to-female ratio of 1:5

There are various forms of gallstone enteric obstruction, related to the sites of gallstone impaction, with various clinical characteristics and degrees of severity. These include Bouveret syndrome (<10% of cases) which occurs in bilioduodenal or biliopyloric fistula, and the more common Barnard’s syndrome (5-75% of cases) in which the site of impaction is in the terminal ileum right before Bauhin’s valve

For diagnosis the radiological signs which make up Rigler’s triad (pneumobilia, small bowel obstruction, ectopic calcified gallstone usually in the right iliac fossa) or tetrad (dilated loops), and are best visible on magnetic resonance imaging are essential, but the gold standard is contrast-enhanced computed tomography scan.

Surgical management, possible emergency surgery for the oldest patients and in severe obstruction, is one-stage simple enterolithotomy, currently more easily performed in video-laparosurgery, when possible.

Treatment of the obstruction is the essential, together with video-laparoscopic cholecystectomy. leaving the SBEF to a second stage surgery after a few weeks.

In more favorable cases radical treatment of the occlusion, of the SBEF and the source of the biliary lithiasis as one-stage procedure is recommended.

Introduction

Spontaneous biliary-enteric fistula (SBEF) is an abnormal communication between the biliary tree and the gastrointestinal tract which develops as a result of biliary or gastrointestinal disease. Iatrogenic fistulas, due to surgery or heavy handed instrumental exploration, are not included in this definition.

R.Colombo, in 1559, was the first to describe SBEF as an occasional finding during an autopsy, but it was J. Cruvelhier, who, almost three centuries later (1829), provided the first, nearly complete classification of SBEF. In 1890 L. Courvoisier published the first multicenter study on SBEF and in 1965 H.Mondor established the definitive criteria for clinical assessment and diagnosis of this pathology (1,2)

The etiology of SBEF is extremely varied. The most common cause is a complication of biliary lithiasis and the second most common is penetrating peptic ulcer. The percentage of SBEF due to penetrating ulcers has steadily decreased over the years, from 50% in early studies to 3% today (3). Other conditions involved in the development of SBEF are biliary inflammation and perihepatic abscess. Less frequently, neoplasia and trauma are direct causes of SBEF (4)

SBEF is more commonly found in women than in men. In fact, given the decreasing number of SBEFs caused by peptic ulcer disease, the gap between the sexes is increasing.

Pathophysiology

There are different pathophysiological mechanisms involved in the development of SBEF, depending on the primary pathology.

In the case of biliary lithiasis, inflammation, if it is localised or even more so if it evolves into a perihepatic abscess, is a key step towards SBEF.

After trauma, internal bleeding or transmural hematoma, can be the *primum movens*. If the initial cause is an ulcer, in particular an ulcer on the posterior wall of the first part of the duodenum, the mechanism is penetration of the ulcer. In the case of neoplasia the fistula is the final result of a process that begins with adhesions and sclerosis and then reaches the phase of infiltration and penetration.

The pathophysiological process which begins with impaction or adhesions, enhanced by inflammation and the toxic effect of bile pigments on tissues,

inevitably leads to infiltration followed by erosion, intramural thrombosis, ischemia and ultimately necrosis (3).

Morphologically, a SBEF is “direct” if there is a close entero biliary connection with mucosal continuity and “indirect” if there is a tortuous tract, often with sacculations containing purulent fluid, between the two anatomical entities

The fistula orifice in the biliary tree is often rather small and located in the anterior part of the gallbladder or the posterior wall of the main bile duct behind the duodenum. The fistula orifice in the bowel is easier to see, normally 1-2 cm in diameter and located to the pylorus, to duodenal bulb or the right colon (5). The most common type of SBEF is the cholecystoduodenal fistula (70% of all SBEF). Other types are the cholecystogastric fistula (0-13.3%), cholecystocolic fistula (0-10.9%), cholecysto-ileal (0-2.5%), and the much less common choledochoduodenal fistula (6)

Symptoms and complications

Symptoms are entirely nonspecific. Usually patients present with a history (1-10 years) of abdominal pain, fever, jaundice, vomiting, diarrhea and crisis of intestinal occlusion (generally isolated episodes that resolve spontaneously).

However, a careful history taking can detect, even in a long clinical history, the acute episode that led to fistulization. Typically this is an episode of especially severe pain with high fever, chills, and subicterus or jaundice followed by diarrhea (3).

According to various authors fistulization is associated with anemia due to bleeding from the fistula tract, maintained by sclerosis, erosion and the anticoagulant activity of bile (1,7)

Other authors state that fistulization, with expulsion of small stones and purulent material, can result in unexpected and often lasting improvement of a patient's symptoms, which explains why in many cases SBEFs elude diagnosis and are only discovered during autopsy(8).

There are various, often severe, complications associated with SBEF. Very often an acute complication leads to the diagnosis of these fistulas. The most frequent complication, which, however, does not always result in hospital admission and may therefore remain unrecognized, is recurrent cholangitis. This consists of repeated episodes of fever, even hyperpyrexia, chills and subicterus or jaundice which often resolve after self-prescribed medical therapy and fasting, only to recur later on, sometimes after a long time interval. The contamination by intestinal contents which come through the fistula, is especially serious in cases of fistulas between the biliary tree and the colon. Not infrequently in these cases biliary inflammation leads

to the formation of an intra- or perihepatic abscess.(1-2). Cholangitis develops in 20-60% of patients with SBEF.

The incidence of gastrointestinal bleeding, is more often reported in older studies, when peptic ulcer was more common, and has fallen from 50%, in the 1960s-70s, to 13% today. The mechanism is now predominantly stoneimpaction, although peptic ulcer disease, which can cause severe bleeding, should be kept in mind (9).

Hemobilia occurs in 1-5% of SBEF, especially in those due to lithiasis. The classic Quincke's triad (jaundice, melena and upper abdominal pain) is not always present but if present is often decisive for a diagnosis of SBEF.

Peritonitis is rare, occurring in ca.1% of cases. It is most commonly localized and associated with the loss of direct communication between the two parts of the fistula or with a possible leak along the fistula tract.

The most common and most severe complication, which is virtually pathognomonic for SBEF, is gallstone ileus. It occurs in 0.5-45% of cases and is, obviously, linked to the percentage of SBEF due to lithiasis. (10)

Gallstone ileus

Gallstone ileus is a mechanical intestinal obstruction caused by a gallstone that has migrated into the gastrointestinal tract and become impacted.

The first description of this condition has been attributed to T.Bartholin, who, in 1654, published an autopsy study in which there was one case of gallstone ileus.

Gallstone ileus is a rather rare complication of a rather common disease: it occurs in 0,000015% of hospitalized patients but 0,0003 % of surgical patients. It is most common in patients over age 65, especially women (male-to-female ratio 1:5) (11).

It is a complication of biliary lithiasis in only 0,4% of cases overall but in 3% of patients over 65. It is the cause of 1.5% of cases of mechanical intestinal occlusion but the incidence reaches 25% in patients over 65.(12)

Overall, the mean age of patients with gallstone ileus is 74 years and the male-to-female ratio is 1:3-1:7.

In spite of the increased use of laparoscopic surgery for cholelithiasis, gallstone ileus has not become less common no doubt because of the ageing of the population and the accompanying chronic diseases. In fact its incidence has risen to ca. 10% in the Unites States and Western countries(13).

The multiple comorbidities and great fragility of these patients explains the high morbidity rate associated with gallstone ileus which has remained 7-30% over the years (14)

As regards the impaction of the stone, there are various forms of occlusion with varying clinical characteristics and varying degrees of clinical severity

Starting from the proximal end of the gastrointestinal tract, there is a gastroduodenal form (<10% of cases), Bouveret syndrome (first described by Bouveret in 1896), which involves impaction in the pylorus or proximal duodenum causing gastric outlet obstruction.

The site of impaction can also be in the proximal ileum (20-40%) or in the distal ileum, which is without doubt the most common site (50-75%). The latter, Barnard's syndrome, is due to the reduction in the size of the lumen right before Bauhin's valve and the chronic parietal lesions in this part of the intestine and causes obstruction of the terminal ileum.

There is also a much less frequent form (2-8%) in which the site of impaction is in the colon, and this is called "gallstone colic" by some authors (15)

As for the characteristics of the occlusion, the most important factor is the size of the obstructing stone, which is usually 2.5-3 cm in diameter but can be 4.5 cm or more especially in cases of proximal obstruction. Moreover, very often more than one stone is involved. In fact, Noriega-Maldonado and colleagues have reported that more than 2 stones were observed in the intestinal lumen in 22% of cases (16).

Other factors for gallstone ileus should be kept in mind. The caliber of the intestinal lumen may be reduced, due anatomical factors or disease, especially chronic inflammatory disease and neoplasia, or due to a former iatrogenic injury. Another common contributing factor is parietal edema which is sometimes pre-existent but is often due to the response of the mucosa to the abrasion and irritation caused by an impacted stone. Changes in intestinal motility caused by the presence of the stone should not be overlooked. Peristalsis pushes the stone forward but when the stone stops moving, due to a reduction in the size of the lumen or some obstacle, this gives rise to an antiperistaltic movement which may move the stone backwards. This back and forth motion can be repeated over and over resulting in intermittent subacute occlusion, (i.e. the occlusion seems to resolve spontaneously only to recur shortly afterwards), which is called "tumbling obstruction" by Anglo-Saxon Authors. (17)

In this complex physiopathological mechanism a factor that assumes great importance is the time it takes the gallstone to move along the gastrointestinal tract. Although many small gallstones that pass through a biliary enteric fistula are expelled without producing clinical significance. Medium sized stones tend to increase in size during their trip through the intestinal lumen which can take several days, because intestinal material accumulates on their surface turning them into stones large enough to obstruct the lumen. In many cases, if a recovered stone is cut

in half, one can see a central nucleus, the primary gallstone, which is different for color and consistency density from the rest, wrapped with a more friable, darker rind made up of enteric material and whose thickness directly proportional to the time of the stone sliding through the intestinal lumen (10)

Current problems

To date there are still questions to be resolved regarding the management of gallstone ileus.

First, a more modern approach is required: evaluation of patients should be more rapid and individualized, the diagnostic pathway should take advantage of modern technology— mainly CT and MNR - and treatment strategies should take advantages from new methods of stone retrieval

Unfortunately patients with gallstone ileus are still not hospitalized promptly enough. There is a delay of 4-7 days and this results in an average delay in treatment of 3.7 days (range 3-8 days), which has a negative effect on the prognosis.

Twenty percent of these patients have had prior episodes of cholecystitis but only 50% are aware that they have biliary lithiasis and 25% have not been diagnosed. In 35% only a clinical diagnosis is possible (2).

Gallstone ileus can be acute, i.e. classic obstruction, or subacute if there is a clinical picture of subocclusion or chronic occlusion (Karewsky syndrome) characterized by recurrent episodes of pain due to the passage of stones lodged in the intestine for a long period of time (18)

Typical symptoms are abdominal pain, present in 100% of cases, vomiting that is sudden and unrelated to food intake, hyperperistalsis meteorism and abdominal distension. (19)

Bouveret syndrome (gastric outlet obstruction) is associated with uncontrollable vomiting and at times hematemesis due to lesions of the gastric mucosa (similarly to Boerhaave's syndrome) This syndrome can be easily diagnosed, in 75% of cases, with endoscopic assessment and computed tomography (CT) scan, and the diagnosis is strengthened by the presence of gastric dilatation and an inflammatory mass under the right diaphragm.(20)

At the first visit it is important to carefully evaluate each patient's risk factors: notably age, clinical status, and presence of subocclusion or occlusion, keeping in mind that 4% of patients experience spontaneous resolution of their symptoms that may be definitive, and that in an emergency setting preoperative diagnosis is possible in 24-73% of cases (2).

The American Society of Anesthesiologists (ASA) class of these patients must be determined because they are usually fragile individuals with various comorbidities such as electrolyte imbalance (70%), renal insufficiency (50%) overt systemic inflammatory response syndrome (SIRS) (40%) and liver disease (15%)(4).

Diagnostic imaging

Diagnostic imaging is the key to a rapid diagnosis with only minimal discomfort for the patient.

Plain abdominal X-ray is an important (and less costly) first step to confirm the diagnosis of small bowel obstruction specially in elderly patients with abdominal pain and vomiting without clinical evidence of strangulation such inguinal hernias or previous surgeries. (21)

X-rays, especially with contrast studies, make it possible to demonstrate pneumobilia (the Grotta -Mentschlersign) and evidence of occlusion, as well as indirect and direct signs of the location of the gallstone and the biliary- enteric fistula (22)

Rigler's triad (pneumobilia+ectopic calcified gallstones+abdominal distentionfor dilated ileal loops), which is pathognomonic for gallstone ileus, is seen on plain x-rays in 17-87% of patients.

However only 10 % of gallstons are sufficientlycalcified to be visualized radiographically. (22)

Two out of the three of these signs, present in 50% of cases are considered sufficient for a diagnosis. Rigler's tetrad, i.e. Rigler's triad with evidence of a change in the position of the stone on a repeat x-ray,appears in 35% of cases. (23)

Ultrasound is very important toconfirm the presence of pneumobilia, abdominal effusion and ectopic gallstones : in association with X-ray achieves a sensitivity of the 74 % (24)

Contrast-enhanced CT scan is still the gold standard for the diagnosis of this pathology. It has 98% sensitivity and 100% specificity for pneumobilia and free fluid and can also reveal what type of SBEF is present and the actual site of stone impaction. The only defect of CT scan is underestimation of the size of the stone. (20)

MRI is currently the gold standard for identification and characterization of the SBEF and has a specificity of 100% for Rigler's triad. (23)

Endoscopy is essential in cases of Bouveret syndrome, not only for diagnosis but also during treatment.

Some recent publications have described the use of endoscopic removal after lithotripsy of obstructing gallstones located at the ileocecal valve in order to avoid surgery (25)

Treatment strategies

The treatment strategy should take into account various factors.

A key factor is the site of impaction. In addition the age and clinical condition of each patient should be carefully evaluated and any electrolyte imbalances and metabolic abnormalities should be treated and homeostasis re-established.

For patients who remain in critical condition every solution that avoids surgery should be considered. However if it is at all possible, with reduced risk, a radical surgical approach should be used although it should be kept in mind that minimally invasive surgery can be an option.

As regards surgical timing, the procedure should be considered delayed urgent surgery, to be performed after adequate metabolic homeostasis has been re-established. Emergency surgery should be reserved for the rare critical cases of hemorrhage, perforation and biliary sepsis.

The surgical procedure indicated in case of the advanced age, fragility and/or critical condition of the patient is simple enterolithotomy with removal of the stone and closure of the intestinal wall

As regards technique, once the site of the obstruction has been identified it is advisable to delicately slide the stone upwards, exploiting the proximal dilatation, make a longitudinal incision on the antimesenteric border of the loop, carefully extraction of the stone and then make a transverse suture. Before closure of the incision, palpation of the whole proximal tract of the intestine is recommended so that any other stones present can be identified and extracted. (2)

If the general clinical condition of the patient permits and the risk not high, radical surgery can be performed to treat the occlusion, the biliary disease and the fistula. This solution can be used in younger patients, as a one-stage procedure, especially in cases of concomitant biliary sepsis or gangrene. In other patients, radical surgery should be planned as a two-stage procedure, with the first stage reserved for simple enterolithotomy and definitive treatment of the fistula and biliary disease performed in a second procedure, 3-6 weeks later. At that time intraoperative ultrasound scan can be used to identify the SBEF or, the more modern technique of indocyanine green tracer can be used to mark the fistula tract. (4,22).

The two-stage procedure should be used only for patients who are young and are in fairly good clinical condition but at risk of biliary complications including recurrent

gallstone ileus. The latter complication, experienced by 5-20% of patients, usually occurs during the first 30 days after enterolithotomy, but cases of recurrent gallstone ileus even after 2-6 months have been reported (26,27). Because of this it has recently been proposed that enterolithotomy be associated with cholecystolithotomy to prevent migration of additional gallstones through the fistula tract. The approach proposed involves making an incision in the fundus of the gallbladder to extract all residual stones and then closing the incision with absorbable sutures. This procedure can be performed laparoscopically and can be combined with minimally invasive enterolithotomy. (28).

The value of this approach is reinforced by the observation that in the absence of residual gallstones the fistula tract closes spontaneously after a certain lapse of time (29)

The laparoscopic technique, first performed by Montgomery and published in 1993 (30), is used increasingly often and has been shown to be effective for radical one-stage procedures to treat the occlusion, the biliary disease and the SBEF(31).32

The procedure used to treat Bouveret syndrome is almost always endoscopy (70%) since most patients are elderly with several comorbidities. This can be either simple endoscopy with a net/basket or endoscopy with some form of lithotripsy surgical extraction has been described in cases where endoscopy failed(2, 33,).

Moreover, cases have been described in which Bouveret syndrome was also associated with the presence of a second stone in a more distal location. (34)

The average hospital stay is over 10 days. The most common complications are local infection (35%) and systemic complications (22%). (28)

The morbidity rate after conservative procedures is 29% but rises to 67% after radical surgery

The mortality rate has fallen over the years, from 44% (Mondor, 1965) to 30% in the prelaparoscopic era, to 10-13% today (28)

In very old, fragile patients in critical condition for whom surgical treatment was not possible, the mortality rate remains high:25%. After simple enterolithotomy, particularly laparoscopic enterolithotomy, the mortality rate is 11.7% and after one-stage radical surgery it is 16.9% (20-28)

Conclusions

SBEF is a fairly rare pathology which very often remains unrecognized for a long period of time, usually until complications appear.

Gallstone ileus is the most important complication, and is virtually pathognomonic for SBEF. It manifests clinically as acute or chronic mechanical intestinal occlusion (2).

Gallstone ileus is a pathology found most commonly in individuals over 65 years old, and is more common in women (male-to-female ratio 1:5). It is characterized by a high percentage of cholelithiasis, a preoperative diagnosis rate of less than 50%; and the delay in hospital admission and treatment. The diagnostic gold standard is contrast-enhanced CT scan and for proximal occlusions, endoscopy. (23)

Non-surgical management should be reserved for old patients who are frail and in critical condition, especially those with stones less than 2 cm in diameter on ultrasound and CT scan. However the mortality rate for these patients reaches 26.5% (28, 35).

The surgical treatment most commonly used today is totally laparoscopic or laparoscopic assisted enterolithotomy, via minimally invasive access, which is associated with a mortality rate of almost zero but a 5-20% rate of recurrent gallstone ileus (2,28). Moreover, a 15% rate of gallbladder cancer and persistence of SBEF is reported after simple enterolithotomy (36).

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RIASSUNTO

La fistola biliare-enterica spontanea (SBEF) è una comunicazione anormale tra l'albero biliare e il tratto gastrointestinale che si sviluppa a causa di una malattia biliare o gastrointestinale.

Le fistole iatrogene, dovute a chirurgia o esplorazione strumentale, non sono incluse in questa definizione. R.Colombo, nel 1559, fu il primo a descrivere l'SBEF come un reperto occasionale durante un'autopsia.

In quasi il 90% dei casi la causa della SBEF è la colelitiasi cronica ricorrente. Cause meno comuni sono le ulcere peptiche penetranti e l'infiltrazione neoplastica dal tratto biliare o gastrointestinale

Il tipo più comune di SBEF è la fistola colecistoduodenale e la meno comune è la fistola colecoduodenale. Ci sono varie complicazioni associate all'SBEF, ma spesso queste non vengono prontamente riconosciute dai pazienti o dai medici e la diagnosi e il trattamento possono essere ritardati di anni.

La complicanza più importante, che può essere considerata patognomonica per SBEF, è l'ileo dei calcoli biliari che si manifesta clinicamente come un'ostruzione intestinale meccanica acuta o cronica

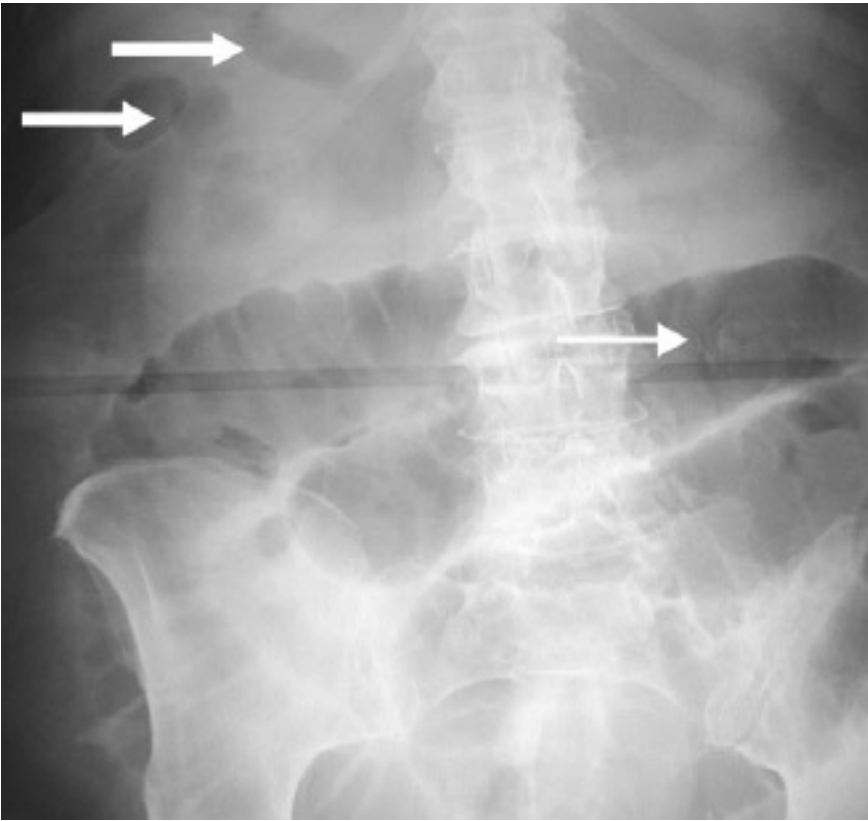
Ileo di calcoli biliari, complicanza piuttosto rara di una patologia piuttosto comune, la litiasi biliare, si riscontra nello 0,000015% dei pazienti ospedalizzati ma nello 0,0003% dei pazienti chirurgici. Si trova principalmente nelle donne di età superiore ai 65 anni, con un rapporto maschi/femmine di 1:5

Esistono varie forme di occlusione, legate alle sedi di occlusione del calcolo biliare, con diverse caratteristiche cliniche e gradi di gravità. Questi includono la sindrome di Bouveret (<10% dei casi) che si verifica nella fistola bilioduodenale o biliopilorica e la più comune sindrome di Barnard (5-75% dei casi) in cui il sito di occlusione è nell'ileo terminale subito prima della valvola di Bauhin

Per la diagnosi sono essenziali i segni radiologici che compongono la triade o tetrade di Rigler e che sono meglio visibili sulla risonanza magnetica, ma il gold standard è la tomografia computerizzata con mezzo di contrasto.

Il trattamento chirurgico, riservato ai pazienti più anziani e ai casi più gravi, è l'enterolitotomia semplice in uno stadio, che oggi viene eseguita sempre più frequentemente per via laparoscopica. Nei casi più favorevoli trattamento radicale dell'occlusione; la litiasi biliare e la SBEF; è raccomandato, sia come procedura in una fase o in due fasi con la seconda procedura dopo alcune settimane.

Pict. 1



Rigler's triad :pneumobilia + ectopic gallstone + abdominal distentionfor dilated ileal loops

Pict. 2



Characteristic target mark of ectopicgallstone

Pict. 3



Macroscopic formation of the ectopic gallstone: Central nucleus is the primitive biliary stone wrapped by more friable enteric material