Turk J Gastroenterol 2010; 21 (2): 163-167

Ectopic opening of the common bile duct into the stomach

Koledoğun mide'ye ektopik açılımı

Orhan SEZGİN, Engin ALTINTAŞ, Enver ÜÇBİLEK

Department of Gastroenterology, Mersin University, School of Medicine, Mersin

Ectopic opening of the common bile duct into the stomach is an extremely rare congenital anomaly of the biliary system. Thus far, only three similar cases have been reported in the literature, and none of them had accompanying gastric cancer. In this report, we present a case with ectopic opening of the common bile duct into the stomach, which is an extremely rare anomaly, associated with gastric cancer, and we discuss the possible relation between the direct flow of bile into the stomach through the ectopic bile duct and the development of gastric cancer.

Key words: Bile duct anomalies, ectopic biliary drainage, stomach, gastric carcinoma

Koledoğun mideye ektopik açılımı biliyer sistemin son derece nadir kongenital bir anomalisidir. Şimdiye kadar tıp literatüründe benzer sadece üç olgu bildirilmiştir. Bunların hiçbirisi mide kanseriyle birlikte değildi. Bu yazıda koledoğun mideye ektopik açılımı ile birlikte mide kanseri de bulunan olguyu sunarak anormal direkt safra akışının mide kanseri gelişimindeki olası rolü üzerinde de durmayı amaçladık.

Anahtar kelimeler: Safra kanalı anomalisi, ektopik safra akışı, mide, mide kanseri

INTRODUCTION

Ectopic opening of the common bile duct (CBD) into the stomach is an extremely rare congenital anomaly of the biliary system. Although ectopic opening of the biliary system into the stomach via the accessory bile duct has been reported previously, until recently there have been only three case reports in the worldwide literature of the CBD opening directly into the stomach (1-6). In the first case, reported by Laennec in 1820, the CBD entered the lesser curvature of the stomach just below the cardia, and in the other case, reported from Bleifus in 1839, the CBD entered the stomach, but the exact site was not mentioned (6). In 1974, Vazquez Quintana et al. (6) reported a patient with jaundice and bile stones in whom the CBD entered the lesser curvature of the stomach about 6 cm above the pylorus. However, none of these cases had accompanying gastric carcinoma. Here, we report a case of ectopic opening of the CBD into the stomach associated with gastric carcinoma, and we review the possible relation between the two conditions.

CASE REPORT

A 76-year-old male was referred to our clinic for endoscopic examination. He had a history of abdominal pain, nausea and weight loss. On gastroscopic examination, surprisingly, we determined cholesterol and black gallstones in the stomach (Figure 1). Additionally, there was a slit-like fistulous opening into the antrum (Figure 2), and a few millimetric bile stones fell into the stomach via this orifice during the endoscopy. A depressed ulcerative lesion 2x1 cm in diameter on the incisura angularis was also observed (Figure 3). The bulbus duodeni and the second part of the duodenum were normal, but there was no major papilla in the duodenum. All these findings suggested an anomaly of biliary opening. Biopsies were taken from the ulcerative lesion, which appeared to be malignant. In another session, we performed cholangiography via the fistulous orifice at the antrum with a gastroscope. When we cannulated the fistulous orifice (Figure 4), the bile duct was opacified and the distal part of the CBD appeared hook-shaped, tapered, short, and dilated. The intrahepatic bile ducts

Address for correspondence: Orhan SEZGİN
Mersin Üniversitesi Tıp Fakültesi
Gastroenteroloji Bilim Dalı Mersin, Turkey
Fax: + 90 324 337 43 05
E-mail: sezginorhan@ttmail.com, orhansezgin@mersin.edu.tr

Manuscript received: 15.10.2008 **Accepted:** 08.07.2009

doi: 10.4318/tjg.2010.0076

Figure 1. On endoscopy, cholesterol and black bile stones are seen in the stomach.

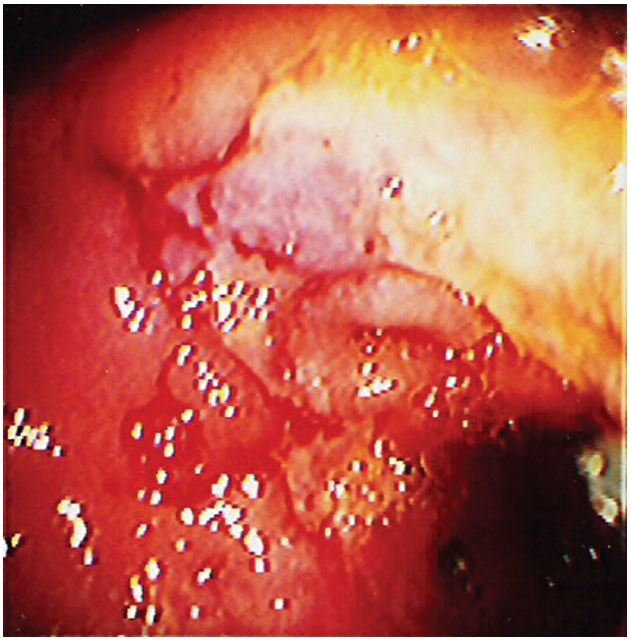

Figure 3. A depressed ulcerative lesion at the incisura angularis, lesser curvature.

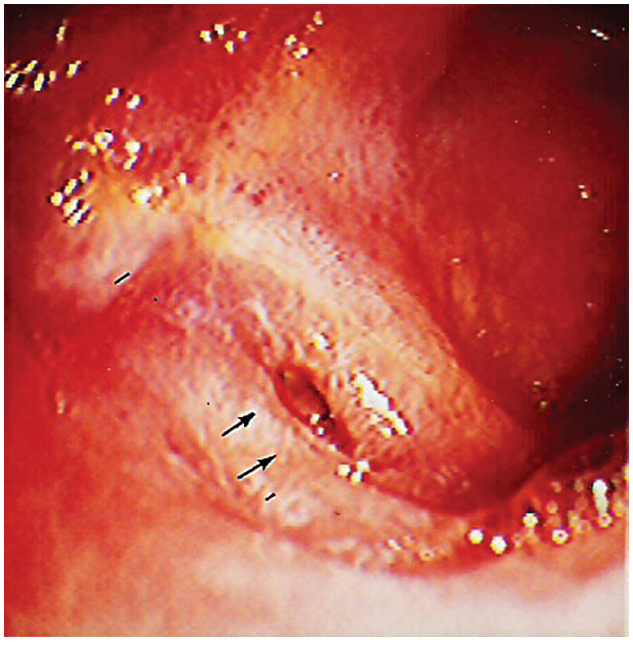

Figure 2. Slit-like ectopic opening of the common bile duct into the antrum is seen (arrows).

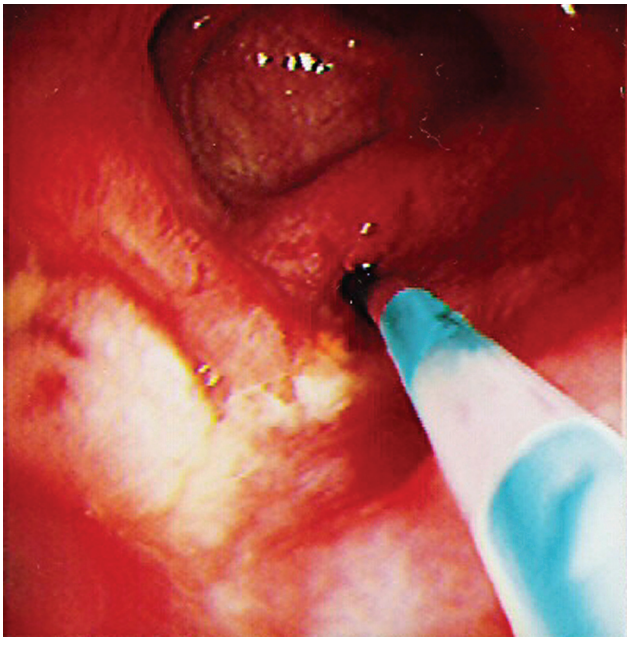

Figure 4. Insertion of the ERCP catheter into the ectopic biliary orifice.

were dilated (Figure 5a, b). Absence of any communication between the biliary system and the duodenum or any other part of the gastrointestinal system suggested ectopic opening of the CBD into the stomach. Some sludge was removed from the bile ducts by balloon-catheter without sphincterotomy. No complication occurred following endoscopic retrograde cholangiography. Biopsy obtained from the ulcerative lesion showed adenocarcinoma. Surgical therapy for gastric cancer was recommended to the patient, but he refused and was lost to follow-up.

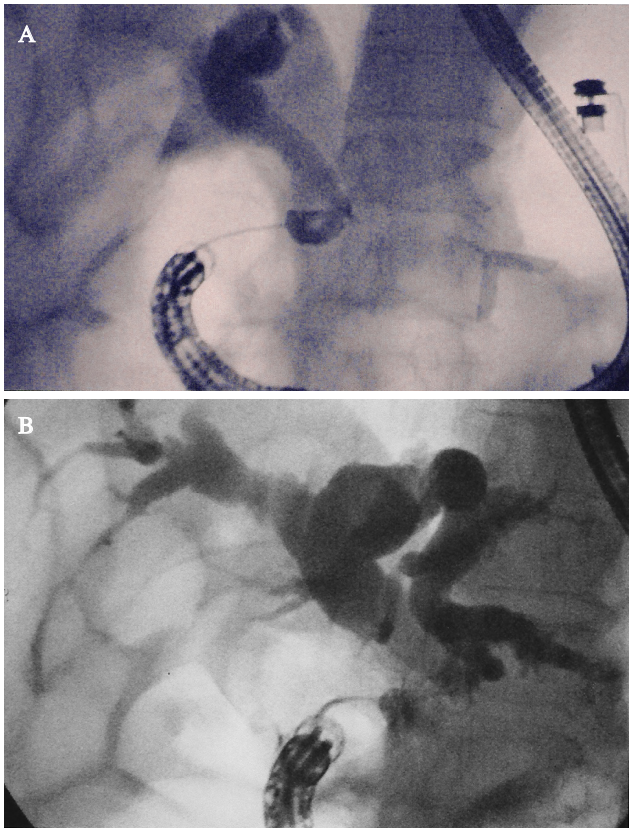

Figure 5. a. When the injection of contrast medium was started, the common bile duct was short with hook-like configuration. Note the shape of the gastroscope. **b.** Following injection of contrast medium, the common bile duct and intrahepatic biliary ducts were dilated.

DISCUSSION

The CBD typically enters the posteromedial aspect of the second portion of the duodenum. Ectopic opening of the CBD into any site other than the second portion of the duodenum is quite rare. However, it can terminate at anomalous sites, including the third or fourth portion of the duodenum, the pyloric canal or duodenal bulb (7-20). Although ectopic accessory bile ducts entering the stomach have been reported rarely, there have been only three cases of CBD opening directly into the stomach (1-6).

The cause of anomalous drainage of the CBD has been ascribed to unidentified errors in embryogenesis. Boyden (21) formulated a hypothesis to explain the generation of ectopic drainage of the CBD in organogenetic stages. Briefly, ectopic bile duct drainage occurs because of disproportional elongation and early subdivision of the primitive hepatic furrow as it develops the pars hepatica and pars cystica (6, 21-23). Ectopic opening of the CBD is commonly associated with biliary tract disease (19). In the present case, the patient had bile duct stones, but there was no identifiable history of severe biliary colic or cholangitis. Plausible explanations for the development of biliary disease are as follows. First, a hook-shaped configuration of the distal CBD, which may result from an acute angulation of the CBD caused by premature drainage of bile into the stomach, may be associated with bile stasis. This hook-shaped configuration of the distal CBD should be considered the characteristic finding for this anomaly (16). Second, a typical configuration of the major duodenal papilla was absent, but a slit-like orifice was present. This deformed configuration of the ectopic opening may reflect a poorly developed sphincter of Oddi or absence of the sphincter. Histologic studies have confirmed absence of sphincteric musculature at the entrance of the accessory bile ducts into the gastrointestinal tract (16, 24). Malfunction of the valve mechanism in the distal CBD permits reflux of intestinal bacteria and gastric contents into the biliary system, which could cause transient obstruction and recurrent cholangitis and liver abscess (6, 8, 12, 25). However, in the present case, absence of the biliary sphincter might have caused free stone passage, and this may account for the lack of biliary symptoms.

There is an anecdotal report that CBD opening into the stomach might cause gastric mucosal injury or gastric ulcer (9). Bile acids usually precipitate in response to acidic pH of the stomach, rendering them innocuous (26). However, there has been a case of periodic symptomatic bile gastritis in which the accessory bile duct had an opening into the stomach. The patient's symptoms subsided following biliary diversion (5). However, when pH increases, bile acids can induce gastric mucosal damage (27). It is not clear whether bile damage causes cancer. Kondo et al. (3) found hyperplasia of the gastric foveolas and atrophy of the fundic gland with its cystic dilatation around the orifice in two patients with ectopic biliary drainage via the accessory bile duct into the stomach associated with gastric carcinoma. Thereafter, Yamashita et al. (25), in their study on double CBDs, reported that concomitant gastric cancer was frequently noted when the accessory CBD opened into the stomach. Although such an association does not prove causation, and a frequent gastrointestinal cancer and a rare anatomic variation may coexist without a cause and effect relationship, Mason and Filipe (28) demonstrated that prolonged exposure of the gastric mucosa to bile may be respon-

sible for atrophic gastritis and predispose to development of gastric cancer. Although we can not prove it, prolonged drainage of excessive bile into the stomach may have played a role in the development of gastric cancer. The advanced age of the patient may also have been a contributory factor.

If an ectopic opening of the CBD is suspected at endoscopy, it should be inspected carefully for flow of bile. In such cases, the opening may be examined by plain upper gastrointestinal barium X-ray, computerized tomography, magnetic resonance cholangiopancreatography or endoscopic ultrasound, but endoscopic retrograde cholangiopancreatography (ERCP) is the "gold standard" (12, 29, 30). On ERCP, the orifice should be cannulated cautiously, and contrast medium should be injected to visualize the CBD and/or the pancreatic duct. Fistula secondary to ulcer or choledocholithiasis, spontaneous or iatrogenic surgical fistula, and surgical choledochoenteric diversion should be included in the differential diagnosis (11). The key to diagnosis is to search for the presence of the major duodenal papilla in its normal location or any other drainage route into the duodenum on cholangiography (16). In the present case, on cholangiography, there was no connection between the bile ducts and any other part of the gastrointestinal tract, and the CBD opened directly into the stomach. If the CBD with ectopic drainage into the stomach is confirmed on cholangiography, a careful endoscopic examination and biopsy of the gastric mucosa surrounding the opening should be performed to rule out cancer.

Management of this anomaly is controversial, with most symptomatic patients being treated surgically or endoscopically. When anomalous drainage of the CBD is associated with choledocholithiasis, it may be extremely difficult to remove the stones by endoscopic methods. The intramural portion of the duct is not developed fully. Therefore, there is a high risk of perforation or bleeding during endoscopic sphincterotomy. For this reason, endoscopic sphincterotomy should be avoided in these patients. Although endoscopic balloon dilation may be practicable, satisfactory placement of a balloon catheter through an ectopic opening is exceptionally difficult because of the acute angulation of the distal CBD (15, 16). However, we successfully performed deep cannulation of the bile ducts and balloon extraction of bile sludge. Surgical procedures to treat these patients and to prevent recurrences or other complications include resection of the ectopic duct and choledochojejunostomy between the biliary section and gastrointestinal tract, and a Roux-Y loop appears to be the best procedure for the prevention of biliary complications and potential risk of gastric carcinoma (5).

An important point about ectopic opening is that it may cause severe damage during the surgical procedure if it goes unrecognized. In fact, the case presented here had stomach cancer. If the patient had had gastrectomy for stomach cancer before the anomalous biliary drainage was recognized, it might have caused severe complications.

In conclusion, in view of this and previous reports, there is at present insufficient scientific evidence to state that a gastric drainage of the CBD may cause gastric cancer. However, it should be kept in mind that although ectopic opening of the CBD into the stomach is extremely rare, it may cause bile duct disease such as gallstones and play a role in the development of gastric cancer.

REFERENCES

1. Kanematsu M, Imaeda T, Seki M, et al. Accessory bile duct draining into the stomach: case report and review. Gastrointest Radiol 1992; 17: 27-30.
2. Sieber WK, Wiener ES, Chang J. Double choledochus with ectopic drainage into the stomach. A rare congenital anomaly of the biliary ductal system. J Pediatr Surg 1980; 15: 817-8.
3. Kondo K, Yokoyama I, Yokoyama Y, et al. Two cases of gastric cancer-bearing double choledochus with ectopic drainage into the stomach. Cancer 1986; 57: 138-41.
4. Horsmans Y, De Grez T, Lefebvre V, Witterwulghe M. Double common bile duct with ectopic drainage of the left lobe into the stomach. Case report and review of the literature. Acta Gastroenterol Belg 1996; 59: 256-7.
5. Bernard P, Le Borgne J, Dupas B, et al. Double common bile duct with ectopic drainage into the stomach. Case report and review of the literature. Surg Radiol Anat 2001; 23: 269-72.
6. Vazquez Quintana E, Labat R. Ectopic drainage of the common bile duct. Ann Surg 1974; 180: 119-23.
7. Lindner HH, Pena VA, Ruggeri RA. A clinical and anatomical study of anomalous terminations of the common bile duct into the duodenum. Ann Surg 1976; 184: 626-32.
8. Kubota T, Fujioka T, Honda S, et al. The papilla of Vater emptying into the duodenal bulb. Report of two cases. Jpn J Med 1988; 27: 79-82.
9. Rosario MT, Neves CP, Ferreira AF, Luis AS. Ectopic papilla of Vater. Gastrointest Endosc 1990; 36: 606-7.

10. Keddie NC, Taylor AW, Sykes PA. The termination of the common bile duct. Br J Surg 1974; 61: 623-5.
11. Pereira-Lima J, Pereira-Lima LM, Nestrowski M, Cuervo C. Anomalous location of the papilla of Vater. Am J Surg 1974; 128: 71-4.
12. Lee HJ, Ha HK, Kim MH, et al. ERCP and CT findings of ectopic drainage of the common bile duct into the duodenal bulb. AJR 1997; 169: 517-20.
13. Lehmann H, Popken H, Schlaak M. The short choledochus syndrome. Case report and retrograde endoscopic visualization of the biliary system. Acta Hepatogastroenterol (Stuttg) 1978; 25: 158-61.
14. Guivarc'h M, Benhamida F. Short common bile duct or high implantation. Apropos of 2 cases. J Chir (Paris) 1976; 112: 237-52.
15. Özaslan E, Sarıtaş Ü, Tatar G, Şimşek H. Ectopic drainage of the common bile duct into the duodenal bulb: report of two cases. Endoscopy 2003; 35: 54.
16. Lee SS, Kim M-H, Lee S-K, et al. Ectopic opening of the common bile duct in the duodenal bulb: clinical implication. Gastrointest Endosc 2003; 6: 679-82.
17. Sfairi A, Farah A. An abnormality of the biliopancreatic junction associated with an ectopic anastomosis of the common bile duct into the 3rd section of the duodenum. Ann Gastroenterol Hepatol (Paris) 1996; 31: 346-8.
18. Li L, Yamataka A, Yian-Xia W, et al. Ectopic distal location of the papilla of Vater in congenital biliary dilatation: implication for pathogenesis. J Pediatr Surg 2001; 36: 1617-22.
19. Doty J, Hassall E, Fonkalsrud EW. Anomalous drainage of the common bile duct into the fourth portion of the duodenum. Arch Surg 1985; 120: 1077-9.
20. McQuillan TC, Castles L. Can the common bile duct drain into the fourth part of the duodenum? Arch Surg 1989; 124: 984-5.
21. Boyden EA. The problem of the double ductus choledochus: an interpretation of an accessory bile duct found attached to the pars superior of the duodenum. Anat Rec 1932; 55: 71-94.
22. Boyden EA. Congenital variations of the extrahepatic biliary tract: a review. Minn Med 1944; 27: 932-3.
23. Moore KL, Persaud TVN. The developing human. 6th ed. Philadelphia: W.B. Saunders, 1998; 276-82.
24. Foulon JP, Hardy JM, Batellier JL, Detrie P. Ectopic opening of the common bile duct. A contribution to the physiological study of the sphincter of Oddi. J Chir (Paris) 1980; 117: 293-8.
25. Yamashita K, Oka Y, Urakami A, et al. Double common bile duct: a case report and a review of the Japanese literature. Surgery 2002; 131: 676-81.
26. Gu JJ, Hofmann AF, Ton-Nu HT, et al. Solubility of calcium salts of unconjugated and conjugated natural bile acids. J Lipid Res 1992; 33: 635-46.
27. Northfield TC, McColl I. Postprandial concentrations of free and conjugated bile acids down the length of the normal human small intestine. Gut 1973; 14: 513-8.
28. Mason R, Filipe I. The aetiology of gastric stump carcinoma in the rat. Scand J Gastroenterol 1990; 25: 961-5.
29. Holz K. Atypical orifice of the common bile duct during localization of the Vater papilla on the rear wall of the duodenal bulb. Fortschr Geb Rontgenstr Nuklearmed 1970; 112: 409-10.
30. Kristic M, Stimec B, Kristic R, et al. EUS diagnosis of ectopic opening of the common bile duct in the duodenal bulb: a case report. World J Gastroenterol 2005; 11: 5068-71.