

ORIGINAL ARTICLE

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Dieulafoy's lesion as a rare cause of gastrointestinal bleeding: a single-center experience

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**Abstract**

To define the clinical characteristics of Dieulafoy's lesion (DL), determine the localization features, and investigate endoscopic treatments and their results. This retrospective study included 20 patients who presented to the Department of Gastroenterology due to gastrointestinal (GI) bleeding between January 2015 and December 2019 and were diagnosed with DL. Age, gender, comorbidities, medications used, presentation findings, hemoglobin and hematocrit values at the time of presentation, number and type of transfusions if applied, length of hospitalization in days, mortality/survival, and endoscopic procedure parameters, including the number of procedures, localization of DL, type of bleeding stigmata, and the applied technique were recorded for all patients. Ten (50%) of the patients were women, and the mean age of all patients were 66.8 ± 17.8 years. One or more comorbidities were present in 90% of the patients. The use of acetyl salicylic acid (ASA) was present in five (25%) patients, ASA+ clopidogrel in five (25%), warfarin in two (10%), and non-steroidal anti-inflammatory drugs in two (10%). The most common complaint was observed to be hematemesis and/or melena in 16 patients (80%). DL was most frequently seen in the stomach ($n=10$; 50%) where it was most commonly located in the proximal corpus ($n=6$; 60%). The most prevalent bleeding stigma was active bleeding, which was seen in nine (45%) patients during endoscopy. Eight (40%) patients received endoscopic treatment combined with a mechanical method, and nine (45%) patients only received a mechanical treatment (endoscopic band ligation or hemoclip). The median number of hospitalizations was five days (2-22). Transfusion was required by 13 patients (65%). The mortality rate was determined as 5%. DL should be considered especially in older patients with chronic diseases who present with recurrent GI bleeding. An endoscopic examination is the first method to be applied in the diagnosis and treatment of DL. Mechanical methods should be prioritized in endoscopic treatment.

Keywords: Gastrointestinal system, bleeding, Dieulafoy's lesion, endoscopic treatment**Introduction**

Although advances in the treatment of causes leading to acute gastrointestinal (GI) bleeding have significantly reduced the number of bleeding cases, it is still a major cause of morbidity and mortality. The annual incidence of GI bleeding ranges from 50 to 150 per 100,000 [1], and related mortality is around 5-10% [2]. The most common cause of GI bleeding is peptic ulcer, and one of the rare causes is Dieulafoy's lesion (DL). DL is a submucosal arterial formation that protrudes from a small mucosal defect anywhere in the GI tract and leads to massive GI bleeding, especially in the elderly [3]. Also known as a 'caliber persistent artery', DL has an incidence of 0.3% to 6.7% [1,4,5].

DL was first described as an aneurysm in the stomach by Gallard in 1884, but the actual clinician that named this lesion and demonstrated its clinical characteristics was Paul Georges Dieulafoy [6]. The majority of DLs occur in the proximal stomach, but they can also be localized in the esophagus, small intestine, and large intestine [7,8].

The first step in the diagnosis of DL is GI endoscopy. However, even experienced endoscopists can overlook the diagnosis of DL. In cases where no focus is detected on endoscopy, angiography and/or endoscopic ultrasonography can be used [5]. The treatment options include band ligation, thermoregulation, bipolar electrocoagulation, sclerotherapy and endoscopic hemoclip. The success rate with endoscopic treatment methods is 75-100% [9]. In cases where treatment cannot be successfully provided by endoscopic techniques, surgical treatment options should be considered [10,11].

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The purpose of this retrospective study was to define the clinical characteristics of DL, determine the localization of this lesions, and investigate the endoscopic treatments applied and their results.

Materials and Methods

This retrospective single-center study included patients that underwent endoscopy due to GI bleeding and were diagnosed with DL at the Department of Gastroenterology between January 2015 and December 2019. Patients were screened by searching the term 'Dieulafoy' lesion' in the Endocam system, and their endoscopy reports were obtained. Using the esophagogastroduodenoscopy and ileocolonoscopy reports of the patients, the data concerning the location of DL, bleeding stigma type, localization in the stomach, endoscopic treatment method applied, number of hemoclips used, and number of endoscopic procedures required were recorded. In addition, age, gender, comorbidities, medications used, presentation findings, hemoglobin, hematocrit, platelet and international normalized ratio values, number and type of transfusions if applied, length of hospital stay in days, and mortality/survival status were obtained from the hospital electronic database and recorded in the case report forms.

Endoscopic examinations were performed using Olympus GIF-HQ190 esophagogastroduodenoscopy and Olympus CFH-170L-CFQ-150 Lileocolonoscopy devices by a gastroenterology specialist or a sub-branch specialist under the supervision of a gastroenterology specialist, accompanied by an endoscopy nurse. DL, usually diagnosed by endoscopic examination, is defined as a pigmented protuberance from the vessel stump with minimal surrounding erosion and no ulceration but sometimes caused actively bleeding or oozing [12] (Figure 1). The treatments were classified as endoscopic sclerotherapy, endoscopic band ligation (EBL), and endohemoclip application. Endoscopic sclerotherapy was performed using adrenaline diluted with saline to 1/10,000 and/or 1% Aethoxysklerol, EBL using a Speedband Superview Super 7™ Boston multiple band ligament set, and the endoscopic hemoclip was Quick Clip 2™ Olympus hemoclip. EBL and endoscopic hemoclip application were accepted as mechanical methods. Endoscopic treatment combined with a mechanical method was defined as adrenaline + hemoclip, adrenaline + Aethoxysklerol + hemoclip, adrenaline + Aethoxysklerol + EBL, or adrenaline + EBL.

For the statistical analyses, SPSS Statistics ver. 22.0 (SPSS Inc. Chicago, IL, USA) was used. The conformance of variables to normal distribution was examined by visual (histogram) and analytical methods (Shapiro-Wilk test). Numerical data were expressed as median, standard deviation, maximum and minimum values and categorical data were given as descriptive statistics, such as rate and percentages. Student's t-test was conducted to determine whether there was a statistical difference in the mean values of the numerical variables of age and hemoglobin according to gender. Values with a p value below 0.05 were considered statistically significant.

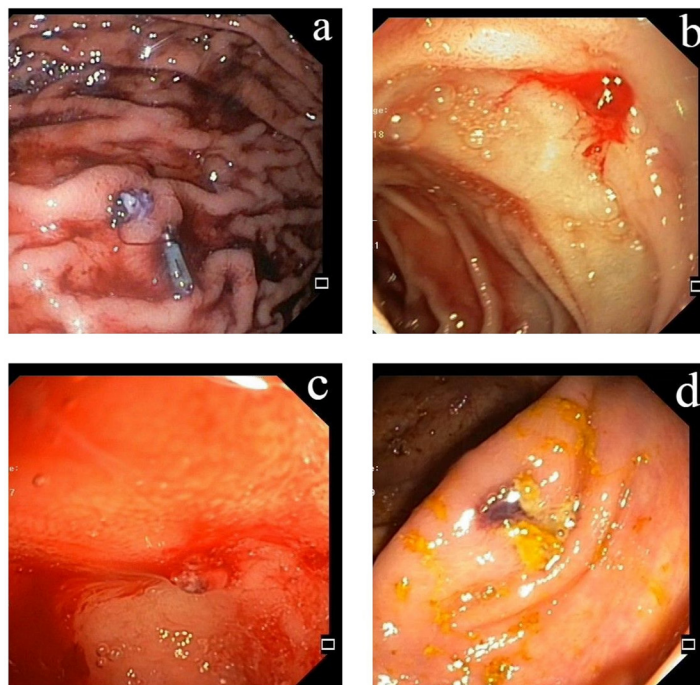


Figure 1. Location of Dieulafoy's lesions in gastrointestinal system. a-stomach, b- duodenum, c-ileum, d-colon (Achieved from the archive of the Ege University Gastroenterology Department Endoscopy Unit)

Results

Of the 20 patients diagnosed with DL, 10 (50%) were female and 10 (50%) were male, with a mean age of 66.8 ± 17.8 years. The mean age of women was 68.4 ± 23.4 years, and that of men was 65.2 ± 10.8 years. One or more comorbidities were present in 90% of the patients. The presentation findings were hematemesis in eight patients (40%), hematochezia in four (20%), hematemesis and melena in three (15%), melena alone in three (15%), and hematemesis and hematochezia in two (10%). At the time of presentation, the median values of hemoglobin and hematocrit were measured as 8.10g/dL and 24.8%, respectively. Table 1 presents the demographic, clinical and laboratory features of the patients.

When the distribution of DLs in the GI system was examined, it was most frequently seen in the stomach ($n = 10$; 50%). The localization of DL was determined as the second segment of the duodenum in three (20%) patients, colon in three (15%) patients, bulb in two (10%) patients, terminal ileum in one (5%) patient, and esophagus in one (5%) patient. Within the stomach, the most common localization of DL was the proximal corpus ($n = 6$; 60%), followed by the fundus ($n = 2$; 20%), antrum ($n = 1$; 10%), and cardia ($n = 1$; 10%). During endoscopy, active bleeding was present in nine (45%) patients, protuberance from the vessel stump in eight (40%), and vessels with clots in three (15%). The endoscopic data of the patients are shown in Table 2.

When the endoscopic treatments were examined, combination therapy with a mechanical method was applied in eight (40%) patients, hemoclip alone in seven (35%), EBL alone in two (10%), sclerotherapy with 1/10,000 adrenaline alone in two (10%), and

sclerotherapy with 1/10,000 adrenaline+1% Aethoxysklerol in two (10%). The number of median hemoclip was three [1-5] in patients who underwent this procedure. There was no statistically significant difference in the comparison of the EBL and hemoclip method. The median number of hospitalization days was five [2-22]. Thirteen (65%) patients required a transfusion, for which only erythrocyte suspension (ES) was used in 11 (55%) patients, ES + apheresis platelet suspension in one patient (5%), and ES + fresh frozen plasma replacement in another patient (5%). For those requiring a transfusion, the median (min-max) unit of ES given was three [1-8]. The mortality rate among the patients hospitalized due to DL was 5%. Table 3 presents the treatment and clinical follow-up data of the patients.

Table 1. Demographic, clinical and laboratory characteristics of the patients

	n (%)
Age (mean± SD)	66.80±17.86
Gender (n=20)	
Female	10 (50)
Male	10 (50)
Comorbidities	
Coronary artery disease	10 (50)
Hypertension	9 (45)
Diabetes mellitus	5 (25)
Medications used (n=20)	
Acetylsalicylic acid (ASA)	5 (25)
ASA+Clopidogrel	5 (25)
Warfarin	2 (10)
NSAID	2 (10)
Unknown	6 (30)
Presentation complaint (n=20)	
Hematemesis	8 (40)
Hematochezia	4 (20)
Melena	3 (15)
Hematemesis and melena	3 (15)
Hematemesis and hematochezia	2 (10)
Laboratory parameters	Median (min-max)
Hemoglobin (g/dl)	8.1 (3.6-12.9)
Hematocrit(%)	24.75 (11.7-39.2)
Platelet (10 ³ /μL)	241.5 (93-371)
INR	1 (0.9-3.1)
INR: International normalized ratio, NSAID: Non-steroidal anti-inflammatory drug.	

Table 2. Endoscopic data of the patients

Parameters	n (%)
Location of lesion (n=20)	
Stomach	10 (50)
Duodenum second segment	3 (15)
Colon	3 (15)
Bulbus	2 (10)
Esophagus	1 (5)
Terminal ileum	1 (5)
Localization of lesions within the stomach (n=10)	
Proximal corpus	
Greater curvature	3 (30)
Lesser curvature	3 (30)
Cardia	1 (10)
Fundus	2 (20)
Antrum	1 (10)
Endoscopic findings (n=20)	
Active bleeding	9 (45)
Pigmented protuberance from the vessel stump	8 (40)
Vessels with clots	3 (15)

Table 3. Treatment and clinical follow-up data of the patients

Treatments applied (n=20)	n(%)
Combination with a mechanical method	8 (40)
Adrenalin+hemoclip	3 (15)
Adrenalin+Aethoxysklerol+hemoclip	3 (15)
Adrenalin+Aethoxysklerol+EBL	1 (5)
Adrenalin+EBL	1 (5)
Hemoclip	7 (35)
EBL	2 (10)
Adrenalin+Aethoxysklerol	2 (10)
Adrenalin	1 (5)
Outcome (n=20)	
Mortality	1 (5)
Surviving	19 (95)
Transfusion requirement (n=20)	
Present	13 (65)
ES	11 (55)
ES+platelet suspension	1 (5)
ES+FFP	1 (5)
Absent	7 (35)
	Median (min-max)
Number of ES units	3 (1-8)
Number of hemoclip used	3 (1-5)
Number of endoscopic procedures applied	1 (1-2)
Number of hospitalized days	5 (2-22)
EBL: endoscopic band ligation, ES: erythrocyte suspension, FFP: fresh frozen plasma	

Discussion

DL is a rare emergency gastroenterological condition that leads to massive bleeding, sometimes overlooked on endoscopy. It is more common in older men and often accompanied by comorbidities.

Mortality in DL has been reduced with the widespread use of endoscopy and is around 5-10% in the literature [2,13]. In studies conducted in Turkey, DL-related mortality has been reported to range from 3.7 to 38.9% [14,15]. In a study conducted in our center in 1998, the mortality rate was determined to be 8% in patients with DL localized in the stomach [16]. In our study, we calculated the mortality rate as 5%. Only one of our patients died, and the cause of mortality in this patient was not bleeding; it was hospital-acquired pneumonia. The reasons for the lower mortality rate in our study compared to the literature may be that our hospital is a tertiary reference center, endoscopy is performed early in cases of active GI bleeding, and an advanced endoscopy specialist and academician is on call when needed for the endoscopy procedure.

In the literature, it has been reported that DL is more common in men (55.6%-66.6%) [3,14-16]. In our study, this rate, unlike the literature, was equal in males and females, which was probably due to the limited number of our patients. Advanced age has been determined as a risk factor for DL [17]. In studies conducted, the mean age has been reported to be 73-79 years [3,14]. In our study, the mean age of the patients was calculated as 66.8 years, similar to the literature. The possible reasons for this finding may be the elongation of the submucosal artery, development of mucosal atrophy in the stomach, and the increased use of non-steroidal anti-inflammatory drugs and ASA that can cause ischemic damage in the stomach, with age [12].

Comorbidities have been reported in approximately 70.8% of patients with DL, most commonly in the form of diabetes mellitus, hypertension, chronic kidney failure, and coronary artery disease. In addition, the most frequently used drugs among these patients have been determined as NSAIDs, ASA, and warfarin [1,9,15]. Similarly, in our study, 90% of patients had one or more comorbidities, with the most common being coronary artery disease, hypertension, and diabetes mellitus. The information on the medications used by the patients was obtained for 70% of the cases, and the types of drugs used were, in order of frequency, ASA, ASA + clopidogrel, warfarin, and NSAID, which is in agreement with the literature.

Most of the DL cases present with melena and/or hematemesis. In many studies, melena (75-33%) has been evaluated as the most common reason for presentation to hospital in this patient group [1,9,14,15]. In one study, hematemesis (60%) was the most common presentation finding [3]. In our study, consistent with the literature, the most common patient presentation was hematemesis and/or melena (70%), which is probably because in the majority of our cases, DLs originated from the upper GI.

In previous studies, the average requirement of transfusion has been reported as 2-3 units of ES [3,15], which is similar to the value we obtained in our study (2.5 units of ES). The possible reason for this transfusion requirement is massive bleeding.

The most common location of DL in the GI system is the stomach [18]. Within the stomach, 46.6-66.6% of DLs are reported to

be localized in the corpus and 59.2% in the fundus [3,14,15]. However, approximately one-third of the lesions are extragastric, which are most often located in the duodenum, followed by the colon [1,7,8]. In addition, there are cases in the literature defined in the bronchi other than the GI system [13]. In our study, the most common location of DLs in the GI system was the stomach (50%). Within the stomach, DLs were most frequently seen in the proximal corpus at 60%, and as extragastric locations, the duodenum and colon were most observed. One of the possible reasons for the frequent occurrence of DLs in the stomach may be that vibrations in a large submucosal vein in this area lead to the disruption of the upper epithelium, while another reason may be considered as gastric abrasion and wear increasing thrombosis within the artery and subsequently leading to necrosis [18,19]. In the colon and rectum, solid content can lead to mucosal stercoral ulcers, resulting in rupture and hemorrhage [1].

There is no consensus on the treatment of DL. The location of the lesion depends on the presence of active bleeding and the technical competence and experience of the specialist. With the development of endoscopic methods, the need for surgery in DL has significantly decreased. In endoscopic treatment, there are thermal (electrocoagulation and argon plasma coagulation), local injection (epinephrine and Aethoxysklerol) and mechanical (EBL and hemoclip) methods [20]. Using endoscopic treatment methods, the success rate has been shown to vary between 75 and 100% [21,22]. In the literature, it has been reported that the rates of recurrent bleeding and hemostasis failure were higher in patients who underwent epinephrine injection or heater probe coagulation alone compared to those also receiving mechanical treatments. This has brought endoscopic mechanical methods to the fore in DL treatment [10,23,24]. The success rates of DL with hemoclip treatment have been observed to range from 94 to 95.2% [25-27]. In our study, similar to the literature, there is no significant difference between EBL and hemoclip in terms of efficacy or safety [9,21]. Today, most endoscopists prefer the combined treatment of hemoclip and injections as the primary treatment [21,28,29]. Similarly, in our study, the most preferred treatment methods were mechanical and/or combination therapy, and our success rate was 95%.

In the literature, the median duration of hospitalization has been reported as 10.5 (1-28) days, and the mean duration as even days (3,14). In our study, the median and mean lengths of hospital stay were five (2-22) days and 7.6 days, respectively, which is similar to the literature.

Since our center is more experienced in mechanical methods, thermal coagulation is not used sufficiently. One of the limitations of our study is that we do not have data on thermal coagulation method, so mechanical methods cannot be compared with thermal coagulation methods. Other limitations of our study can be considered as retrospective design and small sample size.

DL is a rare cause of GI bleeding, which had high morbidity and mortality before the widespread use of endoscopy. The pathogenesis of DL has not yet been fully elucidated. DL should be considered in patients with chronic diseases presenting with GI bleeding for whom an etiological cause cannot be identified on the first endoscopy, as well as in those presenting with recurrent bleeding. An endoscopic examination is the first method to be

preferred in the diagnosis and treatment of these patients since it is effective, inexpensive, and reliable. Mechanical methods should be prioritized in endoscopic treatment. Further studies are needed to raise clinicians' awareness of these rare lesions.

Conflict of interests

The authors declare that they have no competing interests.

Financial Disclosure

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Ethical approval

The local ethics committee approved the study. 20-5T/7.

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