

Uremic Bleeding in 2 Patients with Dieulafoy's Lesion: Case Report and Literature Review

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Abstract

Dieulafoy's lesions are rare vascular malformations of the gastrointestinal tract. A Dieulafoy's lesion is an aberrant vessel that does not reduce in caliber when it extends from the submucosa to the mucosa. Damage to this artery can cause severe and intermittent arterial bleeding from small vascular stumps that are difficult to visualize. Furthermore, these catastrophic bleeding episodes frequently result in hemodynamic instability and the need for transfusion of multiple blood products. Recently, uremic syndrome has been identified as a risk factor for gastric mucosal lesions. We present two clinical cases of acute digestive bleeding due to Dielafoy lesion with chronic kidney disease as the main cause, where two different therapies were performed endoscopically. We concluded with the results of our patients that the best therapy was the application of the hemostatic hemoclip on the injury vs the injection with adrenaline on the wound site. Uremia is identified as a risk factor for upper gastrointestinal bleeding in patients with pre-existing Dieulafoy's lesion, as well as a higher incidence of new bleeding.

Keywords

Dieulafoy's Lesion, Gastrointestinal Bleeding, Endoscopy, Endoscopy, Hemostatic Clips, Kidney Disease, Adrenaline, Thermal Coagulation, Dieulafoy

1. Introduction

Dieulafoy's lesion, first reported by Gallard in 1984 and then described in detail by Georges Dieulafoy in 1898, is a rare vascular malformation characterized by an abnormally dilated submucosal artery (1 to 3 mm). Since its description first, it has rarely been reported in the literature. These lesions can be found in any location of the gastrointestinal tract; however, they occur more frequently in the stomach and rarely in the colon [1].

The incidence of acute gastrointestinal bleeding ranges from 50 - 150 per 100,000 of the population each year [2].

The two largest retrospective series, both from North America, found Dieulafoy's lesion as the source of hemorrhage in 1.9% and 1.2% of all endoscopies performed for acute GI bleeding [3].

Upper gastrointestinal bleeding is approximately four times more frequent than lower gastrointestinal bleeding. Almost 80% of bleeding cases are caused by ulcerative disease, varicose disease or telangiectasis, with occult hemorrhage occurring in less than 5% of cases [1]. Dieulafoy's lesion accounts for 6.5% of the causes of upper gastrointestinal hemorrhage [4]. The objective of this work is to search for a correlation between uremic bleeding with Dieulafoy's lesion as well as to reveal the best therapy for our patients.

2. Clinical Cases

2.1. Patient 1

An 87-year-old patient was admitted to the emergency room due to neurological deterioration, accompanied by dyspnea on minor exertion as well as fever; recently diagnosed with chronic kidney injury without replacement therapy due to refusal of treatment by the same family members and the patient; with a history of having had an episode of non-variceal upper digestive tract bleeding 4 years ago without a determined etiology, recently diagnosed Type 2 Diabetes Mellitus under treatment with metformin, systemic arterial hypertension of 40 years of evolution under treatment with losartan, hydrochlorothiazide and nifedipine, acute myocardial infarction 9 years ago with catheterization and stent placement. Laboratories upon admission: Na: 131, Ca: 7, Mg: 1, P: 3.9, Albumin: 1.8, Eri: 3.2, Hb: 8.7, Hct: 28.1, Plaq: 371, Leu: 15.16, Neut: 11.97, Crea: 2.7, Urea: 132.23, Ttp: 28.8, Tp: 12.4, Inr: 1.08, Glu: 99. During her stay she was diagnosed with sepsis of urinary focus with with positive culture for Pseudomonas aeruginosa, treated with multiple antibiotic regimens with significant improvement in his septic process.

1 week later he started with melena accompanied by hematemesis on multiple occasions, new laboratory studies were taken reporting Hb: 6, Hct: 21, Plaq: 243, Leu: 17.2, Neu: 15.84, Crea: 3.5, Urea: 210. Due to the gross decrease in hemoglobin, 2 red blood cells were transfused and when the patient was stabilized, endoscopy was performed where a Dieulafoy's lesion was reported (**Figure 1**). Hemostasis was performed with adrenaline and hemoclip placement, successfully stopping the bleeding. Without evidence of rebleeding, he remained under surveillance with laboratory studies with an increase in hemoglobin after 3 days of 9 g, which is why it is concluded that he is progressing correctly, scheduling endoscopy within 5 days for post-endoscopic evaluation. During the hospital stay,

the patient continues to deteriorate his real functional status with an increase in creatinine and urea, being refractory to medical treatment and contracting hospital-acquired pneumonia, for which antibiotic therapy based on carbapenems is given. The patient began with uremic encephalopathy and acute respiratory distress syndrome secondary to pneumonia, with a poor response to treatment. Due to hemodynamic instability with the use of vasoactive amines, control endoscopy was canceled and the patient died due to acute respiratory failure.



Figure 1. Endoscopy of the second portion of the duodenum on its posterior surface with visible vessel with adhered clot and bleeding in a layer with perilesional mucosa with edema and generalized hyperemia, with the presence of two hemoclips.

2.2. Patient 2

A 47-year-old male patient came to the emergency room with abdominal pain and melena of 1 week's duration, with asthenia, adynamia, and generalized weakness. The patient was known to have Type 1 diabetes mellitus for 25 years with pharmacological treatment with insulin glargine 30 IU every 24 hours, chronic kidney disease G5 for 9 months with replacement treatment with peritoneal dialysis having 3 exchanges a day with bags of 1.5% of 2000 cc with poor adherence to treatment, having residual urine of 750 mL per day. Uremic gastropathy for 15 years. Laboratories upon admission of ERI: 4.16, Hb: 11.9, Hct: 37.4, Plaq: 210, Leukos: 8.88%, Neutral: 87.6, Urea: 231, Crea: 16. The internal medicine floor is admitted to continue the approach. An endoscopy team is presented who wanted to improve the hemodynamic status to transfer it to endoscopy.

During the stay, bloody stools and hemodynamic instability continued, adding a deterioration in functional status with acute pulmonary edema due to fluid overload. New laboratory studies were taken, reporting ERI: 3.89, Hb: 8, Hct: 35.4, Plaq: 170, WBC: 13.88%, Neutral: 87.6, Urea: 288, Crea: 12. The nephrology



Figure 2. Endoscopy of the second portion of the duodenum on its anterior surface with visible active vessel and bleeding in a layer with perilesional mucosa with edema and generalized hyperemia.

team suggested a change in treatment from renal to hemodialysis, a Mahurkar catheter was placed and the patient proceeded to his first hemodialysis session for 2 hours. With partial improvement in functional status, another globular package is placed during the session. A later day, new laboratories were performed reporting ERI: 3.3, Hb: 7.5, Hct: 35.4, Plaq: 164, Leukos: 15.53%, Neutral: 87.6, Urea: 224, Crea: 8, so an endoscopic procedure was scheduled. During upper video endoscopy, abundant fresh superficial duodenum, bleeding in layers and a small visible vessel with active bleeding were observed on the anterior surface (Figure 2). It was decided to infiltrate adrenaline in the area with a 1:10,000 dilution of 2 cc and then thermal coagulation was performed with thermal biopsy forceps, observing the characteristics of the Dieulafoy's lesion with active bleeding. During the hospital stay, the hemoglobin continued to drop to 6.3 g, a red blood cell packet was passed and it was decided to perform a second endoscopy in which a hemoclip was placed in the bleeding area, causing the bleeding to subside. New laboratories were taken, reporting hemoglobin of 7.6 g, being constant during the next 4 days without a decrease in it. Endoscopy is scheduled within 1 week of follow-up.

Patient continues with exacerbation of his chronic kidney disease refractory to hemodialysis, begins with uremic encephalopathy, respiratory distress syndrome secondary to fluid overload. He went on hemodialysis without being able to tolerate the session. Patient on the internal medicine floor begins with severe respiratory failure, desaturating to 70%, so it is decided to protect via airway with orotracheal intubation, which the family refuses. Patient dies due to acute respiratory symptoms.

3. Discussion

The pathogenesis is not clear, but likely secondary to a congenital abnormality of the blood vessels or a thrombotic event of the arteriole causing necrosis [5]. In the case of our second patient, the first cause of the digestive tract hemorrhage was the increase in uremia secondary to not accepting replacement treatment in the first patient, and in patient 2 not correctly performing his dialysis outside the hospital.

Direct endoscopic visualization of the Dieulafoy's lesion is the gold standard method for diagnosis. Most patients are diagnosed after a single endoscopy; however, poor visualization of the GIT due to food particles or blood may require multiple endoscopies. The endoscopic visual diagnostic criteria that are necessary for a diagnosis of Dieulafoy's lesion is as following:

Normal mucosa around the small defected mucosal lesions which has active pulsatile bleeding smaller than 3 mm.

The visualization of a protruding vessel from a slight defect or normal mucosa The observation of a fresh clot attached to a defect of normal mucosa [4].

In patients with kidney disease, the risk of digestive bleeding increases due to platelet dysfunction associated with uremia, the intermittent use of heparin, and an increase in the incidence of gastric, duodenal, and esophageal ulcers, as well as angiodysplasias [6].

Main mechanisms have been found that are involved in the pathophysiology of uremic bleeding: Decreased hematocrit, intrinsic alterations in platelet functionalism, toxic effect of plasma factors (uremic toxins), Decreased generation of platelet thromboxane A2, increased synthesis of vascular prostacyclin, quantitative and/or qualitative alterations of the von Wille-brand factor, increased nitric oxide synthesis, hyperparathyroidism, deleterious effect of hemodialysis on platelet function [7].

Jutabha *et al.* [8], in a prospective series of 1000 cases of upper gastrointestinal bleeding, reported the following distribution of causes: peptic ulcer, 55%; eso-phagogastric varices, 14%; arteriovenous malformation (angiodysplasia), 6%; Mallory-Weiss tears, 5%; tumors and erosions, 4%; Dieulafoy's lesion, 1%; others, 11%.

Garcia *et al.* The relationship between kidney lesions and chronic kidney disease with anemia is discussed. 9658 patients with CKD were analyzed, of which 286 (2.9%) had anemia; 198 had a positive fecal occult blood test (47% men, 71.1 \pm 11.8 years). The endoscopic study revealed 255 lesions, with at least one lesion in 68.2%, the most prevalent being: adenomatous colorectal polyps (39.6%), acute lesions of the gastric mucosa (22.6%), neoplastic lesions (15.1%), angiodysplasias (14.4%), esophagitis (8.4%), inflammatory bowel disease (4.8%) and ischemic colitis (3.1%). Uremia and acetylsalicylic acid were identified as risk factors for acute lesions of the gastric mucosa [9].

There is no consensus on the treatment of a Dieulafoy's lesion. Treatment options are dependent on the mode of presentation, site of the lesion and available expertise (**Table 1**). Endoscopic treatment is safe and highly successful in terms of achieving initial hemostasis and hemostasis success rates reach 75% to 100%. Advances in endoscopic techniques have reduced mortality in patients with Dieulafoy's lesion—from 80% to 8%—and consequently, the need for surgical intervention has been reduced [10].

Table 1. Advantages and disadvantages of endoscopic techniques for Dieulafoy's lesion[10] (Jeon & Kim, 2015).

	Advantages	Disadvantages
Epinephrine injection	Simple and inexpensive	High risk of rebleeding
	Safe (avoids damage to the bowel wall)	
Argon plasma coagulation	Easy to use	Possibility of only superficial
	Safe	coagulation (inaccessible to larger, deeper vessels)
Clipping	Low risk of rebleeding	Requires more experience
		Difficulty in applying clips to specific sites
Band ligation	Easy to use	Poor visual field
	Accessible to difficult sites Low risk of rebleeding	Possibility of delayed bleeding or perforation

Dieulafoy's lesion has a vast number of differential diagnoses based on the patient's age, comorbidities, and overall health. Angiodysplasia is an important differential due to similar presenting symptoms. It can be differentiated on angiography, by the presence of arteriovenous shunting and vascular ectasia, and by histological examination of the lesion, which reveals the presence of abnormal submucosal vessels [4].

4. Conclusion

Patients who present with gastrointestinal bleeding should be treated with a multidisciplinary approach to reduce mortality and provide a more meaningful treatment. One must consider other systems aside from gastrointestinal causes as a rare condition for adults, appropriate diagnostic and treatment modalities should be done. Endoscopic therapy is still the first-line diagnostic and/or treatment option for Dieulafoy's lesion. Treated patients still need to be closely monitored for rebleeding episodes. We conclude with the results of our patients that the best therapy was the application of the hemostatic hemoclip to the injury. Uremia is identified as a risk factor for upper gastrointestinal bleeding in patients with pre-existing Dieulafoy's lesion, as well as an increased incidence of rebleeding.

Ethical Considerations

The authors declare that they have met all ethical responsibilities regarding data protection, right to privacy and informed consent.

Authorization from the institution's ethics committee is not necessary since at no time do they fail to comply or violate patient anonymity rules, nor is any experimental procedure performed that puts the patient's integrity at risk.

The authors declare that this article does not contain personal information that would allow the patient described to be identified, which makes the patient's informed consent unnecessary for the publication of the article.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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