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Massive Lower GI Bleeding from Dieulafoy-Like Lesion: A Case Report

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Abstract **A case of 70 year-old woman with massive rectal bleeding from a Dieulafoy lesion of rectum was presented. Five years previously she underwent emergency operation because of massive upper gastrointestinal bleeding from active arterial spurting through a minute ulcer at lesser curvature of proximal stomach. The operative finding was compatible with visual criteria of gastric Dieulafoy's lesion. Suture ligation was performed. Reported cases of rectal Dieulafoy's lesion in the literature were reviewed.**

She received successful endoscopic treatment by application of monopolar electrocoagulation probe for the bleeding lesion in the rectum and had no recurrent bleeding for up to 20 months. This patient was believed to have Dieulafoy's lesions in the stomach and the rectum that were responsible for each episod of upper and lower gastrointestinal bleeding.

Dieulafoy's lesion is uncommon cause of gastrointestinal bleeding but can be a potentially life threatening if unrecognised.[1] It was first reported by Gallard in 1884 as "Miliary aneurysm". Dieulafoy has been credited for fully characterized this lesion in 1897 which he coined the term "Exulceratio simplex". Characteristic of the lesion is a large tortuous submucosal artery or caliber persistent artery associated with small mucosal defect. Massive intermittent gastrointestinal bleeding suddenly occur when the exposed artery ruptures.[2-4] Emergency endoscopy is essential for diagnosis but repeat endoscopies are often necessary. Selective angiography and scintigraphy may be helpful if endoscopy fails to identify the bleeding lesion. Therapeutic endoscopy is the treatment of choice with acceptable safety and effectiveness.[5,6] Selective arterial embolization yields less effective hemostasis and is useful in selected or complicated cases.[7] Surgery is the most effective hemostatic method and is indicated when therapeutic endoscopy has failed or unavailable. This lesion can be found anywhere along the gastrointestinal tract, but the lesser curvature of proximal stomach within 6 cm from esophagogastric junction is the most common location.[1-6] Twenty six cases of rectal Dieulafoy's lesion have been reported since 1991.[8-24]

Case Report

A 70 year old woman came to the hospital 4 hr after she passed large amount of clotted and fresh blood per rectum. On admission, she had normal vital signs but with observable orthostatic change. Physical examination was otherwise unremarkable. Digital rectal examination and proctoscopy revealed clotted blood in rectum without active bleeding. The initial hematocrit was 24%. Two units of packed red cell were transfused. Emergent esophagogastroduodenoscopy (EGD) revealed normal finding. Colonoscopy was performed within 24 hr later after bowel preparation. The findings showed a firm blood clot adherent to a minute mucosal defect of rectum at 8 cm from anal verge (Figure 1). When the adherent clot was removed, micropulsatile bleeding from a protruded vessel was observed. Hemostasis was achieved by application of monopolar electrocoagulation probe directly to

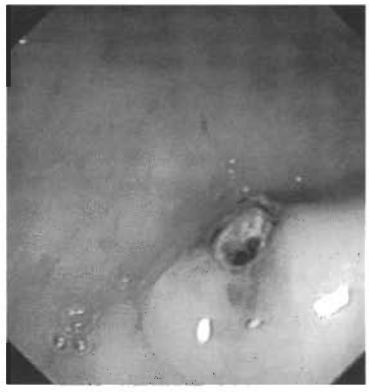

Fig. 1 Typical rectal Dieulafoy's lesion showing adherent clot on minute mucosal defect with normal appearance of surrounding mucosa.

Fig. 2 Endoscopic visualization after application of monopolar electrocoagulation probe

bleeding point with firm compression (Figure 2). She had no recurrence of bleeding in 20 months of follow-up period.

Five years previously the author performed emergency operation for this patient due to massive upper gastrointestinal bleeding with hemodynamic instability at initial presentation. Preoperative esophagogastroduodenoscopy showed huge clotted blood and fresh blood in the stomach which obscured endoscopic visualization. The operative findings demonstrated an active arterial spurting through a minute ulcer at the lesser curvature of proximal stomach which was compatible with the visual criteria of a Dieulafoy's lesion. Suture ligation of the bleeding point was performed without tissue for histologic confirmation.

Discussion

Currently the diagnosis of Dieulafoy's lesion mainly base on endoscopic visualization, Dy et al[25] proposed endoscopic criteria for diagnosis of Dieulafoy lesion which were (1) Active arterial spurting or micro-pulsatile from a minute mucosal defect or through normal surrounding mucosa; (2) Visualization of a protruding vessel with or without active bleeding within a minute mucosal defect or through normal surrounding mucosa; (3) Fresh, densely adherent clot with a narrow point of attachment to a minute mucosal defect or to normal appearing mucosa. Therapeutic endoscopy can be achieved in three methods,[5,6,25-28] namely (1) Injection sclerotherapy with epinephrine, polidocanol, ethanol or orther kinds of sclerosing agents; (2) Coagulation therapy with monopolar or bipolar electrocoagulation (BICAP) or heater probe or laser photocoagulation; (3) Mechanical therapy by hemoclipping or band ligation. Therapeutic endoscopy achieved permanent hemostasis in 85-96 per cent[6,26] of cases, therefore most of recently reported cases had no histologic confirmation. Endoscopic ultrasound with Doppler examination may be used in confirmation of diagnosis by demonstration of vascular lesion and also guiding injection therapy or assessment of therapeutic efficacy.[29] Because the large artery take a variable tortuous course about 2-4 cm in submucosa,[30] surgical treatment should be wide oversew ligation or wide wedge resection or segmental resection.

In this case presentation we used monopolar electrocoagulation probe to arrest the bleedig point because it is readily available in the operating room. It can be used with safety due to thick wall of rectum and it's retroperitoneal location although monopolar electrocoagulation can produce more deeper tissue destruction than other thermal modality. Biopsy that includes the pathologic artery may cause torrential bleeding and precludes endoscopic treatment. We believe this patient developed Dieulafoy's lesion both in the stomach and rectum.

All reported cases of rectal Dieulafoy's lesion including this case presented are summarized in Table 1. Although detailed information of each individual case were incomplete and unavailable in some literature, they were enough data for overview of this rare entity. Age incidence ranges from 5-78 years (median 69 years) male are more affected than female (5.7:1). Comorbidities were found in 10 cases (50%) and three patients[13,22,24] underwent colectomy from complication of diverticular disease. All cases presented with abrupt onset of hematochezia and 10 cases (50%)

Table 1 Summary of rectal Dieulafoy's lesion (1991-2002)

Author/year	Case	Age/Sex (year)	Diagnostic procedure (No of attempt)	Location from Anal verge	Stigmata of recent hemorrhage	Initial treatment (Retreatment)
Franko E 1991[8]	1	20/M	Sig (1)	11 cm.	Spurting	EC (oversew ligation)
Abdulian JD 1993[9]	2	43/M	Col (1)	7 cm.	Protuberance	Inj. Ep. (Inj. AA, STD)
Toosoon JD 1995[10]	3	5/F	Col (3)	4 cm	Protuberance	Inj. HSE + HP
Yeoh KG 1996[11]	4	66/M	Col (1)	6 cm	Protuberance	Inj.Ep.
Harrison JE 1996[12]	5	68/M	Angio. (1)	4 cm	Spurting	Suture ligation
Egushi S 1997[13]	6	78/M	Col (2), Angio, Scin	13 cm	Visible vessel	Suture ligation
Abdelmalek MF1997[14]	7	76/M	Sig, Col, Scin, Sig.	5 cm	Pulsatile	Inj. Ep + HP + Suture ligation
Tan MPC 1997[15]	8-12	Detailed information of each case was not available				
Meister TE1998[16]	13	74/M	Sig (2)	3 cm	Pulsatile	Inj. HSE + HP
	14	77/M	Col (1)	4 cm	Adherent clot	HP
	15	67/M	Sig (1)	10 cm	Pulsatile	Inj. HSE + HP
	16	7/M	Col (1)	7 cm	Visible vessel	Inj. HSE + HP
Nozoe T 1999[17]	17	65/M	Col (1)	5 cm	Spurting	Hemoclip
Amaro R. 2000[18]	18	73/M	Col (1)	7 cm	Oozing	Inj. Ep + HP (Ing. Ep + HP)
Kayali Z.2000[19]	19	77/M	Scin, Col, Sig.	NS	Protuberance	Inj. AA,Ep
	20	73/M	Col, Scin, Col	NS	Visible vessel	Inj. AA,Ep
Chunk IK 2000[20]	21-22	Detailed information was not available except treatment by Hemoclip and Inj. HSE.				
Rajendra T 2000[21]	23	78/M	Col (1), Sig (1)	6 cm	Protuberance	Inj. Ep + BICAP (oversew ligation)
Matsuoka J 2000[22]	24	54/F	Col.(1), Angio, Sig.	3 cm	NS	Inj. AA + EC
Guy RJ 2001[23]	25	18/M	Sig (2), Angio.	3 cm	Spurting	Embolization (Underun suture)
Enns R 2001[24]	26	72/M	Col (2)	NS	Spurting	Inj. Ep. + BICAP.
Treesaranuwattana S 2002	27	70/F	Col (1)	8 cm	Adherent clot	EC

Sig. = Sigmoidoscopy, Col = Colonoscopy, Angio = Angiography, Scin = Scintigraphy, EC = Electrocoagulation, Inj = Injection, HP =Heater probe, AA = Absolute alcohol, STD = Sodium tetradecyl sulfate, HSE = Hypertonic saline and epinephrine solution, Ep = Epinephrine, NS = Not stated, M = Male, F = Female.

had hemodynamic instability at initial presentation. Blood transfusion was required in 14 cases (74%), ranging from 1-14 units. Lesion located at 3-13 cm from and verge, of which 13 cases (81%) were below 8 cm. Initial endoscopy with colonoscopy or sigmoidoscopy can identify the lesion in 9 cases (45%). Selective angiography performed in 4 cases but in only two case that the site of bleeding were demonstrated. Scintigraphy was performed in 4 cases with 3 cases showed active bleeding.

Failure of initial endoscopic diagnosis in seven cases led to second episode of bleeding within 1-12 days (median 2 days).[10,14,19,21,22,24] One of these cases[22] developed the third episode of bleeding on the fourth day. Two cases[10,14] required three endocospic attempts to find the bleeding lesion. Coincidence of diverticular disease was in found in 3 cases (18%).

Endoscopic appearance of stigmata of recent hemorrhage were spurting or pulsatile bleeding 8 cases (44%), oozing 1 case (6%), protuberance or visible vessel 7 cases (39%), and adherent clot 2 cases (11%). Endoscopic treatment modalities were injection therapy alone 5 cases, injection plus coagulation therapy 8 cases, coagulation therapy alone 3 cases, and hemoclipping 2 cases. Sclerosing agents were use as single, multiple agents or mixed solution. Initial hemostasis was achieved in 17 cases (89%). Two cases[9,18] had recurrent bleeding in 3rd and 4th day after initial endoscopic treatment. Permanent hemostasis was achieved in 14 cases (87%), one patient[11] died of pneumonia on the fourth day. Surgical treatment by suture or oversew ligation was performed as initial treatment in 2 cases,[12,13] retreatment of endoscopic failure in 2 cases [8,21] and retreatment in one case after failure of selective arterial embolization.[24] One case[14] was treated by injection and followed by heater probe and suture ligation without recurrent bleeding. Endoscopic and surgical treatment achieved hemo-

stasis in all cases without procedure related mortality and morbidity in follow-up period 2-72 months (mean 14.2 months). All reported cases had no histologic diagnosis.

Conclusion

Rectal Dieulafoy's lesion is a rare cause of lower gastro-intestinal bleeding. Pathology, clinical presentation, diagnostic and treatment modalities of rectal Dieulafoy's lesion are similar to gastric Dieulofoy' s lesion. Hemostasis was achieved in all cases by therapeutic endoscopy and surgical treatment without procedure related mortality and morbidity. This probably due to location of rectal lesion is easily accessible for diagnostic and therapeutic procedure, and thick wall of rectum allows more aggressive endoscopic treatment with low incidence of perforation. By visual criteria, we believe that this patient had two Dieulafoy' lesions in the stomach and rectum.

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