Case Report

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An Unusual Cause of Chronic Abdominal Pain

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Introduction

Periampullary duodenal diverticula (PAD) has incidence around 1-27% and the most common cause of Lemmel's syndrome. PAD arises within a 2-3 cm radius from ampulla of Vater. PAD is mostly asymptomatic, but in 5% of cases it can cause many complications, like right upper quadrant pain, elevated bilirubin, abnormal liver enzymes and/or pancreatic enzymes levels [1]. Endoscopic retrograde cholangiopancreatography (ERCP) is gold standard diagnostic test. Incidental and asymptomatic PAD needs no treatment or intervention, and surgery should be performed in complicated cases [2]. Here i report a case of symptomatic Lemmel syndrome secondary to two giant PAD, arrived at my OPD clinic with intermittent upper abdominal pain since 2016. A 72-yr-old woman was admitted for ERCP on 28-9-22. She was unwell since 2016, with symptoms of recurrent episodes of upper abdominal pain (biliary) with nausea, vomiting and mostly with fever and evidence of jaundice (thrice) during this period. She informed that these symptoms occurred once in every month or sometime, after two months. She was treated for these symptoms many times during this period with intravenous fluids, antibiotics and some pain killer, with improvement of her symptoms. Her previous investigations revealed, OGD in 2016, with duodenal (2nd) diverticula(two), Ultrasonography revealed, dilated CBD (11mm) without any evidence of stone, CT abdomen showed, dilated CBD (compressed by periampullary diverticulas), without any stones in GB & CBD. She has leukocytosis, raised CRP, normal bilirubin & enzymes with raised ALP (with abnormal enzymes & bilirubin, thrice during this time period since 2016), normal pancreatic enzymes. Now before ERCP, her investigations revealed including, complete White blood count raised 12,350/ml (neutrophil 74%). Liver functions were total bilirubin,0.4mg/dl, alanine aminotransferase 26 IU/L, alkaline phosphatase 132 IU/L (32-92). Abdominal CT showed large duodenal diverticulum from second part with resultant upstream dilatation of intrahepatic and extrahepatic bile ducts, MRCP revealed, mildly dilated CBD without any calculus or mass. Mild narrowing seen at its distal end with multiple periampullary duodenal diverticula. ERCP showed two diverticula with central papillary orifice (BOIX IId, classification, Figure 1) Selective Biliary cannulation with endoscopic sphincterotomy (EST) was performed to see CBD for other causes but no stone, stricture or obstruction by tumor could be found and plastic stent 10 Fr x 10cm was placed. After ERCP, patient was discharge on 2nd day. She visited OPD clinic after 3 months, her symptoms were resolved(completely) with normal liver function tests.

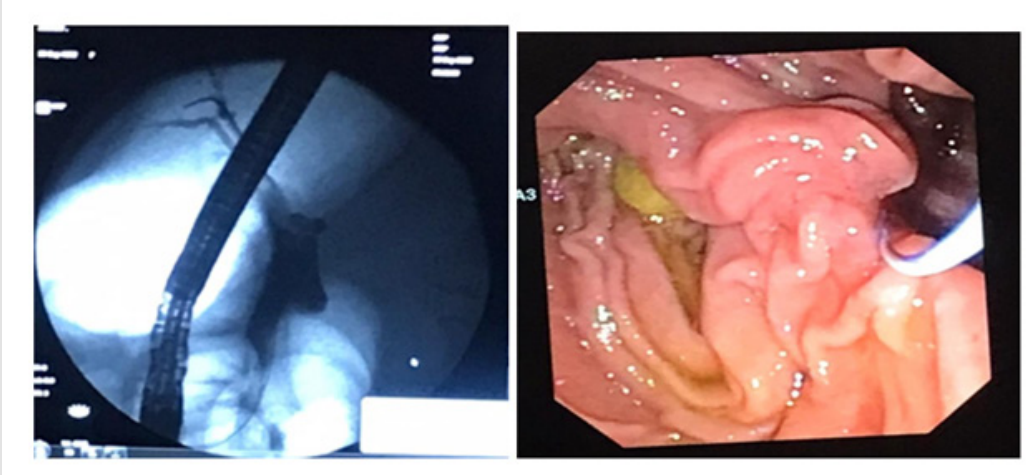

Figure 1

Discussion

Gastrointestinal tract diverticula are outpouchings of all or part of intestinal wall which can be anywhere in the GI tract. Colon is the most common site of gastrointestinal diverticula followed by duodenum [3]. Duodenal diverticula detection rate ranges from 1% to 27% depending on age of the patient and diagnostic modalities used for diagnosis [1]. PAD is most common type around 70% to 75% of all duodenal diverticula. PAD are mostly asymptomatic but in 5% of cases different complications can occur and they include choledocholithiasis, cholangitis, jaundice, pancreatitis, diverticulitis, enterolith or bezoar formation, bleeding, perforation, SOD dysfunction and intestinal obstruction. Hepatocholangiopancreatic disease can rarely occur in the absence of choledocholithiasis and is called Lemmel's syndrome [2]. There are three possible reasons for Lemmel's syndrome to occur. First, direct mechanical irritation of PAD or diverticulitis. Second, PAD may cause dysfunction in the SOD. Third, PAD can cause mechanical compression of distal CBD or ampulla [4]. In our case, patient has intermittent symptoms ranging from biliary colic with or without cholangitis and jaundice during last six years. PAD usually have wide orifice, food material, enterolith, or bezoar within the PAD is frequently evacuated and symptoms could be intermittent. This may be likely reason in our patient who has intermittent symptoms.

In our case, there was no enterolith or bezoar in PAD. Primary biliary stone occur more frequently in the presence of PAD [5]. CBD was explored in our case, no etiology of dilated CBD could be identified except extrinsic compression by distended PAD (Figure 1). Diagnosing Lemmel's syndrome could be challenging, but awareness of this condition is important to avoid mismanagement. PAD are best identified by a side-viewing endoscope during ERCP. On CT scan and MRCP, PAD identified as thin-walled cavity lesions on medial wall of 2nd portion of deodenum that typically contain gas. However, PAD are sometimes filled with fluid and can frequently be mistaken for pancreatic pseudocyst, pancreatic abscess, cystic neoplasm in the pancreas head or even metastatic lymph node [6]. PAD treatment is generally not recommended in asymptomatic patients or would be conservative in pauci-symptomatic patients. Most patients of Lemmel's syndrome present with symptoms related to biliary obstruction (i.e. jaundice, abdominal pain, and cholangitis) probably due to extrinsic compression of CBD, so some form of treatment is required. Therapeutic options in Lemmel's syndrome ranges from endoscopic intervention / extraction, extracorporeal shock wave lithotripsy to surgery (diverticulectomy or biliodigestive anastomosis) [7]. In our case, the patient was successfully treated endoscopically by EST and putting plastic stent. It is noted that not all forms of Lemmel's syndrome are caused by extrinsic compression of CBD by PAD. If the reason of Lemmel's syndrome is papillitis, chronica fibrosa or sphincter of Oddi dysfunction as in our case, the simple and appropriate treatment would be EST [8].

Conclusion

Lemmel's syndrome is a rare cause of chronic abdominal pain with or without obstructive jaundice and should be included in the differential diagnosis of biliary obstruction when PAD is present. Symptomatic patient can be successfully treated endoscopically.

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