# Periampullary Diverticulum: A Case of Bleeding from a Periampullary Diverticula

JEH Kua,<sup>1</sup>MBBS, A Seah,<sup>1</sup>MBBS, FRCS (Edin), JBY So,<sup>1</sup>FRCS (Edin), FRCS (Glas), FAMS

## Abstract

Introduction: Duodenal diverticula are uncommon and usually asymptomatic. Complications like bleeding, perforation and biliary fistulae are rare. The management of a bleeding duodenal diverticulum can be challenging. With the improvement of endoscopic techniques, many of the cases reported in the literature were managed with endoscopic methods. We present a case report of bleeding duodenal diverticulum. <u>Clinical Features</u>: The patient was treated successfully with endoscopic haemostasis during her first epidsode when she initially presented with bleeding duodenal diverticulum, but recurred after 2 months. <u>Treatment</u>: Despite initial endoscopic haemostasis during her second episode, she rebled after 2 days, necessitating surgical management. <u>Outcome</u>: After suture ligation of the ulcer, the patient recovered and there was no more recurrence. <u>Conclusion</u>: Periampullary diverticulum is a rare source of gastrointestinal bleeding, which can be challenging to diagnose and treat. A multidisciplinary approach encompassing radiology, endoscopy and surgery is most effective.

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### Introduction

Duodenal diverticula were reported for the first time by Chomel in 1710.<sup>1</sup> They can be classified as either congenital or acquired and intraluminal or extraluminal. They typically occur in the periampullary region, along the medial aspect of the second and third part of the duodenum. Periampullary diverticula (PAD) are extraluminal outpouchings of the duodenum arising within a radius of 2 to 3 cm from the ampulla of Vater. The true prevalence in the general population is difficult to ascertain, with reported rates varying between 0.16% and 27%, according to the diagnostic tests. For upper gastrointestinal radiological contrast studies, incidence rates range from 2% to 5%, whereas for endoscopic retrograde cholangiopancreatography (ERCP) and autopsies, incidence rates have been reported at about 22%.<sup>2</sup> As most of these patients are asymptomatic, these incidence rates may not be reflective of the true prevalence.

Although PAD are usually asymptomatic, complications can occasionally develop. They can be classified as pancreaticobiliary or non-pancreaticobiliary. Pancreaticobiliary complications include an increased incidence of choledocholithiasis and pancreatitis. Non-pancreaticobiliary complications of PAD are haemorrhage, perforation and enterolith formation. PAD are a rare source of gastrointestinal bleeding. We report a rare case of periampullary diverticulum bleeding which was initially treated endoscopically, but subsequently required surgical intervention.

#### **Case Report**

A 69-year-old woman presented with haematochezia and haematemesis for 1 day. She did not have a history of nonsteroidal anti-inflammatory drug (NSAID) ingestion or liver disease. Examination did not reveal any stigmata of chronic liver disease. She had mild epigastrium tenderness on examination. Haemoglobin on admission was 8.3 g/dL and she had 2 units of packed cells transfusion in the ward.

Initial oesophagogastroduodenoscopy (OGD) localised the bleeding to around the junction of the first and second part of duodenum. The source of bleeding was from an ulcer at the apex of a postero-medial periampullary diverticulum (Fig. 1). The bleeding slowed down after an injection of 8 cc of 1:10,000 of adrenaline into the area adjacent to the ulcer.

<sup>&</sup>lt;sup>1</sup> Department of Surgery

National University Hospital, Singapore

Address for Reprints: Dr Jimmy BY So, Department of Surgery, National University Hospital, 5 Lower Kent Ridge Road, Singapore 119074.



Fig. 1. Endoscopic view with a side-viewing duodenoscopy of the bleeding periampullary diverticulum.

Better visualisation of the ulcer was done by switching to a side-viewing duodenoscope, which revealed a bleeding vessel at the base of the ulcer. Argon plasma coagulation was then applied to the vessel to secure haemostasis. She remained stable post-procedure and was discharged after 3 days with proton pump inhibitors.

Two months later, she was readmitted with upper gastrointestinal bleeding. She had been asymptomatic in the interim. Haemoglobin at that time was 10.6 g/dL. With the previous pathology in mind, the OGD revealed a blood clot in the PAD. On removing the clot, there was a slow ooze from the vessel in the same ulcer at the apex of the periampullary diverticulum. This was injected with adrenaline, achieving haemostasis. A computed tomography (CT) scan done excluded a lesion from the pancreas encroaching into the second part of the duodenum. It did reveal some air pockets in the retroperitoneal, peri-pancreatic space suggestive of a micro perforation. However, she was otherwise asymptomatic, with no leukocytosis.

Two days later, she exhibited clinical signs of a recurrent bleed, with a haemoglobin drop of 2 g/dL, associated hypotension and melena. Since she had had endoscopic haemostasis twice, we opted for surgical management.

An anterior duodenotomy was made in the second part of the duodenum, the diverticulum was identified at the medial wall of the duodenum, embedded with the pancreatic tissue (Fig. 2). The diverticulum was everted using stay



Fig. 2. Intraoperative view of periampullary diverticulum.

sutures. After visual identification of the ampulla of Vater, the common bile duct and pancreatic duct, a transduodenal suture-ligation of the bleeding ulcer was done with cessation of the bleeding. Tissue glue was applied to the base of the ulcer and then the duodenotomy was closed with single-layer interrupted PDS 30. She recovered well from the surgery and was discharged from hospital 1 week later. She was seen 9 months later at the outpatient clinic with no bleeding episodes in the interim.

## Discussion

Duodenal periampullary diverticulum is a rare source of gastrointestinal bleeding.<sup>3</sup> The true prevalence of PAD is uncertain. A study at our institution has found PAD in 11.5% of our patients at ERCP.<sup>4</sup> However, endoscopic and radiological examinations are only performed in symptomatic patients and so do not represent the general population. Extraluminal diverticula are outpouchings of mucosa, submucosa and scattered muscle cells. The pathogenesis may involve herniations of mucosa through mural weak points, presumably at sites of vascular or ductal entry into the submucosa. The pathogenesis of PAD in particular may involve inherent weakness in the duodenal wall where the common bile duct and pancreatic duct enter. Coexistent diverticula within the colon may also be present in up to 30% of patients.

Haemorrhage has been reported to occur most commonly with diverticula located in the third and fourth portions of the duodenum and rarely from PAD.<sup>5</sup> There are no reported cases of death from bleeding but patients may present with haematemesis, haematocheizia, melena and anaemia. The cause of bleeding is usually inflammation leading to the erosion of a branch of the superior mesenteric artery, but other possible causative factors include stasis-induced ulceration within the pouch, Dieulafoy's lesions, intradiverticular polyps, aortoenteric fistulas, erosion by bezoars and ectopic gastric mucosa. Haemorrhage from PAD is a rare complication. The incidence of haemorrhage from PAD is not reported and only a few case reports of bleeding are reported in literature.<sup>5,6</sup> It is not always easy to diagnose a bleeding duodenal diverticulum as the bleeding site is usually within the diverticulum. However, diagnosis has been facilitated with the advent of endoscopy, especially with side-viewing duodenoscopy.<sup>7</sup> In our experience, a side-viewing duodenoscope provides a front-on view of the periampullary area and it also provides good perpendicular access for endoscopic haemostatic devices. Alternative investigations include upper gastrointestinal series, CT scan or angiography.

Due to its rarity, experience with management of bleeding PAD is limited and controversial. Treatment can comprise prompt operative methods, definitive endoscopic management or endoscopic haemostasis followed by definitive surgery.

With the advent of therapeutic endoscopy, the diagnosis and treatment of duodenal diverticula-associated bleeding has changed. Definitive endoscopic management involving adrenaline injection,<sup>5,6</sup> multipolar coagulation or hemoclip application<sup>8</sup> has been reported.

The risk of rebleeding episodes after endoscopy treatment alone is unknown. Moreover, complications from endoscopic treatment have also not been reported. Our patient had a rebleeding episode 2 months after endoscopic management. The bleeding was refractory even after a second endoscopic application of adrenaline. The narrow stoma and thin inflamed mucosa limited the use of hemoclips, necessitating surgical intervention.

The role of elective surgery after endoscopic control of bleeding remains undefined and likely unnecessary. The commonest type of surgery involved is a diverticulectomy (excision of the diverticulum and repair of the duodenum). Diverticulectomy for asymptomatic diverticula is controversial and not recommended. The commonest indication for diverticulectomy of duodenal diverticula is bleeding and perforation.

Diverticulectomy of PAD has its risks. It is important to identify the pancreatic and common bile ducts poor to resection to avoid inadvertent injury to these important structures. In 1952, Cattel and Mudge<sup>9</sup> reported an 8% mortality during elective excision of 25 symptomatic duodenal diverticula. Both deaths occurred after significant pancreatic injury induced during surgery, emphasising the difficult dissection of the duodenal diverticulum due to its

close relation to the head of the pancreas. Moreover, symptomatic relief was only achieved in 50% of patients.

At this point, our surgical options included an anterior duodenostomy, with excision of the diverticulum, or invagination of diverticulum with suture-ligation of vessel. Excision of the diverticulum is the treatment of choice, but is technically difficult with the risk of injury to pancreas, biliary or pancreatic ducts. Yin et al<sup>10</sup> reported a 50% duodenotomy leak rate with excision of the diverticulum and postulated that poor tissue condition could be the cause. In our case, the patient had had endoscopic injection of adrenaline into the diverticular wall with inflamed mucosa, as well as hypotension. In view of these factors and the emergent nature of the surgery, we opted for the simpler option of invagination and suture-ligation of the diverticular ulcer. Tissue glue was applied to the base of the ulcer, filling up the diverticulum.

In conclusion, PAD are a rare source of gastrointestinal bleeding but a potential cause of significant morbidity. Diagnosis and treatment can be difficult. A multidisciplinary approach including endoscopy, radiology and surgery is required for successful management.

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