Spontaneous Choledochoduodenal Fistula Secondary to Long-Standing Ulcer Disease

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ABSTRACT

There are several varieties of bilioenteric fistulae. They are usually incidental findings, but once detected, various modalities can then be employed to further delineate the fistula. The fistulae usually arise as a complication of chronic duodenal ulcer disease, cholelithiasis or previous instrumentation to the biliary system. The presence of a fistula per se does not immediately equate to necessity for surgery. The treatment is dependent on its aetiology.

Keywords: fistula, bilioenteric, choledochoduodenal, duodenal ulcer, cholelithiasis

INTRODUCTION

Bilioenteric fistulae are usually incidental findings that may confound results of certain procedures. A case involving an oesophagogastroduodenoscopy (OGD) and pyloric stenting will be presented. We would like to highlight the benefits of having a high index of suspicion for such fistula as well as provide a general discussion of these fistulae.

CASE REPORT

This is a case of a 66-year-old Chinese lady with background cardiovascular disease who had a gastric ulcer diagnosed on OGD many years ago. She denied having any abdominal surgery and was no longer on follow-up for her gastric ulcer disease. She was admitted twice in the last year for recurrent vomiting. OGD showed gastritis and duodenitis. The pylorus was oedematous and tight and there was a diverticulum at the anterior wall of D1. CLO test for Helicobacter pylori infection was positive. Barium meal demonstrated gastritis and a D1 diverticulum (possibly a pseudodiverticulum). Ultrasonography of the hepatobiliary system (HBS) revealed debris in the gallbladder and portal triads that were more echogenic than usual.

She presented this time with vomiting and diarrhoea lasting two to three days. The abdomen was soft with a succussion splash and a per-rectal examination revealed soft stools. Electrolytes showed a hypochloraemic, hypokalaemic metabolic alkalotic picture. The abdominal radiograph did not show dilated bowel loops. Urine culture grew Klebsiella pneumoniae. She was started on intravenous hydration and antibiotics in the ward. Nasogastric tube (NGT) feeding was implemented with initial success. Unfortunately, she was unable to tolerate an increase in her NGT intake and a large amount of residual feeds was aspirated a few days later. Suspecting a gastric outlet obstruction, the patient subsequently underwent two OGDs where pyloric stenosis secondary to ulceration was diagnosed. On both occasions, balloon dilatation of the stenosis was carried out and biopsy of the ulceration revealed adenoma with severe dysplasia.

Fig. 1 Opacification of the first part of the duodenum (D) and the biliary tree via contrast injection through a ball-tipped catheter which was passed through a pyloric stenosis from the stomach.
Finally a third OGD, with a view to deploy an enteral stent, was performed due to persistently high NGT aspirate, histology of severe dysplasia and a poor surgical candidate status. Again, the pylorus had re-stenosed and we were unable to pass the scope through. A ball-tipped catheter was passed through the stenosis and diluted, water-soluble contrast was injected. Surprisingly, opacification of the biliary tree was showed on fluoroscopy then (Fig. 1). As far as we knew, the patient had no prior history of any abdominal surgery and there was no abdominal scar noted on physical examination. At that point of time, we were wondering whether the catheter was pushed too hard and had penetrated the common bile duct (CBD). A fistula, as a complication from her recent balloon dilatations, was another possibility. As we rummaged through the old notes and films, we were relieved to discover that a fistulous tract from the duodenum to the CBD was already present during her previous barium study (Fig. 2). Satisfied that this was a spontaneous choledochoduodenal fistula (CDF) and that we had inadvertently cannulated its orifice, we re-adjusted the catheter to the duodenum and a Microvasive Enteral Wallstent (Boston Scientific Microvasive, Natick, MA, USA) was deployed successfully across the stenosis over a long guidewire. Subsequently, the patient responded well and could tolerate soft diet adequately.

DISCUSSION

Bilioenteric fistulae are usually incidental findings because they seldom produce clinical symptoms(1). Some are found only during surgery thus forcing surgeons to change a planned surgical procedure without adequate pre-operative preparation. This could result in a higher morbidity rate(2). A study of 81 patients over a 50-year period had attempted to list the incidences of several types of spontaneous bilioenteric fistulae. They include (a) cholecysto-duodenal (68%), (b) cholecysto-colic (13.6%), (c) choledochoduodenal (8.6%), (d) cholecysto-gastric (4.9 %) and (e) duodeno-left hepatic (4.9%)⁴⁶.

CDF are located on the longitudinal fold of the papilla (70.6 %) and on the posterior wall of the duodenal bulb (29.4 %)⁴⁶. Rarely is a CDF located on the anterior wall of the duodenal bulb⁴⁶. An attempt was made at differentiating between these parapapillary choledochoduodenal fistulae (PCDF) from separate openings of the pancreatic and biliary ducts. This study concluded that the prevalence of a PCDF was between 2-4%⁴⁶.

Diagnosis is usually made incidentally on radiography or barium meal and follow-through. The radiograph may show the presence of air in the biliary tract or gallbladder⁷. The barium study may show a scarred duodenal cap with a fistulous communication to the CBD. This finding becomes conclusive if the distal part of the CBD is not seen at that time⁶⁸. Once suspected, CDF can be easily diagnosed on OGD with a side-viewing scope. Fistulography can also be performed. An endoscopic retrograde cholangiopancreatogram (ERCP) will show the relation of the fistula to the bile duct system⁴⁹.

Cholecystoduodenal fistulae are generally associated with calculous gallbladder disease and occur predominantly in women. In contrast, CDF usually occurs as a complication of duodenal ulcer disease and is thus seen more often in men⁹¹⁰,¹¹. However, another study played down the contribution of duodenal ulcers towards causing CDF. The rarity stems from the fact that duodenal ulcers typically occur within 4 cm distal to the pylorus whereas the CBD is about 7 cm distal to the pylorus⁹⁶. CDF is also a known complication of choledocholithiasis. In these cases, they seem to arise from stone erosion through the bile duct into the duodenum (a form of Mirizzi’s syndrome). In patients who have previously undergone exploration of the CBD, instrumentation of the CBD is the most likely cause of their fistulae¹¹. One article reported the incidence of iatrogenic CDF as being 9.3 % with this figure rising to 23% if instrumentation involved a rigid probe⁴⁹. Unfortunately, it may even be due to a technical mistake whilst performing sphincteroplasty¹⁴. There have been two rare instances of duodenal tuberculosis¹⁰¹⁶ and one of a primary adenocarcinoma of the duodenum¹⁷ causing a CDF.

Treatment of uncomplicated fistulae appears unnecessary¹². Surgical treatment may have been performed for ulcer disease in the past but medical treatment is now the treatment of choice with the
avilability of potent anti-ulcer agents\textsuperscript{18}. The majority of these fistulae heal spontaneously with intensive medical management\textsuperscript{19}. When surgery is recommended, the operation of choice is truncal vagotomy with distal gastrectomy (antrectomy) and gastroenterostomy by Bilroth II method, leaving the fistula intact\textsuperscript{20,21}. Endoscopic sphincterotomy has shown good results where choledolithiasis is complicated by CBD\textsuperscript{15,18}. A more sophisticated means of stone extraction involves a papillotome which is used to cut the CBD between the papilla and orifice of the fistula. There will be free outflow of bile and any residual stone can then pass spontaneously or be removed with a basket catheter. This method is termed endoscopic papillotomy (EPT)\textsuperscript{14} or endoscopic fistulotomy (EFT)\textsuperscript{22}. In the group of patients whose CDF was caused by instrumentation, spontaneous healing of the fistula was noted\textsuperscript{13,15,16}. Choledectomy, CBD exploration and biloenteric reconstruction may be done but are reserved in cases of a biliary stricture, which rarely occurs\textsuperscript{23,24}.

**CONCLUSION**

This case presented demonstrates how biliary tree under unsuspected circumstances can cause some panic initially for the endoscopist. We hope this article will serve as a reminder to the readers the rarely occurring CDF as a differential diagnosis of perforation of the CBD. Sometimes, the presence of a spontaneous CDF might even be a blessing as it could provide an access to the biliary tree for therapeutic procedures when cannulation of the papilla proves to be difficult. For patients requiring upper gastrointestinal or biliary surgery, it would be prudent to suspect and then demonstrate the presence of a biloenteric fistula in any patient who fits the appropriate clinical scenario as this will lower perioperative morbidity, especially prior to a planned procedure.

**REFERENCES**