

Periampullary Dieulafoy's Lesion: A Diagnostic ChallengeLok Yuh Ing^{1*}, Lee Yuk Loong²

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Abstract: Dieulafoy's lesion is a rare etiology of recurrent gastrointestinal (GI) bleeding and periampullary location is very uncommon for a bleeding Dieulafoy's lesion. Endoscopy is the current standard method for diagnosis and treatment but difficult to diagnose when bleeding is inactive. A case of a sixteen-year old male patient was presented with recurrent massive upper GI bleed. Multiple endoscopic findings were unremarkable and angiography showed dilatation of gastroduodenal artery but no sign of active bleeding. Endoscopy was repeated during active bleeding and it showed bleeding periampullary Dieulafoy's lesion but failed endoscopic hemostasis. Ultimately, hemostasis was achieved by transcatheter arterial embolisation (TAE) of gastroduodenal artery. Thus, periampullary Dieulafoy's lesion represents a diagnostic and therapeutic challenge for clinicians.

Keywords: Periampullary Dieulafoy's lesion, endoscopy, diagnosis, challenge.

INTRODUCTION

Dieulafoy's lesion is characterised by dilated submucosal artery which protrudes through tiny mucosal defect [1] and leads to massive GI bleed when it ruptures spontaneously. It is commonly found at stomach. Dieulafoy's lesion at periampullary location is very unusual [2]. It is diagnosed at endoscopy especially during active bleeding.

However, Dieulafoy's lesion is difficult to identify due to absence of ulcerations. This is a case report of a 16-year-old male patient presented with recurrent massive upper GI bleed and was diagnosed as periampullary Dieulafoy's lesion after multiple endoscopies and hemostasis was successfully achieved by TAE. This case report is to increase the awareness to this lesion, to discuss the pitfalls in the diagnosis and management of this rare lesion.

CASE PRESENTATION

A 16-year-old male patient with history of three hospital admissions (shown in table below) for recurrent episodes of fresh melaena and symptomatic anaemia in the past 18 months. He has no medical illness and has no history of taking alcohol, analgesia or traditional medications.

Date of admission	Class of hemorrhage upon admission	Endoscopic findings	Others
15/2/13-19/2/13	Class 1	Oesophagogastroduodenoscopy (OGDS): Forrest Ib ulcer at D1/D2 junction, Forrest 3 ulcer at distal corpus, gastric erosion at fundus	
17/2/14-19/2/14	Class 2	OGDS: Forrest IIc ulcer at D1/D2 junction (no stigmata of recent bleeding)	Repeated procedures after 1 month as outpatient: 1) OGDS: healed ulcer 2) Merkel scan: no Merkel diverticulum
14/10/14-20/10/14	Class 3	OGDS: normal Colonoscopy: normal	Capsule endoscopy as outpatient (14/11/14): normal

1 month after third admission, patient was admitted for fresh melaena, class 2 hemorrhage. OGDS showed blood tracking from distal stomach to D3,

unable to visualise bleeding point. Post-OGDS, patient persistently presented with fresh melaena, dropping hemoglobin level and hemodynamically unstable.

Hence, proceeded with laparotomy, enterotomy and on table endoscopy, it showed multiple ulcers and blood clot at proximal jejunum (40cm to 70cm from DJ flexure). Therefore, elemental jejunal resection with primary anastomosis was performed. HPE of resected jejunal segment revealed serositis.

6 days post operation, patient presented with fresh melaena. Repeated OGDS and colonoscopy showed normal finding. Proceeded with CT

angiography and noted prominent and tortuous of gastroduodenal artery but no sign of active bleeding. The next day, he presented with another 2 episodes of hematemesis. OGDS showed bleeding periampullary Dieulafoy's lesion near ampulla but no ulcer seen. 1 hemoclip was applied to secure hemostasis. Nevertheless, patient passed out fresh melaena the following day, 3 hemoclips and heater probe were applied at bleeding point.



Fig-1: CT angiography showed prominent and tortuous gastroduodenal artery but no active contrast extravasation

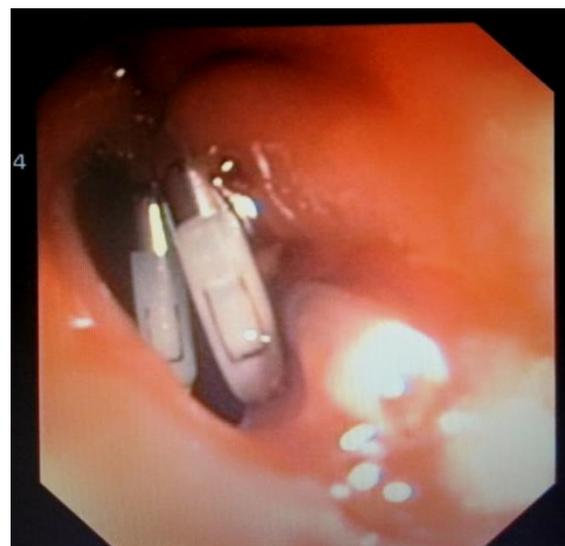


Fig-2: OGDS showed protrusion of submucosal vessel at duodenum. No mucosal ulceration seen. Hemoclips were applied to secure hemostasis

Patient was well until 4 days later, he developed 1 episode of hematemesis. Hence, he was sent for TAE of gastroduodenal artery, a 2cmx2cm coil was inserted, and hemostasis secured. No sign of re-bleeding after 4 years of follow up.

DISCUSSION

Dieulafoy's lesion is characterised by exteriorisation of a large submucosal arteriole through a minimal mucosal tear surrounded by normal mucosa causing massive and recurrent upper GI bleeding in 1-2% of all upper GI bleeding. 75-95% of Dieulafoy's

lesion is found within 6cm of the gastroesophageal junction, predominantly on the lesser curvature. Extragastric lesions include duodenum (14%), colon (5%), jejunal (1%), and esophagus (1%). Periampullary location is very unusual.

Dieulafoy's lesion affects mainly male patient (M:F=2:1). Age varied from 20 months to 90 years old, with mean age 50 years old. It presented more commonly in elderly population with multiple comorbidities in up to 90% of patients. It also affecting previously healthy individuals as in our case. The exact

etiology and pathogenesis of Dieulafoy's lesion remained unknown.

Diagnosis of Dieulafoy's lesion is typically by endoscopy. The endoscopic criteria are visualisation of an active arterial bleed, or of vessel protruding from a small mucosal defect (2-5mm) or normal mucosa without ulcerations, or of a clot that is adherent to a small mucosal defect[3]. About half of the lesions were identified during first endoscopy especially within 2 hours of active bleeding. In our case, failure to identify lesion during initial endoscopy was due to the site of bleeding covered by clots, the intermittent nature of the bleeding, and the absence of ulcerations. Besides, Dieulafoy's lesion may overlook as concomitant lesions eg. Ulcers which may responsible for hemorrhage, as in our patient during his first hospital admission. Otherwise, angiography may provide useful information for site of bleeding. Although our patient's angiography showed prominent and tortuous gastroduodenal artery without extravasation of contrast, it did provide important information for location of lesion and site of embolisation later.

Therapeutic endoscopy is the mainstay of treatment. It is successful in 85% of cases in achieving permanent hemostasis [4]. However, 10% require a repeat endoscopic therapy. Alternatively, TAE is used in patients who fail endoscopic therapy or lesions beyond the reach of therapeutic endoscope, clinical success rate ranges from 70-80% [5]. The role of TAE is to selectively reduce blood supply at the source of bleeding while maintaining sufficient collateral blood flow to maintain intestinal viability. In the case of failed endoscopic hemostasis or embolotherapy, surgical exploration with intraoperative endoscopy can achieve excellent result by combining the advantages of endoscopic visualisation with open surgical ligation of bleeding vessel.

The long term prognosis of Dieulafoy's lesion is good. The recent mortality rate is 8.6% which is

almost the same in the endoscopically and surgically treated patients.

CONCLUSION

Periampullary Dieulafoy's lesion can represent a diagnostic and therapeutic challenge. A high index of suspicion for Dieulafoy's lesion should be given when common causes of upper GI bleed have been ruled out by routine endoscopy. Hence, increased awareness and early endoscopy during a bleeding episode are essential for accurate diagnosis.

Lists of abbreviations used

GI - gastrointestinal
TAE - transcatheter arterial embolisation
OGDS - Oesophagogastroduodenoscopy
DJ - duodenojejunal
NSAIDS - non steroidal anti-inflammatory drug

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