Double common bile duct with an ectopic drainage into the stomach

Jeong-Ik Park, Sang-Hoon Oh¹

Department of Surgery, Inje University Haeundae Paik Hospital, Inje University College of Medicine, Busan, ¹Department of Surgery, Inje University Busan Paik Hospital, Inje University College of Medicine, Busan, Korea

Double common bile duct (DCBD) is a rare congenital anomaly among biliary anomalies. The anomaly has an important clinical implication because of its association with anomalous pancreaticobiliary ductal union (APBDU) and upper gastrointestinal cancers. In addition, if one of the two common bile ducts is mistaken for the cystic duct during surgery, bile duct injury is likely to occur. Treatment depends on the coexistence of concomitant cancer and APBDU. A 54-year-old male diagnosed of gastric cancer was referred for surgery. During surgery, we incidentally detected bile leak from the tubular structure around the hepatoduodenal ligament. We performed intraoperative cholangiogram by cannulizing into the tubular structure, and confirmed the tubular duct as the accessory bile duct with an ectopic drainage into the stomach, which was connected to the proximal common bile duct. In this study, we report a rare case of DCBD with an ectopic drainage into the stomach and review the literature.

[Ann Surg Treat Res 2015;88(4):229-231]

Key Words: Bile ducts, Abnormalities, Common bile duct, Gallstones

INTRODUCTION

The anomalies of the biliary system are various and frequent, however, double common bile duct (DCBD) is an extremely rare congenital anomaly [1]. One usually has normal drainage into the duodenum and the other, termed accessory common bile duct (ACBD), opens in different parts of the upper gastrointestinal (GI) tract. This anomaly has great significance because it is often associated with bile duct stone, anomalous pancreaticobiliary ductal union (APBDU) and upper GI tract malignancies [2]. We experienced a very rare case of DCBD with an ectopic drainage into the stomach in a gastric cancer patient. Here we report the case and review the literature.

CASE REPORT

A 54-year-old male presented to a primary care physician with a complaint of indigestion. He underwent gastrofiberscopy, which revealed a small circumferential ulceroinfiltrative ill-defined mucosal lesion at the anterior wall of the antrum. Biopsy specimens were taken from the ulceroinfiltrative lesion, and showed adenocarcinoma. He was diagnosed with gastric cancer and referred to our hospital for an operation. We didn't detect any abnormal findings physically or in the laboratory. Abdominal CT scan showed mild gastric mucosal thickening in the anterior wall of the gastric antrum. We decided to perform laparoscopic assisted distal gastrectomy. During the surgery, we incidentally detected bile leak from the tubular structure around the hepatoduodenal ligament, and

Received September 29, 2014, Reviewed October 27, 2014,
Accepted October 28, 2014

Corresponding Author: Sang-Hoon Oh

Department of Surgery, Inje University Busan Paik Hospital, Inje University College of Medicine, 75 Bokji-ro, Busanjin-gu, Busan 614-735, Korea

Tel: +82-51-890-6500, Fax: +82-51-894-7993

E-mail: gsshoh@paik.ac.kr

This study was presented as a poster at 11th World Congress of the International Hepato-Pancreato-Biliary Association (IHPBA), 22-27 March, 2014.

Copyright © 2015, the Korean Surgical Society

© Annals of Surgical Treatment and Research is an Open Access Journal. All articles are distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0/>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

Fig. 1. Operative finding. There is a tubular structure on the left side of the hepatoduodenal ligament which was ligated on the stump (arrow).

Fig. 2. Intraoperative cholangiogram shows that the accessory common bile duct was connected with the proximal common bile duct and linked with the left intrahepatic duct.

therefore converted to open surgery (Fig. 1). We performed intraoperative cholangiogram by cannulizing into the tubular duct, and confirmed that the tubular duct was the accessory bile duct with an ectopic drainage into the stomach, which was connected with the proximal common bile duct and linked with the left intrahepatic duct (Fig. 2). In the specimen, the ACBD was independent of the cancer lesion (Fig. 3). The ACBD was resected afterward. The patient was discharged without complications, and is doing well until now.

Fig. 3. Specimen shows that the accessory common bile duct (ACBD) opening is located in the lesser curvature of the stomach, and ACBD site is away from the gastric cancer lesion of the antrum.

DISCUSSION

The anomalies of the biliary tract are various and common. One of the common encountered biliary anomalies in upper abdominal surgery is the accessory bile ducts opening into the extrahepatic bile ducts or into the gallbladder. Other than classic drainage into the duodenal papilla, ectopic opening of the biliary system into the upper GI tract via the accessory bile duct is an extremely rare anomaly [1]. According to a report by Boyden [3], Vesarius reported the first case of DCBD draining into the duodenum and stomach in 1543. According to Teilum [1], only 24 cases were recognized in the western literature during the 500 years leading up to 1986. On the other hand, Yamashita et al. [2] reviewed 47 cases of DCBD reported in the Japanese literature from 1968 to 2002. Goor and Ebert [4] classified DCBD into seven configuration types according to the anatomical appearance of the anomaly. Saito et al. [5] and Kanematsu et al. [6] modified the classification based on Goor's morphological grouping consisting of four configuration types of DCBD regardless of the site of the ACBD opening. Type I, common bile duct with a septum within the lumen; type II, common bile duct which bifurcates on the way with independent drainage; type III, double biliary drainage without a communicating channel; and type IV, double biliary drainage with one or more communicating channels. Our case was categorized as type II according to this classification. The symptoms of DCBD include epigastric pain, nausea or vomiting, right upper quadrant pain, heart burn, fever, and jaundice though it may also be asymptomatic [6]. Our patient complained of indigestion, but it is unclear if the symptom was due to the gastric cancer, rather

than the DCBD. Yamashita et al. [2] divided ACBD into 5 groups according to the site the duct opened into; the stomach, the first portion of the duodenum, the second portion of the duodenum, the pancreatic duct, or the septum. He also emphasized that the opening site of the ACBD is clinically more important than its anatomical appearance. This is due to two clinical significances of DCBD. First, the duplication of the common bile duct can lead to severe intraoperative injury to one of the two common bile ducts, which can be mistaken for the cystic duct and be ligated [7]. Second, APBDU and concomitant cancers are serious conditions in DCBD patients [2]. According to Yamashita's review, gallbladder cancer, gastric cancer, ampullary cancer and pancreatic cancer developed more frequently in DCBD patients. Gastric cancers developed only in patients with ACBD opening into the stomach; prolonged exposure of the gastric mucosa to components of duodenal juice, bile and pancreatic juice may be responsible for atrophic gastritis and a predisposition to the development of gastric cancer, but this relationship remains uncertain [8]. Gallbladder cancer and ampullary cancer usually developed in patients with ACBD openings into the second portion of the duodenum and pancreatic duct. Most of all, there was a very high incidence of concomitant APBDU with DCBD. In other words APBDU has never been seen in cancer patients with ACBD openings into the first portion of the duodenum or stomach [2]. Due to these two concomitant conditions, preoperative diagnosis is very important for DCBD patients. Unfortunately, preoperative recognition of DCBD is very rare. Adequate preoperative diagnosis is based on radiological and endoscopic findings.

magnetic resonance cholangiopancreatography is useful for demonstrating anomalies and anatomic variants of the biliary and pancreatic duct [9]. In some cases, gastrofiberscopy shows an intragastric biliary opening with or without bile flow [6]. In our case, gastrofiberscopy was performed by the primary care physician, so we couldn't confirm if the intragastric accessory bile duct opening was identified. Treatment of DCBD depends on the coexistence of concomitant cancer and APBDU. In cases without cancer, surgical resection of the ACBD is recommended. In addition, when APBDU is also present, surgical separation of the flow of bile and pancreatic juice into the GI tract should also be performed to prevent cancer in the biliary system [10]. Furthermore, when DCBD without cancer or coexistent APBDU is incidentally detected, a careful endoscopic biopsy of the gastric mucosa surrounding the opening is recommended. If atypical and dysplastic lesion is found, gastrectomy should be performed. Otherwise, periodic endoscopic examination is recommended due to the risk of cancer [8].

In conclusion, we reported a case of DCBD with ectopic drainage into the stomach. Because the opening site of the ACBD in DCBD cases has very important clinical implications with concomitant APBDU and cancers, adequate diagnosis and proper surgical treatment should be considered.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES

1. Teilum D. Double common bile duct. Case report and review. *Endoscopy* 1986;18: 159-61.
2. Yamashita K, Oka Y, Urakami A, Iwamoto S, Tsunoda T, Eto T. Double common bile duct: a case report and a review of the Japanese literature. *Surgery* 2002;131:676-81.
3. Boyden EA. The problem of the double bile ductus choledochus (an interpretation of an accessory bile duct found attached to the pars superior of the duodenum). *Anat Rec* 1932;55:71-93
4. Goor DA, Ebert PA. Anomalies of the biliary tree. Report of a repair of an accessory bile duct and review of the literature. *Arch Surg* 1972;104:302-9.
5. Saito N, Nakano A, Arase M, Hiraoka T. A case of duplication of the common bile duct with anomaly of the intrahepatic bile duct. *Nihon Geka Gakkai Zasshi* 1988;89:1296-301.
6. Kanematsu M, Imaeda T, Seki M, Goto H, Doi H, Shimokawa K. Accessory bile duct draining into the stomach: case report and review. *Gastrointest Radiol* 1992;17:27-30.
7. Paraskevas G, Papaziogas B, Ioannidis O, Kitsoulis P, Spanidou S. Double common bile duct: a case report. *Acta Chir Belg* 2009;109:507-9.
8. Kondo K, Yokoyama I, Yokoyama Y, Harada A, Nagayo T. Two cases of gastric cancer-bearing double choledochus with ectopic drainage into the stomach. *Cancer* 1986;57:138-41.
9. Amano Y, Takahashi M, Oishi T, Kumazaki T. MR cholangiopancreatography of double common bile duct with ectopic drainage into stomach. *J Comput Assist Tomogr* 2002;26:141-2.
10. Djuranovic SP, Ugljesic MB, Mijalkovic NS, Korneti VA, Kovacevic NV, Alempijevic TM, et al. Double common bile duct: a case report. *World J Gastroenterol* 2007;13: 3770-2.