Dieulafoy’s lesions of the rectum:
A rare cause of lower gastrointestinal bleeding

Robert Enns MD FRCP

Massive hematochezia typically occurs in elderly patients (average 60 years old) (1), carries a mortality rate of up to 30% (2-5) and accounts for about 0.7% of all discharges from hospital (6). Although there are many possible causes of lower gastrointestinal bleeding (7), the etiology of massive lower gastrointestinal bleeding is much more limited. The most common cause of massive lower gastrointestinal bleeding is diverticulosis, which accounts for approximately 30% to 40% of all cases (7-13). Although arteriovenous malformations are responsible for only 2% of all lower gastrointestinal bleeding (7), they account for 15% to 30% of massive gastrointestinal hemorrhage (8,9,11-19). Other causes of massive lower gastrointestinal bleeding include (in order of decreasing frequency)
cancer, polyps, inflammatory bowel disease and ischemia.

Dieulafoy's lesion, also known as 'caliber-persistent artery of the stomach', was originally reported by Gallard (20) and subsequently was described further by the French surgeon Dieulafoy (21) in 1889. It is usually a gastric lesion found in the proximal one-third of the stomach, near the esophagogastic junction. Histologically, it is defined as a thick-walled arterial vessel surrounded by a very shallow ulcer (22). The presentation is relatively uniform, with patients presenting with massive upper gastrointestinal hemorrhage (sometimes recurrent) and melena. The lesion is uncommon (1% to 2% of upper gastrointestinal hemorrhages) (23-25) and sometimes difficult to locate endoscopically. Once located (usually in the body or fundus of the stomach), endoscopic therapy is the treatment of choice. Originally confined to the stomach, Dieulafoy's lesions have now rarely been described in the esophagus (23), small bowel and colon (26-29). We describe a case of massive lower gastrointestinal hemorrhage secondary to a rectal Dieulafoy's lesion, which was treated successfully endoscopically.

CASE PRESENTATION
A 72-year-old white man presented to hospital with shortness of breath. He had an extensive cardiac history, and had been managed on amiodarone, nitroglycerin and furosemide. Investigations on his shortness of breath were extensive and included computerized tomography of his chest, bronchoscopy and subsequent open-lung biopsy. Bronchiolitis obliterans with organizing pneumonia and interstitial fibrosis (likely secondary to amiodarone) was diagnosed. Intravenous steroids were administered with some improvement in his respiratory parameters. Surgical history
His creatinine was also noted to be elevated to 205 µmol/L, included a sigmoid colon resection for diverticular disease. His creatinine was also noted to be elevated to 205 µmol/L, and a renal biopsy did not demonstrate any evidence of vasculitis. Three weeks after admission, passage of bloody stools was noted. His hemoglobin decreased from 104 g/L to 92 g/L, but he did not develop any orthostatic changes. Although he was transfused with 2 U of blood that evening, his hemoglobin fell further the following day to 84 g/L. Although he had no upper gastrointestinal symptoms, an upper endoscopy was performed, which demonstrated superficial esophageal ulcers. These were biopsied and proved to be herpes simplex esophagitis. Because no blood was visualized in the upper intestinal tract, a colonoscopy was performed immediately after the upper endoscopy. This demonstrated dark blood throughout the colon (with the surgical anastamosis visible at 20 cm from the anal verge) but no evidence of active bleeding. There was no blood within the terminal ileum.

Because there was no evidence of active bleeding, the patient was managed supportively. The following day, rectal bleeding recurred; this time it appeared bright red in colour. A total of 8 U of packed red blood cells were transfused. A repeat unprepared colonoscopy was performed. Initially, visualization was challenging within the rectum, because bright red blood coated the entire region up to the surgical anastamosis. Careful irrigation showed what appeared to be a 'spurting' site of bleeding 5 cm distal to the anastamosis (Figure 1, Top left). Three millilitres of 1/10,000 adrenaline were injected into the bleeding region, with 'blanching' of the mucosa and subsequent cessation of bleeding. The site was then localized and determined to be Dieulafoy's lesion of the rectum (Figure 1, Bottom left). Using a 10 French BICAP probe (bipolar probe, Circon ACMI Corporation, USA) electrocaugulation (20 J, 5 s intervals, five applications), the lesion was cauterized (Figure 1, Top right, Bottom right). Over the next 12 months, there was no recurrence of bleeding.

### DISCUSSION

‘Exulceratio simplex’ was a term coined by Dieulafoy (21) to describe a superficial gastric mucosal lesion that he believed to be the initial stages of a gastric ulcer whose progression was interrupted by the occurrence of bleeding. The ‘Dieulafoy’ lesion has now, however, been characterized histologically as an unusually large artery coursing just beneath the gastric mucosa (22). Once thought to be a rare cause of gastrointestinal hemorrhage, the widespread use of emergency endoscopy has led to increasing numbers of reports of this lesion in various parts of the gastrointestinal tract. Although usually considered an acquired abnormality, a congenital etiology has been suggested by authors who have discovered the lesion in patients as young as 20 weeks old (30). The most common site of these lesions remains the stomach, with most lesions located in the body (67%) and a smaller number (25%) in the fundus of the stomach (25).

Dieulafoy’s lesions of the colon were first reported by Barbier et al (31) in 1985, when three cases of lower gastrointestinal hemorrhage from ‘Dieulafoy-like’ lesions were published. Since that time, 19 other cases of lower gastrointestinal bleeding from Dieulafoy’s lesions have been reported (Table 1). What actually causes these lesions to bleed is a source of speculation. No correlation has been noted with alcohol, smoking, nonsteroidal anti-inflammatory drugs or peptic ulcer disease. Constipation has been suggested as an initiating factor in colonic Dieulafoy’s lesions (27,32,33), as has arterial compression of the mucosa (leading to a mucosal erosion) (30).

<table>
<thead>
<tr>
<th>Author (reference)</th>
<th>Age (years), sex</th>
<th>Bleeding site (n)</th>
<th>Diagnosis (n)</th>
<th>Treatment (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Barbier et al (31)</td>
<td>59, male; 63, male; 74, male</td>
<td>Right colon</td>
<td>Surgical (2); endoscopic (1)</td>
<td>Surgical resection (3)</td>
</tr>
<tr>
<td>Richards et al (39)</td>
<td>60, male</td>
<td>Right colon</td>
<td>Angiography</td>
<td>Surgical resection</td>
</tr>
<tr>
<td>Schmitt et al (40)</td>
<td>21, female</td>
<td>Right colon</td>
<td>Endoscopic</td>
<td>Endoscopic (adrenaline and athoysklerol)</td>
</tr>
<tr>
<td>Schmid et al (41)</td>
<td>69, male</td>
<td>Right colon</td>
<td>Endoscopic</td>
<td>Endoscopic polyectomy (cauterized)</td>
</tr>
<tr>
<td>Ma et al (42)</td>
<td>60, male</td>
<td>Right colon</td>
<td>Radiological (angiogram)</td>
<td>Surgical resection</td>
</tr>
<tr>
<td>Franko and Chadovoyne (33)</td>
<td>20, male</td>
<td>Rectum</td>
<td>Endoscopic</td>
<td>Failed adrenaline and heater probe; surgical (oversewn)</td>
</tr>
<tr>
<td>Abdullian et al (32)</td>
<td>43, male</td>
<td>Rectum</td>
<td>Endoscopic</td>
<td>Endoscopic (adrenaline and alcohol and tetradocyl sulphate)</td>
</tr>
<tr>
<td>Tooson et al (36)</td>
<td>5, female</td>
<td>Rectum</td>
<td>Endoscopic (colonoscopy × 3)</td>
<td>Endoscopic (adrenaline and heater probe)</td>
</tr>
<tr>
<td>Dy et al (26)</td>
<td>65, female; 70, male; 73, female; 76, female; 94, female</td>
<td>Right colon (4); transverse colon (1)</td>
<td>Endoscopic</td>
<td>Endoscopic (adrenaline and heater probe)</td>
</tr>
<tr>
<td>Abdemalek et al (35)</td>
<td>76, male</td>
<td>Rectum</td>
<td>Endoscopic</td>
<td>Endoscopic (adrenaline and heater probe), then surgical (oversewn)</td>
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<tr>
<td>Meister et al (27)</td>
<td>5, female; 7, male; 67, male; 74, male; 77, male</td>
<td>Rectum</td>
<td>Endoscopic</td>
<td>Endoscopic (adrenaline and heater probe)</td>
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<tr>
<td>Eguchi et al (37)</td>
<td>78, male</td>
<td>Rectum</td>
<td>Red blood cell scintigraphy</td>
<td>Surgical (oversewn)</td>
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the rectum. Seventeen of the 22 lesions were diagnosed by endoscopic means. Similar to Dieulafoy’s lesions of the stomach, approximately twice as many men were affected as women (15 versus seven) (34). The mean age was 58 years old. Eight lesions (three rectal, five right colon) were treated surgically (three oversewn, five right hemicolecotomies). The other 14 lesions were treated endoscopically with a combination of adrenaline and heater probe (n=7); heater probe alone (n=3); adrenaline and yttrium-argon-garnet laser (n=1); adrenaline and a sclerosing agent (n=2); and snare polypectomy (n=1). Two patients treated endoscopically subsequently had recurrent bleeding. One had been treated with adrenaline injection, and a repeat endoscopic treatment with a sclerosing agent (alcohol and sodium tetradecyl sulphate) successfully stopped the bleeding (32). Another had a lesion at the hepatic flexure treated initially with heater probe coagulation and adrenaline. Repeat therapy with the same modalities was successful when he rebled three days after the initial treatment (26). One of the lesions treated surgically (oversewn) had previously failed endoscopic therapy (33). The other two rectal lesions were treated surgically (despite successful endoscopic treatment in one) (35), presumably on the assumption that endoscopic therapy would not result in long term success in the cessation of bleeding.

This case of a Dieulafoy’s lesion is the 11th discovered in the rectum. All presented with hematochezia. Only two were female (both five years old) (36). Overall, 10 of 11 patients with rectal Dieulafoy’s lesions were diagnosed endoscopically (one by nuclear scintigraphy). Three patients subsequently had surgical over-sewing performed, but only one of the surgical cases had failed endoscopic management (33). In another, surgical over-sewing was performed intra-operatively when the lesion was discovered within the rectum (37). The case presented is the only one formed intraoperatively when the lesion was discovered. Levine et al. (20) also presented a case of a rectal Dieulafoy’s lesion that bled consistency with an arterio-venous malformation but only one of the surgical cases had failed endoscopic therapy (33).

REFERENCES


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