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An Unusual Case of Pathological Bile Reflux: A Case Report

[1]Shah SP, [2]Sharma P, [3]Gautam R, [4]Kumar S, [5]Pandey AK, [6]Tandon V, [7]Govil D

[1-7]Department of Surgical Gastroenterology, Apollo Hospitals- New Delhi

Corresponding Author: Shah SP, Department of Surgical Gastroenterology, Apollo Hospitals- New Delhi

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Abstract

Background: Bile reflux refers to reflux of the contents in the duodenum such as bile, pancreatic juice, and duodenal juice back to stomach. The Incidence of bile reflux after cholecystectomy is reported between 40-60%. However severe and symptomatic bile reflux is rare. Choledocho-duodenal fistula formation after cholecystectomy is even more infrequent.

Case: A forty eight year lady with previous history of open cholecystectomy for gall stone disease, presented to Indraprastha Apollo Hospital with complaint of episodes of upper abdominal pain and regurgitation that started after one year of the surgery. She underwent multiple evaluation over past seven years without any conclusive pathological diagnosis. Upper Gastrointestinal endoscopy revealed voluminous amount of bile in the esophagus. MRCP and Endoscopic ultrasonography of upper Gastrointestinal tract that was suggestive of fistulous communication between CHD and first part of duodenum. She underwent exploratory laparotomy, the fistulous tract between proximal CHD and first part of duodenum was delineated and biliary diversion by Roux-en-Y hepaticojejunostomy was done. Patient has resolution of all the symptoms after surgery.

Conclusion: We have a rare case of severe bile reflux after open cholecystectomy with anatomical pathology of choledochoduodenal fistula formation. Biliary diversion in the form of Roun-en-Y hepaticojejunostomy

Keywords: MRCP, CHD, Reflux.

Introduction

The incidence of bile reflux after cholecystectomy is reported between 40-60% (1,2). However severe and symptomatic bile reflux is rare. Choledocho-duodenal fistula formation after cholecystectomy is even more infrequent. Bile reflux refers to reflux of the contents in the duodenum such as bile, pancreatic juice, and duodenal juice back to stomach, leading to inflammation. Spectrum of bile reflux can vary from duodenogastric reflux to duodeno-gastro-esophageal reflux. In pathological bile reflux, there is a continuous reflux at a greater rate than in normal subjects mainly due to gastroduodenal dysmotility, disorder of gastroduodenal hormones, or

surgical alteration of pylorus. It is also seen in association with previous biliary procedures, sphincterotomy and stenting.

A case of choledochoduodenal fistula following open cholecystectomy that presented with atypical chronic cough, has been reported in PubMed in year 2021 (3). We report a case of severe duodeno-gastro-esophageal bile reflux secondary to a spontaneous choledochoduodenal fistula following open cholecystectomy.

Case Report

A forty eight year lady, with no medical comorbidity, with previous history of open cholecystectomy for gall stone disease eight years back in Ethiopia, presented to Indraprastha Apollo Hospital (IAH) Delhi. She had complains of occasional episodes of upper abdominal pain and discomfort ,regurgitation and heart burn that started after one year of the surgery. The symptoms were progressively worsening over the period to the severity that she had episodes vomiting of bile in the recent days.

She underwent multiple evaluation over past seven years without any conclusive pathological diagnosis. With the background of persistent and worsening severe symptoms, that affected her quality of life and day to day activity, she was thoroughly evaluated. Upper Gastrointestinal endoscopy revealed voluminous amount of bile in the esophagus with Los Angeles Grade A esophagitis. PH impedance study was done , reported De-Meester score of 12 however it is insensitive to alkaline reflux (figure 1).

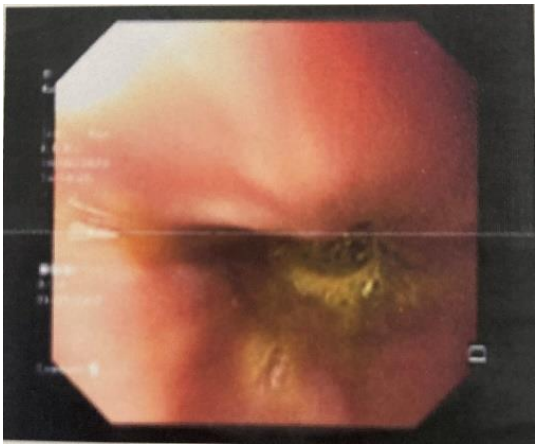

Figure 1

Figure 2

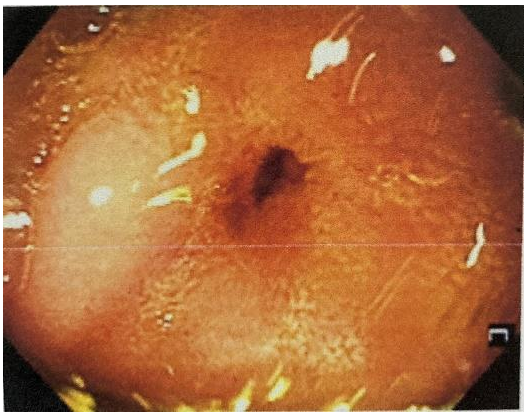

Figure 3

MRCP was done, that showed diffuse mural thickening of CHD and CBD, the first part of duodenum was closely adherent to CHD, that raised the suspicion of possibility of communication between first part of duodenum and CHD without definable communication (figure 2). As we

were not able to reach to a definite diagnosis, we further went ahead with endoscopic ultrasonography of upper Gastrointestinal tract that was suggestive of fistulous communication between common hepatic duct and first part of duodenum (figure 3).

All the details of the radiological and endoscopic findings were discussed with the patient. Keeping in view, the severity of the symptoms affecting her day to day activity and patient's choice for definitive management, she was planned for surgery. She underwent Exploratory Laparotomy. Intraoperatively, there was dense adhesions between stomach, duodenum and anterior parietal wall. After meticulous dissection, a fistulous tract between proximal CHD and first part of duodenum was delineated (figure 4).

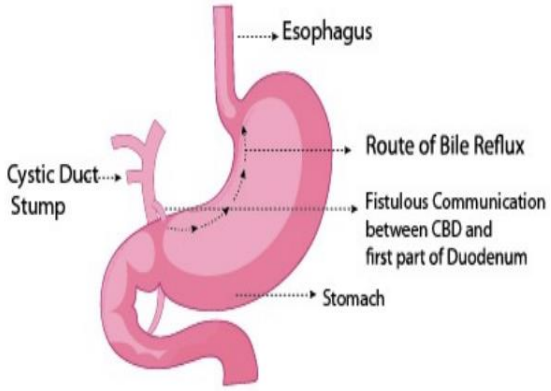

Figure 4

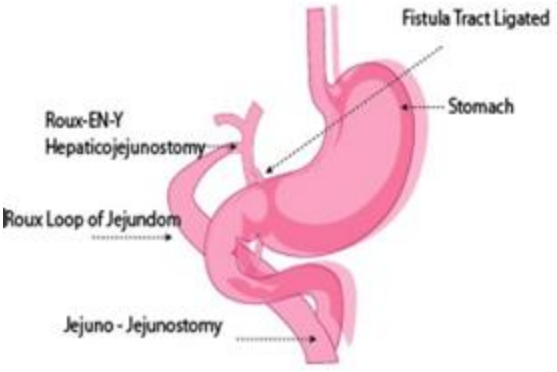

Figure 5

The fistula tract was ligated and Roux-en-Y side to side hepaticojejunostomy was done (figure 5). In the post operative period, she had uneventful recovery. She is being followed up regularly since last 2 years. Currently she has resolution of all the symptoms.

Dicussion

Some degree of bile reflux is frequently observed in post cholecystectomy patient, however severe pathological bile reflux is rare. The diagnosis of bile reflux gastritis is still challenging for its atypical clinical symptoms and nonspecific auxiliary examinations. Abdominal pain, dyspepsia, nausea with bilious vomiting, bitter taste, poor appetite, and heartburn are seen in some patients even while other do not have symptoms. Moreover, the severity of these symptoms is not found to be proportional to the amount of bile in the reflux. Secondary bile reflux post cholecystectomy may have various mechanism of development. Most explained mechanism are: a) malfunction of the sphincter of Oddi that may be caused by injury of the direct neural pathways between gall bladder and duodenum, b) Pressure of the bile duct relatively increases after cholecystectomy, leading to powerful discharge of bile from the bile duct to the duodenum, that can produce a continuous flow and exceed the clearing capacity of the duodenum (4).

Sphincter of Oddi may also be disturbed in previous biliary procedures like sphincterotomy and stenting. Development of choledocho-duodenal fistula further bypasses the regulation of sphincter of Oddi and add to development of pathological bile reflux (5). Our patient presumably had chronic biliary tree inflammation that might have led to gradual adhesion and formation of fistula between CHD and duodenum that possibly added to disturbance in bile flow and subsequent development bile reflux.

Although there is no gold standard for diagnosis of Bile reflux, four vital techniques including hepatobiliary scintigraphy, gastroscopy with aspiration of gastric juice, fiberoptic bilirubin monitoring, and esophageal impedance-pH testing are commonly recognized to be most accurate tools. Hepatobiliary scintigraphy shows radiotracer in the stomach to prove reflux. It is deemed as the least invasive investigation with good tolerability and sensitivity. However, price and radiation exposure limit its application (6). The grading of Duodenogastric reflux is done on the basis of classification by Thomas WEG (7). After insertion of gastroscope for over one-minute, continuous gastrointestinal reflux can be seen in patients. Visualization of bile stain more effectively indicates the retention of a large volume of bile juice in the stomach. (8)(9) However, lack of specificity in terms of endoscopic manifestations imposes restriction on its practical value. Aspiration of gastric fluids enables chemical analysis of the composition of fluids to testify the presence of bile acids. It also allows for histological determination of Bile reflux (10). Bilitec probe, based on the theory that bilirubin absorbs light at a specific wavelength, can deduce the presence of bile (4)(11). Esophageal impedance-pH testing is highly sensitive for all kinds of reflux. It uses a combination of reflux data as a marker for the presence of bile reflux (12). Although it is an advancement in the detection of reflux, it represents a measure of entire reflux instead of a particular measure of bile reflux. Diagnosis of CDF depends on some suspicious radiological findings i.e. pneumobilia on USG, opacification of bile duct on barium study and demonstration of fistulous communication in CECT abdomen. Endoscopy and or Endoscopic Ultrasound helps to confirm the diagnosis.

To date, there no unified therapeutic regimen is documented for the patients with Bile reflux. Related studies on the treatment of BRG are ongoing, and the first priority is to eliminate risk factors. Patients ought to quit cigarettes and wine, control blood glucose, stick to a healthy and regular diet. Use of ursodeoxycholic acid (UDCA), hydrotalcite, proton pump inhibitors (PPIs), and prokinetic agents are widely accepted for the treatment of Bile reflux. UDCA, which has been proved to have explicit curative effects, plays a role both in protecting gastric mucosa and reducing reflux. UDCA can also promote the excretion of endogenous bile acids, reduce bile viscosity, and accelerate the flow of bile (13). Hydrotalcite relieves abdominal discomfort to some extent by neutralizing bile acids and enhancing the effect of the mucosal barrier (14). Proton pump inhibitors inhibit can relieve digestive symptoms caused by acid reflux. Prokinetic agents aim to enhance gastric and duodenal peristalsis and accelerate gastric emptying. The efficacy of medical management varies from patient to patient. It is found that the efficacy of a single drug for secondary BRG is poor, hence combination therapy such as UDCA combined with hydrotalcite are found to be superior to other options. If none of the above works, surgical management of bile reflux with the purpose of diverting bile away from the stomach has been advocated. In our view, Roux-en-Y Hepaticojejunostomy should can be taken as first choice of surgery.

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