

Case report

Rectal dieulafoy-like lesion complicated by an ischemic ulcer after successful endoscopic injection-therapy with dilute epinephrine in hypertonic glucose water

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SUMMARY

Dieulafoy's lesion is an unusual source of massive lower gastrointestinal hemorrhage. It is characterized by severe bleeding from a minute submucosal arteriole that bleeds through a punctate erosion in an otherwise normal mucosa. Although Dieulafoy's lesions were initially described only in the stomach and upper small intestine, they are being identified with increasing frequency in the colon and rectum. We describe a young patient who presented with severe lower gastrointestinal bleeding caused by a rectal Dieulafoy-like lesion. This is the first report of a rectal Dieulafoy-like lesion treated successfully with endoscopic epinephrine plus hypertonic glucose water (50%GW) injection and complicated by an ischemic ulcer.

Key words: Dieulafoy's lesion, Epinephrine, Hypertonic Glucose Water (50%GW)

INTRODUCTION

Dieulafoy's disease is a well known entity characterized by a bleeding or clot bearing artery protruding into the intestinal lumen without surrounding ulceration.¹ The exact process that causes this tortuous superficial vessel to become eroded and bleed is unknown.

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Most cases have occurred in the gastro-esophageal junction and in the stomach along the lesser curvature. Less commonly cases are encountered in the small intestine, colon and rectum.¹ Only nine cases of rectal Dieulafoy-like lesion have been reported in the English medical literature. We present a case of massive rectal bleeding secondary to a rectal Dieulafoy-like lesion in a young woman. This was treated successfully by an injection of 8ml 1:10.000 epinephrine plus hypertonic glucose water (50%GW) and complicated by an ischemic ulcer.

CASE REPORT

A 40-year-old woman presented to the emergency room with a 4h history of bright red blood per rectum. For many hours before the onset of bleeding, she had been aware of anorectal discomfort and a sensation of wanting to defecate. There was no history of previous gastrointestinal bleeding or gastrointestinal disease. The patient had tachycardia and orthostatic changes, but remained normotensive. The abdomen was soft and non-tender without hepatosplenomegaly, whereas rectal examination revealed bright red blood. The initial Ht was 36% and decreased to 31% with fluid resuscitation. The prothrombine time (PT), partial thromboplastin time (PTT), and the platelets count were normal. Colonoscopy undertaken 2h after admission revealed fresh blood in the rectum and sigmoid colon. On withdrawing the scope blood was seen to be oozing from a small mucosal defect in the superior valve of Houston. We injected 8cc of epinephrine plus hypertonic glucose water (50%GW) 1:10.000 to the lesion resulting in blanching around the mucosal defect (Figure 1A). Hemostasis was secured and bleeding did not recur. A flexible sigmoidoscopy, five days

later revealed an ulcer with thrombus in the injected area (Figure 1B). We suggested to our patient to increase her intake of dietary fiber to 20g/day, and a new colonoscopy one month later showed complete healing of ulcer.

DISCUSSION

Exulceration simplex was first reported by Gallard in 1884,² and was characterized 14 years later by the French surgeon Dieulafoy.³

Dieulafoy's lesion have been typically associated with the upper GI tract. More than 75 percent of these lesion are located along the lesser curve in the cardia or proximal body of the stomach.¹ Less than one-fourth are encountered in the duodenum, and there are incidental reports of Dieulafoy's lesions of the jejunum⁴ small intestine⁵ and colon.^{6,7}

Nine cases with rectal Dieulafoy's lesions were described previously in the literature.⁸⁻¹⁴ All cases presented with acute, massive, lower gastrointestinal bleeding. All of them were diagnosed endoscopically, three with rigid sigmoidoscopy, the other six with colonoscopy. The first case reported in the literature by Franco et al⁸ was a 20-year old male who required transanal surgical ligation after multiple attempts to control the bleeding endoscopically, including coagulation, failed. The case described by Abdulian et al⁹ was treated initially with epinephrine but rebleeding occurred after four days. A sec-

ond therapeutic endoscopy was performed using a combination of alcohol and sodium tetradecyl sulfate with good effect. Tooson et al¹⁰ described a five-year-old female with intermittent rectal bleeding who required three colonoscopies to find the rectal Dieulafoy's lesion. She was treated with a combination of epinephrine and 3.6 percent NaCl followed by thermocoagulation (30J). The fourth case¹¹ was treated initially only with epinephrine injection with good hemostatic effect, but the vessel was still present on a sigmoidoscopic examination performed the next day. There was also evidence of rectal bleeding. The patient could not be followed up, because he died of pneumonia four days later.

Amaro et al¹² reported an elderly patient who presented with severe lower gastrointestinal bleeding caused by a rectal Dieulafoy's lesion. The lesion was treated with epinephrine injection followed by heater probe coagulation as recommended for gastric lesions. After treatment there was an episode of rebleeding which could have been the result either of sloughing of the eschar over the vessel or from the ulcer created by the first coagulation therapy. They consider the latter more likely, knowing the great vascularity of the rectal mucosa and the nonvisualization of the vessel at the time of the second endoscopy. Two cases of gastrointestinal hemorrhage resulting from Dieulafoy-like lesion of the rectum are described by Kayali et al.¹³ Both patients developed acute episodes of massive lower gastrointestinal hemorrhage.

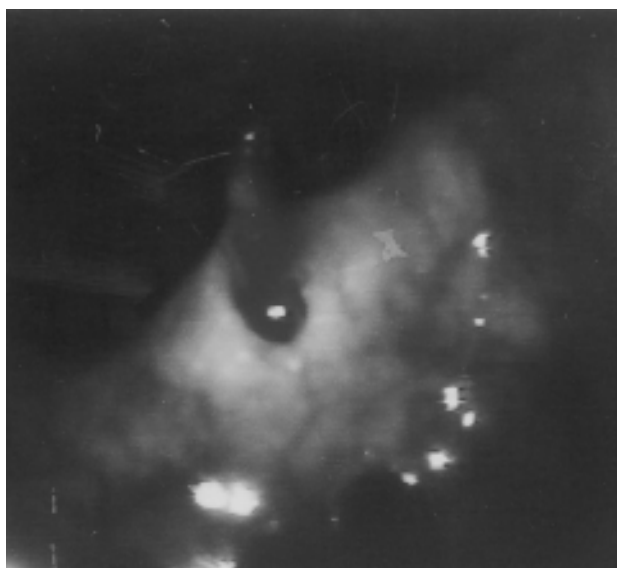


Figure 1A. Submucosal injection of dilute epinephrine in hypertonic glucose water stops bleeding from Dieulafoy-like lesion and causes mucosal blanching.



Figure 1B. A deep ulcer with thrombus has developed in the injection area 5 days following therapy.

The patients were successfully treated by alcohol and epinephrine injection. Colonoscopy assisted in earlier diagnosis and added therapeutic options to the treatment regimen for this lesion.

The last two cases¹⁴ were two Dieulafoy's lesions of the anal canal who presented with sudden onset of massive hemorrhage. In first patient the bleeding was controlled by oversew ligation of the pumping arteriole. The second patient was taken to the operating room and the lesion was excised in continuity with the hemorrhoidal complex under local anesthesia. No further bleeding was encountered, in both cases, at a follow-up of three months.

Various forms of endoscopic therapy have been reported with Dieulafoy's lesion, including heater probe, bipolar coaptive electrocoagulation, non-contact laser photocoagulation, injection epinephrine plus ethanol or sodium tetradecyl-sulfate, and rubber band ligation.¹⁵ When endoscopic therapy is unsuccessful in stopping bleeding from Dieulafoy's lesion, surgery is the keystone with wedge resection or oversewing of lesion.

Dilute epinephrine 1:10,000 or 1:100,000 with 7.2% or 3.6% hypertonic saline, 0.9% saline and hypertonic dextrose water is highly effective in stopping active bleeding of non-variceal gastrointestinal lesions. Submucosal injection of epinephrine cause local tissue tamponade, vasoconstriction and platelets aggregation. Unlike sclerosants, epinephrine does not cause tissue damage and large volumes (up to 20ml) can be safely injected.¹⁵

Chung et al¹⁶ reported that treatment with hypertonic glucose water (50%GW) in gastric variceal bleeding was superior to treatment with 1.5% sodium tetradecyl sulfate (STS) in controlling bleeding and in achieving variceal obliteration, less rebleeding, and a lower complication rate. The rate of occurrence of secondary gastric ulcers after injection sclerotherapy was significantly higher (92%) in the STS group than in the GW group (46%) ($p < 0.05$). They believe that hemostasis due to osmotic dehydrating and sclerosant effect of hypertonic glucose water, despite less mural necrosis and fibrotic obliteration of varices occurs.

Histopathologically, the Dieulafoy-like lesion is characterized by a small mucosal defect with minimal inflammation and a large tortuous, thick-wall artery at the base of the defect with rupture of the artery into the lumen.¹ Endoscopic criteria for diagnosis of Dieulafoy's lesion include a minute, less than 3mm, mucosal defect in combination with any of the following: 1) a protruding small (1-2mm) blood vessel; 2) active arterial bleeding; or 3)

fresh adherent clot with a narrow point of attachment.¹⁴ The minute mucosal defect typically occurs within an area of otherwise normal mucosa. There is no surrounding inflammation exudates, or ecchymosis. Our case fulfilled those visual criteria in view of both the presence of a minute mucosal defect without surrounding inflammation, and active arterial bleeding.

We believe that there was synergic effect of epinephrine and hypertonic glucose water producing permanent thrombosis of the vessel responsible for hemorrhage, and ischemic necrosis of injected area.

In conclusion our case underscores that submucosal injection of dilute epinephrine in hypertonic glucose water (50%GW) is very effective in stopping bleeding of non variceal gastrointestinal lesion although non complicated.

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