

CASE REPORT

Periampullary Dieulafoy's Lesion: An Unusual Cause of Gastrointestinal Bleeding

Surinder S Rana¹, Deepak K Bhasin¹, Rajesh Gupta²,
Thakur Deen Yadav², Vivek Gupta³, Kartar Singh¹

Departments of ¹Gastroenterology, ²General Surgery, and ³Radiodiagnosis,
Post Graduate Institute of Medical Education and Research (PGIMER), Sector 12. Chandigarh, India

ABSTRACT

Context Dieulafoy's lesion is an unusual cause of gastrointestinal bleeding with the most common location being the stomach. A periampullary location is rare for a bleeding Dieulafoy's lesion. **Case report** We present the case of a 52-year-old female who presented with intermittent painless melena. Her upper gastrointestinal endoscopy and colonoscopy were normal. She was a diagnostic challenge as no definite lesion could be identified on capsule endoscopy. However, as there was presence of fresh blood in the proximal jejunum, a push enteroscopy was performed which revealed the presence of fresh blood in the duodenum and proximal jejunum. But no bleeding lesion could be identified. A side view endoscopy was performed which revealed a bleeding periampullary Dieulafoy's lesion. Immediate hemostasis was achieved with an injection of adrenalin. Other episodes of bleeding occurred and the patient was finally treated surgically. **Conclusion** A periampullary Dieulafoy's lesion presenting with obscure gastrointestinal bleed is a diagnostic challenge and can be missed on capsule endoscopy.

INTRODUCTION

Dieulafoy's lesion (exulceratio simplex, cirroid aneurysm or caliber-persistent submucosal vessel) is an unusual but important and potentially life threatening cause of gastrointestinal bleeding responsible for up to 5% of acute upper gastrointestinal bleeds [1, 2, 3]. A Dieulafoy's lesion can occur anywhere in the gastrointestinal tract. The most common location is the stomach (64-82% of cases) and the proximal lesser curvature within 6 cm of the gastroesophageal junction is the classic site [3]. The duodenum is the second most common site for a Dieulafoy's lesion (14-18% of cases) and the majority of these lesions are located in the duodenal bulb (53%) followed by the third part of the duodenum (29%) and the junction of the first and second parts of the duodenum (18%) [3, 4, 5]. A periampullary location is rare for a bleeding Dieulafoy's lesion and has rarely been reported in the published literature [6]. We report a patient with a

periampullary Dieulafoy's lesion who presented with obscure overt gastrointestinal bleeding; it was a diagnostic challenge since this lesion could not be identified on capsule endoscopy.

CASE REPORT

A 52-year-old female presented with a history of intermittent painless melena of 6 months duration. She had been evaluated at a private health centre and had received multiple blood transfusions. Her upper gastrointestinal endoscopy and colonoscopy did not reveal any abnormality and she was referred to us for a capsule endoscopy. Her clinical examination was non-contributory except for the presence of mild pallor. The hematological examination was suggestive of iron deficiency anemia with a normal platelet count and coagulation profile. Renal function, liver function tests and ultrasound of the abdomen were normal. Capsule endoscopy (Given Imaging Ltd., Yoqneam, Israel) was performed after overnight fasting and preparation with polyethylene glycol ingestion and revealed the presence of fresh blood in the proximal jejunum along with a pool of fresh blood with no underlying ulcer (Figure 1). The major papilla was not identified on the capsule endoscopy. A push enteroscopy was subsequently performed which revealed the presence of fresh red blood in the distal duodenum and proximal jejunum. No bleeding lesion was identified. The push enteroscope was immediately withdrawn and a side viewing endoscope (TJF 160,

Received March, 10th, 2010 - Accepted April 7th, 2010

Key words Ampulla of Vater; Angiography; Capsule Endoscopy; Endoscopy

Correspondence Surinder Singh Rana

Department of Gastroenterology, Postgraduate Institute of Medical Education and Research (PGIMER), Chandigarh, India-160012
Phone: +91-172.274.9123; Fax: +91-172.274.4401

E-mail: drsurinderrana@yahoo.co.in; sonalisurinder@yahoo.co.in

Document URL <http://www.joplink.net/prev/201005/23.html>

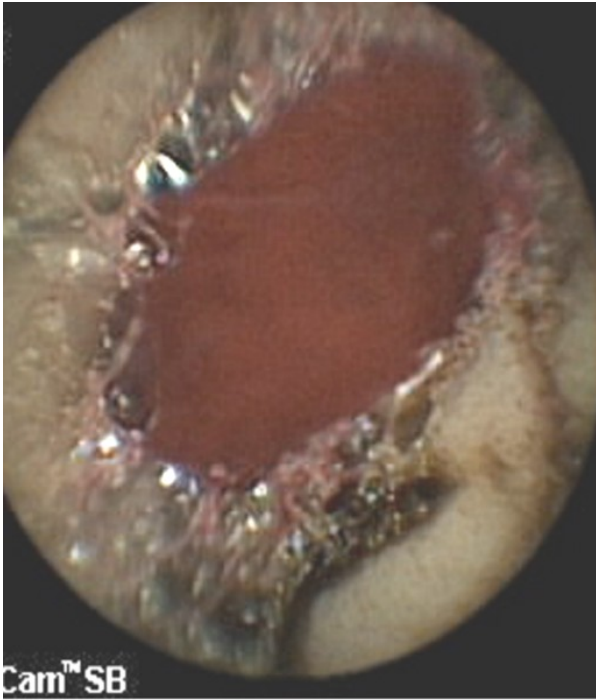


Figure 1. Capsule endoscopy: a pool of fresh blood in the proximal jejunum with no underlying ulcer.

Olympus Optical Co. Ltd., Tokyo, Japan) was inserted. On side view endoscopy, a pool of fresh blood was localized near the papilla (Figure 2). Using a cannula, the area was washed with distilled water and revealed an ooze of fresh blood from a pinpoint defect in the periampullary region. There was no surrounding ulcer or reddish area suggestive of a Dieulafoy's lesion (Figure 3). Immediate hemostasis was achieved with an endoscopic injection of adrenalin.

An episode of bleeding occurred two days after adrenaline injection and a repeat side view endoscopy was performed which revealed fresh bleeding from an

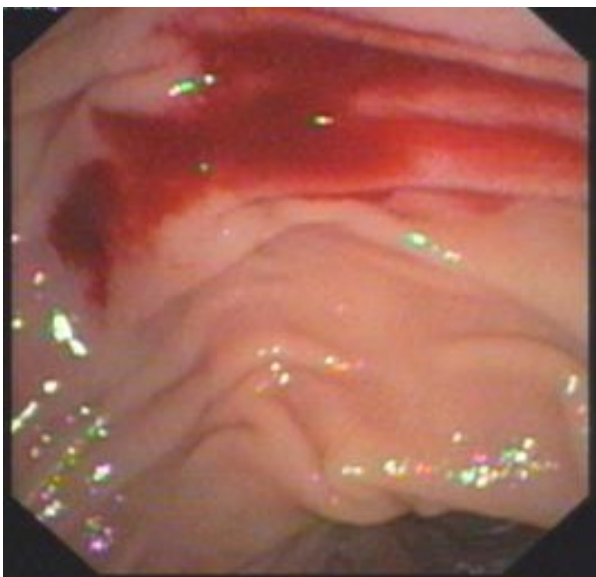


Figure 2. Side view endoscopy: Fresh blood near the papilla.



Figure 3. Side view endoscope: oozing of fresh blood from a pinpoint defect in the periampullary region, suggestive of a Dieulafoy's lesion.

periampullary Dieulafoy's lesion; immediate hemostasis was achieved by the application of a hemoclip (Figure 4). After remaining asymptomatic for one week, another episode of bleeding occurred and an urgent angiography was performed. The angiogram obtained after selective catheterization of the superior mesenteric artery revealed an area of abnormal vascular blush in the second part of the duodenum



Figure 4. The application of a hemoclip on the Dieulafoy's lesion.

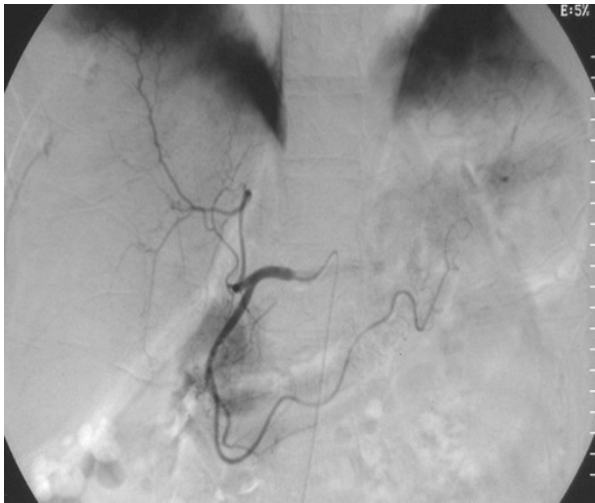


Figure 5. Superior mesenteric angiogram: abnormal area of blush in the second part of the duodenum.

arising from the gastroduodenal artery (Figure 5). As the feeder vessels were of very small caliber, super selective catheterization and embolization was not feasible. Therefore, the patient underwent surgery and oversewing of the abnormal vessel was carried out. The patient had an uneventful postoperative course and is asymptomatic with no recurrence of gastrointestinal bleeding at 18 months of follow-up.

DISCUSSION

Although the duodenum is the second most common site after the stomach for a Dieulafoy's lesion because of its unique blood supply which consists of end arteries, a periampullary location is very rare for a Dieulafoy's lesion and has rarely been reported [6, 7]. A Dieulafoy's lesion is difficult to diagnose and repeated endoscopies may be necessary for diagnosis because of the small point of bleeding, intermittent bleeding or the small lesion being covered by a clot or blood [3]. Endoscopic hemostatic therapy is the treatment of choice in endoscopically accessible Dieulafoy's lesions with more than 90% hemostasis rates [1, 2, 3, 8, 9, 10]. Endoscopic hemostatic therapy includes monotherapy with an injection of epinephrine, sclerosant, alcohol, glue or hypertonic saline or the use of thermal probe monotherapy. These therapies can also be used in combination for better results. Other endoscopic hemostatic measures include mechanical hemostasis using bands or clips. Endoscopic ultrasound can show the dilated pulsating submucosal artery and can also help to guide endoscopic treatment [11]. Angiography and embolization can also be attempted for hemostasis in a bleeding Dieulafoy's lesion, although some studies have shown disappointing results with high rebleed rates [3, 12, 13, 14, 15]. Surgery is the last resort for a bleeding Dieulafoy's lesion and, in spite of the high success of endoscopic therapy, 3-16% of the patients still required salvage surgical therapy according to published studies [1, 3, 8, 12, 13].

Capsule endoscopy has improved our ability to diagnose small intestinal diseases and has thus helped considerably in the management of patients with obscure gastrointestinal bleeding [16]. However, the study by Clarke *et al.* [17] and an earlier study by Kong *et al.* [18] have stressed the fact that capsule endoscopy does not visualize the major papilla in the majority of the cases. The visualization rate of the major papilla varied from 10.4% in the Clarke *et al.* study to 43.6% in the Kong *et al.* study [17, 18]. As a result of the poor visualization rates of the major papilla on capsule endoscopy, it has been suggested that a non-diagnostic capsule endoscopy should not be interpreted as evidence of a normal small bowel especially if periampullary lesions are suspected. The case presented of a periampullary Dieulafoy's lesion also points out the limitation of capsule endoscopy in detecting periampullary lesions, although, in our case, the capsule endoscopy enabled us to detect the presence of fresh blood in the proximal small bowel, thus locating the source of the bleeding.

Conflict of interest The authors have no potential conflict of interest

References

1. Kasapidis P, Georgopoulos P, Delis V, Balatsos V, Konstantinidis A, Skandalis N. Endoscopic management and long-term follow-up of Dieulafoy's lesions in the upper GI tract. *Gastrointest Endosc* 2002; 55:527-31. [PMID 11923766]
2. Sone Y, Kumada T, Toyoda H, Hisanaga M, Kiriya S, Tanikawa M. Endoscopic management and follow up of Dieulafoy lesion in the upper gastrointestinal tract. *Endoscopy* 2005; 37:449-53. [PMID 15844024]
3. Lee YT, Walmsley RS, Leong RW, Sung JJ. Dieulafoy's lesion. *Gastrointest Endosc* 2003; 58:236-43. [PMID 12872092]
4. Veldhuyzen Van Zanten SJO, Bartelsman JF, Schipper ME, Tytgat GNJ. Recurrent massive haematemesis from Dieulafoy vascular malformations-a review of 101 cases. *Gut* 1986; 27:213-22. [PMID 3485070]
5. Ibrarullah M, Waghlikar GD. Dieulafoy's lesion of duodenum: a case report. *BMC Gastroenterol* 2003; 3:2. [PMID 12581456]
6. Hyun CB. Periampullary Dieulafoy's lesion. *Hospital Physician* 2005; 23-7.
7. Womack N. Blood flow through the stomach and duodenum. Clinical aspects. *Am J Surg* 1969; 117:771-80. [PMID 5794863]
8. Dy NM, Gostout CJ, Balm RK. Bleeding from the endoscopically-identified Dieulafoy lesion of the proximal small intestine and colon. *Am J Gastroenterol* 1995; 90:108-11. [PMID 7801908]
9. Romaozinho JM, Pontes JM, Lérias C, Ferreira M, Freitas D. Dieulafoy's lesion: management and long-term outcome. *Endoscopy* 2004; 36:416-20. [PMID 15100950]
10. Yamaguchi Y, Yamato T, Katsumi N, Imao Y, Aoki K, Morita Y, et al. Short-term and long-term benefits of endoscopic hemoclip application for Dieulafoy's lesion in the upper GI tract. *Gastrointest Endosc* 2003; 57:653-6. [PMID 12709692]
11. Levy MJ, Wong Kee Song LM, Farnell MB, Misra S, Sarr MG, Gostout CJ. Endoscopic ultrasound (EUS)-guided angiotherapy of refractory gastrointestinal bleeding. *Am J Gastroenterol* 2008; 103:352-9. [PMID 17986314]
12. Reilly HF 3rd, al-Kawas FH. Dieulafoy's lesion. Diagnosis and management. *Dig Dis Sci* 1991; 36:1702-7. [PMID 1748038]

13. Schmulewitz N, Baillie J. Dieulafoy lesions: a review of 6 years of experience at a tertiary referral center. *Am J Gastroenterol* 2001; 96:1688-94. [PMID 11419815]
 14. Durham JD, Kumpe DA, Rothbarth LJ, van Stiegmann G. Dieulafoy disease: arteriographic findings and treatment. *Radiology* 1990; 174:937-41. [PMID 2305095]
 15. Ashour MA, Millward SF, Hadziomerovic A. Embolotherapy of a Dieulafoy lesion in the cecum: case report and review of the literature. *J Vasc Interv Radiol* 2000; 11:1059-62. [PMID 10997471]
 16. Bhasin DK, Rana SS. Gastrointestinal bleeding: From overt to obscure. *Endoscopy* 2006; 38:116-21. [PMID 16479417]
 17. Clarke JO, Giday SA, Magno P, Shin EJ, Buscaglia JM, Jagannath SB, Mullin GE. How good is capsule endoscopy for detection of periampullary lesions? Results of a tertiary-referral center. *Gastrointest Endosc* 2008; 68:267-72. [PMID 18378233]
 18. Kong H, Kim YS, Hyun JJ, Cho YJ, Keum B, Jeon YT, et al. Limited ability of capsule endoscopy to detect normally positioned duodenal papilla. *Gastrointest Endosc* 2006; 64:538-41. [PMID 16996345]
-