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Dieulafoy's lesion of the anal canal: Report of a case

Anal kanalda dieulafoy lezyonu: Olgu sunumu

Özgür FIRAT[1], Yüksel KARAKÖSE[1], Cemil ÇALIŞKAN[1], Özer MAKAY[1], Ömer ÖZÜTEMİZ[2], Mustafa Ali KORKUT[1]

Departments of [1]General Surgery and [2]Gastroenterology, Ege University, Faculty of Medicine, İzmir

Background/aims: *Dieulafoy's lesion of the anal canal is a very rare clinical case. Although it was first described in the stomach, there has been an increasing frequency, especially in the last decade, of reports of the lesion in the colorectal region.* ***Methods:*** *Herein, we report one case presenting with massive hematochezia requiring multiple blood transfusions due to a Dieulafoy's lesion in the anal canal. To our knowledge, this is the fourth case in Medline. Surgical oversewing was attempted twice but rebleeding occurred, and local excision through the anal canal was performed.* ***Results:*** *The patient was treated successfully with mucosectomy including the lesion.* ***Conclusions:*** *Sclerotherapy, alcohol and epinephrine injection, thermocoagulation and selective arterial embolization are the options of therapeutic endoscopy and interventional radiology. As for surgical management, oversewing is an alternative technique. However, in our opinion, because of the recurrent and life-threatening manner of this arterial bleeding pattern, local excision, if possible, is the most reliable management of the disease.*

Key words: Dieulafoy's lesion, gastrointestinal hemorrhage, anal canal

Amaç: *Dieulafoy lezyonunun anal kanalda görülmesi oldukça nadirdir. İlk kez mide de görülmüş olmasına karşın, özellikle son yıllarda bu lezyonların kolorektal bölgede görüldüğünü bildiren yayınların sayısı giderek artmaktadır.* ***Yöntem:*** *Bu bildiride, anal kanalda bulunan Dieulafoy lezyonu nedeniyle masif hematokezya ile başvuran ve multipl kan transfüzyonlarına ihtiyaç duyan bir hasta sunulmaktadır ve bilgimize göre bu hasta Medline'da bildirilen dördüncü olgudur. Lezyon iki kez sutüre edilmiş ancak kanamanın tekrarlaması üzerine anal kanaldan girişim ile lokal eksizyon uygulanmıştır.* ***Bulgular:*** *Hasta, lezyonu içerecek şekilde uygulanan mukozektomi ile başarılı bir şekilde tedavi edilmiştir.* ***Sonuç:*** *Terapötik endoskopi ve girişimsel radyoloji bizlere skleroterapi, alkol ve epinefrin enjeksiyonu, termokoagülasyon ve selektif arteryel embolizasyon gibi seçenekler sunmaktadır. Cerrahi tedavide ise lezyonun sutüre edilmesi alternatif bir yöntemdir. Ancak bizim görüşümüz; arteryel özellikte, hayatı tehdit eden ve tekrar kanama olasılığı yüksek olan bu lezyonlarda mümkün olduğu takdirde lokal eksizyon en güvenilir tedavi metodu olacaktır.*

Anahtar kelimeler: Dieulafoy lezyonu, gastrointestinal kanama, anal kanal

INTRODUCTION

Dieulafoy's lesion is one of the rare causes of massive lower gastrointestinal bleeding. The lesion typically consists of a submucosal arteriole bleeding through a very small mucosal defect usually on the lesser curve of the stomach; similar lesions have also been described in the esophagus, small intestine, colon, rectum and anal canal (1-3). In this paper, we report a patient with massive hematochezia and our surgical management.

CASE REPORT

A 67-year-old female patient with a past medical history significant for essential hypertension, chronic renal failure and tuberculosis was admitted to our hospital with massive hematochezia causing hemodynamic instability. Following the resuscitation, rectal digital examination revealed bright red blood with clots. Anoscopy revealed a mucosal lesion covered with clots 5 cm above the anal verge corresponding to 5 o'clock in prone position, and it was seen that the bleeding had stopped spontaneously. Colonoscopy was performed and no other lesion was found. The patient had no active bleeding during a follow-up of 4 days. On day 5, rebleeding occurred and she again developed symptoms of hemorrhagic shock. Repeated anoscopy revealed the round mucosal defect with a 5 mm diameter corresponding to 5 o'clock position, 3 cm proximal to the pectineal line including a

Address for correspondence: Özgür FIRAT
Department of General Surgery, Ege University, Faculty of Medicine, 35100 Bornova, İzmir, Turkey
Phone: +90 232 390 40 20 • Fax: +90 232 339 88 38
E-mail: ozgur.firat@ege.edu.tr

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brisk, spurting arteriole. Dieulafoy's lesion was considered and initial control of the bleeding was obtained with a gauze pack. After resuscitation, hemodynamic stability was achieved and the lesion was oversewn with 2.0 polyglactin (Vicryl®) for hemostasis. The patient did well for 8 days and had normal stools. After 8 days, active bright red rebleeding was encountered again. This time the lesion was oversewn with 2.0 silk and the patient had no hematochezia for 7 days and had normal brown stool several times. After 7 days, rebleeding occurred with the same pattern. Local excision was performed with electrocautery with 2 cm around the lesion, and the locus was sutured with 1.0 chromic catgut for hemostasis. There was no recurrence of bleeding after 6 months of follow-up. The report of the histopathological examination was "Dieulafoy's lesion with a morphology of wide caliber, thick-walled artery with a superficial mucosal erosion" (Figure 1).

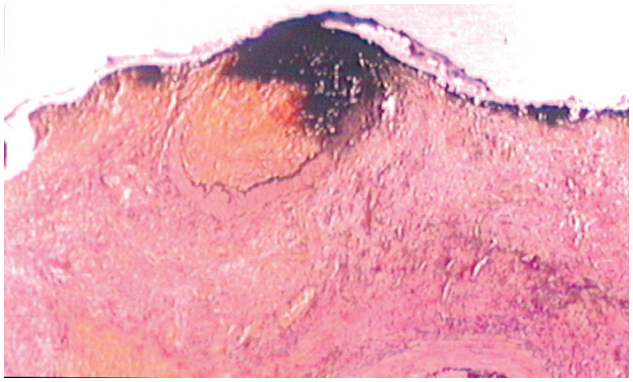

Figure 1. Histopathological view of the characteristic non-reducing arteriole protruding from a tiny mucosal defect (Elastic – van Gieson X 40)

DISCUSSION

After the first description by two French surgeons, Gallard and Dieulafoy, in the late 19th century (4, 5), Dieulafoy's lesion took its place among the most fatal causes of lower gastrointestinal bleeding. Although it was first described as a gastric lesion existing along the lesser curvature, similar lesions were found in the esophagus, duodenum, small intestine, colon, rectum1 and finally the anal canal (2, 3).

Dieulafoy's lesion has a characteristic clinical presentation. Patients develop sudden, massive, painless and recurrent gastrointestinal bleeding and admit to hospital in the case of hemodynamic instability usually requiring transfusions. The lesion has no tendency for heredity or any age group, but has a male predominance (2).

The pathogenesis of the lesion is not clear and in many authors' opinion, it is congenital in origin. It typically consists of an abnormally large, thick-walled and tortuous submucosal artery which protrudes from a tiny mucosal defect without surrounding ulceration. The characteristic non-reducing caliber of the arteriole underlying the mucosa and the absence of a true aneurysm are the histologic evidences of Dieulafoy's lesion after the microscopic examination (1).

Advances in endoscopic methods and interventional radiology have resulted in diagnostic methods having an important role in treatment. The diagnosis usually becomes certain after visualization of active arterial bleeding, and after adequate visualization, injection therapy or thermocoagulation may be performed (6). Endoscopic clipping is another method which has recently been performed successfully in these kinds of lesions (7). In cases of massive bleeding, colonoscopy has a high failure rate and angiography or labelled red cell scintigraphy may be more helpful in localizing the site of bleeding. Selective arterial embolization is also a therapeutic option after mesenteric angiography (8).

Despite the great advances in therapeutic endoscopy and radiology, surgical management still retains its importance. As for the lesions in the stomach, small intestine and colon in which endoscopic therapy failed, segmental resection should be considered; however, the lesions in the rectum or anal canal will require a major procedure (8). Therefore, surgical oversewing is appropriate for these accessible lesions. In cases where oversewing fails, a local excision including the bleeding vessel may have to be applied.

In our case, the patient was treated with local excision after two unsuccessful attempts to oversew. In our opinion, particularly in the case of lesions in the anal canal, the mechanical damage of the mucosa will be greater because of the hard nature of the stool and recurrence will be more likely; therefore, local excision is the most reliable management of Dieulafoy's lesion in this region.

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