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CASE REPORT

An uncommon benign cause of a dilated common bile duct

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Introduction

Obstruction of the common bile duct with consecutive jaundice usually occurs in the setting of choledocholithiasis and pancreatico-biliary tumours (Assy et al, 1999). Other causes, some rare, include intraluminal pathology, mural causes and extrinsic compressions. Periampullary duodenal diverticulum (known as Lemmel syndrome) is a rare benign cause of common bile duct obstruction.

Periampullary duodenal diverticulum (duodenal diverticulum arising within a 2–3 cm radius of the major papilla) is reported in 17.2% in patients undergoing endoscopic retrograde cholangiopancreatography, with a higher prevalence in older people (Lobo et al, 1999). Periampullary duodenal diverticulum is usually asymptomatic and incidentally detected on endoscopy or cross-sectional imaging done for other reasons, but rarely it can cause complications. One complication is obstructive jaundice caused by compression of the common bile duct, in the absence of choledocholithiasis or periampullary neoplasia – Lemmel syndrome. This article reports a case of Lemmel syndrome that was successfully managed by endoscopic clearance of food residue from the periampullary duodenal diverticulum.

Case report

A 60-year-old woman, who was an ex-smoker, with a medical history of diabetes, cholecystectomy and coronary heart disease, presented with moderate intensity pain in the right upper quadrant, with accompanying low-grade fever, lasting for about 3 days. She reported similar episodes in the last year, which improved with fasting and antispasmodics. Her family history was unremarkable. Physical exam revealed upper abdominal tenderness with equivocal Murphy's sign. Laboratory workup showed mild leucocytosis – 12 300/mm^3 (normal range 4000–10 000/mm^3) with neutrophilia 78.3%, altered liver function tests with aspartate aminotransferase 104 U/litre (3–35 U/litre), alanine aminotransferase 133 U/litre (3–35 U/litre), gamma-glutamyl transpeptidase 208 U/litre (5–38 U/litre), alkaline phosphatase 140 U/litre (30–120 U/litre), total bilirubin 1.93 mg/dl (0.3–1.2 mg/dl) with conjugated bilirubin 0.58 mg/dl (0–0.2 mg/dl), slightly elevated amylase 130 U/litre (normal range 28–100 U/litre), lipase within normal limits 55 U/litre (7–60 U/litre) and elevated C-reactive protein level 43 mg/litre (0–5 mg/litre). On abdominal ultrasound, a dilated common bile duct (12 mm) with mild bilateral intrahepatic bile duct ectasia was noted. Cholangitis was diagnosed and the patient was admitted for parenteral antibiotics and further workup. A contrast-enhanced computed tomography scan was ordered, which confirmed common bile duct dilatation without any evidence of obstruction; moreover, a 3 cm duodenal diverticulum was noted (Figure 1). Magnetic resonance cholangiopancreatography excluded lithiasis and revealed mass effect of the diverticulum on the common bile duct (Figure 2). Oesophagogastroduodenoscopy showed a periampullary duodenal diverticulum impacted with food residue, with a normal appearing papilla major. Impacted food was removed from the diverticulum endoscopically, providing symptom relief and resolution of cholangitis.

Discussion

Lemmel syndrome refers to a periampullary duodenal diverticulum which compresses the common bile duct, causing cholangitis, pancreatitis or jaundice, in the absence of lithiasis or malignancy (Lemmel, 1934).

Besides the mechanical compression of the common bile duct by inflamed or impacted periampullary duodenal diverticulum, as seen in this case, recurrent episodes of cholangitis in Lemmel syndrome have also been attributed to chronic inflammation of the ampullary area from the adjacent diverticulum, which may lead to fibrosis of the papilla or dysfunction of

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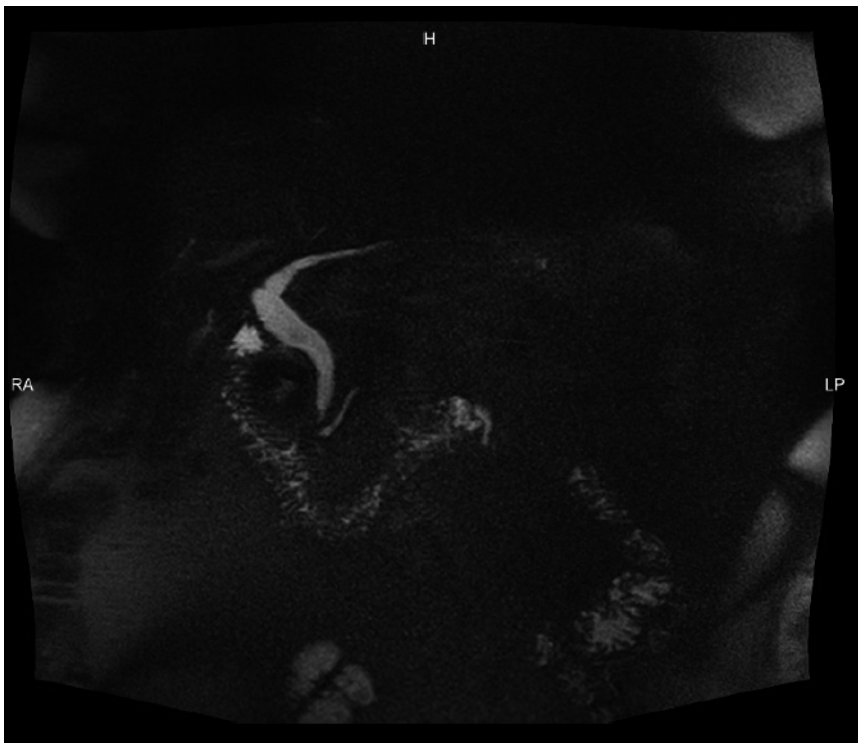

Figure 1. Magnetic resonance image showing mass effect of periampullary diverticulum on common bile duct.

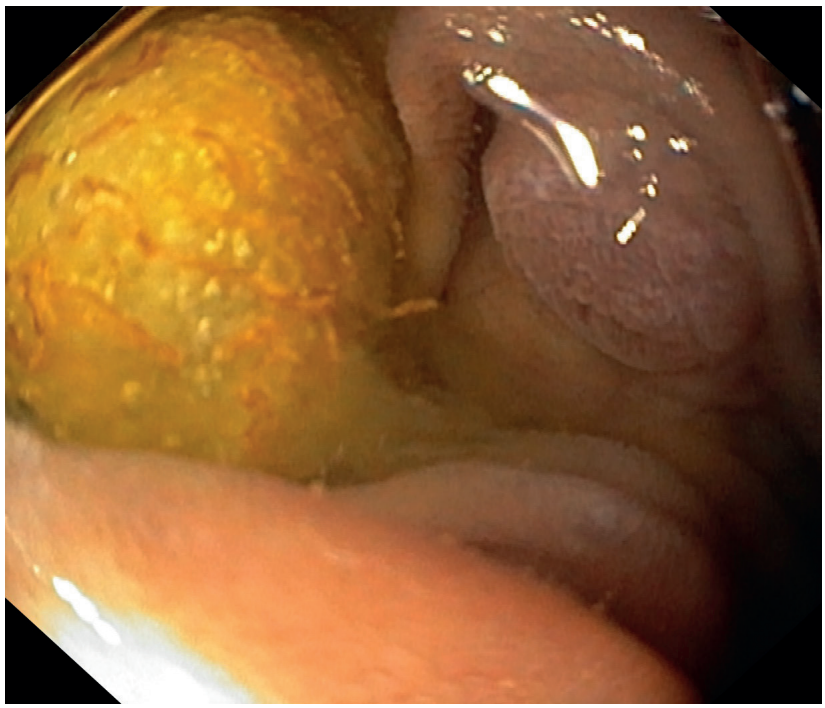

Figure 2. Endoscopic image showing food impaction of periampullary duodenal diverticulum.

the sphincter of Oddi (Desai et al, 2017). Nevertheless, chronic stasis in the periampullary duodenal diverticulum with consequent common bile duct obstruction can also lead to development of primary choledocholithiasis; this may pose diagnostic challenges as the lithiasis could be considered to be the primary cause of the biliary obstruction.

Most cases of periampullary duodenal diverticulum are usually asymptomatic and incidentally detected on oesophagogastroduodenoscopy or imaging done for other purposes. Rarely they can cause complications, categorised as pancreaticobiliary (pancreatitis,

Key points

- Periampullary duodenal diverticulum are usually asymptomatic and detected incidentally.
- Rarely, periampullary duodenal diverticulum can cause biliary obstruction and cholangitis, a condition entitled Lemmel's syndrome.

cholangitis) or non-pancreaticobiliary (diverticulitis, fistula, haemorrhage, small bowel obstruction by enterolith, perforation) (Alzerwi, 2020). Lemmel syndrome is characterised by periampullary duodenal diverticulum complicated with obstructive jaundice by common bile duct compression. Cholangitis in Lemmel syndrome can lead to further complications such as hepatic abscesses (Rouet et al, 2012).

A forward-viewing scope can usually detect the periampullary duodenal diverticulum, with a sensitivity of 75%, but examination with a duodenoscope is considered the diagnostic gold standard (Khan et al, 2017; Alzerwi, 2020). On imaging, mixed air/fluid contents within the periampullary duodenal diverticulum can mimic cystic lesions, abscesses or even malignancy. Endoscopic ultrasound can be useful for a differential diagnosis and can provide real time visualisation of the diverticulum compressing the common bile duct (Somani and Sharma, 2017).

Management usually requires endoscopic retrograde cholangiopancreatography with sphincterotomy, which is technically more difficult in the setting of periampullary duodenal diverticulum. However, some authors have reported no differences in cannulation rates or endoscopic retrograde cholangiopancreatography-associated complications in patients with and without periampullary duodenal diverticulum (Egawa et al, 2010). As seen in this case, biliary obstruction can also be relieved by endoscopic removal of the diverticulum content (Imao et al, 2003). Endoscopic treatment is needed in cases of stenosing fibrosing papillitis or sphincter of Oddi dysfunction, where common bile duct compression is not the main pathogenic mechanism of biliary obstruction. If endoscopic retrograde cholangiopancreatography fails, surgery can be indicated (Alzerwi, 2020). Surgical options include diverticulectomy (associated with significant morbidity and mortality), inversion or reconstruction of the diverticulum, and bypass procedures such as biliodigestive anastomosis (Yoneyama et al, 2004). There is no consensus regarding the best surgical therapeutic approach, as large series with surgically treated Lemmel cases are lacking.

Conclusions

Although periampullary duodenal diverticulum is frequently an incidental finding and asymptomatic, complications can occur. Lemmel's syndrome should be considered as a differential diagnosis of common bile duct obstruction in the presence of periampullary duodenal diverticulum. Better awareness of this condition is needed to avoid mismanagement.

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