CAS CLINIQUE/CASE REPORT

RECTAL DIEULAFOY’S LESION
Report of two cases treated successfully by endoscopic means

Rita SLIM, César YAGHI, Khalil HONEIN, Rami MOUKARI, Raymond SAYEGH

ABSTRACT : Rectal Dieulafoy’s lesion is an unusual cause of abrupt and massive lower gastrointestinal hemorrhage. It is characterized histologically by a caliber-persistent submucosal artery that protrudes through a minute mucosal defect. Various theories and risk factors have been proposed to explain the occurrence of bleeding but none is completely satisfying.

We present two cases of rectal Dieulafoy’s lesion which were treated efficaciously by a simple injection of a sclerosing agent in the first case and by a combination of epinephrine injection and thermal probe coagulation in the second leading to a complete and rapid disappearance of the abnormal vessel.

INTRODUCTION

Initially described by Gallard [1] in 1884, then characterized by Dieulafoy [2] in 1898, Dieulafoy’s lesions (DL) have been identified in different parts of the digestive tract. The first case of rectal Dieulafoy was reported in 1991 [3] and since then twenty to twenty-five cases were reported in the literature [4-8]. Traditionally, surgery was the treatment of choice, more recently endoscopic management using different modalities proved to be efficacious [8-10]. We describe two patients who presented with massive lower gastrointestinal bleeding due to a rectal DL, which were diagnosed, treated and controlled endoscopically with complete disappearance of the visible vessel, 24 hours later.

CASE REPORTS
Case 1

A 58-year-old female with a past medical history of hypertension, diabetes mellitus and left hemiplegia, treated with clopidogrel (Plavix®), was admitted in the emergency room for massive bright rectal bleeding. The patient had had no recent history of abdominal pain, enemas administration or rectal thermometer use and previous stools had been normal.

Her hemoglobin on admission was 6.5 g/dl and blood pressure 100/60 mmHg.

After hemodynamic resuscitation with intravenous fluids and two packed blood cells units, an upper gastrointestinal endoscopy was performed and revealed congestive gastritis. A flexible rectosigmoidoscopy showed a large amount of blood in the rectum without identifying any lesion.

The bleeding stopped spontaneously and the patient remained hemodynamically stable. A bowel preparation was immediately started.

Twenty-four hours later, a massive rectal bleeding reoccurred and the patient received 8 units of packed blood cells. A colonoscopy was then performed revealing red blood and clots in the rectum. No blood was noted in the entire colon which was free of any lesion causing massive lower gastrointestinal bleeding. While withdrawing the colonoscope, an adherent clot was identified at 3 cm above the anal margin over a visible vessel protruding from a normal surrounding rectal mucosa (Figures 1, 2). The lesion was endoscopically treated with 8 ml of 1/10000 epinephrine and polidocanol 1% with good hemostatic effect. A flexible rectosigmoidoscopy done 24 hours later showed a clear scar (Figure 3) in the treated area with no evidence of the incriminated arteriole and complete absence of blood. There was no recurrence of bleeding after 23 months of follow-up.

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Case 2

A 72-year-old woman, with a past medical history of coronary bypass treated with clopidogrel (Plavix®) and bisoprolol (Concor®), was admitted in the emergency room for a brisk rectal bleeding developed 4 hours before admission. There was no history of abdominal pain nor use of any non steroidal anti-inflammatory medication and previous stool were normal.

On admission, her blood pressure was at 110 over 60 mmHg and her pulse rate at 100 beats/mn. Digital rectal exam showed fresh blood clots. Her hemoglobin level on admission was at 11 g/dl. Renal and hepatic function tests were normal. A flexible rectosigmoidoscopy, demonstrated at 5 cm above the anal margin an adherent clot overlying a visible vessel. The surrounding mucosa as well as the 40 last centimeters of the sigmoid colon were normal. The lesion was treated with 6 ml of epinephrine diluted at 1/10000 followed by coagulation with bipolar thermal probe (gold probe® by microvasive).

A second flexible rectosigmoidoscopy done 24 hours later showed only a scar in the treated area with no blood. The patient was discharged on the second day after admission and there was no recurrence of bleeding after 18 months of follow-up.

DISCUSSION

An isolated protuberant vessel visualized in the rectum without any ulcer or erosion with a normal surrounding mucosa is diagnostic of a rectal DL. The pathogenesis of this lesion is still debated, but histologically it is characterized by an aberrant artery coursing beneath the mucosal surface without any change of the caliber as it traverses the muscularis propria toward the muscularis mucosa [11]. It’s believed now that this lesion is congenital in origin and could be seen in all age groups, albeit rarely in neonates and young infants [12-13]. Various theories have been proposed to explain the occurrence of bleeding. The presence of dysplastic changes, which are seen at the point of rupture of the vessel supports the theory that the final pathologic process is one of slow weakening of the vessel wall with perhaps an eventual localized dilatation [11].
ties, particularly cardiovascular disease, hypertension, chronic renal failure, diabetes, and excessive use of alcohol, have been described in almost 90% of all the patients with DL [14]. Our first patient was diabetic and had evidence and risk factors of atherosclerosis whereas the second one had stable cardiovascular disease. Both of our patients were taking platelets aggregators inhibitors. Most cases of rectal DL that have been described couldn’t find any clear association between the occurrence of bleeding and the use of such medications, even though the use of aspirin, warfarin, or nonsteroidal anti-inflammatory drugs have been reported in more than 50% of patients with gastric DL [15-16].

Dieulafoy’s lesion is considered as a rare cause of gastrointestinal bleeding. It has a male predominance with a sex ratio of about 2 to 1 [11-12, 14]. The age of predilection is between 50 to 70 years of age [14-15, 17]. It’s incidence along the gastrointestinal tract varies between 1.2 and 3.4% [14, 18-19]. Although it is typically found in the proximal stomach, physicians should be aware that it must be included as a potential cause of massive rectal bleeding where it’s most frequently found in the proximal colon and rectum [3, 7, 14, 16, 18, 20]. Rectal localization of DL is atypical. Azmuuddin et al. [21] described two cases of DL involving the anal canal. Several cases of rectal DL have been so far reported in the literature [5-10, 22]. The classic presentation was that of an abrupt onset of painless rectal bleeding. Endoscopy has been able to localize the lesion in almost all the cases, sometimes after repeated examinations [6, 14]. Angiography was rarely necessary in contrast to colonic lesions where massive bleeding makes the endoscopic diagnosis more difficult [23]. Our first patient had two endoscopic examinations, the second one with better conditions allowing the visualization of the lesion. We think that a standard bowel preparation, when feasible, along with the awareness of the physician facilitate the diagnosis of rectal DL.

In the era of therapeutic endoscopy, transanal suture ligation [3] or resection [22, 24] of rectal DL is considered as the last resort of treatment when endoscopic therapy fails. Multiple therapeutic modalities had been used, including injection techniques, coagulation methods, band ligation and hemoclipping (Table I). Although there are no randomized controlled studies comparing the available modalities, the different endoscopic therapies are reported as safe and efficacious in controlling bleeding from a rectal DL. Combination therapy with injection followed by thermal probe coagulation is the predominant treatment in the reported cases. Meister et al. [6] reported 5 patients who received this treatment and who where followed clinically between 8 to 25 months without recurrence of bleeding. Amaro et al. [7] reported another case treated similarly with a colonoscopic control 3 months later showing complete disappearance of the lesion.

Injection therapy using epinephrine and/or a sclerosing agent (polidocanol, absolute alcohol or Na tetradecyl) is considered a good therapeutic choice [4, 8] echoing the more extensive accumulated data for the treatment of gastric DL [32], where initial reported hemostasis rates were as high as 95% [33-34], but with reported rates of recurrent bleeding as high as 55% [17]. An isolated injection of epinephrine has been reported in one case of rectal DL, the bleeding stopped but the vessel was still visible on the sigmoidoscopic examination performed the next day [5]. Clipping is being used in the colon in the prevention and hemostasis of post polypectomy hemorrhage and the repair of iatrogenic perforation. It also has been effective in controlling massive hemorrhage from a rectal Dieulafoy’s lesion in four recent cases [25-27]. Endoscopic elastic band ligation has been used successfully to treat DL in different parts of the digestive tract, especially in the rectum [28-31], with hemostasis rates ranging between 75% and 100% and no recurrence of bleeding [35].

In our cases, the first lesion was treated with a combination injection of epinephrine and polidocanol and the second one with epinephrine injection followed by thermal probe coagulation. The sigmoidoscopic control, done only 24 hours later, showed a complete disappearance of the abnormal vessels leaving instead a little scar. The long-term follow-up – approximately 2 years – with no recurrence of bleeding, confirms the efficacy of either treatments which were rapidly curative in our two cases. The main advantage of an endoscopic control, soon after the therapeutic approach, is the detection of a scar that facilitates the localization of the treated region in order to assess the complete disappearance of the vascular abnormality. If the vessel is still visible one should proceed to a second combined treatment option to prevent recurrence of bleeding.

In conclusion, we emphasize that although rectal DL lesions are unusual sources of rectal bleeding, they must be included in the differential diagnosis especially in the occurrence of massive lower intestinal bleeding. Our data support the use of endoscopic therapy as safe and effective in the treatment of rectal DL lesions. We further recommend a control rectosigmoidoscopy to detect lesions susceptible of recurrent bleeding and eventually propose a complementary treatment.

<table>
<thead>
<tr>
<th>TABLE I</th>
<th>RECAPITULATIVE TABLE INCLUDING ALL CASES OF RECTAL DIEULAFOY’S LESIONS PUBLISHED IN THE LITERATURE TREATED BY ENDOSCOPIC MEANS</th>
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R. SLIM et al. – Rectal Dieulafoy’s lesion
REFERENCES


