Dieulafoy lesions, a rare cause of gastrointestinal hemorrhage, are caliber-persistent submucosal arteries without surrounding ulcers or mucosal lesions. Dieulafoy lesions rarely bleed profusely and may be missed during diagnostic endoscopy. Most of these lesions are located in the stomach; they have been reported only rarely in other parts of the gastrointestinal tract, including the small bowel. We report a case of gastrointestinal hemorrhage secondary to a Dieulafoy lesion in the afferent limb of a Billroth II gastric bypass.

Case Report

A 63-year-old African American woman with a history of hypertension, asthma, and osteoarthritis presented to the emergency room with a 3-day history of melena associated with upper abdominal discomfort. The patient denied experiencing nausea, vomiting, chest pain, dyspnea, fever, or chills. She did not have a history of smoking or substance or alcohol abuse. Her medications included diclofenac sodium/misoprostol (Arthrotec, Pfizer), lisinopril, and a fluticasone/salmeterol inhaler. Three years ago, she had presented to another institution with an episode of gastrointestinal bleeding, at which time she underwent a Billroth II gastrojejunostomy to control the bleeding.

On examination, the patient was not in acute distress. Her vital signs were stable, her oral mucosa was dry, and her conjunctiva appeared pale. Cardiovascular and pulmonary examinations were unremarkable. The patient had a large midline abdominal scar. Bowel sounds were present in all 4 quadrants, and her abdomen was soft. Mild epigastric tenderness was found without guarding or rigidity, but no organomegaly was detected. A rectal examination revealed a normal sphincter tone and black, loose stool that tested positive for occult blood. The remainder of the examination was unremarkable.

On admission, the patient’s hemoglobin level measured 10.3 g/dL, with a hematocrit of 34% and a mean corpuscular volume of 103.2 fl. Other laboratory tests revealed severe nonanion gap acidosis, with a bicarbonate level of 6 mEq/L, a blood urea nitrogen level of 85 mg/dL, and a creatinine level of 1.9 mg/dL. A computed tomography scan of the abdomen showed evidence of prior bowel surgery; however, there was no evidence of bowel obstruction, free gas or fluid, or discrete mass lesions.

Several hours after admission, the patient became hypotensive and was treated with fluid resuscitation. She was admitted to the intensive care unit and was started on a continuous infusion of pantoprazole and normal saline. Eight hours after admission, a repeat blood test found a hemoglobin level of 6.9 g/dL. The patient underwent a transfusion of 2 units of packed red blood cells (PRBC). An urgent esophagogastroduodenoscopy (EGD) revealed a Billroth II anatomy, with blood clots in the gastric pouch and the afferent loop of the patient’s prior gastrojejunostomy (Figure 1). Active bleeding and ulcers were not found. After the EGD, the patient experienced multiple episodes of melena and 1 episode of coffee-ground emesis. Her hemoglobin level dropped to 5.8 g/dL, and she received a transfusion of 4 more units of PRBC. A repeat EGD revealed blood clots and oozing from the afferent limb of the gastrojejunostomy, with no identifiable source of bleeding. We were unable to examine the entire length of the afferent limb. Although the patient was hemodynamically stable, she required additional transfusions due to her low hemoglobin level. At this point, a selective angiography was performed, but it did not reveal an active source of bleeding. The patient continued to have melena. By this time, she had received 11 units of PRBC, and she was taken to the operating room for intervention.
Prior to surgical intervention, an EGD with push enteroscopy was performed using a pediatric colonoscope. During this procedure, we were able to examine most of the afferent loop of the gastrojejunostomy, which showed persistent oozing of blood. The efferent loop appeared to be clean. After thorough irrigation, an oozing site that was suspicious for a Dieulafoy lesion was identified in the afferent loop of the jejunostomy (Figure 2). A total of 6 mL of epinephrine solution (1:10,000) was injected submucosally, and 3 endoclips were placed to achieve complete cessation of the bleeding. The area was tattooed with India ink to facilitate any future surgical interventions. Due to the successful endoscopic treatment, surgical intervention was not needed. The patient remained stable upon extubation. No additional episodes of bleeding were reported, and the patient’s hemoglobin and hematocrit levels stabilized. Her diet was advanced, and she was discharged home in good condition.

Discussion

Although Dieulafoy lesions are rare, they are an important cause of gastrointestinal bleeding. These lesions were first described by Gallard in 1884.1 When reporting on 3 patients with these lesions in 1898, the French surgeon Dieulafoy used the term “exulceratio simplex” because of the lesion’s small size and large artery with normal histology.2 A Dieulafoy lesion consists of a caliber-persistent arteriole that protrudes through a tiny mucosal defect, usually within 6 cm of the gastroesophageal junction on the lesser curve of the stomach.3,5 Similar lesions have also been described in the distal esophagus, duodenal bulb, colon, rectum, and jejunum.6-26 In approximately 4–9% of massive upper gastrointestinal bleeding episodes, a demonstrable cause cannot be found. A Dieulafoy lesion is thought to cause acute and chronic upper gastrointestinal bleeding in approximately 1–2% of these cases.27 Dieulafoy lesions are more common in males, occur at a median age of 54 years, and are not related to alcohol or nonsteroidal anti-inflammatory drug use.28 The etiology of Dieulafoy lesions is unknown, although they may be related to congenital or acquired vascular malformation.

It has been suggested that mechanical pressure from an abnormal vessel may progressively erode the thin mucosa overlying a pulsating artery, causing it to bleed.29,30 This artery, which protrudes through a solitary, tiny mucosal defect (2–5 mm) usually located in the upper part of the stomach, may spontaneously rupture and cause massive bleeding. These bleeding episodes can present as recurrent, massive hematemesis associated with melena (51%), hematemesis alone (28%), or melena alone (18%). On average, initial resuscitation requires a transfusion of up to 8 PRBC units.31

The majority of Dieulafoy lesions (75–95%) are located within 6 cm of the gastroesophageal junction. These lesions can easily be missed during endoscopy due to the intermittent nature of the bleeding. Approximately 49% of these lesions are identified during an initial endo-
scopic examination; however, 33% of lesions require more than 1 endoscopy for identification.\textsuperscript{31,32} If an EGD is performed within 2 hours of admission, there is a greater likelihood of identifying the lesion. The remaining cases are identified via angiography during active bleeding or via laparotomy. An endoscopic ultrasound (EUS) may also confirm the diagnosis by revealing a tortuous submucosal vessel near the mucosal defect. EUS can also help in the surveillance and assessment of endoscopic treatment.\textsuperscript{33}

Therapeutic endoscopy is the initial treatment of choice. Different endoscopic modalities have been used, including bipolar electrocoagulation, injection sclerotherapy, heater probe, laser photocoagulation, epinephrine injection, hemoclipping, and banding.\textsuperscript{29} The first endoscopic intervention is successful in 85% of patients, while 10% of patients need a repeat endoscopy, and 5% of patients may require surgical intervention.\textsuperscript{14} Angiography may have a therapeutic role, although this technique is usually reserved for patients who are not amenable to endoscopic therapy and who are poor surgical candidates.\textsuperscript{34} For duodenal and proximal jejunal lesions, surgical exploration can be performed via intraoperative endoscopy to avoid unnecessary bowel resection. This exploration enables endoscopic visualization with open surgical suture ligation.\textsuperscript{35} Following endoscopic management, 1 series found no recurrence of bleeding from Dieulafoy lesions over a mean follow-up period of 28 months; another series found no recurrence of bleeding over a mean follow-up period of 36 months.\textsuperscript{7,31}

\textbf{Conclusion}

To the best of our knowledge, this case study is the first report of a Dieulafoy lesion in the afferent limb of a gastrojejunostomy. This case study demonstrates the diagnostic problems inherent to these lesions (due to the intermittent nature of the bleeding); these problems include failure to detect a lesion during the first 2 endoscopies and a negative angiogram. This case study also illustrates successful endoscopic management that obviated the need for surgery.

Although Dieulafoy lesions are rare, they are an important and potentially fatal cause of gastrointestinal bleeding. Advanced endoscopic techniques, including capsule endoscopy, single- or double-balloon enteroscopy, and spiral enteroscopy, will increase the number of diagnosed cases and lead to better management strategies by providing access to longer segments of the small bowel. Careful examination of the mucosa during endoscopy remains the most important factor for detecting these lesions, providing appropriate therapeutic modalities, and decreasing morbidity and mortality.

\textbf{References}


Dieulafoy-Like Lesion Bleeding: In the Loop

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Eddi and associates present a patient with a Dieulafoy-like lesion in the afferent loop of a Billroth II (BII) gastric resection.1 This case is both novel and interesting, as it covers much clinical turf. Acute upper gastrointestinal hemorrhage has an incidence of 50–150/100,000 cases each year and a mortality rate of 7–10%.2,3 The etiology of the vast majority of upper gastrointestinal hemorrhages is peptic ulcer disease. In approximately 5% of cases, the cause of bleeding cannot be identified with a standard diagnostic method, such as endoscopy, in which case the condition is referred to as “obscure gastrointestinal bleeding.”4

When a Dieulafoy lesion (DL) is suspected, the following diagnostic—though relatively nonspecific—criteria may be used: “active arterial spurting or micropulsatile bleeding from a minute mucosal defect less than 3 mm or normal mucosa; visualization of a protruding vessel with or without bleeding within a small mucosal defect or normal mucosa; and the appearance of a fresh, adherent clot to a minute mucosal defect or normal mucosa.”4,5 For definitive pathologic diagnosis of a DL, it is necessary to obtain a surgical specimen, and herein lies the rub; most recent reports rely almost exclusively on endoscopic criteria, as surgery is not usually needed in these patients.

The case report by Eddi and coworkers examined a patient’s disease course, which started as locally obscure bleeding that can be defined as: “bleeding from the gastrointestinal tract that persists or recurs after a negative initial evaluation using endoscopy and radiologic imaging.”1,7 This definition of obscure bleeding is modified from the definition used by Pasha and colleagues, which applied to the entire gastrointestinal tract.1,7 In the case study by Eddi and colleagues, endoscopic diagnosis of the DL was established during push enteroscopy just prior to emergency surgery, which would have likely yielded a definitive pathologic diagnosis.1 Surgery in this case would not have been trivial, as the bleeding site was putative and an intervention likely would have involved either bowel resection in the antemesenteric duodenum or jejunum or the mesenteric side of the duodenum, as well as oversewing of the lesion with or without limited local excision. Performing these interventions in the previously surgerized abdomen of a blood-depleted older patient would have been a daunting prospect. It is therefore not surprising that the mortality associated with DLs exceeds the average mortality for gastrointestinal bleeding more than 2-fold.8

Issues with definitions have troubled this condition since its beginning. The lesions were first reported by Gallard in 1884; however, these lesions were named after Dieulafoy—whose own report came more than a decade later—because Gallard’s description was deemed lacking.9 Dieulafoy used the term “exulceratio simplex” as the key descriptor of the lesion not merely because of its small size and large artery, as suggested by Eddi and associates, but also because he thought that the lesion represented an initial stage of a gastric ulcer.1,9 In the modern endoscopic era, his description would be called into question, as the term “ulceration” connotes a component of chronic inflammation that is absent from DLs by definition.

Now that the diagnosis and definitions of DLs have been outlined and the gravity of the patient’s initial condition has been placed into context, we can consider the novel and evocative elements of the case report by Eddi and colleagues.1

The occurrence of a DL in the BII afferent limb appears to be novel, but as the authors correctly affirm, no region of the gut is immune. DLs may also occur in the bronchus of the respiratory system, which is not surprising given the fact that the gastrointestinal tract

References

and pulmonary system are evaginations of the same embryologic bud.\textsuperscript{10,11} DLs have been found after gastric resections, but the lesions are usually located proximal to the anastomotic site in the stomach.\textsuperscript{6,15} In a Swiss series of nonvariceal bleeding in 480 patients, 5.8% of all cases of bleeding were attributed to a DL; in 1 case (4%), 3 separate endoscopic attempts were needed to establish a DL diagnosis, as in the case report by Eddi and coworkers.\textsuperscript{1,12} The occurrence of DL bleeding after a BII procedure has been reported in a series from Greece that consisted of a slightly smaller number of patients.\textsuperscript{6} Twelve instances of bleeding were reported in patients who underwent a BII procedure; of these reports, 10 occurred at the anastomotic site. The locations of the remaining 2 cases were not described, making it impossible to confirm the novelty of this aspect of the case report. Nevertheless, the Greek study was germane to the present case report, as patients in the study bled from a different location in at least 10 of the 12 BII reconstruction patients. Some of the reasons advanced for the pathogenesis of the DL are based on this surgically altered anatomic location.

Although we do not know the cause of the bleeding that necessitated the BII procedure in this case report, the greatest delay for a DL rebleed in the Swiss study was 6 months, making it unlikely that this patient’s original source of bleeding was a DL. In the Greek study, a majority of patients had gastrectomies, but it appears that none of these patients had prior DL bleeding. In this study, endoscopic banding ligation (EBL) was used to control DL bleeding rather than hemostatic clips. DL treatment with epinephrine vasoconstriction injection and hemostatic clipping are fairly contemporary therapeutic options; the report by Eddi and colleagues outlines all contemporary treatment modalities except for argon plasma coagulation.\textsuperscript{13}

The second interesting element of the case report by Eddi and coworkers deals with the nature of the bleeding lesion.\textsuperscript{1} A DL diagnosis is likely if a tortuous or ectatic artery is seen via angiography or pathology of the resected sample after laparotomy.\textsuperscript{4} Endoscopic ultrasound can also reliably confirm the diagnosis.

There are 3 common scenarios of DL bleeding. The first scenario involves the presentation of a DL with intermittent, recurrent, profuse bleeding. In this scenario, the initial endoscopy and arteriography commonly fail to detect the bleeding source. If repeated endoscopies and arteriographies fail to diagnose the source of obscure bleeding in an unstable patient, intraoperative endoscopy can be performed with a diagnostic yield of 70–100%; however, bleeding in the small intestine requires a laparotomy or laparoscopy.\textsuperscript{2,14} In the case report by Eddi and associates, performing an endoscopic ultrasound in the afferent loop would have been challenging and it likely would have required a miniprobe examination.\textsuperscript{1,15}

In the second bleeding scenario, only an oozing site is visible—no protruding vessels, clots, or minute mucosal defects are seen—and the source of bleeding is thought to most likely be a DL-like bleed.

In the third scenario, the patient has a history of obscure bleeding that requires gastric resection; there is a high suspicion of a DL-like lesion; repeat surgery is an option due to the failure of endoscopic treatment; and there is a role for preoperative angiography for lesion localization.

In the 2 case series previously discussed, the proportion of DL bleeding episodes at gastrointestinal anastomoses ranged from 10% to 58% and occurred predominantly after BII procedures; the setting was usually massive gastrointestinal blood loss, as in the case report by Eddi and coworkers.\textsuperscript{1,6,12} Although the reason for the occurrence of a DL at or near the anastomotic site is unknown, the authors postulated that the explanation may involve bile reflux, an incomplete vagotomy, or vascular malformation after gastrectomy.\textsuperscript{5} These etiologies may have applied to the patient in the case report; however, somewhat unusually, the afferent loop appeared to be the source of the bleeding. The diagnosis of a DL in this scenario was likely, though a DL was unusual enough that other entities be considered in the differential diagnosis.

Other possible etiologies include marginal ulcers, which are increasingly being reported after gastric bypass surgery or vertical-banded gastroplasty.\textsuperscript{16} In a study of patients taking nonsteroidal anti-inflammatory drugs who presented with obscure bleeding, 12% had ileal and/or jejunal ulcerations ranging from small to large punched-out lesions with oozing blood or scarring that could be mistaken for a DL.\textsuperscript{1,17}

Various other causes of upper gastrointestinal hemorrhage have morphologic features that differ from those associated with DLs; nevertheless, these causes should be considered in the differential diagnosis. They include hemobilia, submucosal tumors, ischemic small-bowel necrosis, benign and malignant tumors, telangiectatic lesions, arteriovenous fistulas, aortoenteric fistulas, and Mallory-Weiss lesions. Primary and secondary aortoenteric fistulas most commonly occur at the distal duodenum or jejunum and can cause massive upper gastrointestinal hemorrhage. The latter group of fistulas is usually associated with prior aortic surgery, aortic aneurysms, and severe atherosclerosis.\textsuperscript{18} Arterial-type bleeding from diverticula is also interesting from the point of view of overlap, as a DL within a gastric diverticulum has been reported.\textsuperscript{19}

Vascular malformations, on the other hand, usually cause chronic blood loss, unlike a DL, which typically has intermittent, recurrent, upper gastrointestinal acute hemorrhage. Vascular ectasias are typically seen in patients with renal failure, aortic valve disease, a history
of radiation treatment, scleroderma, collagen diseases, or CREST syndrome (calcinosis, Raynaud phenomenon, esophageal dysmotility, sclerodactyly, and telangiectasia). The patient treated by Eddi and associates did not appear to have features of a connective tissue disorder.1 Bleeding from a Mallory-Weiss tear—a laceration in the mucosa at the gastroesophageal junction extending into the gastric cardia or, less commonly, the distal esophagus—is often self-limited, compared to the recurrent bleeding usually seen with a DL.

Some studies have shown an association between DL-like bleeding and alcohol intake or advanced liver disease, but no association has been seen with nonsteroidal anti-inflammatory drugs.4,20-22 However, concomitant administration of these agents may conceivably prolong and increase the severity of bleeding in patients with a DL.

Another newly described entity that can be diagnosed using the previously mentioned endoscopic criteria is DL-like bleeding caused by isolated gut spider nevi in patients with advanced liver disease. Spider nevi are central arterioles with small vessels radiating from the center.23,24 Both DLs and bleeding from isolated spider nevi may share a common endoscopic appearance in patients with advanced liver disease. Pressure within a central arteriole can reach as high as 70 mmHg, and erosion of thin overlying mucosa can give rise to arteriolar spurting bleeding, which is indistinguishable from a classic DL.25 There are many other forms of arterial or arteriolar bleeding that may qualify as DL bleeding based on endoscopic criteria; examples of these forms include aortoesophageal fistulas and splenic artery aneurysms.26,27

Because endoscopic features are shared with other entities, many authors now characterize bleeding or lesions as “DL-like.” In fact, a PubMed search of the term “DL-like” found 27 results, more than 10% of the results obtained by a search of the term “Dieulafoy” (240). The patient described in the case report by Eddi and colleagues did not have a previous aortic surgery, past penetrating trauma, or advanced liver disease, making DL the likely diagnosis by exclusion.1 We suggest that “DL-like” be used to describe bleeding when angiographic, endoscopic ultrasound, or pathologic data are unavailable or when less than 50% of cases in a series follow definitive lines of evidence for a DL. These guidelines would promote a more homogeneous population and allow for the creation of needed meta-analyses, as most series are quite small and unlikely to inform recommendations for definitive treatment in different clinical scenarios.

The distribution of these lesions is important and may help to determine a likely common pathway of causation, as previously discussed for a postgastrectomy DL. Currently, endoscopy is the initial step for diagnosing a DL, as this procedure can be both diagnostic and therapeutic.28,29 When endoscopic investigation fails, angiography, video capsule endoscopy (VCE), technetium-99m-labeled red blood cell scan, and endoscopic ultrasonography can be useful for identifying the correct diagnosis.2,14,15 In the case presented by Eddi and associates, VCE would not have contributed to the diagnosis.1 Endoscopic ultrasound can reveal a large-caliber vessel in the submucosa, suggesting a diagnosis of DL in the absence of bleeding.15 Angiography can particularly aid in the diagnosis of a DL in the colon or rectum, where endoscopic visualization can be obscured due to poor bowel preparation or active bleeding.30

The success rates of various endoscopic interventions for achieving hemostasis of DL bleeding range from 79% to 100% (Table 1).11,29,31,32 Several studies have reported that EBL, one of these interventions, is inexpensive, easy

Table 1. Initial Success Rates of Various Endoscopic Methods for Treatment of Bleeding Caused by Dieulafoy Lesions

<table>
<thead>
<tr>
<th>Modality</th>
<th>Hemostatic success rate</th>
<th>Rebleeding rate</th>
<th>Reference*</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ethanol injection</td>
<td>98%</td>
<td>11%</td>
<td>Asaki S, et al36</td>
</tr>
<tr>
<td>Norepinephrine and polidocanol</td>
<td>96.4%</td>
<td>11%</td>
<td>Baettig B, et al12</td>
</tr>
<tr>
<td>Thermal probe</td>
<td>79%</td>
<td>9%</td>
<td>Norton ID, et al39</td>
</tr>
<tr>
<td>Hemostatic clip placement</td>
<td>94.1%</td>
<td>9.3%</td>
<td>Yamaguchi Y, et al37</td>
</tr>
<tr>
<td>Endoscopic band ligation</td>
<td>100%</td>
<td>None</td>
<td>Alis H, et al31</td>
</tr>
<tr>
<td>Argon plasma coagulation</td>
<td>82%</td>
<td>6%</td>
<td>Lacopini F, et al39</td>
</tr>
<tr>
<td>Nd:YAG laser</td>
<td>80%</td>
<td>Not available</td>
<td>Skok P38</td>
</tr>
</tbody>
</table>

*These references are a representative—not an exhaustive—list of applicable studies.

to use, and as effective as bipolar coagulation or injection. Likewise, endoscopic hemostatic clip application is as effective as EBL for achieving hemostasis of a DL. In difficult-to-treat cases, angiography can not only aid in the diagnosis of a DL, but it can also be useful for achieving hemostasis by embolization.

When endoscopic and angiographic interventions fail, surgical treatment in the form of wide-wedge resection of the affected portion or oversewing of the lesion can be performed. Newer, minimally invasive laparoscopic surgery has also reportedly been successful for treatment of a DL located in the jejunum and stomach; preoperative endoscopic tattooing of the DL may be a valuable adjunct to the laparoscopist.

Conclusion

DLs and DL-like bleeding should be included in the differential diagnosis of patients presenting with recurrent, intermittent, massive upper gastrointestinal bleeding in all age groups as well as in patients with multiple comorbidities. Endoscopy remains the diagnostic and therapeutic tool of choice, with high success rates. The case reported by Eddi and coworkers reflects this statement and serves to reinforce many clinical considerations.

References