Spontaneous Choledochoduodenal Fistula Secondary to Long-Standing Ulcer Disease

M W C H'ng, H B Yim

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

Singapore Med J 2003 Vol 44(4):205-207

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.

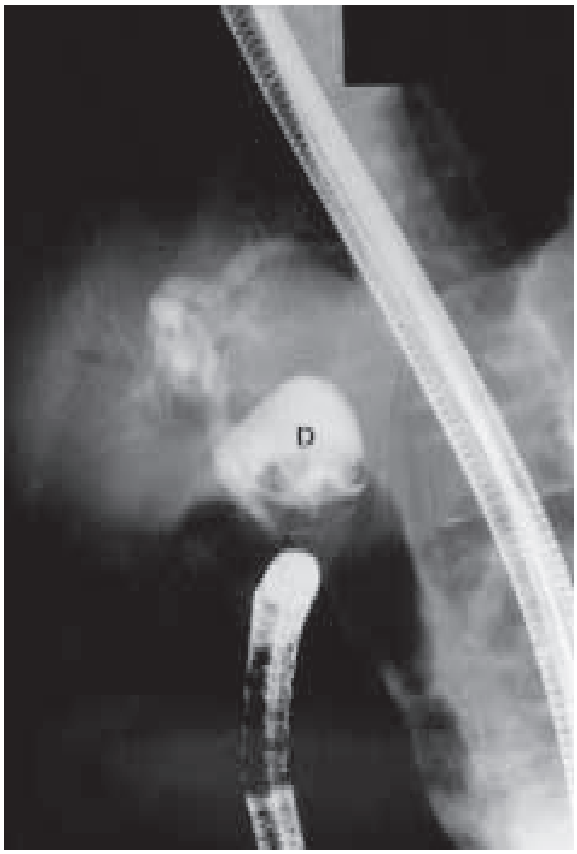

Fig. 1 Opacification of the first part of the duodenum (D) and the biliary tree via contrast injection through a ball-tipped catheter which was passed through a pyloric stenosis from the stomach.

Department of Radiology
Tan Tock Seng Hospital
11 Jalan Tan Tock Seng
Singapore 308433

M W C H ng, MBBS (Melbourne)
Medical Officer

Gastroenterology Unit
Department of General Medicine
Tan Tock Seng Hospital
11 Jalan Tan Tock Seng
Singapore 308433

H B Yim, MBBS, MRCP (UK), FAMS
Consultant Gastroenterologist

Correspondence to:
Dr Yim Heng Boon
Tel: (65) 6357 7893
Fax: (65) 6357 8355
Email: heng_boon_yim@ttsh.com.sg

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.

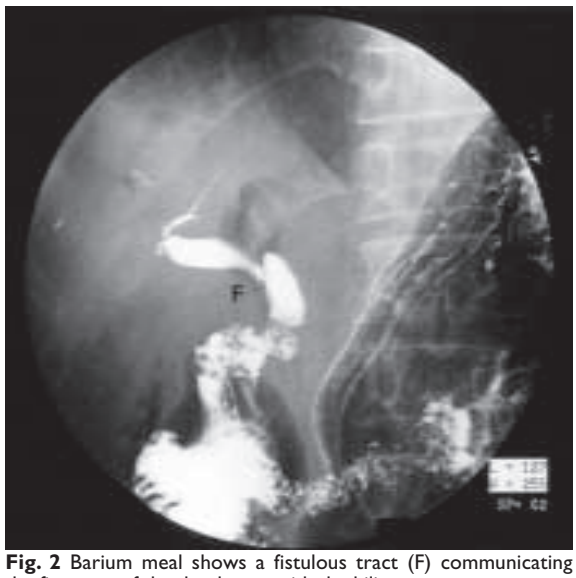

Fig. 2 Barium meal shows a fistulous tract (F) communicating the first part of the duodenum with the biliary tree.

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-colonic (13.6%), (c) choledocho-duodenal (8.6%), (d) cholecysto-gastric (4.9 %) and (e) duodeno-left hepatic (4.9%)[3].

CDF are located on the longitudinal fold of the papilla (70.6 %) and on the posterior wall of the duodenal bulb (29.4 %)[4]. Rarely is a CDF located on the anterior wall of the duodenal bulb[5]. 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%[6].

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 gallbladder[7]. 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 time[8]. 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 system[4].

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 men[9,10]. 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 pylorus[8]. 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[11] (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 fistulae[12]. One article reported the incidence of iatrogenic CDF as being 9.3 % with this figure rising to 23% if instrumentation involved a rigid probe[13]. Unfortunately, it may even be due to a technical mistake whilst performing sphincteroplasty[14]. There have been two rare instances of duodenal tuberculosis[15,16] and one of a primary adenocarcinoma of the duodenum[17] causing a CDF.

Treatment of uncomplicated fistulae appears unnecessary[12]. Surgical treatment may have been performed for ulcer disease in the past but medical treatment is now the treatment of choice with the

availability of potent anti-ulcer agents[18]. The majority of these fistulae heal spontaneously with intensive medical management[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[20,21]. Endoscopic sphincterotomy has shown good results where cholelithiasis is complicated by CDF[14]. 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)[4] or endoscopic fistulotomy (EFT)[22]. In the group of patients whose CDF was caused by instrumentation, spontaneous healing of the fistulae was noted[13]. Cholecystectomy, CBD exploration and bilioenteric reconstruction may be done but are reserved in cases of a biliary stricture, which rarely occurs[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 bilioenteric fistula in any patient who fits the appropriate clinical scenario as this will lower perioperative morbidity, especially prior to a planned procedure.

REFERENCES

1. Michowitz M, Farago C, Lazarovici I, Solowiejczyk M. Choledochoduodenal fistula: a rare complication of duodenal ulcer. Am J Gastroenterol 1984; 79:416-20.
2. Mittelstaedt WE, Carone Filho E, Sekini JH, Deutsch CR, Neri JR, Pires PW, et al. Bilio-digestive fistulas. Arq Gastroenterol 1984; 21:3-12.
3. Stagnitti F, Mongardini M, Schillaci F, Dall Olio D, De Pascalis M, Natalini E. Spontaneous biliodigestive fistulae. The clinical considerations, surgical treatment and complications. G Chir 2000; 21:110-7.
4. Bethge N, Hintze RE. Spontaneous and iatrogenic choledochoduodenal fistula — endoscopic diagnosis and therapy Z Gastroenterol 1988; 26:704-7.
5. Shimao K, Yamaue H, Nishimoto N, Terasawa H, Saigan S, Onishi H, et al. Choledochoduodenal fistula at the anterior wall of the duodenal bulb: a rare complication of duodenal ulcer. Hepatogastroenterology 1999; 46:261-4.
6. Nakano T, Yabuta I, Shimomura H. Clinical evaluation of choledochoduodenal fistula near the duodenal papilla: association with separate openings of the pancreatic and biliary ducts. Nippon Shokakibyo Gakkai Zasshi 1994; 91:57-65.
7. Amar A, Ribeyre D, Lombard F, Yonneau L, Egarnes M, Valyi L, et al. Choledochoduodenal fistula: an unusual complication of duodenal ulcer. Gastroenterol Clin Biol 1996; 20:394-6.
8. Shah P, Ramakantan R. Choledochoduodenal fistula complicating duodenal ulcer disease (A report of 3 cases). J Postgrad Med 1990; 36:167-8.
9. Topal U, Savci G, Sadikoglu MY, Tuncel E. Choledochoduodenal fistula secondary to duodenal peptic ulcer. A case report. Acta Radiology 1997; 38:1007-9.
10. Page JE, Dow J, Dundas DD. Ulcerogenic choledochoduodenal fistula. Clin Radiology 1989; 40:58-60.
11. Van Linda BM, Rosson RS. Choledochoduodenal fistula and choledocholithiasis: treatment by endoscopic enlargement of the choledochoduodenal fistula. J Clin Gastroenterol 1984; 6:321-4.
12. Martin DF, Tweedle DE. The aetiology and significance of distal choledochoduodenal fistula. Br J Surg 1984; 71:632-4.
13. Rimer U, Zederfeldt B, Fork FT. Juxtapapillary choledochoduodenal fistula following choledochal exploration and diagnosed by early duodenoscopy. Acta Chir Scand 1986; 152:363-6.
14. Costamagna G, Coppola R, Belli P, Masetti R, Magistrelli P, Nuzzo G. Juxtapapillary choledochoduodenal fistula. Diagnosis and treatment in 19 cases. Ital J Surg Sci 1987; 17:347-53.
15. Miyamoto S, Furuse J, Maru Y, Tajiri H, Muto M, Yoshino M. Duodenal tuberculosis with a choledocho-duodenal fistula. J Gastroenterol Hepatol 2001; 16:235-8.
16. Al Nakib B, Jacob GS, Al Liddawi H, Commen J. Choledochoduodenal fistula due to tuberculosis. Endoscopy 1982; 14:64-5.
17. Tsai CJ. Primary adenocarcinoma of the duodenum with choledochoduodenal fistula. Scand J Gastroenterol 1994; 29:930-3.
18. Parekh D, Segal I, Ramalho RM. Choledochoduodenal fistula from a penetrating duodenal ulcer. A case report. S Afr Med J 1992; 81:478-9.
19. Aitken RJ, Bornman PC, Dent DM. Choledochopancreatoduodenal fistula caused by duodenal ulceration. A case report. S Afr Med J 1986; 69:707-8.
20. Iso Y, Yoh R, Okita K, Murakami N, Nozoe T, Funahashi S, et al. Choledochoduodenal fistula: a complication of a penetrated duodenal ulcer. Hepatogastroenterology 1996; 43:489-91.
21. Dubois F, Berthelot G, Levard H. Choledochoduodenal fistula of ulcerous origin. Two cases. Presse Med 1985; 14:879-81.
22. Urakami Y, Kishi S. Endoscopic fistulotomy (EFT) for parapapillary choledochoduodenal fistula. Endoscopy 1978; 289-94.
23. Fowler CL, Sternquist JC. Choledochoduodenal fistula: a rare complication of peptic ulcer disease. Am J Gastroenterol 1987; 82:269-71.
24. Misra MC, Grewal H, Kapur BM. Spontaneous choledochoduodenal fistula complicating peptic ulcer disease — a case report. Jpn J Surg 1989; 19:367-9.