

Dieulafoy's Lesion in the Anal Canal: A Rare Cause of Massive Gastrointestinal Hemorrhage

Anal Kanalda Dieulafoy Lezyonu: Masif Gastrointestinal Kanamanın Nadir Görülen Bir Nedeni

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ABSTRACT

Dieulafoy's lesion is rare but life-threatening cause of lower gastrointestinal hemorrhage. In this study it is aimed to present a case of Dieulafoy lesion in anal canal which causes massive lower gastrointestinal hemorrhage and mortality. A 72 year-old male patient with massive lower gastrointestinal bleeding applied to the hospital. Since no lesion was detected on colonoscopy and scintigraphy, emergency laparotomy was performed for the unstable patient. On the intraoperative colonoscopy, a lesion was located proximal to anal canal and clipped. Despite the absence of rebleeding, the patient died due to respiratory complications associated with massive transfusion at the eighth day of his postoperation. For this kind of lesions, mortality rates were reported as up to 80%. In this case, the lesion was not found on evaluation performed under emergency setting. Although it is rare, Dieulafoy lesion should be kept in mind in patients with massive lower gastrointestinal hemorrhage. For treatment, conservative methods such as selective arterial embolisation, sclerotherapy, epinephrine injection, thermocoagulation and clips application can be used. When the location of bleeding was not revealed in preoperative evaluation, colonoscopy can be used during surgery. In case of delayed diagnosis the hemorrhage may be kept under control; however, morbidity and mortality may increase with massive transfusion.

Keywords: Anal canal, blood transfusion, colonoscopy, Dieulafoy lesion, hemorrhage, surgery

ÖZ

Dieulafoy lezyonu nadir görülmesine rağmen alt gastrointestinal sistem kanamalarının hayatı tehdit eden nedenlerindendir. Masif alt gastrointestinal kanamaya ve mortaliteye neden olan anal kanaldaki Dieulafoy lezyonunu sunmayı amaçladık. Yetmiş iki yaşındaki erkek hasta masif kanama nedeniyle hastaneye başvurmuştur. Kolonoskopi ve sintigrafide odak saptanamadığından, instabil durumda olan hastaya acil olarak laparotomi yapılmıştır. İntraoperatif olarak yapılan kolonoskopide anal kanal proksimalinde lezyon saptanarak klipslenmiştir. Kanama tekrarlamamasına rağmen, masif transfüzyona bağlı solunum problemleri nedeniyle postoperatif sekizinci günde hasta eksitus olmuştur. Bu tür lezyonlarda %80'e kadar varan oranlarda lezyon ilişkili mortalite oranları bildirilmiştir. Acil şartlarda yapılan bu değerlendirmelerde lezyon görülebilmiştir. Nadir karşılaşılsa da masif gastrointestinal kanamalarda Dieulafoy lezyonunun akılda tutulması gerekmektedir. Tedavide selektif arteriyel embolizasyon, skleroterapi, epinefrin enjeksiyonu, termokoagülasyon ve klipsleme gibi konservatif yöntemler kullanılabilir. Kanama odağının preoperatif dönemde bulunamadığı durumlarda lokalizasyon için cerrahi sırasında kolonoskopiden faydalanılabilir. Tanıda gecikme olan durumlarda kanama kontrol altına alınsa da masif transfüzyonlara bağlı morbiditenin ve mortalitenin artabileceği unutulmamalıdır.

Anahtar Kelimeler: Anal kanal, kan transfüzyonu, kolonoskopi, Dieulafoy lezyonu, kanama, cerrahi

Introduction

Dieulafoy's lesion is among the rare and life-threatening causes of bleeding from the lower gastrointestinal system (GIS).¹ It usually occurs due to submucosal arterial bleeding from the mucosal defect in the lesser curvature of the

stomach. Likewise, in addition to the stomach, it may be encountered in the esophagus, small intestine, bowel, rectum and anal canal.^{1,2,3} Herein, we aimed to present a case with Dieulafoy's lesion in the anal canal, which caused massive lower GIS hemorrhage and mortality.



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Case Presentation

A 72-year-old male patient without documented systemic disease has been referred to the gastroenterology clinic from an external center for lower GIS hemorrhage lasting for 24 hours. Although the urgent endoscopic evaluation of the upper and lower GIS was suboptimal, no focus of active bleeding was detected. Angiographic examination was not performed because of high creatinine value (1.7 mg/dL) and creatinine clearance of <50 ml/min. The scintigraphic examination, which was performed with 20 mCi Tc-99m-marked erythrocyte, demonstrated probable bleeding from the left colon. The patient received a total of 12 units of erythrocyte suspension with 6 units given within the 24 hours after hospitalization, and 6 units of fresh frozen plasma. The case was discussed with the general surgery clinic because of continuing bleeding, poor general status, and suspicious bleeding focus in the left colon. Overall examination of the patient revealed an arterial blood pressure of 80/50 mmHg, arterial pulse of 110/minute, hemoglobin value of 8.3 g/dL (11.7-16), hematocrit concentration of 24.3% (35-47), platelet count of $90 \times 10^3/\mu\text{L}$ (150-400), and international normalized ratio value of 1.3 (0.8-1.2). The patient underwent laparotomy because he was hemodynamically unstable despite massive transfusion. The exploration demonstrated no extraluminal pathology. Appendectomy was performed, the stump was not sutured, but seromuscular continuous circular suture around (purse suture) was placed. The colon was accessed through the appendectomy stump by a colonoscope and the colonoscope was extraluminally pushed forward by hands towards to the proximal aspect; no intraluminal pathology was detected. Colotomy was performed through the sigmoid colon as the focus suggested on scintigraphy and the entire colon was examined accessing by a colonoscope. A Dieulafoy's lesion of 1 cm was detected in the proximal aspect of the anal canal and a clip was placed endoscopically (Figures 1 and 2). The bleeding was kept under control; thereafter, appendectomy stump was closed, but colostomy area was not considered available for primary suturing because of iatrogenic injury caused during colonoscopy procedure. The surgery was completed after partial sigmoid colon resection and anastomosis. Bleeding did not recur in the postoperative period, but the patient developed respiratory failure due to Acute respiratory distress syndrome (ARDS) during follow-up and he was monitored on the postoperative second day as attached to the mechanical ventilator. The patient died due to ARDS on the postoperative day 8.

Discussion

Dieulafoy's lesions are among the fatal causes of lower GIS hemorrhage with a lesion-associated mortality rate reported

up to 80%.¹ As was in the present case, it is more prevalent in males.¹ The pathogenesis is unclear; in addition to the studies reporting that the lesions are usually encountered in the patients with multiple comorbidities and are associated with non-steroid anti-inflammatory (NSAID), aspirin and warfarin use¹, there are also studies reporting that the lesions are not associated with alcohol, smoking or NSAIDs but may be associated with constipation.² Abnormally wide, tortuous submucosal arterioles with thick wall, which protrude under the thinned mucosa without ulceration, lead to serious bleeding.³ For the lesions in the colorectal region, it has been dwelled on the theorem that solid content leads to laceration and bleeding due to luminal stercoral effect causing necrosis on the dilated submucosal lesion.¹ Even though there is no consensus, mucosal injury and ischemia due to aging and cardiovascular diseases are considered important in the pathogenesis.¹ In the literature, there are cases reported to have hypovolemic shock as was in the present case.^{4,5} It can be overlooked during improper evaluations performed under emergent situations or may be misdiagnosed as hemorrhoidal disease.⁵ Endoscopic and



Figure 1. Close view of Dieulafoy's lesion
Dieulafoy's lesion in the proximal anal canal, which has been detected by endoscopy during intraoperative period and underwent clipping (black arrow)



Figure 2. Clipped Dieulafoy's lesion in the proximal anal canal (black arrow)

angiographic methods can be used for diagnosis as was in the present case. The diagnosis is made detecting the focus of bleeding and the lesion.^{3,4,6} Endoscopically, the diagnosis can be made in the event of; 1) detecting a lesion accompanied by <3 mm mucosal defect with micropulsatile or gushingly active bleeding surrounded by normal mucosa, 2) observing a vessel surrounded by normal mucosa or protruded from a small mucosal defect either with or without bleeding, and 3) detecting a solid clot on the small mucosal defect or on the mucosa that appears to be normal.¹ Treatment includes conservative methods such as selective arterial embolization, sclerotherapy, epinephrine injection, thermocoagulation and clipping.^{1,2,3,4,5,6,7} Surgical suturing or segmental resection can be performed in the situations where the bleeding cannot be brought under control radiologically or endoscopically.^{1,2,3} Larger resections may be necessary for the lesions in the anal canal, rectum or esophagus. There are studies recommending local resection as the surgical procedure because of solid structure of the stool in the anal canal.³ In the present case, the focus could have been hardly detected by intraoperative colonoscopy and bleeding was brought under control by endoscopic clipping. Despite a large resection such as subtotal colectomy that can be performed in nonlocalized GIS hemorrhage, lesions in the anal canal would have not been included in the specimen as was in the present study. Hence, the risk of the lesion's not being resected or continuation of bleeding is in question despite large resection.

Conclusion

Although Dieulafoy's lesions are rarely encountered in the anal canal, they should be kept in mind as they cause life-threatening bleeding. It should be also kept in mind that Dieulafoy's lesions in the anal canal may be endoscopically overlooked as they are localized in a region that could be rapidly passed through during endoscopy procedure, anal canal is not reevaluated by retroflexion before the procedure is completed, a clear view cannot be provided because of continuing bleeding, the procedure is performed suboptimally under emergent situations, or they are uncommon lesions. Perioperative colonoscopy can be beneficial in the event the focus of bleeding could not be detected preoperatively. Systemic complications and

mortality can be encountered due to massive transfusion even the bleeding is brought under control.

Ethics

Informed Consent: It was taken.

Peer-review: External and Internal peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: Mustafa Berkeşoğlu, Aydemir Ölmez, Mehmet Kasım Aydın, Mehmet Özgür Türkmenoğlu, Tahsin Çolak, Concept: Mustafa Berkeşoğlu, Aydemir Ölmez, Mehmet Kasım Aydın, Mehmet Özgür Türkmenoğlu, Tahsin Çolak, Design: Mustafa Berkeşoğlu, Aydemir Ölmez, Mehmet Kasım Aydın, Mehmet Özgür Türkmenoğlu, Tahsin Çolak,

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