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Cholecystolithiasis Complicated with Common Bile Duct Duodenal Fistula: A Case Report

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Abstract

Rationale: Common bile duct duodenal fistula (CDF) is a relatively rare type of internal fistula. It is a pathological fistula formed between the common bile duct and the duodenum. Due to its deep anatomical position, CDF is not easy to be found. In recent years, with the continuous improvement of imaging diagnostic level, more and more CDF has been diagnosed. **Patient concerns:** The patient was a 69-year-old woman who came to our hospital with recurrent epigastric pain and discomfort for 1 year.

Diagnosis: She had upper abdomen contrast-enhanced computed tomography (CECT) scan which showed cholecystolithiasis with cholecystitis; The biliary tract had accumulated gas and was dilated; There was partly intestinal gas and liquid accumulation and gas liquid level; The spleen was found to be large. Magnetic resonance cholangiopancreatography (MRCP) was undertaken which showed suboptimal intrahepatic and extrahepatic bile duct development, suboptimal gallbladder development, slightly rough wall, and patchy filling defect. Duodenal microscopy suggested that the sinus tract above the duodenal papilla was formed, and a 0.3cm×0.3cm opening was observed on the side of the papillary mouth, and bile was discharged after suction and the bile samples indicated elevated amylase and phospholipase A2.

Interventions: The patient were enjoined from eating water and food, Intravenous infusion of vitamin C, vitamin B6 and amino acids was used for symptomatic and supportive treatment. Meanwhile, lansoprazole was given for acid inhibition to protect gastric mucosa, levofloxacin and cefminol sodium for anti-infection, so as to relieve the symptoms of abdominal pain. Due to low blood potassium, the patient was given intravenous infusion to replenish potassium and maintain water electrolyte balance.

Outcomes: The patient's family members voluntarily discharged the patient after consultation considering the patient's older age.

Lessons: In elderly women with a long history of cholecystitis, ERCP should be performed to exclude CDF if bile duct pneumatosis is suggested in addition to upper abdominal CT examination, and surgical treatment is preferred when CDF is confirmed. Those who are not surgically fit to undergo major surgery could be treated with intravenous alimentation by means of total parenteral nutrition and gastro-jejunostomy bypass or gastro-lower duodenostomy by-pass.

Keywords: Common bile duct duodenal fistula; Diagnosis; treatment.

Abbreviations:

CDF: Common bile duct duodenal fistula ERCP: Endoscopic Retrograde Cholangiopancreatography MRCP: Magnetic resonance cholangiopancreatography CT: computed tomography EST: Endoscopic sphincterotomy **ENBD:** Endoscopic nasobiliary drainage **ERBD:** Endoscopic retrograde biliary drainage **EMBE:** Endoscopic metal biliary endoprosthesis

Introduction

Common bile duct duodenal fistula is a special form of choledochoenteric fistula, which is a pathological channel connecting the common bile duct to the duodenum. Common bile duct duodenal fistula has a low incidence. Patients often Come to the hospital presenting with the symptoms of acute cholangitis including fever, right upper abdominal pain and jaundice (we Called this Charcot triad). Due to the atypical clinical manifestations and the inability of some imaging examinations to directly confirm the diagnosis, the diagnosis of choledoco-enteric fistula often tends to be difficult and its misdiagnosis could be easy.

Case presentation

The patient was a 69-year-old female who had been having episodes of recurrent epigastric pain and discomfort over the preceding one year. About 1 year preceding her presentation, the patient had developed pain and discomfort in her upper abdomen without inducement, mainly within her right upper abdomen, with persistent distension pain which was accompanied by paraphylactic colic, and which was ensued by radiating pain in her shoulder and back, and occasional nausea. She did not have any episodes of vomiting, fever, chills, chest pain, chest tightness or any other discomfort. She had colour-Doppler ultrasound scan of her abdomen in her local hospital which had indicated that she had "cholecystitis", and because of which she had received "anti-inflammatory" treatment with infusion, but the effect of the treatment was not obvious. Pursuant to the aforementioned treatment, the patient had been suffering from pain and discomfort within her right upper abdomen. The doctor in the local hospital suggested that she should undergo surgical treatment, but the patient refused. Upon her admission, her abdominal examination revealed that her abdomen was flat and soft, with deep tenderness in the right upper quadrant of the abdomen, which was obvious at the intersection of her right midclavicular line and costal margin, without rebound tenderness. Relevant auxiliary tests were improved. The results of her routine haematology and biochemistry blood tests indicated a slight increase in glutamic-oxalacetic transaminase and indirect bilirubin, and a slight decrease in serum potassium. She had contrast-enhanced computed tomography (CECT) scan of her upper abdomen which suggested presence of gallstone with cholecystitis, intrahepatic and extrahepatic bile duct and gallbladder gas with extrahepatic bile duct dilatation, gas and fluid accumulation within part of the intestinal tract, visible gasliquid level, and enlarged spleen (Figure 1 B - C). She had

magnetic resonance cholangiopancreatography (MRCP) which suggested suboptimal development of intrahepatic and extrahepatic bile ducts, with no definite filling defect, dilatation, or stenosis. The gallbladder was poorly displayed, with a slightly rough wall, patchy filling defect and unclear pancreatic duct (Figure 1 D). She underwent gastroscopy, duodenal endoscopic examination and endoscopic ultrasonography which demonstrated chronic non-atrophic gastritis, the formation of the sinus tract above the duodenal papilla, the main papilla was found in the descending segment, nodular, good opening, no ulcer, diverticulum and new organisms. A 0.3cm×0.3cm opening was observed on the side of the papillary orifice, and bile outflow was observed after suction, we collected bile samples and performed laboratory tests, which indicated elevated amylase and phospholipase A2 (Figure 1 A). The patient was therefore diagnosed as having the following: 1. Choledochoduodenal fistula 2. Gallstones with cholecystitis 3. Splenomegaly 4. chronic atrophic gastritis 5. hypokalemia.

Active treatment was given, we enjoined the patient from eating water and food, Intravenous infusion of vitamin C, vitamin B6 and amino acids was used for symptomatic and supportive treatment. Meanwhile, lansoprazole was given for acid inhibition to protect gastric mucosa, levofloxacin and cefminol sodium for anti-infection, so as to relieve the symptoms of abdominal pain. Due to low blood potassium, the patient was given intravenous infusion to replenish potassium and maintain water electrolyte balance. After the symptoms of abdominal pain were improved, the patient was instructed to have a liquid diet, eat less and have more meals, and gradually increase food intake. After the patient's general condition had improved, we explained the patient's condition with her family, and suggested active surgical treatment should be carried out. The surgical treatment option that was recommended was cholecystectomy plus exploration of the common bile duct and the patient's management included keeping the gastrointestinal tract unblocked, and oral laxative drugs and venting drugs were given to treat intestinal gas accumulation. The family members of the patient considered that the patient was old, the operation risk was high, and the operation effect could not be guaranteed, we communicated with the patient again, and suggested endoscopic duodenal papillary sphincterotomy (EST), which could remove the stone while opening the fistula. Finally, the patient's family chosed conservative treatment and was discharged automatically. The patient was discharged automatically, and no follow-up was performed after her discharge.

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Figure 1. The patient had enhanced upper abdominal CT, duodenal endoscopic and MRCP [**A:** Endoscopic peripapillary fistula about 3mm in diameter; **B:** Upper abdominal CT showed gallstone and atrophic gallbladder; **C:** Intrahepatic bile duct gas accumulation; **D:** The development of MRCP cholangiography system is not obvious (indicating the presence of gas)]

Discussion

The incidence of CDF accounts for 5-25% of all biliary internal fistulas [2,3]. Since the first cases of CDF were reported in 1840, few cases have been reported in the world literature. Singh et al mentioned in their report that cholecystoduodenal fistula was relatively common in choledochoenteric fistula [4], while related studies in China found that choledochoduodenal fistula was relatively common, and the differences were considered as follows: When patients develop choledocholithiasis with chronic cholangitis, Chinese people tend to tolerate it. Long-term chronic inflammatory stimulation leads to fistula of the common bile duct, and CDF occurs when the fistula is opened in the duodenum.

Our patient was an elderly female, who had recurrent episodes of epigastric discomfort over a period of 1 year. Duodenal endoscopic examination and endoscopic ultrasound examination of the patient suggested that the sinus tract above the duodenal papilla was formed with bile outflow, and the bile samples indicated elevated amylase and phospholipase A2, while endoscopic ultrasound suggested that the fistula was connected to the common bile duct. It was considered that the CDF was caused by long-term chronic stimulation of biliary calculi.

At present, there are two main classification methods for common bile duct duodenal fistula, respectively proposed by professor ikeda and professor jianping gong [5-7]. The former is divided into type i and type ii according to the location of the fistula. Type i refers to a fistula which is located on the longitudinal plica of the duodenal papilla, while type ii refers to a fistula which is located on the duodenal bulb. According to Professor Ikeda's classification, our patient 's fistula is type I. The latter is divided into A, B and C according to the location of the fistula. Type A fistula is more than 2 cm away from the duodenal papilla, type B fistula is less than 2 cm away from the duodenal papilla, and type C fistula is also called common bile duct para-duodenal fistula, and the fistula is located at the fold of the duodenal papilla. According to the classification provided by Professor Jianping Gong, our patient was classified as having type C: para-duodenal fistula.

It has been stated that the incidence of choledocoduodenal fistula is related to age and sex of the patient, and that the older the person is, the higher the incidence is, which may be related to long-term biliary tract diseases in the elderly. The prevalence rate of choledocoduodenal fistula in women is higher in comparison with that of men, which is considered to be related to higher incidence of cholelithiasis in women [6]. It has also been documented that CDF is most common in choledocholithiasis, cholelithiasis, or perforation of duodenal ulcer [6-12].CDF has also been reported as a complication of recurrent biliary tract infection, adjacent tumours, tuberculosis, radiotherapy and iatrogenic injury [13-15]. Our patient was in line with the conditions of elderly women, cholelithiasis and repeated biliary stimulation. It was considered that after long-term cholecystolithiasis, falling stones were incarcerated in the stenosis of the lower section of the common bile duct, long-term stimulation of the bile duct caused inflammatory oedema of the duct wall, and longterm ischemic necrosis of the duct wall formed internal fistula with the intestine.

It has been documented that abdominal pain is a common clinical manifestation of CDF, followed by fever and jaundice [16], in addition to nausea, vomiting, diarrhoea, abdominal distension and other common gastrointestinal discomfort, which are often misdiagnosed as digestive tract ulcer, cholecystitis and other diseases, thus delaying treatment.

CDF is mainly differentiated from cholecystoduodenal fistula. The main difference between the two lies in the different location of occurrence. The X-ray examination of cholecystoduodenal fistula can reveal the accumulation of gas in the gallbladder. Barium meal examination shows that the contrast agent enters the gallbladder; Upper abdominal CT scan will indicate Riger triad: ansa dilatation, bile duct gas accumulation, and unclear boundary between gallbladder and duodenal wall; In clinical manifestations, cholecystoduodenal fistula is more prone to calculous ileus. Our patient met the common clinical manifestations of CDF in that she had recurrent epigastric discomfort, gallbladder pneumatosis, but the gallbladder wall was intact, and a parapillary fistula was seen by during her duodenoscopy. In addition, CDF should also be differentiated from parapillary diverticulum. The diverticulum has an opening near the duodenum, but the edge is neat, the openings are usually round, and food residues are common in the holes. After the catheter is inserted into the holes, it will fold back to the intestinal cavity, and all the overflow can be seen after the injection of contrast agent.

It has been iterated that at present, CDF can be diagnosed directly or indirectly through auxiliary examinations: imaging examinations such as upper abdominal Bultrasound scan, CT scan, MRCP and other imaging examinations often suggest biliary pneumatosis, and 14% to 58% of CDF patients have biliary pneumatosis [17], but biliary pneumatosis is not unique to CDF. In cases of choledochoduodenal fistula, plain erect x-ray would tend to demonstrate air under the diaphragm and the patient may also have evidence of right shoulder tip pain. Additionally, plain abdominal x-ray would tend to demonstrate air within the para-duodenal fossa. During digestive tract radiography, the patient is allowed to take the semi-upright prone position, which makes it easier for the contrast agent to enter the fistula. When there is a common bile duct duodenal fistula, the examiner can see the deformation of the duodenum and the contrast agent retrogradely passing into the biliary duct through the fistula, but the size of the fistula cannot be observed by digestive tract radiography. At this time, the examiner can observe the size of the fistula through duodenal endoscopy, but it cannot be determined whether it is an internal fistula formed between the bile duct and the intestinal wall. Therefore, ERCP has become the most accurate auxiliary examination for the diagnosis of CDF, through which not only the fistula size but also the fistula location can be accurately observed [19]. Our patient had a long-term history of cholecystitis with cholecystolithiasis. CT examination of the upper abdomen suggested that there was gas accumulation in the bile duct. The features of the CT scan findings enabled us to suspect the existence of a fistula. At this point we could diagnose CDF rather than cholecystoduodenal fistula or duodenal diverticulum.

There is no uniform standard for the treatment of CDF, and there are many treatment methods, which are mainly determined according to the etiology, the severity of the disease and the general condition of the patients. Choi et al. [18] believed that CDF is prone to retrograde biliary tract infection, and in order to reduce the risk of recurrent biliary tract infection and biliary stenosis after fistula healing, CDF should be actively treated even if there are no symptoms or small fistulas. Moreover, CDF is a high risk factor for cholangiocarcinoma, which requires surgical treatment even if patients have no obvious clinical symptoms [19].

The treatment of CDF depends on the etiology and classification of internal fistula [20]. The researchers' treatment of CDF is summarized as follows:

1.Endoscopic treatment of CDF: Since incarceration of common bile duct stones is the most important cause of CDF, endoscopic duodenal papillary sphincterotomy (EST) can be used for the treatment of CDF, it can remove the stones while cutting fistula, which has become the most commonly used method for treating CDF [21]. Specifically, including: ① fistula expansion: suitable for small fistula which can't unobstructed drainage, in order to achieve the purpose of unobstructed drainage, change the condition of not smooth. The method of fistula enlargement can be directly expanded from the fistula along the inner segment of the bile duct wall, or from the main nipple up to the fistula, if necessary, continue to open up, in order to smooth drainage, the papillary sphincter should be completely cut open.⁽²⁾ Elimination of lesions: if combined with bile duct stones, bile duct stones can be removed at the same time.(3) Smooth drainage: ENBD, ERBD, EMBE are feasible. The above methods can be used either alone or in combination, so that CDF endoscopic therapy can replace partial surgical treatment, thus creating a new approach for CDF therapy. West and Shellenberger [22] reported an elderly CDF patient with choledocholithiasis who underwent EST plus calculus removal under endoscopy and biliary stent implantation at the same time. Six weeks later, the fistula of the patient was completely healed and the stent was removed. Shao and Chen [23] believed that endoscopic papillary balloon dilation for stone removal was safer than EST, with fewer complications such as postoperative bleeding and pancreatitis. The above treatment methods provide a new scheme for the endoscopic treatment of CDF, but the closure of postoperative fistula and the occurrence of reflux cholangitis need further study.

2. Biliary reconstruction treatment of CDF: Due to the difficulty of self-healing of CDF sinus tract, biliary reflux is still the main cause of recurrent cholangitis. Endoscopic therapy is mainly applicable to patients with small fistulas; for patients with large fistulas or poor endoscopic effect, biliary enterostomy can effectively solve the reflux of food and gastroenteric fluid. Li et al. [24] proposed the following strategies : (1) When fistula greater than 1cm, CBD greater than 2cm and associated with reflux cholangitis, after removing the stones, Roux-en-Y choledochojejunostomy or Roux-en-Y intrahepatic cholangiojejunostomy to build an effective drainage, a transection of the CBD was applied to prevent the reflux of duodenal juice; (2) A side-to-side choledochojejunostomy without transecting the duct and an effective biliary drainage was applied when fistula diameter was between 0.5 and 1.0cm, CBD greater than 2cm, no biliary complications or duodenal fluid reflux. ③ For CDF secondary to cholecystolithiasis and fistula diameter larger than 0.5cm, CBD larger than 1.2cm, cholecystectomy plus biliary probe was performed, the latter aims to check Whether residual stones falled in the bile duct or not. ④When the orifice fistula was less than 0.5 cm and there were no complications of biliary tract, nonoperative treatment was used. Historically, CDF has

been an indication for conversion to laparotomy in laparoscopic cholecystectomy [25]. However, laparoscopic surgery has become more and more mature. The experience collected from 19 surgeons in this paper shows that with the continuous success of laparoscopic methods,

Conclusion

CDF is common in elderly women with a long-term history of cholecystitis, clinical manifestations of repeated upper abdominal symptoms and other symptoms, and imaging examination showed biliary gas and fluid accumulation. CDF can be diagnosed through ERCP, and the treatment method should be determined according to the type of fistula and the etiology of the patient. If CDF is misdiagnosed or missed, bile will repeatedly stimulate the bile duct wall, leading to recurrent cholangitis in patients, and long-term inflammation will lead to bile duct cancer. High entero-cutaneous fistula could also result with loss of excessive fluid and electrolytes and there could be development of right sub-phrenic abscess as well as paraduodenal abscess. Therefore, with the continuous improvement of auxiliary examination technology, the diagnostic rate of CDF is also gradually improved. Its treatment is also gradually clear, so as to improve its clinical cure rate, so that it has become a common and easily cured disease. In areas where ultrasound scan is not immediately available, erect chest x-ray would show air under the diaphragm, and plain abdominal x-ray would tend to demonstrate para-duodenal gas which should alert the clinician to the possibility of CDF for which reason the patient should be referred on to a tertiary hospital where facilities for ultrasound scan of abdomen, including Doppler ultrasound scan, contrast-enhanced ultrasound (CEUS) scan, Computed Tomography Scan, Magnetic Endoscopic Resonance Imaging, retrograde cholangiopancreatogram, as well as per-cutaneous cholangiopancreatogram are available for the appropriate investigation which would then be ensued by the most appropriate management of the patient who has CDF.

Author contributions

Han Li and Chenglong Guo contributed to literature research and draft the manuscript. Dongdong Ji and Xuewen Wang contributed to provide the relevant images. Xingyuan Zhang contributed to collect clinical data. Lingqun Kong contributed to corrections to the manuscript and figure editing. All authors have approved the final version of the manuscript.

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CDF can also be closed and the conversion rate of some other methods to open has been reduced. The fistula was removed laparoscopically, stitched manually, and closed with titanium clips. No death or postoperative complications were reported [26].

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