

Submucosal Juxtapapillary Duodenal Diverticulum Complicated with Lemmel's Syndrome: A Case Report and Literature Review

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Abstract

Introduction: Juxtapapillary duodenal diverticulum can cause chronic inflammation of the ampulla, swelling of the nipples, compression of the common bile duct and pancreatic duct openings, and leading to Lemmel's syndrome. Juxtapapillary duodenal diverticulum complicated with Lemmel's syndrome can cause severe signs, such as persistent abdominal pain, cholangitis, pancreatitis, and obstructive jaundice, which is easily confused with other diseases (such as pancreatic tumors and bile duct stones), the rate of misdiagnosis has increased. Therefore, timely identification, diagnosis and treatment to alleviate the symptoms of patients pose certain challenges for clinicians.

Case presentation: An 81-year-old patient had a history of hypertension, diabetes mellitus, recurrent pancreatitis, and gallstone disease. Admitted to hospital for half an hour with persistent mid-upper abdominal pain. At that time, we considered that it was caused by choledocholithiasis. However, after preliminary CT examination, a mixed high-density shadow was found near the descending part of the duodenum, the gallbladder was enlarged and stones were formed, and no choledocholithiasis was found. After admission, abdominal pain was relieved after the treatment of fasting, acid suppression, fluid replacement, and anti-infective, but the next day, abdominal pain recurred, and bilirubin increased significantly. In order to further clarify the cause of this disease, MR cholangiopancreatography was performed, and high density of masses in the descending part of duodenum was found. Dilatation of common bile duct and pancreatic duct, and no choledocholithiasis were found. Due to the patient's signs were atypical, we were somewhat confused in the diagnosis. After our comprehensive evaluation and discussion, further duodenal endoscopy and tissue biopsy were performed and diagnosed as juxtapapillary duodenal diverticulum, which originated from submucosa. Due to the particularity of this disease and a comprehensive evaluation of the patient, we performed the treatment of EST (Endo-scopicsphincterotomy), and the patient's symptoms gradually improved after surgery. After follow-up, no obvious abdominal discomfort was found.

Discussion and Conclusion: Juxtapapillary duodenal diverticulum complicated with Lemmel's syndrome may not have typical symptoms and specific complications, and the rate of misdiagnosis is high. When patients have symptoms that cannot explain recurrent abdominal pain, bloating, bleeding, and recurrent pancreatitis, the possibility of this disease should be considered, and relevant examinations (such as CT, angiography, and duodendoscopy) should be performed in time to identify and diagnose to reduce the incidence of obstructive jaundice and pancreatitis.

Keywords: Juxtapapillary duodenal diverticulum; Lemmel's syndrome; Treatment

Introduction

Duodenal diverticulum is divided into primary (congenital dysplasia causing duodenal saccular protuberance) and acquired (such as duodenal ulcer scar traction, causing duodenum to protrude outward to form a cystic process) [1]. Duodenal diverticulum is generally asymptomatic, and most patients can be found during related examinations (such as CT, endoscopy, and angiography). However, duodenal diverticulum is prone to infection, followed by other complications (such as obstruction, bleeding, perforation, cholelithiasis, cholangitis, and pancreatitis) [2,3]. If duodenal diverticulum does not show complications such as obstruction, bleeding or perforation, it can be relieved by the treatment of anti-inflammatory and acid suppression [4]. If the above related complications occur and the drug treatment is not effective, surgical treatment should be considered [5]. The incidence of juxtapapillary duodenal diverticulum is about 75% of duodenal diverticulum, and larger diverticulum can compress the end of common bile duct and pancreatic duct, causing obstruction of bile and pancreatic juice, which can cause obstructive jaundice, cholangitis and pancreatitis, leading to paramastilla syndrome (Lemmel's syndrome), and can cause severe clinical symptoms and even endanger the lives of patients [6,7]. Importantly, the identification and diagnosis of atypical juxtapapillary duodenal diverticulum complicated with Lemmel's syndrome is relatively difficult, which is easily confused with pancreatic tumors and biliary diseases (such as stones and tumors), leading to misdiagnosis and missed diagnosis [8]. Therefore, the identification, diagnosis, and

treatment of Lemmel's syndrome bring certain challenges for clinicians. Here, we reviewed an elderly patient with juxtapapillary duodenal diverticulum complicated with Lemmel's syndrome, and provided some relevant clinical data, test results and related experience, and hope to provide some reference for clinicians in the diagnosis and treatment of this disease.

Case Presentation

An 81-year-old male patient was admitted to hospital with persistent mid-upper abdominal pain for half an hour. Physical examination: T: 36.4°C, P: 110/min, Bp: 112/85 mmHg, yellow staining of skin and sclera, tenderness in mid-upper abdomen, no rebound pain. Blood routine: WBC: $10.42 \times 10^9/L$, N: 89%, Hb 134 g/L, Pit $260 \times 10^9/L$, amylase and bilirubin increased. This patient had a history of hypertension, diabetes mellitus, recurrent pancreatitis, and gallstone disease. Based on the

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patient's medical history and signs, we initially considered the cause of choledocholithiasis. Abdominal CT examination was performed, it was found that mixed high-density shadows near the duodenum, many foreign body masses and enlarged gallbladder with stones were seen, but no obvious stones in the common bile duct (Figure 1). After admission, abdominal pain was relieved after the treatment of fasting, acid suppression, fluid replacement, and anti-infective, but the next day, abdominal pain recurred, and bilirubin increased significantly. In order to further clarified the cause of this disease. Further MR cholangiopancreatography showed that masses of high density in the descending part of duodenum, dilatation of the common bile duct and pancreatic duct (Figure 2). According to the findings of CT and MR examinations, we considered the possibility of a pancreatic head tumor or diverticulum. In order to clarify the specific nature of the mass. Therefore, further duodenal endoscopy was performed, and peripheral foreign bodies and food clumps were cleaned, it was found that inflammation reaction was found on the inner wall of the descending part of duodenum, the nipple was swollen significantly, and the opening of nipple was located at the edge of the mass (Figure 3). On this basis, tissues were further taken for biopsy, fortunately, it was finally diagnosed as juxtapapillary duodenal diverticulum, which originated from the submucosa, the possibility of pancreatic head tumors was excluded.

After admission, the drug treatment was carried out, but the effect was not good. In order to relieve patients' clinical symptoms as soon

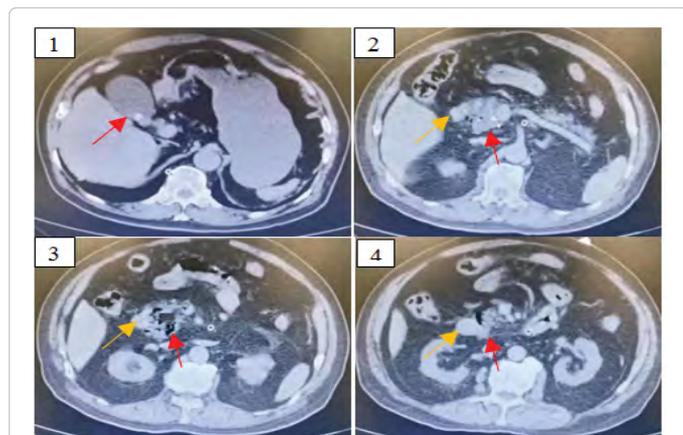


Figure 1 (1-4): Imaging findings of abdominal CT scan. Large diverticulum in the descending part of duodenum with the dilatation of common bile duct and pancreatic duct as shown on CT.



Figure 2 (1 and 2): Schematic diagram of MR cholangiopancreatography, MR cholangiopancreatography showed juxtapapillary duodenal diverticulum complicated with the dilatation of common bile duct and pancreatic duct.

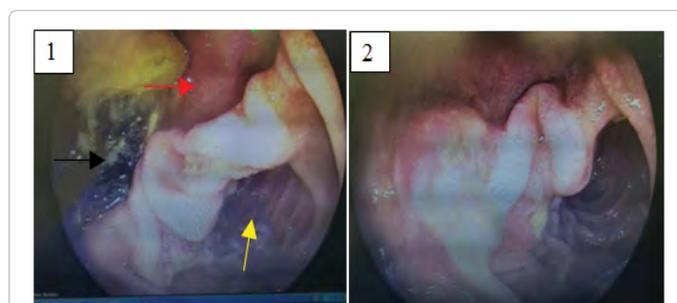


Figure 3 (1 and 2): Schematic diagram of duodenal endoscopy, Schematic diagram of the descending part of duodenum shown by duodenal endoscope. In the first picture: The red arrow showed a larger diverticulum, located at the edge of the big nipple, and a foreign body mass shadow (shown by the black arrow) could be seen in front of the diverticulum to cover the surrounding intestine and nipple. The yellow arrow showed the descending part of duodenum. In the second picture: After the abnormal residue was removed, the structure of the intestine and the nipple were clearly displayed, and the nipple was obviously enlarged and the mucosal edema.

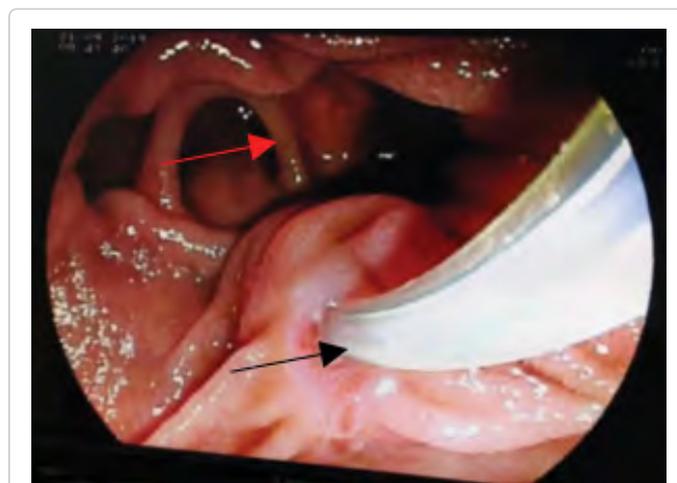


Figure 4: Nipple intubation and drainage after EST operation, the papillary muscle was incised and intubated for drainage. The red arrow indicated the diverticulum, which was located next to the enlarged nipple. The black arrow showed nipple intubation.

as possible, we considered surgical treatment. However, other surgical procedures (such as, direct diverticulectomy and duodenal resection) have more harmful and higher chance of concurrent pancreatic fistula. Therefore, we chosen EST for treatment, the nipple incision knife was inserted into the common bile duct of the descending part of the duodenum through duodenoscopy. Subsequently, the nipple sphincter was incised and intubated for drainage (Figure 4). After operation, the patient's condition gradually improved, the physical signs were stable, and he was discharged on the 5th day, and the patient was followed up, no obvious abdominal discomfort was found.

Discussion

Duodenum is the most common site of digestive tract diverticulum, mostly in the descending part, and the incidence increases gradually with the increase of age, and the incidence rate is 3.8%-20%. While the juxtapapillary duodenal diverticulum refers to a diverticulum that occurs in the 20-30mm around the major duodenal papilla [9,10]. Due to the occurrence of juxtapapillary duodenal diverticulum in special places, it can induce inflammation, bleeding and perforation, as well as induce biliary and pancreatic diseases, however, there are not many

cases complicated with Lemmel's syndrome [11]. Lemmel first reported Lemmel's syndrome in 1934. At present, the universally recognized diagnostic standard is that the duodenum has diverticulum and the common bile duct is dilated, but it is not accompanied by stones [12]. The main clinical manifestations of patients with Lemmel's syndrome are recurrent fever, abdominal pain, cholangitis, cholecystitis, pancreatitis, and gallstones. No biliary stones were found after relevant examinations (such as B-ultrasound, barium meal radiography, and CT), the possibility of Lemmel's syndrome should be considered [13]. Importantly, some patients with Lemmel's syndrome may be free of typical symptoms and specific complications, leading to higher rates of misdiagnosis and missed diagnosis. Therefore, when patients have symptoms that cannot explain such as recurrent abdominal pain, bleeding, pancreatitis, especially for older people, the possibility of Lemmel's syndrome should be considered first. In this case, the patient has abdominal pain and jaundice, and the clinical symptoms are not typical. Therefore, it is easy to misdiagnose, which brings us some obstacles in diagnosis and treatment. Recognized that there are many pathological factors of Lemmel's syndrome, which can mainly include the following three aspects [14,15].

- Direct mechanical stimulation of diverticulum can cause chronic inflammation reaction of the ampulla, leading to nipple fibrosis.
- Diverticulum compresses the opening of the ampulla, resulting in oddis sphincter dysfunction.
- Diverticula around the nipple can mechanically compress the distal common bile duct or ampulla, resulting in high pressure in the bile ducts and pancreatic ducts, and eventually cause biliary and pancreatic diseases.

Therefore, early detection and diagnosis of juxtapapillary duodenal diverticulum, early prevention of related complications, timely treatment of symptomatic duodenal diverticulum, reducing the occurrence of complications and improving the cure rate of patients are another difficult problem faced by clinicians. Imaging (such as CT, MRCP, B-ultrasound and gastrointestinal barium meal radiography) and endoscopy are of great value in the diagnosis and differentiation of juxtapapillary duodenal diverticulum complicated with Lemmel's syndrome [16]. For example, CT and MRCP examination can find thin-walled sac-like protrusions on the wall of the descending part of the duodenum, which can be accompanied by gas or liquid, dilatation of the common bile duct and pancreatic duct, enlarged gallbladder, and exudation around the diverticulum and the pancreas [17]. While MR cholangiopancreatography has higher diagnostic value, which can more clearly observe the dilatation degree of bile duct and pancreatic duct [18]. Compared with imaging examination, duodenoscopy can not only directly observe the anatomical relationship between the nipple opening and the diverticulum, but also can take tissue for biopsy to rule out other diseases (such as cancers). Patients with high suspicion of Lemmel's syndrome can determine the condition of bile duct and pancreas by ERCP [19]. In this case, CT and MR cholangiopancreatography examination revealed that massive structures could be seen in the descending part of the duodenum around the pancreatic head, a large number of foreign bodies can be seen around the mass, and common bile duct and pancreatic ducts were dilated. In order to identify other diseases and confirm our diagnosis, and further duodenal endoscopy and tissue biopsies were performed, and the final diagnosis was juxtapapillary duodenal diverticulum complicated with Lemmel's syndrome, which originated from submucosa.

The anatomy of juxtapapillary duodenal diverticulum is complicated, and its treatment also brings some trouble to clinicians. Juxtapapillary duodenal diverticulum can stretch the sphincter due to food incarceration, and local tissue adhesion caused by repeated inflammation, forced resection often causes severe complications of the biliary and pancreatic and duodenal perforation. Duodenal resection has more trauma and a higher incidence of pancreatic fistula [20,21]. Therefore, EST surgery has become an ideal treatment method. We comprehensively evaluated the condition of this patient, and the medical drug treatment was not effective. So, we decided to perform EST treatment. Symptoms gradually relieved after operation, and no obvious abdominal discomfort was found during follow-up.

Through some of our related experience, for patients with suspected or symptomatic juxtapapillary duodenal diverticulum complicated with Lemmel's syndrome, we recommend the following treatments:

- For patients with suspected juxtapapillary duodenal diverticulum, ERCP or duodenoscopy should be performed early to determine the existence of diverticulum, as well as the location, size and number of diverticula, and to understand its effect on duodenal papilla.
- Medical treatment should be carried out early, such as the treatment of acid suppression, promotion of gastrointestinal peristalsis and anti-inflammatory, reduce the occurrence of complications.
- If the medical treatment is not effective, surgical treatment should be performed, for example, direct resection of duodenal diverticulum, but duodenal fistula is prone to occur, so duodenostomy is recommended.
- If circumstances permit, post-gastrectomy and Billroth II gastrojejunostomy can be considered, which can stop the food from passing through the duodenum, thus relieving the symptoms of the duodenum, and can achieve the purpose of curing the disease.

Ethics approval and consent to participate

Ethical approval has been exempted by our institution's ethics committee (The Ethics Committee of Nanchang University) as this publication is a case report, if patients/patient's next-of-kin have given their informed written consent for the publication of this case report.

Consent for publication

An informed written consent was taken from the patient for reporting this case and the accompanying images.

Conclusion

The red arrow in the first picture showed that the enlarged gallbladder with a round-like high-density stone shadow. The red arrow in the second, third and fourth pictures was the descending part of the duodenum, next to which a mixed high-density mass could be seen. A circular high-density shadow could be seen in the descending part of the duodenum as indicated by the yellow arrow. The large clump-shaped high-density shadow next to the duodenal papilla shown by the red arrow in the first picture, the hepatic-pancreatic ampulla was compressed, and the common bile duct and pancreatic duct were dilated. In the second picture, the descending part of the duodenum (shown by the yellow arrow), the clump-shaped shadow (shown by the red arrow) was more clearly displayed, which obviously compressed the opening of ampulla of hepatopancreas. The common bile duct was

significantly dilated (shown by the purple arrow), and dilatation of the pancreatic duct could also be seen (shown by the green arrow).

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