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Case report

Choledochoduodenal fistula: an unusual complication of penetrated duodenal ulcer disease

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ABSTRACT

Spontaneous choledochoduodenal fistula in the absence of primary biliary disease is a very unusual complication of duodenal ulcer disease. In most cases it is diagnosed incidentally, because it seldom gives clinical manifestations. Although surgical approaches have been the treatment of choice in the past, the use of modern antisecretory drugs turns now management strategy to more controversial issues, as the fistula per se is not an indication for surgery.

Three of our cases are reported herein and the literature on choledochoduodenal fistula, secondary to a penetrating duodenal ulcer is reviewed.

Keywords: Choledochoduodenal fistula, duodenal ulcer, complication

INTRODUCTION

Although hospitalization and surgery for uncomplicated peptic ulcers has decreased over the last 25 to 35 years in USA and Europe, the number of hospital admissions for complications such as ulcer-associated hemorrhage has remained relatively unchanged.[1]

According to the available literature during the last decades the incidence of spontaneous biliary enteric fistulas has been increased. Spontaneous internal biliary fistulas are not an uncommon complication of primary biliary disease, presenting in 3-5% of cases.[2] Choledochoduodenal fistulas are infrequent and are usually secondary to peptic ulcer disease in 80% of cases, and they appear with the signs and symptoms of the underlying peptic ulcer disease.[2] It is important to differentiate between these two types of fistula, as prognosis is poor in the biliary enteric fistula secondary to gall-bladder disease and its treatment of choice is undisputedly surgical, while the prognosis is good in ulcerogenic fistula, although its treatment still remains controversial.[2] In ulcerogenic fistulas, although in the past, surgical approaches have been the treatment of choice, with the introduction of modern antisecretory drug treatments,[3] management now seems to be controversial.

A total of 179 cases of Choledochoduodenal fistula secondary to duodenal peptic ulcer have been reported since 1987, but the actual incidence is probably higher, as the majority of Choledochoduodenal fistulas are asymptomatic.[4,9] Since then, no more than 20 additional cases have been reported in world literature, according to publications sited in Medline database, which currently increases the number to almost 200 cases.[3,10,11]

A three-case series of patients with benign peptic ulcer disease perforating into the common bile duct, who were followed over the last 20 years in our Department are reported herein with review of the literature.

CASE REPORTS

Case 1

A 35-year-old man suffering for more than 20 years from severe epigastric pain, vomiting and heartburn is described. Past surgical history involved appendectomy at the age of 15 years, when typical symptoms of duodenal ulcer were misdiagnosed as acute appendicitis. At the

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age of 18 years the patient suffered a severe episode of massive upper gastrointestinal bleeding and at that time radiological examination showed increased gastric transit time and a markedly deformed duodenal bulb because of a duodenal ulcer. Since that time, the patient has been readmitted three times to other hospitals due to severe upper gastrointestinal bleeding episodes. During hospitalisation the patient was managed with symptom-relieving medical treatment. However the patient continued heavy consumption of cigarettes and alcohol.

In 1985 the patient was hospitalised for 30 days, due to an episode of massive haematemesis and maelena; Upper gastrointestinal endoscopy showed a bleeding duodenal ulcer with a scarring stenotic duodenal bulb. The patient was managed conservatively with antisecretory drugs. Apart from of Hct, RBC, and WBC, all other laboratory tests were within normal limits. Plain abdominal x-ray film showed air in the biliary tree. Barium study of the upper gastrointestinal tract revealed pyloric stenosis and a markedly deformed duodenal bulb from which the barium entered and filled the common bile duct while linear radiolucencies in the liver, representing air in the normal-sized intrahepatic ducts, were also evident (Figure 1). At operation, a chronic duodenal ulcer with pyloric stenosis was found, while liver and gallbladder were normal. A truncal vagotomy and a posterior gastroente-rostomy were performed without intervening into the area of fistula. One year later, x-ray films of the upper gastrointestinal tract showed no barium reflux into the common bile duct.

The patient was admitted again seven years later with acute cholecystitis symptoms. Exploratory surgery was performed, which revealed an acutely inflamed gallbladder, with stones, while the common bile duct was normal. Operative cholangiography was normal, without bile duct dilatation, stones or any evidence of fistula. Cholecystectomy was performed, followed by uncomplicated recovery. At the 10-year follow-up the patient is in good health status without any symptoms suggestive of cholangiitis or peptic ulcer disease.

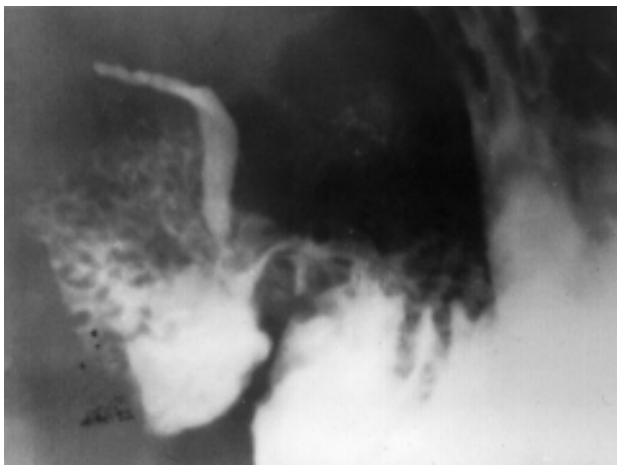

Figure 1. Barium meal study indicating deformed duodenal bulb and presence of barium in the common bile duct.

Case 2

An 87-year-old woman was admitted to the hospital with acute epigastric pain, vomiting and fever up to 38°C. Physical examination revealed mild epigastric and right upper quadrant tenderness. Laboratory tests showed leucocytosis (12500/μI) with 85% neutrophils, while all other laboratory findings were within normal limits. The patient had had similar episodes in the past and was treated with cimetidine for presumed duodenal ulcer. Abdominal ultrasound showed microlithiasis of the gallbladder and presence of air in the common bile duct. Upper gastrointestinal endoscopy showed an active duodenal ulcer with possible fistula formation communicating with the bile duct. A gastrografin follow-through meal further confirmed the existence of a choledocho-duodenal fistula. The patient was initially treated conservatively with antisecretory drugs. An operation was decided on and performed a few days later, resulting in uncomplicated laparoscopic cholecystectomy.

The postoperative course was uneventful. Two years later the patient is alive, with no symptoms of peptic ulcer disease or any fistula-related complaints.

Case 3

A 42-year-old man was admitted to the hospital because of two melaena episodes. On admission blood pressure was 126/76 mm Hg, heart rate was 90/min and temperature normal. Hematocrit was 34.3%, while the other laboratory findings were within normal limits. The patient had a history of three upper gastrointestinal tract-bleeding episodes due to a duodenal ulcer, occasionally treated with cimetidine. During hospitalization, the patient was managed conservatively as vital signs were stable. Upper gastrointestinal endoscopy revealed an active anterior bulb ulcer accompanied by a significant scarring bulb deformity. After two days of conservative treatment melaenas stopped and the patient's condition was significantly ameliorated. At 10th day hospitalisation, the patient reported right upper quadrant abdominal pain and tenderness accompanied by a fever up to 39°C.

Laboratory tests showed WBC=13.940/pl with 80% neutrophils, ALT at 116 IU/1 and γ-GT at 185 IU/1. There were no clinical signs of jaundice. Abdominal ultrasonography revealed cholelithiasis with oedema of the gallbladder and the presence of air in the bile duct. Gastrographin follow-through meal confirmed the presence of a choledochoduodenal fistula (Figure 2). The patient was managed conservatively and his clinical condition improved significantly, to full recovery. A few days later the patient underwent cholecystectomy. The patient was discharged with instructions for conservative oral antisecretory treatment for peptic ulcer disease.

DISCUSSION

Bartholin first described a biliary-enteric fistula in 1654, but duodenal ulcer as a causative communicating mechanism was first recognized and published by Long in 1840 in the London Medical Gazette[12] . Biliary-enteric fistula is a rare complication, occuring in 0.3-0.5 % of patients who have been treated for chronic duodenal ulcer disease.[2,8,12]

Cholecystoduodenal fistulas represent the most common type of bilioenteric fistulas while choledochoduodenal fistulas account for only 1-25% of bilioenteric fistulas cases.[8] Although 75-90% of bilioenteric fistula cases are associated with cholelithiasis,[6,8] only 5-6% of them are associated with duodenal peptic ulcers.[6,13,14] However, 75-80% of choledochoduodenal fistula cases are caused by duodenal peptic ulcer disease in western countries,[6,8,13] while this occurs in only 15% of cases in Japan.[15] Choledochoduodenal fistulas (CDDF) usually occur after peptic ulcer perforation.

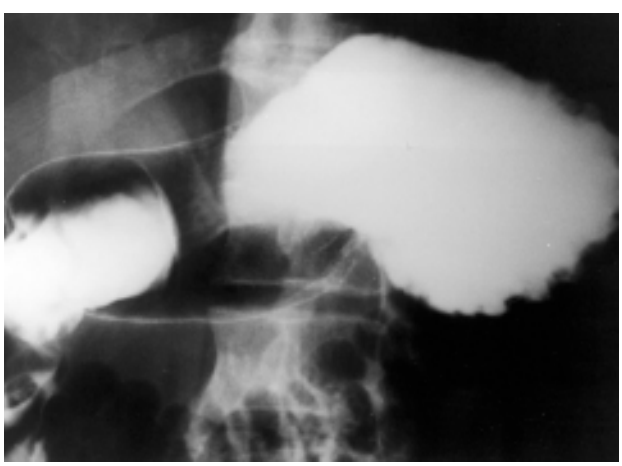

Figure 2. Upper gastrointestinal barium study showing duodenal bulb deformity and part of the choledochoduodenal fistula.

The majority of patients are usually in the fifth or sixth decade of life and have a long history of symptomatic dyspepsia. In addition, men outnumber women by at least 3 to 1 in biliary communications arising from penetrating duodenal ulcer[16]. Symptoms or signs attributable to the fistula itself are exceptional, the most usual symptoms being cholangitis, which occurs in less than 10% of cases.[7,17]

Obstructive jaundice or upper gastrointestinal hemorrhage is rare, however the possibility of biliary fistula existence should be considered in patients with a history of duodenal ulcer disease and jaundice[17,20]. The first sign of this abnormal biliary-enteric communication may be the presence of air in the biliary tree as seen on plain roentgenogram of the abdomen or with ultrasound or CT.[5,21] The presence of pneumobilia is helpful for the final diagnosis, but it is present in only 14-22% of bilioenteric fistulas.[5,7,22] Pneumobilia in nonoperated patients, except in cases of emphysematous cholecystitis and primary or secondary reflux of the ampulla of Vater, is almost pathognomonic of some types of internal biliary fistula.[23,27] The diagnosis is usually not established until the unanticipated finding of contrast material existence in the biliary tree during barium meal evaluation of patients with known or suspected peptic ulcer disease, as occured in our patient.[16,28] This phenomenon seems to occur in both symptomatic and asymptomatic patients. Endoscopic examination, biopsy in appropriate cases, and cannulation of the fistula for precise radiographic delineation helps establish the pathologic features, confirming the presence of a chronic peptic ulcer, excluding tumour or other rare diseases, and guiding the therapeutic intervention choice.[5,10,11,28] Endoscopic retrograde cholangiopancreatography may be unsuccessful in ulcerogenic fistulas as the orifice may be visualized and cannulated, and injection of contrast material may delineate a common bile duct of normal appearance.[10,11,16,28,29]

Most cases of CDDF occur at the posterior wall of the duodenal bulb, as fistulas at the anterior wall of the duodenal bulb are extremely rare.[6,8,10,16,29]

The natural history of CDDF caused by ulcers is determined by features of the underlying chronic duodenal ulcer.[28] Ulcer healing is accompanied in most cases by fistula healing as well.[10,28,29] This contrasts with biliary-enteric fistula caused by gallstones, which more commonly stay open, keeping their asymtomatic clinical course.[20]

In the absence of primary biliary disease, a CDDF resulting from a perforating duodenal ulcer presents a minimal risk of cholangitis or future biliary stricture, although this potential must be acknowledged.[7,17,20] Acute acalculous cholecystitis with jaundice during early postoperative course was observed by Ayyash and Jadallah,[31] while others described a case of acalculous cholecystitis 7 months postoperatively.[32] Although cholecystectomy is not mandatory in every case of acalculous cholecystitis, some investigators believe that it should be initially performed in order to avoid the late complication of cholecystoduodenal fistula, as well as the danger of later cholecystitis.[13,31]

Oral medical treatment of CDDF arising from a duodenal ulcer was formerly performed in high surgical risk patients. The medical management for peptic ulcer has developed with the advent of antacids and proton pump inhibitors.[2,6] Two of the three patients reported here responded well to the medical treatment and only one patient required the surgical approach. Surgery must be reserved for patients with poorly controlled or recurrent ulcer symptoms, major ulcer complications, such as perforation, hemorrhage, or obstruction, or exceptional cases with cholangitis or biliary obstruction.[28,29] The operation of choice seems to vary along with the surgeon's preference, but at present, the great majority of surgeons seem to favour vagotomy and exclusion-type gastrectomy whenever possible.[22] The results of vagotomy and gastrectomy have been generally encouraging.[2]

Fistula management during operation varies considerably. If the bile duct is distally obstructed, most investigators stress the necessity for temporary biliary decompression, using a T-tube in the common duct or some type of internal drainage procedures,[10] such as cholecystoduodenostomy or Roux-en-Y drainage, since there is no way to predict whether biliary obstruction will resolve with ulcer treatment by itself.[17] The gallbladder should be removed when a fistula communicates with it or if a choledochojejunostomy is constructed; in such the cases gallbladder becomes a nonfunctioning liability. The duration of follow-up in most cases has rarely exceeded a whole year. Consequently, the natural history of the disease has not been well enough described. In our patients' series, the follow-up time ranged from 2 to 11 years.

The operative mortality reported in cases of CDDF is low. In 56 cases described up to 1964 no operative mortality was reported.[7] These favorable results contrast with a significant mortality of 12.5-40%, which follows reconstruction procedures of internal fistulas secondary to gallstones. There were no deaths or major complications in our series of patients. A complete follow-up for as long as 12 years confirms the previous observation that jaundice, cholangitis, or abnormal liver function are rarely encountered in ulcerogenic choledochoduodenal fistulas (CDDFs). Treatment should be directed towards peptic ulcer disease relief rather than correction of CDDF. In many patients, optimal results may be achieved by using only oral medical therapy.

The medical treatment of CDDF with antacids and proton pump inhibitors, formerly reserved for high surgical risk patients, seems to be symptom relieving in most cases and should be recommended instead of surgery.

If operation is indicated, a conservative procedure, which corrects the ulcer diathesis and leaves the CDDF intact, may be proved sufficient. Vagotomy and gastroenterostomy will accomplish these goals and obviate the necessity of entering a scarred duodenum.

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