



Case Report

Giant duodenal diverticulum with mucinous carcinoma of distal bile duct, mimicking Lemmel syndrome: A rare case report

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ABSTRACT

Introduction: Giant duodenal diverticulum is a very rare case. There are only few cases reported. We reported a case of giant duodenal diverticulum with biliary obstruction caused by mucinous carcinoma of distal common bile duct (CBD), that mimicking Lemmel syndrome.

Case presentation: A 68-years-old man admitted to hospital with recurrent epigastric pain, jaundice and fever. Magnetic resonance cholangiopancreatography showed dilated intrahepatic and extrahepatic biliary tree, dilated gallbladder and cystic mass in pancreatic head that pushed the pancreatic duct ventrally. Emergency laparotomy was performed. Distended edematous gallbladder with necrotic spot, dilated of CBD and compressible bulging of the pancreatic head were found. Duodenotomy in 2nd-3rd part was made and found a giant duodenal diverticulum filled with food and mucus. Tight adhesion to the ampulla of Vater, common bile duct, and pancreas due to fibrosis, met difficulties in dissection with a lot of bleeding, hence the diverticulum was not removed. Gastrojejunostomy, cholecystectomy and choledocho-duodenostomy were also done. Pathologic examination of CBD mucus was accordance with mucinous carcinoma.

Discussion: Periampullary duodenal diverticulum can cause obstructive jaundice, known as Lemmel syndrome. This case was different as the giant duodenal diverticulum located in the 3rd part filled with food and mucin that compressed both distal CBD and pancreatic duct. The cause of obstructive jaundice could be fibrotic tissue in distal CBD and mucinous carcinoma.

Conclusion: Giant duodenal diverticulum with bile obstruction is very rare and challenging in diagnosis and treatment. The other cause of obstruction should be considered such as mucinous carcinoma of distal CBD.

1. Introduction

Giant duodenal diverticulum is a very rare case. The incidence of duodenal diverticulum is 1–5% in radiologic series and 11–22% in autopsy series. Bierton and Gupta reported a case of giant inflamed duodenal diverticula in the second/third part of duodenum in 72 years old female with non-radiating epigastric pain for less than 12 hours duration [1]. Millard in 1974 reported four cases of giant duodenal diverticulum which consisted of 3 cases arising from the 3rd part of duodenum and a case was from the 4th part [2]. A giant duodenal diverticulum in the second part was reported by Horst [3]. Periampullary giant duodenal diverticulum can cause obstructive jaundice known as Lemmel syndrome [4]. However, the other causes are challenging to

be determined as the cause of obstructive jaundice in giant duodenal diverticulum. We reported a case with giant diverticulum in the 3rd part of the duodenum with obstructive jaundice caused by early stage of mucinous carcinoma of distal common bile duct.

2. Case presentation

A 68-years-old man admitted to hospital by referral from district hospital with chief complaint upper abdominal pain, jaundice, fever, nausea without vomiting and upper abdominal bulging. The abdominal pain was likely to occur around 2–3 hours after meals. The upper abdominal pain, primarily epigastric pain, was recurrent since 18 months before admission, which was treated as chronic gastritis with

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antacid and omeprazole by primary health care physicians. It worsened one month before admission and was accompanied by dark color urine as well as yellowish sclera. Therefore, the patient was hospitalized in district hospital for 10 days and discharged with subsided signs and symptoms. Seven days before admission, the patient admitted to the previous district hospital with abdominal pain and dark color urine then underwent ultrasonography. It showed dilated intrahepatic and extrahepatic biliary tree, gallbladder and cystic mass in the pancreas as shown in Fig. 1. There was no history of melena. The patient was referred to the hospital.

Vital signs showed blood pressure 124/84 mmHg, pulse rate 88 times per minute, respiratory rate 21 times per minute, body temperature 38.8 °C, body weight 47 kg, height 152 cm. Both sclera were icteric. The skin was dark yellowish with scratch marks. Chest, heart and lungs were within normal limit. The upper abdomen was slightly distended, there was painful fixed mass on palpation and no liver enlargement. Laboratory findings were shown in Table 1.

Magnetic resonance cholangiopancreatography (MRCP) showed dilated intrahepatic and extrahepatic biliary tree, dilated gallbladder, around 7 cm cystic mass in pancreatic head that pushed the pancreatic duct ventrally, dilated pancreatic body and tail. The cystic mass in the pancreatic head was filled with semisolid mass and suspected air bubble, as shown in Fig. 2. The differential diagnosis were giant duodenal diverticulum, pancreatic abscess and pancreatic pseudocyst with obstructive jaundice and cholangitis. Emergency biliary drainage with Endoscopic Retrograde Cholangiopancreatography (ERCP) stenting or Percutaneous Transhepatic Biliary Drainage (PTBD) was preferred, but the hospital has no facility for ERCP or PTBD and the referral hospital could not do it immediately. Therefore, the patient underwent emergency laparotomy by digestive surgeon.

After abdominal opening, there was distended edematous gallbladder with necrotic spot, dilated of common bile duct and bulging of pancreatic head. Minimal yellowish fluid came out due to puncture of the mass, hence pancreatic abscess and pseudocyst were ruled out. Small opening in the border of the 2nd-3rd part of duodenum was made. Operator's finger was inserted into duodenum and found an opening through the cyst, supported a giant duodenal diverticulum and there was no papilla of Vater tumor. Kocher maneuver was made and there was a big diverticulum with semisolid content that could be evacuated into the duodenal lumen. The content was semisolid food and mucus. Tight adhesion to the ampula of Vater, common bile duct, and pancreas due to fibrosis met difficulties in dissection with a lot of bleeding. Therefore, the diverticulum was not removed. Duodenotomy was closed, duodenal exclusion by suture obstruction of the pylorus, and gastrojejunostomy with Braun anastomosis was done, as well as choledocho-duodenostomy

Table 1

Laboratory findings.

Lab Parameters	Normal values	Results
Hemoglobin (g/dl)	14.0–16.0	8.9
Leucocyte ($\times 10^3/\mu\text{L}$)	4.0–11.0	25.6
Thrombocyte ($\times 10^3/\mu\text{L}$)	150–400	175
AST (U/L)	<50	103
ALT (U/L)	<50	83
Alkali phosphatase (U/L)	44–147	246
Gamma GT (U/L)	9–40	309
Direct bilirubin (mg/dl)	<0.2	11.67
Indirect bilirubin (mg/dl)	<0.6	2.14
Total bilirubin (mg/dl)	0.2–1.2	13.81
Albumin (g/dl)	3.4–4.8	2.6
Sodium (mmol/L)	138–146	135.9
Potassium (mmol/L)	3.5–4.9	3.53
Chloride (mmol/L)	98–109	96
Calcium (mmol/L)	2.15–2.55	1.84
Bleeding time	1–3	2 min 00 sec
Clotting time	5–15	7 min 30 sec
PPT	10.2–12.1	10.6 sec
APTT	24.8–34.4	27.1 sec
INR		1.07

AST (aspartat aminotransferase); ALT (alanin aminotransferase), Gamma GT (Glutamyl Transferase), APTT (Activated Partial Thromboplastin Time), PPT (Plasma Prothrombin Time), INR (International Normalized Ratio).

and cholecystectomy due to biliary obstruction. Pus and thick mucus came out from the common bile duct during choledocho-duodenostomy. Gallbladder, with no stone within, and thick mucus were sent for pathologic examination. The result was acute recurrent cholecystitis and cancerous cells in the thick mucus that was in accordance with mucinous carcinoma. Since tumor was limited in the distal part of common bile duct, pancreato-duodenectomy (Whipple) surgery was planned after general condition permitted for mayor surgery. The patient got sepsis and bile leak on the 3rd post operative day. Patient's condition worsened and died on the 5th post operative day. This case report has been reported in line with the SCARE Criteria [5].

3. Discussion

Bowel diverticulum is herniation of mucosal and submucosal bowel wall layers through a muscular defect, mostly located in the entering of bowel artery. Colonic diverticulum is the most frequent bowel diverticulum, followed by duodenal diverticulum. There is no gender difference regarding the incidence of duodenal diverticulum. The cause of duodenal diverticulum is mostly acquired as it is generally discovered

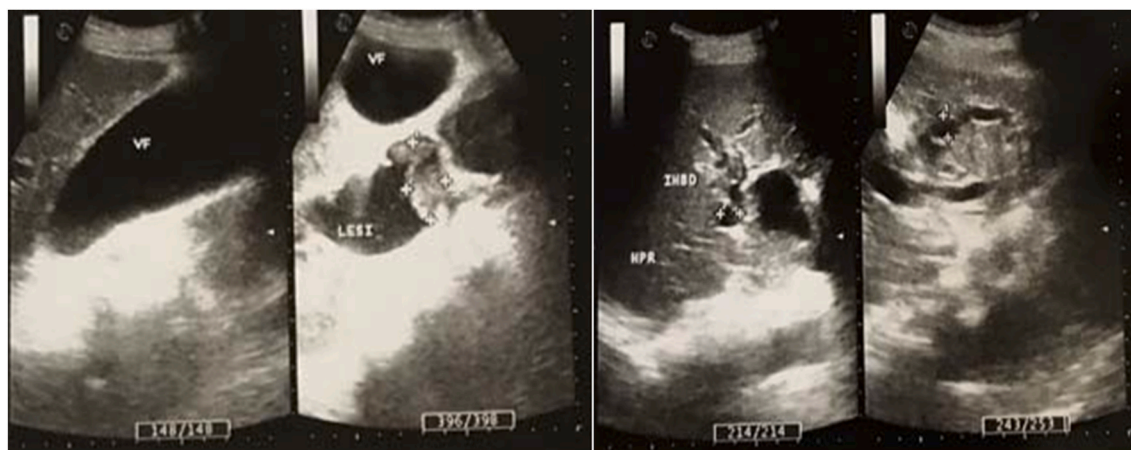


Fig. 1. Ultrasonography showed dilated gallbladder, intrahepatic and extrahepatic bile duct and pancreatic cyst (VF: vesica fellea/gallbladder, IHBD: intrahepatic bile duct, HPR: hepar, yellow arrow: cystic mass). (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

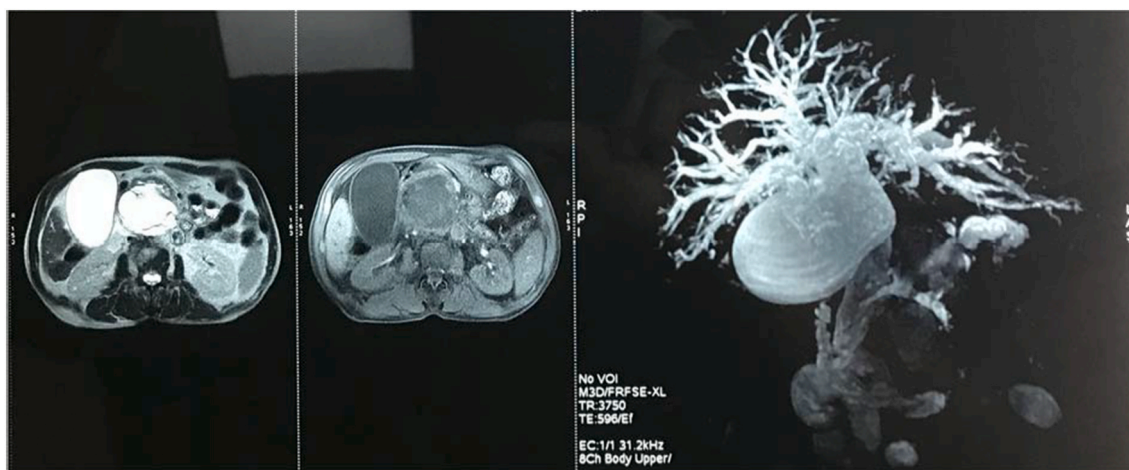


Fig. 2. Magnetic resonance cholangiopancreatography (MRCP) showed dilated gallbladder, intrahepatic and extrahepatic biliary tree. Cystic mass behind pancreatic head was filled with internal debris and suspected air bubble. Pancreatic duct was pushed ventrally. Pancreatic body and tail were dilated.

after the age of 40 and 60% of patients are older than 70 years old [6]. The patient in this case report was 68 years old.

The most frequent location of duodenal diverticulum is medial border of the second part (70%), followed by medial border of the third and fourth part (26%) and lateral border of the second part (4%) [7]. Neither the incidence of giant duodenal diverticulum among the duodenal diverticulum nor the distribution of giant duodenal diverticulum are known. The previous case reports showed that it was located in the 3rd part [2,3], one case in the connection of the 2nd and 3rd part [1] and one case in the 4th part [2]. This case report showed in the 3rd part of duodenum. Duodenal diverticulum can protrude into medial or lateral side of duodenum and mostly arise from the medial site as the entering of artery. This case showed the diverticulum arose from medial site, in line with previous case reports [1,3]. However case reports by Millard reported 2 out of 4 cases arose laterally whereas the other two cases were not stated [2].

Duodenal diverticulum is mostly asymptomatic and discovered incidentally, only 1–5% reported with complications and symptomatic. The complications of duodenal diverticulum are hemorrhage, perforation, gastroduodenal, biliary or pancreatic obstruction [6]. Mostly periampullary duodenal diverticulum is the cause of extra hepatic obstructive jaundice as reported by Lemmel in 1934, furthermore known as Lemmel syndrome. There were some reported cases regarding Lemmel syndrome recently [4,8–12]. Obstruction of distal common bile duct (CBD) can be caused by diverticulitis that can result in papilla of Vater fibrosis, sphincter Oddie dysfunction, and direct compression from distended periampullary diverticulum due to enterolith or bezoar [11]. This case was different with Lemmel syndrome as the giant diverticulum located in the 3rd part of duodenum filled with food and mucin that compressed both distal CBD and pancreatic duct, whereas pathological result of CBD mucus was in accordance with mucinous carcinoma. The cause of obstruction in this case could be both fibrotic tissue in the distal CBD and mucinous carcinoma. This case was also accompanied with obstructive jaundice which was different compared to previous cases of giant duodenal diverticulum [1–3].

4. Conclusion

Giant duodenal diverticulum with bile obstruction is very rare and challenging in diagnosis and treatment. Giant diverticulum rarely cause obstructive jaundice, therefore the other cause of bile obstruction should be considered such as mucinous carcinoma of distal common bile duct as shown in this case.

Ethical approval

This article type (case report) that does not require a formal ethical committee approval. Approval has been given by the Director of St Elizabeth Hospital, Semarang, Indonesia.

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None.

Authors contribution

Bernardus Parish Budiono: Data collection, reference collection and original draft, Melissa Angela Chionardes: Data collection, reference collection and editing the manuscript, Sigit Adi Prasetyo: Data collection, reference collection and editing the manuscript, Ignatius Riwanto: Supervise, operate the patients and final editing of the manuscript.

Conflicts of interest

None.

Research registration number

N/A.

Guarantor

Bernadus Parish Budiono.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Patient perspective

The patient did not present his point of view.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.amsu.2022.103253>.

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