

Congenital left intrahepatic bile duct draining into gastric wall mimicking biliary reflux gastritis

Jian Guan, Ling Zhang, Jian-Ping Chu, Shao-Chun Lin, Zi-Ping Li

Jian Guan, Jian-Ping Chu, Shao-Chun Lin, Zi-Ping Li, Department of Medical Imaging, the First Affiliated Hospital, Sun Yat-Sen University, Guangzhou 510080, Guangdong Province, China

Ling Zhang, Department of Medical Imaging and Interventional Radiology, Cancer Center, Sun Yat-Sen University, Guangzhou 510080, Guangdong Province, China

Author contributions: Guan J and Chu JP designed the report; Zhang L and Li ZP collected the patient's clinical data; Lin SC performed MRCP scan; Chu JP and Guan J analyzed the data and wrote the paper.

Open-Access: This article is an open-access article which was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited and the use is non-commercial. See: <http://creativecommons.org/licenses/by-nc/4.0/>

Correspondence to: Jian-Ping Chu, MD, Department of Medical Imaging, the First Affiliated Hospital, Sun Yat-Sen University, 58th the Second Zhongshan Road, Guangzhou 510080, Guangdong Province, China. truechu@hotmail.com
Telephone: +86-20-87755766
Fax: +86-20-87615805

Received: October 6, 2014

Peer-review started: October 7, 2014

First decision: October 14, 2014

Revised: November 4, 2014

Accepted: December 1, 2014

Article in press: December 1, 2014

Published online: March 21, 2015

including electronic endoscopy, endoscopic ultrasonography, endoscopic retrograde cholangiopancreatography and magnetic resonance cholangio-pancreatography. Finally, congenital ectopic left intrahepatic bile duct draining into the stomach was found, which caused biliary reflux gastritis. The patient did not receive any surgery. Good recovery was achieved by medical treatment.

Key words: Ectopic left intrahepatic bile duct; Endoscopic ultrasonography; Endoscopic retrograde cholangiopancreatography; Magnetic resonance; Cholangiopancreatography

© **The Author(s) 2015.** Published by Baishideng Publishing Group Inc. All rights reserved.

Core tip: Abnormalities and variations of the biliary ducts are not rare. Most aberrant bile ducts eventually drain into the descending part of duodenum through the papilla of Vater. However, drainage of the left hepatic bile duct into the stomach is extremely rare. We report a case with congenital left intrahepatic bile duct draining into gastric wall. The clinical symptoms are similar to bile reflux gastritis and the imaging changes are easily misdiagnosed as gastric tumor. The comprehensive imaging examination is necessary for correct diagnosis.

Guan J, Zhang L, Chu JP, Lin SC, Li ZP. Congenital left intrahepatic bile duct draining into gastric wall mimicking biliary reflux gastritis. *World J Gastroenterol* 2015; 21(11): 3425-3428 Available from: URL: <http://www.wjgnet.com/1007-9327/full/v21/i11/3425.htm> DOI: <http://dx.doi.org/10.3748/wjg.v21.i11.3425>

Abstract

Abnormalities and variations of the biliary ducts are not rare. Most aberrant bile ducts eventually drain into the descending part of duodenum through the papilla of Vater. However, drainage of the left hepatic bile duct into the stomach is extremely rare. A 29-year old man was admitted to the hospital with the diagnosis of biliary reflux gastritis. Comprehensive imaging modalities were performed

INTRODUCTION

Knowledge of the hepatic duct variants is necessary when interpreting images and performing surgical or

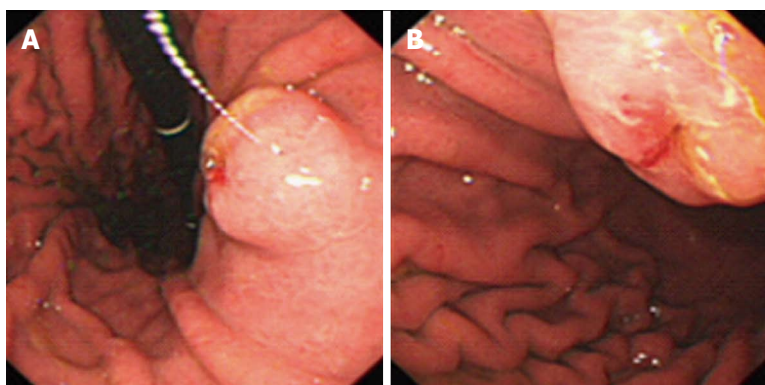


Figure 1 Electronic gastroscopy showed a papillary mass in lesser curvature of the stomach, and some bile on the surface of the mass.

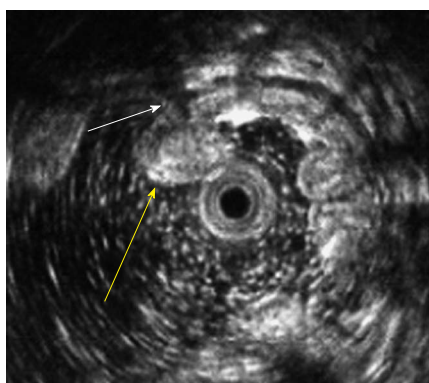


Figure 2 Endoscopic ultrasonography revealed a homogeneously hyperechoic submucosal mass (yellow arrow) with clear margin and a hypoechoic tunnel inside (white arrow).



Figure 3 Endoscopic retrograde cholangiopancreatography showed a tubular structure (white arrow) between left hepatic lobe and lesser curvature of the stomach. GB: Gallbladder.

non-surgical therapeutic procedures. Sharma *et al*^[1] described that the branching pattern of intrahepatic bile ducts (IHD) was atypical in 47% patients. Guerra *et al*^[2] reported that one case of ectopic papilla of Vater in the pylorus. Although anatomical variations of IHD present several types of branching patterns^[1-4], anomalous opening of IHD located outside the biliary tree was rare and only described in case reports. The purpose of this study is to describe an extremely rare anatomic variation of left intrahepatic duct draining into the lesser curvature of stomach.

CASE REPORT

A 29-year old man was admitted to our hospital and presented with upper abdominal pain and burning sensation for more than a year. These symptoms aggravated especially under the circumstance of waking-up in the early morning and starvation, and then relieved after eating and activity. This patient did not have any history of gastrointestinal disease and surgery. Physical examination revealed soft and plain abdomen without tenderness and rebound pain.

The electronic endoscopy showed congestive swelling in gastric antral mucosa and a submucosal papillary mass in the lesser curvature of the stomach.

This mass manifested as smooth surface projecting to gastric lumen with a size of 1.0 cm × 1.0 cm. There was some bile-like liquid draining out from the fissure-shaped opening of the papillary mass (Figure 1). Gastric mucosa biopsy was performed. Endoscopic ultrasonography revealed a homogeneous hyperechoic submucosal mass of around 8 mm in diameter with clear margin. The structure of the affected gastric wall was normal and intact. There was a hypoechoic tunnel crossing the muscularis propria and serous layer of gastric wall (Figure 2). Pathologic result of mucosa biopsy indicated gastric inflammation. Then endoscopic retrograde cholangiopancreatography (ERCP) was performed. Small amount of contrast agent (Ultravist) was injected through the opening of the papillary mass. We observed that contrast agent was traced through the opening into the bile duct-like structures, and then into the left intrahepatic bile duct, and finally into the common bile duct and the duodenum (Figure 3). In order to entirely display the variant bile duct, magnetic resonance cholangiopancreatography (MRCP) was carried out. It showed the variant bile duct arising from the left hepatic duct connected with gastric wall and formed a papillary orifice in the lesser curvature of the gastric body (Figure 4). A hyperintense tubular structure of around 4 mm in diameter was seen

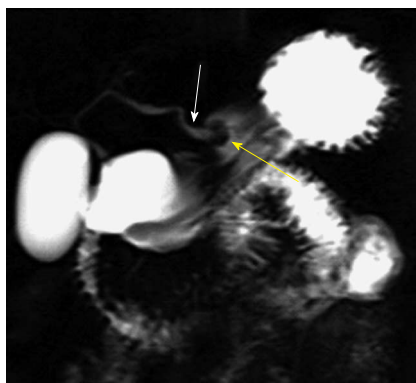


Figure 4 Magnetic resonance cholangiopancreatography revealed the variant bile duct (white arrow) originating from left hepatic duct drained into the lesser curvature of stomach. Note the hyperintense tubular structure (yellow arrow).

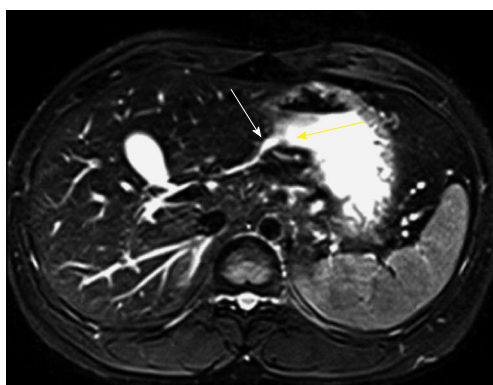


Figure 5 Axial T2WI, a tubular structure (white arrow) with hyperintense signal and its opening (yellow arrow) in stomach can be found as well.

between the left hepatic lobe and stomach on axial T2 weighted imaging (Figure 5). The common bile duct, the right and left hepatic duct, and pancreatic duct were normal without dilatation; and moreover, the gallbladder was also in normal size.

The final diagnosis was congenital ectopic left intrahepatic bile duct draining into stomach, which caused biliary reflux gastritis (Figure 6). The mild biliary reflux gastritis was not indicative for surgery, so the patient was requested to change his dietary habits avoiding greasy food to protect gastric mucosa from biliary stimulation. Prokinetic drugs (Domperidone) and conjugated bile salts drug (Almadrate Sulfate) were prescribed to control the symptoms. The follow-up included periodic monitoring of endoscopy and *Helicobacter pylori* infection. Until now, the symptoms have been alleviated and no evidence of *Helicobacter pylori* infection has been got.

DISCUSSION

Abnormalities and variations of the biliary ducts are not rare. There are different drainage types. Most aberrant bile ducts eventually drain into the descending part

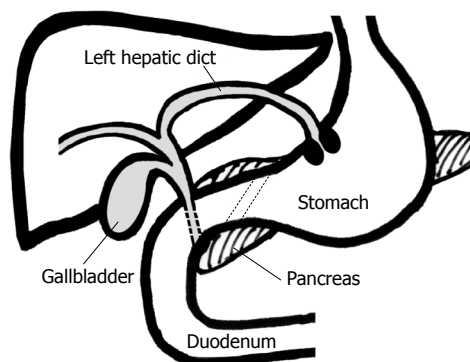


Figure 6 Hand drawing of this case better shows the relationship between variant bile duct and the lesser curvature of the stomach.

of the duodenum through papilla of Vater^[1,2]. Ectopic papilla of Vater located in the duodenal bulb and the pylorus has been described as case reports^[4,5]. However, anomalous opening of the left hepatic bile duct into the stomach is very rare.

In our case, anomalous drainage of the left intrahepatic duct into the lesser curvature of the stomach was found. The biliary duct abnormalities may be explained as the overgrowth of left intrahepatic bile duct extending into the stomach wall in embryonic stage. Part of bile was drained into the stomach *via* this anomalous opening, which caused the symptoms similar to bile reflux gastritis. Therefore, for bile gastritis cases, the common causes include uncoordinated movement disorders of stomach and duodenum, insufficiency of pyloric function and delayed gastric emptying, in addition to the causes mentioned above, the anomalous opening of bile duct in stomach should also be considered.

The anomalous opening of bile duct in the stomach formed papillary structure mimicking submucosal tumors, so it was difficult to make accurate diagnosis only by electronic gastroscopy. It should be differentiated from many submucosal masses, such as interstitialoma, lipoma, carcinoid, schwannoma, ectopic pancreas, etc. Although endoscopic ultrasound can be some helpful in excluding submucosal tumors by demonstrating its internal echo characteristics and location in the gastric wall, especially the tubular hypoechoic structure inside "submucosal papillary mass", it was still difficult to make a qualitative diagnosis as the lack of tracing the tubular structure outside the gastric wall. MRCP is a noninvasive examination as effective as the ERCP in showing the drainage path and opening morphology of ectopic bile duct. The findings by MRCP were in consistent with those by ERCP, which indicates that MRCP might be the first choice for diagnosis.

In our case, the patient didn't receive any surgery and he was well controlled by medical treatment. In addition, periodic follow-up of endoscopy was normal and no *Helicobacter pylori* infection was found. So we continued follow-up.

The ectopic duct is likely to be misdiagnosed as submucosal tumors due to its special structure. Thus, the patient may receive unnecessary surgical procedures or suffer intraoperative accidental injury to the bile duct, resulting in biliary fistula, bile peritonitis or other medical complications. Therefore, it is essential to fulfill comprehensive examinations before proper intervention.

COMMENTS

Case characteristics

This is a rare case about the ectopic opening of the left bile duct into the stomach. The clinical manifestations are very similar to bile reflux gastritis, which presents as abdominal pain and a burning sensation when fasting.

Clinical diagnosis

According to its bile irritation symptoms, this case is diagnosed as bile reflux gastritis.

Differential diagnosis

Clinically, the main differential diagnosis is bile reflux gastritis. The radiological differential diagnosis includes gastric tumor and ectopic pancreas.

Laboratory diagnosis

Laboratory diagnosis is non-specific.

Imaging diagnosis

Imaging findings revealed that the ectopic opening of left hepatic duct situated in the gastric wall, which allows bile to drain into the stomach directly.

Pathological diagnosis

Histopathological examination confirmed gastritis.

Treatment

Treatment including eating little but often, inhibition of gastric acid and gastric mucosa protection can effectively relieve symptoms.

Related reports

As the limit of our knowledge, no related case was reported.

Term explanation

No specific terms were used in our case report.

Experiences and lessons

Bile reflux gastritis is not necessarily caused by abnormal pyloric sphincter. It may be caused by the ectopic biliary opening on the gastric wall, so the comprehensive imaging examination is necessary.

Peer-review

The ectopic opening of the left bile duct into the stomach is a very special anatomic variation of bile duct. The clinical symptoms are similar to bile reflux gastritis and the imaging changes are easily misdiagnosed as gastric tumor. This patient only received symptomatic treatment and long-term follow-up. We still lack of experience in the treatment of the disease since it is a rare condition.

REFERENCES

- 1 **Sharma V**, Saraswat VA, Baijal SS, Choudhuri G. Anatomic variations in intrahepatic bile ducts in a north Indian population. *J Gastroenterol Hepatol* 2008; **23**: e58-e62 [PMID: 18700937 DOI: 10.1111/j.1440-1746.2008.05418.x]
- 2 **Guerra I**, Rábago LR, Bermejo F, Quintanilla E, García-Garzón S. Ectopic papilla of Vater in the pylorus. *World J Gastroenterol* 2009; **15**: 5221-5223 [PMID: 19891024 DOI: 10.3748/wjg.15.5221]
- 3 **Elmunzer BJ**, Taylor JR. Aberrant right hepatic duct with patent ducts of Luschka. *Gastrointest Endosc* 2011; **74**: 196; discussion 197 [PMID: 21531408 DOI: 10.1016/j.gie.2011.02.025]
- 4 **Uchiyama D**, Fujimoto K, Fujimoto N, Hayabuchi N. Anatomic variation of the intrahepatic bile duct. *Intern Med* 2008; **47**: 1631 [PMID: 18797125]
- 5 **Disibeyaz S**, Parlak E, Cicek B, Cengiz C, Kuran SO, Oguz D, Güzel H, Sahin B. Anomalous opening of the common bile duct into the duodenal bulb: endoscopic treatment. *BMC Gastroenterol* 2007; **7**: 26 [PMID: 17610747 DOI: 10.1186/1471-230X-7-26]

P- Reviewer: Intagliata E, Iso Y, Zhong YS

S- Editor: Qi Y L- Editor: A E- Editor: Liu XM





Published by **Baishideng Publishing Group Inc**

8226 Regency Drive, Pleasanton, CA 94588, USA

Telephone: +1-925-223-8242

Fax: +1-925-223-8243

E-mail: bpgoffice@wjgnet.com

Help Desk: <http://www.wjgnet.com/esps/helpdesk.aspx>

<http://www.wjgnet.com>



ISSN 1007-9327



9 771007 932045