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Jejunal Dieulafoy's Lesion: A Systematic Review of Evaluation, Diagnosis, and Management

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Abstract

Jejunal Dieulafoy's lesion is an exceedingly rare but important cause of gastrointestinal bleeding. It frequently presents as a diagnostic and therapeutic conundrum due to the rare occurrence, intermittent bleeding symptoms often requiring prompt clinical action, variability in the detection and treatment methods, and the risk of rebleeding. We performed a systematic literature search of MEDLINE, Cochrane, Embase, and Scopus databases regarding jejunal Dieulafoy's lesio from inception till June 2020. A total of 136 cases were retrieved from 76 articles. The mean age was 55 ± 24 years, with 55% of cases reported in males. Patients commonly presented with melena (33%), obscure-overt gastrointestinal bleeding (29%), and hemodynamic compromise (20%). Hypertension (26%), prior gastrointestinal surgery (14%), and valvular heart disease (13%) were the major underlying disorders. Conventional endoscopy often failed but single- and double-balloon enteroscopy identified the lesion in 96% and 98% of patients, respectively. There was no consensus on the treatment. Endoscopic therapy was instituted in 64% of patients. Combination therapy (34%) with two or more endoscopic modalities, was the preferred approach. With regard to endoscopic monotherapy, hemoclipping (19%) and argon plasma coagulation (4%) were frequently employed procedures. Furthermore, direct surgical intervention in 32% and angiographic embolization was performed in 4% of patients. The rebleeding rate was 13.4%, with a mean follow-up duration of 17.6 ± 21.98 months. The overall mortality rate was 4.4%. Jejunal Dieulafoy's lesion is still difficult to diagnose and manage. Although the standard diagnostic and therapeutic modalities remain to be determined, device-assisted enteroscopy might yield promising outcomes.

Keywords

Dieulafoy's lesion, jejunum, gastrointestinal bleeding, evaluation, diagnosis, management, therapeutic endoscopy, clinical outcomes

Introduction

Dr M. T. Gallard first reported Dieulafoy's lesion as "miliary aneurysms of the stomach" in 1884.[1] Later on, the French surgeon, Paul Georges Dieulafoy (1839-1911), more precisely described this clinical entity and referred to it as an "exulceratio simplex" in 1898.[2] This atypical lesion of undetermined etiology was subsequently termed as "Dieulafoy's lesion." Since then, the definition of this abnormality has been diversified to represent an acute process occurring in several body locations.[3] Currently, the true incidence remains unknown, but it is responsible for up to 5% of gastrointestinal bleeds.[4-6] It shows a predilection toward advanced age, with a male-to-female ratio of 2:1.[6] It typically afflicts the stomach (72%), but extragastric occurrences have also been reported in the duodenum (15%), esophagus (8%), colon (2%), and rectum (2%).[6,7] Clinical presentation is frequently related to a painless, intermittent, massive gastrointestinal hemorrhage.[8] With

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the advent of endoscopy, the diagnosis and treatment are drastically ameliorated. Due to multiple advancements in the nonoperative management, a vast majority of lesions are now amenable to endoscopic manipulation.[8] Therefore, mortality rate of bleeding Dieulafoy's lesion has shown a significant decline from 79% to 8.6%.[9,10]

Jejunal Dieulafoy's lesion is a rare cause of gastrointestinal hemorrhage that can be torrential and life-threatening. Levine and Valk first described jejunal aneurysm with rupture of a submucosal artery in 1944.[11] Moreira-Pinto et al highlighted in their study that only 18 cases of this entity had been reported until 2009.[12] Although jejunal lesions are increasingly documented in the current times, the overall incidence remains considerably low. A plethora of recent studies have divulged that merely 1% of all the confirmed cases of Dieulafoy's lesion are found in the jejunum.[6,12] The remarkable rarity, unrecognized lesions, intermittent nature of bleeding, and the lesion site mostly inaccessible to conventional endoscopes make jejunal lesion a difficult-to-diagnose etiology.[13] Furthermore, the lack of standard treatment, coupled with rebleeding risks, also renders it a therapeutic dilemma.[13-18] In order to highlight the knowledge gap and to help frame specific diagnostic and therapeutic protocols in the future, a systematic review of the existing data has been warranted. To our research, this study represents the first comprehensive review pertaining to the clinical characteristics, diagnostic workup, and treatment of jejunal Dieulafoy's lesion based on the most recent information from the limited number of studies.

Materials and Methods

A systematic search of MEDLINE (PubMed and Ovid), Cochrane, Embase, and Scopus databases was conducted from inception to June 2020. The search terminologies and subheadings such as "Dieulafoy's lesion," "gastrointestinal bleeding," "melena," "hematochezia," and "hematemesis" were combined using the Boolean operators "AND" and "OR" with the terms "jejunum" and "jejunal." A manual search of bibliography lists was performed for additional articles. Furthermore, several conference papers were also enlisted. We initially screened all retrieved titles and abstracts to determine their relevance to our topic. The following inclusion criteria was applied for the final analysis: (1) articles describing cases of jejunal Dieulafoy's lesion that either fulfilled endoscopic criteria or had a biopsy-proven diagnosis; (2) cases having sufficient data regarding patient demographics, clinical presentation, therapeutic approach, and morbidity and mortality information; (3) studies published in the English language; and (4) articles available in the full-text form. The exclusion criteria mainly consisted of duplication, articles written in languages other than English, irrelevance to the topic, and a lack of sufficient patient data.

Results

A total of 257 articles consisting of but not limited to retrospective studies, case series, and case reports were initially collected. Thirteen publications were identified from the reference lists of the search results. The titles and abstracts of all these articles were carefully reviewed. A total of 176 articles were excluded as they were not related to our study, were in a language other than English, and/or the full-text versions were not available. Ninety-four articles were first enlisted for re-review. After further exclusion of duplicate and redundant articles, 76 articles were included in the present study. PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow diagram represents the search methodology for data synthesis and results (Figure 1). A thorough perusal of these articles yielded a total of 136 cases of jejunal Dieulafoy's lesion. The screened articles were downloaded for data extraction and tabulation regarding patient demographics, diagnostic workup, lesion site within the jejunum, management, and clinical outcomes (Supplementary Tables 1 and 2, available online). The descriptive data are provided as mean ± standard deviation, median (range), or percentage, as applicable.

Discussion

Patient Characteristics

Jejunal Dieulafoy's lesion may show a slight male predominance, with a male-to-female ratio of 1.2:1. This observation stands in contrast with the clear male predominance patterns seen in Dieulafoy's lesions in general.[6,13] Furthermore, a significant variation in age distribution, ranging from 10 to 95 years (mean ± standard deviation, 55 ± 24 years; median, 82 years), was observed (Table 1). However, a vast majority of patients were diagnosed in their seventh and eighth decades of life (Figure 2). Therefore, advanced age portends an increased risk of the jejunal disease, which correlates with demographic characteristics of patients with Dieulafoy's lesions from other gastrointestinal locations.[13,19] While a fixed geographical distribution does not exist, most cases included in this review were reported in developed countries (Figure 3). The aforesaid trend may be attributed to the technical advancements in diagnostic modalities and improved patient care in developed nations. Due to the advanced age and underlying chronic medical conditions, a number of patients were on anticoagulants and/or nonsteroidal anti-inflammatory drugs (NSAIDs).[20,21]

Pathogenesis

Dieulafoy's lesion is a vascular abnormality that maintains a caliber of 1 to 3 mm, which is 10-fold larger compared with

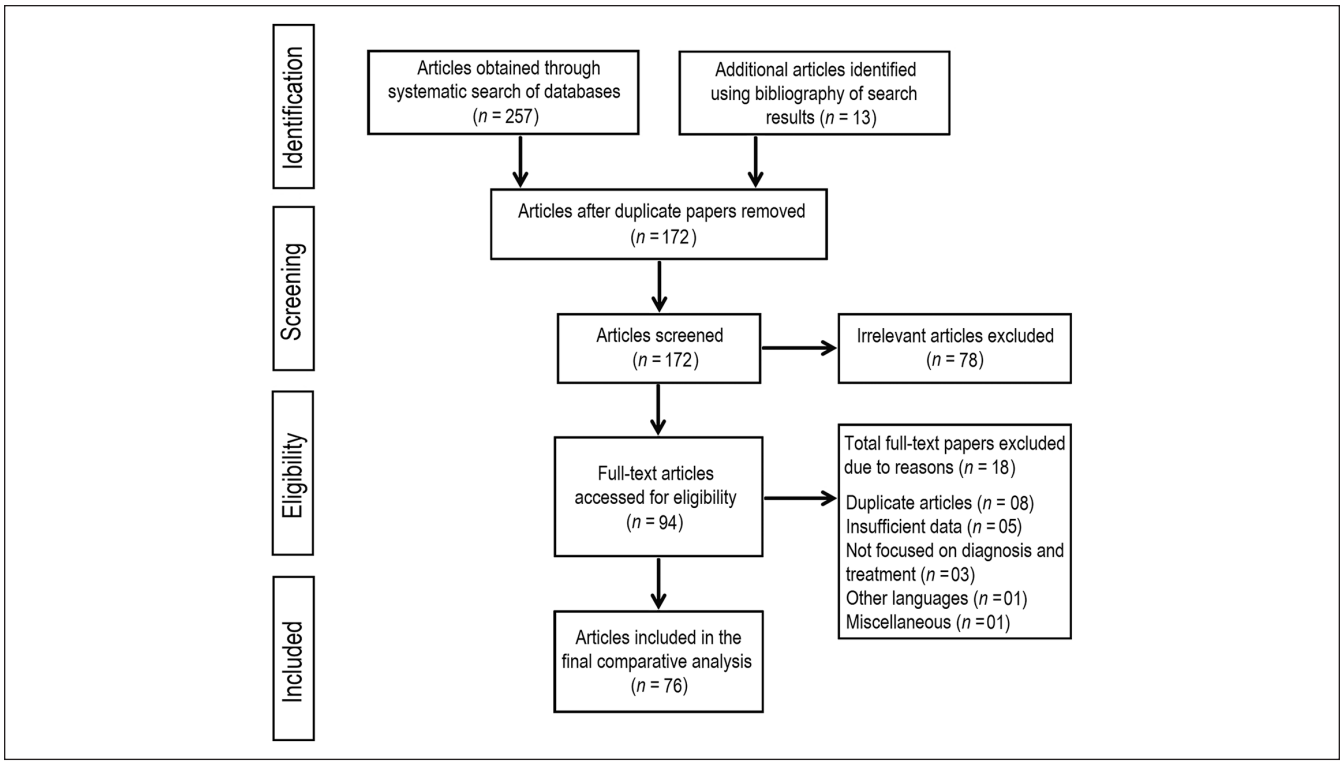

Figure 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram representing the search methodology for data synthesis regarding jejunal Dieulafoy's lesion.

Table 1. Demographic and Key Data of Patients With Jejunal Dieulafoy's Lesion[a].

Patient characteristics	N = 136
Age (years)	
Mean	55
Range	10-95
Age distribution of patients, n (%)	
≥50 years	82 (61%)
<50 years	52 (29%)
Gender, n (%)	
Male	74 (55%)
Female	60 (45%)
Admission type, n (%)	
Nonelective	83 (61%)
Elective	53 (39%)
Anticoagulants/NSAIDs, n (%)	
Aspirin/NSAIDs	17 (15%)
Antiplatelet drugs	13 (12%)
Initial hemoglobin levels, (g/dL)	
Mean	8.4
Range	3.3-14.4

Abbreviation: NSAIDs, nonsteroidal anti-inflammatory drugs.
[a]Two patients did not have age and/or gender data available.

the caliber of a normal submucosal artery.[22,23] It is characterized by a small mucosal defect with no surrounding mucosal inflammation. No aneurismal, arteriosclerotic, or vasculitic changes are associated with this entity. These aberrant vessels frequently assume a tortuous course and tend to project through the mucosal defect (2-5 mm), making them vulnerable to minor mechanical trauma.[24] Several theories have been postulated to account for the spontaneous rupture and bleeding from a Dieulafoy's lesion. One theory posits that the constant pressure exerted by the pulsatile nature of the large-caliber vessel can cause minute mucosal erosion, which may subsequently lead to gastrointestinal hemorrhage.[25] Additionally, age-related wear and tear of the submucosal vessels can lead to thrombosis, which may cause ischemia and hypoperfusion of the surrounding mucosa. This mechanism is termed the "vascular steal phenomenon" due to the presence of a pale mucosal halo observed in angiodysplasia.[26] This theory may explain the relatively increased risk of bleeding from Dieulafoy's lesion in old age, especially in patients older than 70 years.[27] In agreement with these hypotheses, advanced patient age was frequently noted in our analysis, potentially denoting a causal association with the lesion. Given the occurrence of jejunal Dieulafoy's lesion in newborns, congenital origin may also be considered.[28] In a retrospective study, Shin et al showed that antiplatelet agents and alcohol consumption were risk factors associated with the formation of upper gastrointestinal Dieulafoy's lesion.[29] Therefore, the presence of chronic medical conditions and the concurrent use of anticoagulants and NSAIDs may also contribute to senile mucosal atrophy, possibly leading to the formation of this lesion.[30,31] In addition, these

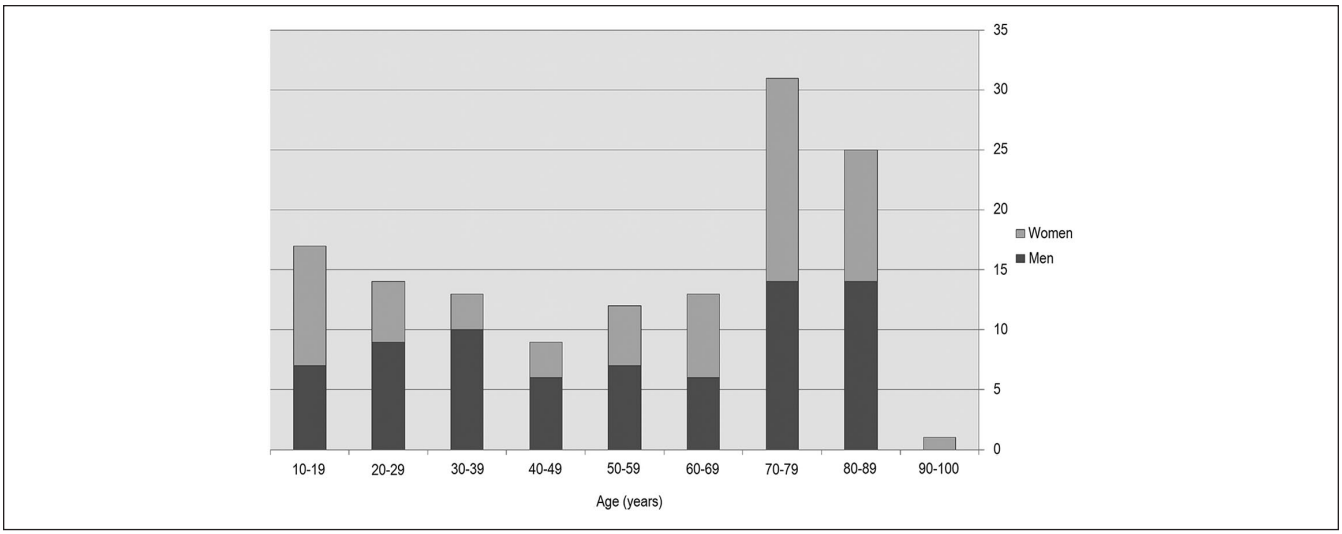

Figure 2. Age and gender distribution of patients with Dieulafoy's lesion of the jejunum.

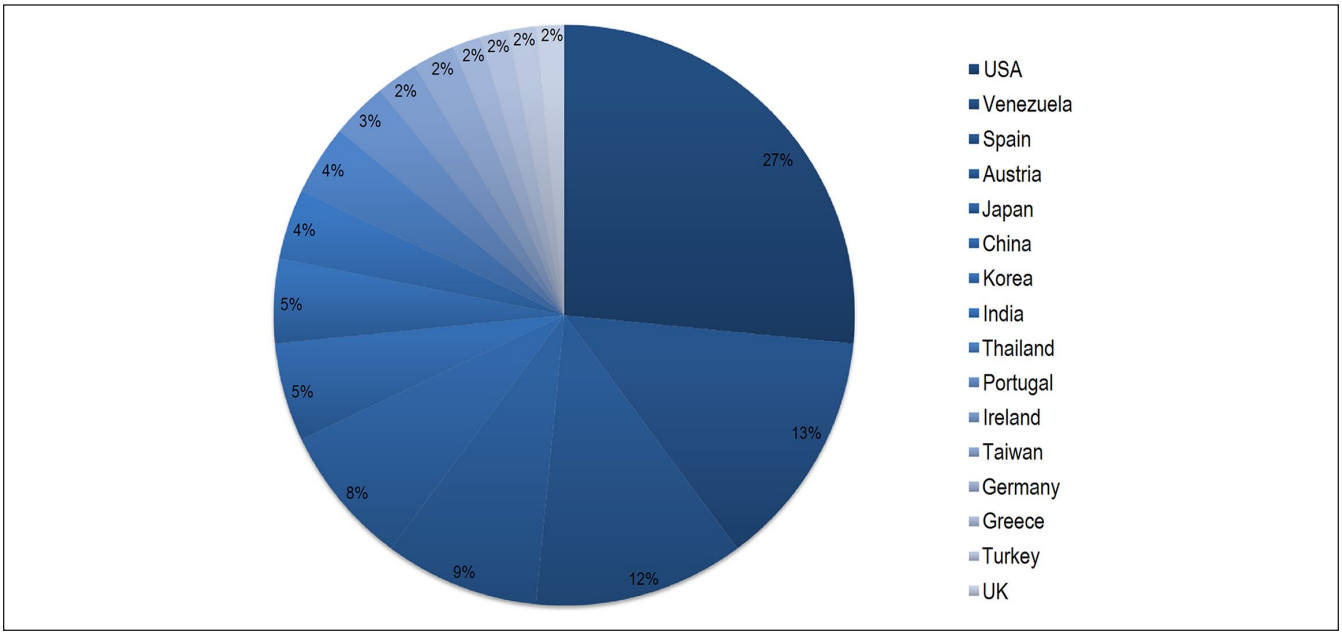

Figure 3. Pie chart showing the geographical distribution of the reported cases of jejunal Dieulafoy's lesion.

factors may also precipitate bleeding from the existing lesions.[32,33]

Clinical Presentation

The classical clinical presentation of jejunal Dieulafoy's lesion includes painless gastrointestinal hemorrhage.[34-37] Patients often present with melena (33%), obscure-overt gastrointestinal bleeding (29%), or hematochezia (13%) (Table 2).[38-44] The bleeding can be particularly severe and intermittent due to the vessel's large diameter and pulsatile nature (Figure 4).[45,46] Therefore, hemodynamic compromise may also ensue in approximately 20% of patients, culminating in tachycardia, hypotension, and even hypovolemic syncope.[47-49] At presentation, the mean hemoglobin level in patients with jejunal lesion was 8.4 g/dL. The number of blood units transfused ranged from 2 to 9. This drop in hemoglobin levels explains the possible presenting symptoms like dizziness/fatigue (11%) and iron-deficiency anemia (10%). Furthermore, nonspecific initial clinical symptoms such as abdominal discomfort can also be encountered.[50] Since these vague presenting complaints can mimic other gastrointestinal pathologies, an accurate diagnostic modality is required to avoid misconstruing the underlying etiology. Notably,

Table 2. Presentation Patterns in Patients With Jejunal Dieulafoy's Lesion.

Clinical presentations	N (%)
Gastrointestinal bleeding	
Melena	45 (33%)
Obscure-overt bleeding	39 (29%)
Hematochezia	17 (13%)
Bright-red blood per rectum	14 (10%)
Hematemesis + melena	6 (4%)
Hematemesis	3 (2%)
Hematemesis + hematochezia	3 (2%)
Sudden hemodynamic compromise	27 (20%)
Dizziness/fatigue	15 (11%)
Blood loss anemia	13 (10%)
Abdominal pain/epigastric discomfort	10 (7%)
Sudden change in conscious status	9 (6%)

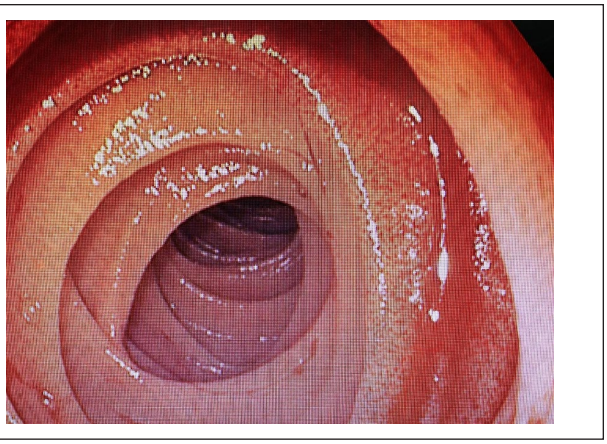

Figure 4. A 35-year-old male who had undergone prior upper endoscopy 6 months ago for acute upper gastrointestinal bleeding that had not identified any culprit lesions, presented with acute-onset melena and an acute drop in hemoglobin level from 12.0 g/dL to 7.6 g/dL. Push enteroscopy revealed an actively bleeding lesion without surrounding erosions or ulceration in the proximal jejunum, consistent with a Dieulafoy's lesion.

Dieulafoy's lesion can also be incidentally detected in patients hospitalized for other medical conditions. Although it has been described in only one case report, jejunal lesion may also be associated with intestinal intussusception.[51] In symptomatic individuals, presentation patterns indicate the general condition of the patient, bleeding site, and the extent and duration of hemorrhage. Therefore, these factors collectively play a key role in gauging the hemodynamic status, which remains exorbitantly focal in both the diagnosis and management.[52]

Comorbid Conditions

Hypertension and prior gastrointestinal surgery were the commonly observed underlying disorders with the frequency of 26% and 14%, respectively. In hypertensive states, there is a constant pulsatile motion of these large tortuous vessels that can introduce a mucosal defect in the jejunum. A direct contact with food contents or minor trauma can rupture these vessels, potentially leading to massive bleeding. As with gastric and duodenal Dieulafoy's lesion, prior gastroenterological surgery may also precipitate bleeding in patients with jejunal disease, indicating the stress injury as a probable inciting factor.[22] Gadenstätter et al highlighted prior Billroth II gastrectomy in subsequent causation of jejunal lesions.[53] Additionally, we posit that the presence of congestive heart failure, resulting in a fluid overload state, might also herald the formation of such lesions involving large, dilated, and tortuous malformed vessels that are susceptible to fluid-pressure alterations.[6,28,31,54] Several other important clinical associations of jejunal Dieulafoy's lesion were also noted in our analysis (Table 3).[21,55,56] Alcoholism, drug abuse, diverticulosis, asthma, diabetes mellitus, chronic kidney disease, ischemic heart disease, rheumatic fever, gastrointestinal angiodysplasia, and hyperlipidemia were among the less common comorbidities.[16,21,34] In a number of patients, preexisting gastrointestinal conditions like peptic ulcer disease,

Table 3. Major Comorbid Conditions Associated With Jejunal Dieulafoy's Lesion[a].

Comorbid conditions	%
Cardiovascular	
Hypertension	26%
Valvular heart disease	13%
Congestive heart failure	10%
CAD/CABG	10%
Atrial fibillation	8%
Unexplained anemia	5%
Cerebrovascular accident	4%
Gastrointestinal and liver	
Prior gastrointestinal surgery	14%
History of gastrointestinal bleeding	7%
Cirrhosis	4%
Diverticulosis	3%
Gastrointestinal angiodysplasia	2%
Substance use disorder	
Alcoholism	3%
Drug abuse	2%
Miscellaneous	
Cancer	7%
Diabetes mellitus	2%
Chronic kidney disease	2%
Asthma	2%
Diabetes mellitus	2%
Hyperlipidemia	2%

Abbreviations: CAD, coronary artery disease; CABG, coronary artery bypass grafting.
[a]In this study, comorbid condition data were documented for 91 of 136 patients. Of the 91 patients, 65 had underlying medical conditions, whereas 26 patients had no comorbidities.

Table 4. Comparison of Diagnostic Yields of Different Modalities Employed in Patients With Jejunal Dieulafoy's Lesion.

Diagnostic modality	N (%)
Endoscopy	
Double-balloon enteroscopy	41/42 (98%)
Single-balloon enteroscopy	25/26 (96%)
Push enteroscopy	8/11 (73%)
Small bowel capsule endoscopy	13/23 (57%)
Conventional upper endoscopy	13/64 (20%)
Colonoscopy	1/41 (2%)
Visceral angiography	21/39 (54%)
Bleeding scan	6/14 (43%)
Surgical methods	
Exploratory laparotomy[a]	28/30 (93%)
Intraoperative endoscopy	10/12 (83%)
Autopsy	2/2 (100%)

[a]The final diagnosis was made after histopathologic analysis of the resected jejunal specimen.

gastroesophageal reflux disease, gastritis, and obesity were also present.[21,40] Similarly, rare associations of jejunal lesion included hypertrophic obstructive cardiomyopathy, endocarditis, pericarditis, congenital heart disease, abdominal aortic aneurysm, arteriovenous malformation, hepatitis, polycystic ovarian syndrome, dysfunctional uterine bleeding, osteoarthritis, obstructive sleep apnea, tuberculosis, and human immunodeficiency syndrome.[18,47,51,56] Ke et al also reported a case of jejunal Dieulafoy's lesion associated with mucosa-associated lymphoid tissue lymphoma, alluding to a possible correlation.[57]

Diagnosis

Jejunal Dieulafoy's lesion continues to pose a diagnostic challenge. It frequently eludes detection with conventional endoscopy due to the intermittent nature of the hemorrhage and possible inaccessibility of jejunum with standard forward-viewing endoscope.[58] Furthermore, endoscopic navigation in the relatively narrow jejunal lumen can be difficult. In order to identify these quiescent lesions, endoscopists may wrongly search for mucosal abnormalities and inflammation owing to a lack of required skillset. The blood can often wind up as clots after a bleeding episode, which may also shroud the underlying lesions.[59] The vague clinical features potentially masquerading as peptic ulcer disease or gastrointestinal tumors can further obscure the diagnosis.[60,61] Therefore, the efficacy of the conventional endoscopy in jejunal Dieulafoy's lesion remains controversial.[62-64] However, endoscopies performed within the first 12 hours of bleeding onset may show a slightly better diagnostic value.[65] In this review, initial upper endoscopy and colonoscopy demonstrated low diagnostic yields of 20% and 2%, respectively (Table 4). Capsule endoscopy and push enteroscopy have also been undertaken with variable efficacies, especially in stable patients after unremarkable initial endoscopies.[66-68] In undiagnosed cases where hemodynamic compromise is evident, mesenteric angiography can be helpful.[41,57] Computed tomography angiography carries particular importance to identify extragastric culprit vessels. A formal heparin therapy with angiography can be added in cases where standard angiography is futile. Furthermore, bleeding scans may also help in small intestinal hemorrhage after nondiagnostic endoscopies.[55,69] Surgical evaluation is requisite in most hemodynamically unstable patients. It is particularly warranted in clinical settings that are bereft of necessary endoscopic expertise. During surgery, intraoperative enteroscopy can be performed to help localize the bleeder.[70]

The findings of this review showed that the single- and double-balloon enteroscopies were the ideal techniques, with the diagnostic yields of 96% and 98%, respectively. These device-assisted enteroscopies are more efficacious in identifying Dieulafoy's lesion as they complement noninvasive techniques like capsule endoscopy.[71] They offer safe and effective deep direct endoscopic access to the jejunum for diagnostic evaluation and therapeutic interventions.[71] In order to determine the oral or anal approach, the clinical presentation and subsequent capsule endoscopy findings are pertinent.[72] Min et al emphasized the need for repeat capsule endoscopy in patients with obscure gastrointestinal bleeding.[67] Lipka et al highlighted that most Dieulafoy's lesions are located in the proximal jejunum.[73] Dulic-Lakovic et al revealed that the average distance of jejunal lesions from the pylorus was 132 ± 115 cm.[74] The oral route is used if the likely culprit bleeder is located in the proximal two thirds of the small bowel on capsule endoscopy. If the oral route is unrevealing of the lesion, the deepest insertion point should be marked before opting for the anal approach.[74] After accurate identification, the lesion site should also be tattooed using India ink to mark it for recurrence or possible presence of more than one Dieulafoy's lesions.[33,75] In this review, most lesions were identified in the proximal jejunum (64%), followed by the distal (24%) and middle parts (13%). In addition, the lesion was also noted in the jejunal side of the anastomotic sites in some cases. Notably, chronic intermittent hemorrhage is a common occurrence while performing device-assisted enteroscopy, potentially warranting multiple diagnostic attempts.[74] Lipka et al reported that all jejunal lesions were identified after the first diagnostic attempt.[73] Dulic-Lakovic and colleagues showed that 1.5 endoscopies were required per patient to diagnose jejunal disease.[74] The temporal relationship between the initial presentation and the diagnostic value of this procedure is also important. Robles et al revealed that the diagnostic yield of emergency double-balloon enteroscopy was remarkably higher than the enteroscopy performed after 24 hours of the symptom onset (detection rate: 40.7% vs 0.9%).[76] In our analysis, the overall mean number of diagnostic attempts in patients with jejunal disease was 3 (range: 1-11). The lack of widespread availability and associated costs can present potential problems in

Table 5. Efficacy of Treatment Modalities in Reported Cases of Jejunal Dieulafoy's Lesion[a].

Therapeutic modality	Hemostasis rate	Rebleeding rate
Endoscopic therapy		
Combination endoscopic therapy	39/46 (85%)	7/46 (15%)
Hemoclipping	23/25 (92%)	2/25 (8%)
Argon plasma coagulation	5/6 (83%)	1/6 (17%)
Injection sclerotherapy	2/4 (50%)	2/4 (50%)
Bipolar electrocoagulation	2/2 (100%)	0/2 (0%)
Band ligation	1/2 (50%)	1/2 (50%)
Heater probe	1/1 (100%)	0/1 (0%)
Angiographic embolization	0/5 (0%)	5/5 (100%)
Surgical resection	43/43 (100%)	0/43 (0%)

[a]Two patients remained untreated and died.

the use of device-assisted enteroscopy, culminating in repetitive diagnostic evaluations. Thus, prompt detection of jejunal Dieulafoy's lesion still remains a major clinical concern due to the limitations of conventional endoscopy.

Management

The therapeutic guidelines for bleeding jejunal Dieulafoy's lesion remain elusive. In this review, 86 (64%) patients were initially treated with endoscopic therapy. Direct surgical treatment was applied in 43 (32%) and angiographic embolization was attempted in 5 (4%) patients. Unfortunately, 2 patients remained untreated, and the lesion was eventually identified on autopsy examination (Table 5).[14,18] With the improved therapeutic prowess of endoscopic techniques, endoscopic therapy has been increasingly recognized as an effective method of treatment. Currently, no consensus on optimal endoscopic monotherapy exists. However, several combinations of endoscopic techniques, including injection sclerotherapy, heater probe, electrocoagulation, and mechanical methods are used. Endoscopic injection sclerotherapy alone has now become a relatively uncommon intervention in patients with Dieulafoy's lesion.[77] Epinephrine monotherapy (initial dose: 2.5 mL; dilution of 1:10 000) is simple, inexpensive, and safe, but it carries a significant risk of rebleeding.[78] In combination therapy, epinephrine injection remarkably improves the hemostasis success rates. It helps achieve better visualization that facilitates the direct application of subsequent definitive treatment.[79] Thermal endoscopic therapy is used to achieve hemostasis by applying contact or noncontact thermocoagulation.[80] Contact thermocoagulation includes bipolar and heater probes, whereas argon plasma coagulation (APC) is an example of a noncontact method. Notably, contact methods may lead to transmural injury in thin-walled gastrointestinal tract segments, such as the small bowel. Therefore, noncontact thermocoagulation is relatively safe as it mostly elicits superficial tissue injury.[81]

Mechanical endoscopic methods such as hemoclipping and band ligation confer a slight therapeutic superiority over other endoscopic techniques.[82-84] Recently, hemoclipping is exacting to come to terms within locations such as the small bowel (Figure 5).[73,82] Major predictors of successful hemoclipping include the presence of surrounding soft tissue and appropriate positioning of hemoclips.[83] Hemoclip application via device-assisted enteroscopy has gained preeminence in the current times.[84-88] In our review, endoscopic hemoclipping alone achieved a relatively higher primary hemostasis rate (92%) and a lower rebleeding rate (8%). Endoscopic band ligation is relatively easier than hemoclipping as its versatility allows it to reach in difficult gastrointestinal sites.[89,90] However, the issues like prolonged procedural time due to technical complexity and rebleeding following ulcer formation in ligated mucosa may be encountered.[91] Prior anecdotal clinical evidence also supports the use of over-the-scope clip (OTSC) in refractory bleeding jejunal Dieulafoy's lesions.[92,93] Combination endoscopic therapy was used in 46 of 134 patients. It consisted of varying combinations of epinephrine

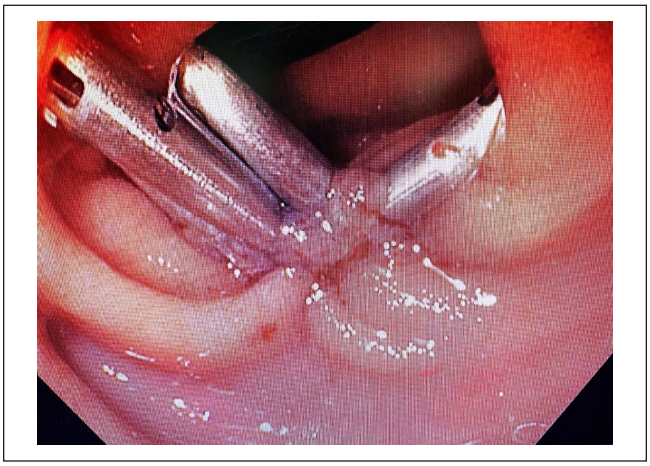

Figure 5. Primary hemostasis was achieved using a combination of epinephrine injection and endoscopic hemoclipping. After 2 days of uneventful hospital course, the patient was discharged home in a stable condition. No further gastrointestinal hemorrhage occurred during 6 months of follow-up.

Table 6. Efficacy of Various Combinations Employed as Endoscopic Combination Therapy in Reported Cases (N = 46) of Jejunal Dieulafoy's Lesion.

Combination of endoscopic modalities	Primary hemostasis rate, n (%)	Rebleeding rate, n (%)
Injection therapy + endoscopic hemoclipping	18/18 (100%)	0/18 (0%)
Injection therapy + APC	8/12 (67%)	4/12 (33%)
Epinephrine + APC + hemoclipping	3/3 (100%)	0/3 (0%)
Epinephrine + endoscopic hemoclipping + site tattooing	2/2 (100%)	0/2 (0%)
Bipolar electrocoagulation + APC + endoscopic hemoclipping	2/2 (100%)	0/2 (0%)
APC + endoscopic hemoclipping	1/2 (50%)	1/2 (50%)
Endoscopic hemoclipping + over-the-scope clip	1/1 (100%)	0/1 (0%)
Epinephrine + bipolar electrocoagulation	1/1 (100%)	0/1 (0%)
Epinephrine + ethyl alcohol	1/1 (100%)	0/1 (0%)
Epinephrine + 50% dextrose water	0/1 (0%)	1/1 (100%)
Bipolar electrocoagulation + endoscopic hemoclipping	0/1 (0%)	1/1 (100%)
Epinephrine + photocoagulation	1/1 (100%)	0/1 (0%)
Epinephrine + endoscopic band ligation	1/1 (100%)	0/1 (0%)

Abbreviation: APC, argon plasma coagulation.

injection, APC, bipolar electrocautery, hemoclipping, and endoscopic band ligation (Table 6).[94-98] Two frequently applied combinations included injection therapy plus hemoclipping and injection sclerotherapy with APC. The technical feasibility of combination endoscopic therapy was good, with a primary hemostasis rate of 85%.

Angiography is therapeutically effective in 40% to 89% cases of nondiverticular bleeding.[99,100] In our review, transarterial embolization was performed in 5 cases but it failed to secure hemostasis.[41,49,57,101,102] Subsequently, these patients were treated with surgery (n = 4) or endoscopic therapy (n = 1). Factors such as relative jejunal mobility and its wall thinness may disrupt an optimal arterial compression following transarterial embolization, leading to treatment failure. Surgical interventions are indicated if other diagnostic modalities fail, especially in patients with hemodynamic compromise.[103] Intraoperative identification in jejunal lesions is difficult as they lack visible fibrosis, inflammation, polyposis, or significant mucosal changes, warranting preoperative localization for a successful resection.[104] Intraoperative enteroscopy using methylene blue dye or Tc-99m red blood cells may help in this regard.[36,105] In terms of surgical techniques, several open and minimally invasive procedures have been employed. Mino et al first reported the laparoscopic treatment for jejunal Dieulafoy's lesion.[106] Prasad et al described a combined capsule endoscopic detection followed by laparoscopic-assisted transumbilical resection.[107] The minimally invasive approach is increasingly used, but segmental resection with end-to-end anastomosis still remains the most common surgical technique.[34,41,108] Simple overlaying is discouraged as it may lead to rebleeding. After precise identification of the lesion site, the culprit bleeder's surgical suture ligation may also be considered.[109] In this review, 43 patients underwent direct surgical intervention and 12 required surgery after failed initial therapeutic attempts. Surgery demonstrated 100% hemostasis rate.

Clinical Outcomes

Although existing data regarding jejunal Dieulafoy's lesion remain limited, previous studies reported rebleeding rates of 12.5% and 20%, respectively.[72,73] In this review, the overall primary hemostasis rate was 87.3% (117 of 134), with a rebleeding rate of 13.4% (18 of 134). The presence of multiple underlying medical conditions, active infection, treatment with adrenaline monotherapy, and arterial spurting were possible precipitating factors for rebleeding. However, it is observed that the recurrence in Dieulafoy's lesion was independent of patient age, gender, or lesion site.[110] The treatment of rebleeding included surgical resection (n = 12), APC plus epinephrine injection (n = 2), hemoclipping (n = 2), APC plus hemoclipping (n = 1), and OTSC plus endoscopic band ligation (n = 1). Of 18 patients who experienced rebleeding, 16 recovered well with retreatment but 2 patients died. The overall mortality rate in patients with jejunal Dieulafoy's lesion was 4.4%. The causes of death included missed diagnosis (n = 2), rebleeding (n = 2), cardiac complications (n = 1), and worsening of Goodpasture syndrome (n = 1).[14,18,21,56,78] In these patients, concomitant cardiovascular disease, respiratory failure, sepsis, and/or cirrhosis were major predictors of mortality.[71] Based on this review's findings, a timely and appropriate treatment regimen is exceedingly important for better long-term prognosis.

Follow-up

The follow-up recommendations in patients with jejunal Dieulafoy's lesion lack consensus, resulting in case-to-case variations.[111] It is important to note that the rebleeding may occur from hours to weeks after initial treatment. In previous research, patients developed recurrence 12, 13, and 49 days, and even 12 weeks after the first treatment.[56,72,73] Therefore, a follow-up duration of at least 6 months may be suggested

and a longer follow-up of years may not bear significant clinical value.[112] In terms of the method of follow-up, surveillance endoscopy was performed in several patients. In this review, follow-up information was documented in 87 patients. The mean duration of the follow-up was 17.6 ± 21.98 months (range: 1-145 months). Even though this article bears limitations as the data are mostly gleaned from reports of single cases and case series, it summarizes the existing clinical evidence regarding jejunal Dieulafoy's lesion. It highlights the diagnostic and therapeutic importance of single- and double-balloon enteroscopy in such patients. These observations should be the forerunner for research utilizing these device-assisted enteroscopy techniques for jejunal Dieulafoy's lesion.

Conclusion

Jejunal Dieulafoy's lesion is a rare but serious clinicopathologic entity. It is imperative for endoscopists to have apt knowledge of this condition as it can cause treacherous and life-threatening gastrointestinal hemorrhage. This disease continues to present a diagnostic dilemma. The probable lesion site within the jejunum and hemodynamic status of the patients are important for the selection of initial intervention. Endoscopic methods are preferred in hemodynamically stable individuals. Jejunal disease can often elude conventional endoscopy, necessitating the uptake of advanced techniques. In addition to diagnosis, single- and double-balloon enteroscopy offers a reasonable therapeutic approach. The optimal endoscopic treatment remains to be determined, but the data presented here support the use of hemoclipping with or without injection sclerotherapy. After successful endoscopic hemostasis, rebleeding episodes are uncommon. In hemodynamically unstable and difficult-to-diagnose patients, exploratory laparotomy remains the eventual diagnostic strategy. Surgical management is advantageous with extremely low risk of rebleeding. The mortality and morbidity related to jejunal lesion has significantly decreased with the improvements in treatment armamentarium. This article illustrates that gastroenterologists should develop a patient-specific strategy for the diagnosis and treatment for jejunal Dieulafoy's lesion, one that also considers the respective clinical settings.

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Supplemental Material

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