# RCS ADVANCING SURGICAL STANDARDS

# **REVIEW**

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# Dieulafoy's lesion: current trends in diagnosis and management

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#### **ABSTRACT**

BACKGROUND Dieulafoy's lesion is a relatively rare, but potentially life-threatening, condition. It accounts for 1-2% of acute gastrointestinal (GI) bleeding, but arguably is under-recognised rather than rare. Its serious nature makes it necessary to include it in the differential diagnosis of obscure GI bleeding. The aim of this study was to review the current trends in the diagnosis and management of Dieulafoy's lesion.

MATERIALS AND METHODS Using Medline, a literature search was performed for articles published in English, using the search words 'Dieulafoy'(s)' and 'gastrointestinal bleeding'. All retrieved papers were analysed and the findings are summarised in this review.

RESULTS There is no consensus on the treatment of Dieulafoy's lesions. Therapeutic endoscopy can control the bleeding in 90% of patients while angiography is being accepted as a valuable alternative to endoscopy for inaccessible lesions. Currently, surgical intervention is kept for failure of therapeutic endoscopic or angiographic interventions and it should be guided by preoperative localisation.

CONCLUSIONS Advances in endoscopy have increased the detection of Dieulafoy's lesions and decreased the mortality from 80% to 8.6%. There are recent encouraging reports on the successful use of laparoscopic surgery in managing symptomatic Dieulafoy's lesions.

#### **KEYWORDS**

Dieulafoy's lesion - Diagnosis - Management - Gastrointestinal bleeding

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The incidence of acute gastrointestinal bleeding ranges from 50–150 per 100,000 of the population each year. It is caused by peptic ulcers and oesophageal or gastroduodenal erosions in almost 80% of patients.¹ Obscure gastrointestinal bleeding is reported to account for up to 5% of all gastrointestinal (GI) haemorrhages. It is defined as overt or occult bleeding from a source that cannot be readily determined by standard investigations, such as barium studies and endoscopic investigation. The source is often difficult to locate because the pathology is anatomically inaccessible, small, or subtle. Dieulafoy's lesion is one of the causes of obscure gastrointestinal bleeding that could result in treacherous and life-threatening gastrointestinal haemorrhage.².5

A Dieulafoy's lesion, also termed 'calibre persistent artery',² is a relatively rare, but potentially life-threatening, cause of haemorrhage from the gastrointestinal tract. It is difficult to determine its true incidence in the general population accurately as they are silent until presentation and, even then, it can pose a diagnostic challenge. They are

believed to account for only 1–2% of acute GI bleeding,<sup>5–5</sup> but are arguably under-recognised rather than being truly rare. The lack of awareness about the Dieulafoy's lesion contributes to its 'rarity', increase in morbidity, and also the previously reported mortality of up to 80% associated with this lesion.<sup>6</sup> Rare or not, the precarious nature of the presentation make it necessary to include them in differential diagnosis of any acute GI haemorrhage. The aim of this article is to present an up-to-date review of literature on Dieulafoy's lesion and the recent trends in its diagnosis, treatment and prognosis.

### **Materials and Methods**

Using Medline, a literature search was performed for papers published in English, using the text words 'Dieulafoy'(s)' and 'gastrointestinal bleeding'. All retrieved papers which were relevant to the study were analysed and the findings are summarised in this review. Overall, 159



Georges Dieulafoy (1839–1911)
The picture is considered to be in the public domain from the National Library of Medicine <a href="http://ihm.nlm.mih.gov">http://ihm.nlm.mih.gov</a>)>

papers were identified and screened; of these, 45 papers were read and analysed in detail.

# Dieulafoy's lesion

Originally described by Gallard in 1884 as 'miliary aneurysms of the stomach', 5,7-10 it was more accurately distinguished by the French surgeon Georges Dieulafoy in 1898 following his study of fatal gastric haemorrhage in three asymptomatic young men. 5,4 He termed these lesions 'exulceratio simplex' based on his belief that these lesions were the early stage of peptic ulceration. 5,7,10,11 In just over 100 years since Dieulafoy's original paper there had been more than 280 reported cases world-wide. 4 The first case of two synchronous bleeding Dieulafoy's lesions, located on the greater curve of the stomach and in the jejunum, was recently reported by Marangoni *et al.*5 in 2008.

# **Pathology**

A normal artery of the GI tract will narrow progressively as it traverses the wall of its end organ.<sup>2</sup> A Dieulafoy's lesion describes what is essentially a histologically normal vessel<sup>5</sup> that has an abnormally large diameter, maintaining a constant width of 1–3 mm.<sup>2,5</sup> It runs a tortuous course within the submucosa<sup>5,8,12</sup> and, typically, the lesion protrudes through a small mucosal defect varying from 2–5 mm,<sup>5</sup> which has fibrinoid necrosis at its base.<sup>8</sup>

The stomach is the most common site for Dieulafoy's lesion.  $^{2,15}$  The mucosal defect occurs most commonly on the lesser curve  $^{5,8,11,14,15}$  with  $80\%^7$  to  $95\%^{15}$  of these lesions being located within 6 cm of the gastro-oesophageal junction.  $^{8,14,17}$  This is attributed to the architecture of the blood supply to the lesser curve of the stomach as the vessels arise

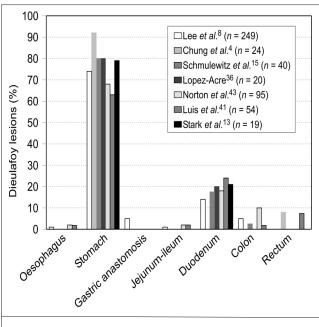


Figure 1 Location of Dieulafoy's lesion reported in literature.

directly from the arterial chain running outwith the lesser curve. In the remainder of the stomach, the blood supply is derived from a submucosal plexus of larger vessels.<sup>18</sup>

Approximately one-third of lesions are extragastric, most frequently in the duodenum followed by the colon. <sup>10</sup> Initially, colonic Dieulafoy's lesions were believed to occur mainly in the right colon but later it has been shown that they occur throughout the colon. <sup>15</sup> Dieulafoy's lesions have also been described in the oesophagus, small intestine, <sup>17</sup> rectum <sup>19</sup> and anal canal. <sup>20</sup> They have been described outside the GI tract with the thirteenth case in the literature of Dieulafoy's lesion of the bronchus described by Gharagozloo *et al.* <sup>21</sup> in 2008 (Figs 1 and 2).

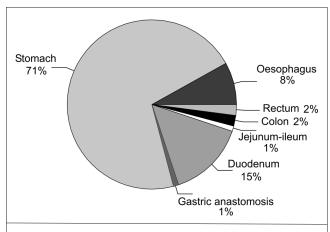


Figure 2 Mean percentage of the location of Dieulafoy's lesion. 4,8,13,15,36,41,43

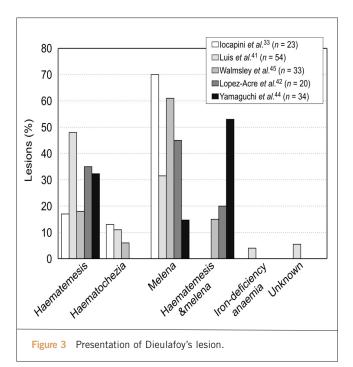
# Table 1 Typical patient profile presenting with Dieulafoy's lesion

- Elderly (M:F = 2:1)
- Multiple co-morbidities
- · Often already hospitalised
- · Often on NSAIDs, aspirin and warfarin
- Presents with massive GI bleeding which might be recurrent
- No previous history of GI pathology

## **Aetiology**

Once believed to be acquired and aneurysmal in nature, <sup>15</sup> pathological reports have failed to associate these lesions to aneurysms, atherosclerosis, arteritis or inflammation. <sup>5,8,10,19</sup> There are cases in the literature of newborns being affected, <sup>5</sup> while rare, this could support the suggestion that these lesions are in fact congenital in nature. <sup>2,5,8,12</sup>

Interestingly, it has been observed from case histories that a large proportion of patients presenting with rupture are already hospitalised. This has led to the suggestion of stress injury and predilection for the infirm. <sup>15,22</sup> While the incidence and aetiology are still uncertain, there is a common patient profile observed from the various case studies. They are twice as common in males than females <sup>3,5,10,14,25,24</sup> and can affect any age group. <sup>5,22</sup> However, they do present



more commonly in the elderly population. <sup>15,22</sup> Co-morbidities are present in 90% of patients, <sup>10,25</sup> most frequently cardiopulmonary dysfunction <sup>8,22</sup> and chronic renal failure. <sup>8</sup> Also, the observation that drugs such as non-steroidal anti-inflammatory drugs (NSAIDs), aspirin and warfarin have been used in over half of patients presenting Dieulafoy's lesion <sup>8</sup> led some researchers to propose a causal link. However, there is little evidence in the literature to support this (Table 1). <sup>5,8,15</sup>

## Theories on spontaneous rupture

Several mechanisms have been proposed to account for the rupture and the subsequent massive haemorrhage. One theory suggests that the pulsations in a large submucosal vessel lead to disruption of the overlying epithelium. This leads to localised ischaemia and exposure to bowel contents which ultimately result in erosion and rupture. 5,10,12,25 Another theory suggests that gastric wear and tear promotes thrombosis within the artery leading to the subsequent necrosis. 5,16

Solid contents in the large bowel and rectum could possibly contribute to the development of mucosal stercoral ulceration over an abnormally dilated submucosal arteriole followed by rupture and haemorrhage. It is also suggested that age-related mucosal atrophy might contribute to the process. The use of NSAIDs or alcohol with resultant mucosal injury has also been suggested but there is no consistent evidence to support this causal relationship.<sup>5,15</sup>

While the exact pathogenesis is poorly understood, the consensus is that there is some form of mucosal erosion or ischaemic injury that is possibly related to ageing or cardiovascular disease which further weakens an intrinsically vulnerable point and unmasks the silent anomaly.<sup>15,25</sup>

#### **Clinical presentation**

Dieulafoy's lesions typically present acutely as massive haemorrhage which is often recurrent<sup>5</sup> and can take the

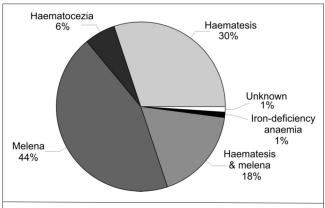


Figure 4 Mean percentage of the presentation of Dieulafoy's lesion.

# Table 2 Diagnostic criteria for Dieulafoy's lesion on endoscopy<sup>4,8,14</sup>

- Active arterial spurting or micropulsatile streaming from a mucosal defect < 3 mm or through normal surrounding mucosa
- Visualisation of protruding vessel with or without bleeding, within a minute mucosal defect or through normal surrounding mucosa
- The appearance of fresh, densely adherent clot with a narrow point of attachment to a minute mucosal defect or to normal appearing mucosa

form of haematemesis, melena, fresh bleeding per rectum or haematochezia (Figs 3 and 4).

# **Investigations**

#### **Endoscopy**

Initial GI endoscopy is effective in diagnosing up to 70% of patients.<sup>5</sup> However, several endoscopies may be required with 6% of patients requiring three or more to establish the diagnosis.<sup>14</sup> Reasons for unsuccessful initial endoscopy were attributed to excessive blood (44%) or the lesion was subtle and overlooked (56%).<sup>10</sup> In the latter situation, aids to endoscopic diagnosis have been suggested. Wright *et al.*<sup>26</sup> have described provocation of bleeding using intravenous bolus of heparin. The use of endoscopic ultrasound has also been used to aid endoscopic diagnosis.<sup>7</sup>

Push enteroscopy, which is an extension of upper GI endoscopy, allows assessment of the small intestine up to a distance of 150 cm from the pylorus. Intra-operative enteroscopy enables direct evaluation of the small intestine with a diagnostic yield of 70–100% in patients with obscure GI bleeding. However, it requires laparotomy or laparoscopy.<sup>6</sup>

Successful localisation of Dieulafoy's lesion has also been achieved by the use of wireless capsule endoscopy,<sup>6,27</sup> which has the benefit of being minimally invasive<sup>6</sup> but it does not allow therapeutic intervention.<sup>28</sup>

The characteristic endoscopic findings of a Dieulafoy's lesion are an isolated protruding vessel surrounded by normal mucosa, which does not have an associated ulcer.<sup>5,11,19</sup> If the lesion is actively bleeding, then blood could be visualised spurting or oozing from the pin-point defect, or in the absence of bleeding a clot without an ulcer might be seen.<sup>5</sup> An agreed diagnostic criterion for Dieulafoy's lesion on endoscopy is composed of three categories which a lesion may fall into as summarised in Table 2.

#### **Angiography**

Angiography is useful when endoscopic methods fail to localise the lesion. It is especially useful for lesions in the colon or rectum where the view could be obscured by active bleeding and poor bowel preparation.<sup>19</sup> There is no specific diagnostic criterion to diagnose a Dieulafoy's lesion on angiography as typical features are variable,29 but the diagnosis is suggested on demonstration of a tortuous and ectactic artery.<sup>5,8</sup> The findings include extravasion of contrast from what is seen as a normal looking blood vessel. For gastric lesions, the demonstration of tortuous vessels in the territory of the left gastric artery lacking early venous return is suggestive. Lesions in the anorectal region can be missed if they are situated below the region supplied by the inferior mesenteric artery. Internal iliac artery visualisation might be necessary in these cases.<sup>25</sup> The use of computed tomography (CT) angiography has also been described in the literature to locate the source of bleeding that cannot be diagnosed by endoscopic methods.<sup>50</sup>

#### Red cell scanning

Technetium-99m labelled red blood cell scans have also been used to identify the location of bleeding Dieulafoy's lesion when endoscopy had failed.<sup>51</sup> It has the advantage that the threshold for detecting extravasation into the gut is only 20% of that required by angiography.<sup>52</sup>

# **Treatment**

There is no consensus on the treatment of Dieulafoy's lesion. <sup>19</sup> Treatment options are dependent on mode of presentation, site of the lesion and available expertise. The evolution of endoscopic methods of haemostasis has markedly reduced the need for surgery in Dieulafoy's lesions.

### **Endoscopic treatment**

Endoscopic methods are the treatment of choice in easily accessible lesions.<sup>15</sup> The reported success rate is in excess of 90%.<sup>4,5,25,24</sup> Endoscopic haemostatic procedures can be classified into three groups: (i) thermal – electrocoagulation, heat probe coagulation and argon plasma coagulation; (ii) regional injection – local epinephrine injection and sclerotherapy; and (iii) mechanical – banding and haemoclip.<sup>2,4,11,15,55</sup>

Each technique has both advantages and disadvantages related to the haemostatic mechanism and the technical procedure itself with varying success rates.<sup>4</sup> There is some evidence in the literature to suggest that endoscopic mechanical haemostatic methods are more effective and successful in achieving haemostasis when compared to injection or thermal treatment methods.<sup>4</sup> Also, combined endoscopic therapies are reported to have lower re-bleeding rate when compared to endoscopic monotherapy.<sup>5,8</sup>

Endoscopic ultrasound (EUS) guided treatment of the underlying vessel have been reported. EUS may help detection of the aberrant vessel in the submucosa. It also can confirm ablation of a Dieulafoy's lesion after injection therapy or elastic band ligation by confirming absence of blood flow.<sup>8</sup>

Due to the small size of the lesion, which is often difficult to locate, tattooing of the lesion is advocated by some to allow for rapid identification of the lesion should re-bleeding occur. Tattooing of the lesion will be particularly useful during minimally invasive surgical intervention.

#### Angiography and embolisation

Angiography can be used to embolise actively bleeding Dieulafoy's lesions. This is useful to deal with lesions that fail to respond to endoscopic methods of haemostasis. <sup>15</sup> However, embolisation carries the risk of ischaemia to the area supplied by the relevant artery. <sup>29</sup> If the bleeding lesion is supplied by multiple collaterals, extensive embolisation may be required, <sup>9</sup> which sometimes make it an unsuitable choice of treatment because of the risk of the resultant ischaemia. Table 3 summaries the situations in which selective embolisation may be considered as the treatment of choice in actively bleeding Dieulafoy's lesions in the GI tract. Angiography with embolisation is the preferred treatment for bronchial Dieulafoy's lesions. <sup>12</sup>

#### Surgical treatment

Surgical resection was historically the first-line treatment of Dieulafoy's lesions, usually taking the form of gastrotomy and wide-wedge resection or gastrectomy. This has now been overtaken by advances in endoscopic procedures. Surgical resection is currently reserved for the 5% of cases that are refractive to endoscopic or angiographic methods. Surgical procedures currently employed include under-running of the lesion or a wedge resection of the affected section of gut. 5.8

It is suggested by some authors that surgical resection is still preferable, especially in lesions that may be exposed to hard stool, perhaps increasing their likelihood of re-bleeding.<sup>20</sup>

#### **ROLE OF MINIMALLY INVASIVE SURGERY**

While the majority of patients requiring surgery would have undergone laparotomy, more recently laparoscopic surgery for removal of these lesions has been described.<sup>29</sup> Laparoscopic surgery is an attractive option for treating these lesions as it offers a cure while being minimally invasive to the patient. However, successful laparoscopic resection relies on accurate localisation of the bleeding.<sup>55</sup>

There are several case reports in the literature describing successful laparoscopic wedge resection of bleeding Dieulafoy's lesions in the jejunum<sup>6</sup> and in the stomach following pre-operative or intra-operative localisation.<sup>54,56–58</sup>

# Table 3 Indications for selective angiography in bleeding Dieulafoy's lesion<sup>10</sup>

- Failed endoscopic therapy
- Lower GI bleeding or lesions beyond reach of therapeutic endoscopy
- · Poor candidates for surgery

Laparoscopic transgastric resection of Dieulafoy's lesion involving an anterior gastrotomy and resection of the lesion, without the need for endoscopy, has been performed.<sup>59</sup> However, such procedures involve an unnecessary gastrotomy.

Accurate intra-operative localisation of these lesions could represent a challenge.<sup>55</sup> Several methods have been described to ensure precise localisation. This was first tackled by intra-operative endoscopy that allowed for real-time localisation. Following induction of pneumoperitoneum, upper GI endoscopy was then performed. The laparoscopic light was then dimmed or turned off. The gastroscope was used to visualise and locate the bleeding lesion. The area was marked by clips or suture laparoscopically and resection was then performed.<sup>54</sup> Mixter *et al.*<sup>40</sup> described the use of combined endoscopy and laparoscopy to identify and ligate the artery that feeds the bleeding Dieulafoy's lesion without the need for resection.

Laparoscopic wedge resection is reported to have lower re-bleeding rates when compared with oversewing the Dieulafoy's lesion. <sup>56</sup> However, wedge resection may not be feasible for lesions situated within 6 cm of the gastro-oesophageal junction; in these circumstances, oversewing the lesion or laparoscopic clip placement on the vessel feeding the lesion may be a better option. <sup>56,40</sup>

Alva *et al.*<sup>54</sup> described pre-operative localisation of the lesion by both tattooing with India ink and using clips, thus allowing accurate localisation whilst alleviating the need for intra-operative endoscopy. The most frequent difficulty with clip placement is that it could fall out prior to surgical intervention. Clips used for pre-operative localisation of the lesion(s) have not been known or reported to interfere with the stapling devices or to cause disruption of the staple line during wedge resection.<sup>54</sup>

# Re-bleeding

The risk of re-bleeding from Dieulafoy's lesions is reported to be between 9–40% and is higher in endoscopic monotherapy compared with combined endoscopic therapies.<sup>5,8</sup> It should be noted that the endoscopic methods of haemostasis are the preferred treatment in re-bleeding

#### Table 4 Key facts about Dieulafoy's lesions

- · Should be included in the differential diagnosis of obscure GI bleeding
- Accounts for less than 2% of all cases of acute GI bleeding
- Life-threatening bleeding occurs in 10% of cases
- · Described in all parts of the GI tract and all age groups
- · Awareness and careful endoscopy are essential for diagnosis
- Repeat endoscopies may be necessary to establish diagnosis
- Therapeutic endoscopy is successful in over 90% of cases
- Tattooing at initial endoscopy is advisable to aid localisation if re-bleeding occurs
- Endoscopic methods of haemostasis are the preferred treatment in re-bleeding
- Angiography and embolisation is reserved for lower GI tract lesions and failed therapeutic upper GI endoscopy
- There are recent encouraging reports on the successful use of laparoscopic surgery
- Mortality decreased from 80% to 8.6%

from a Dieulafoy's lesion.<sup>25</sup> Re-bleeding has also been reported after embolisation because of collateral circulation or incomplete embolisation of the feeding artery.<sup>10</sup>

#### **Prognosis**

Advances in endoscopy have increased the detection rate of Dieulafoy's lesions and have significantly decreased the mortality from 80%. 8.25 to 8.6%. 10 Improvement in the prognosis might also be explained by the increasing use of endoscopic rather than surgical intervention to control the bleeding which is reported to be effective in more than 90% of patients. 4 Luis *et al.* 41 reported that the outcome of acute GI bleeding due to Dieulafoy's lesion has more favourable outcome when compared to acute bleeding from gastric or duodenal ulcer.

#### **Conclusions**

Managing active Dieulafov's lesion poses a diagnostic and therapeutic challenge. Dieulafoy's lesion should be included in the differential diagnosis of obscure GI bleeding in all age groups. GI endoscopy has proven to be highly effective diagnostic and therapeutic tool in the majority of patients. Increasingly, angiography is being accepted as a valuable alternative to endoscopy, depending on the clinical situation and the initial outcomes of therapeutic endoscopy. Currently, surgical intervention is kept for failure of therapeutic endoscopic and angiographic interventions and it should be guided by pre-operative localisation. Surgical intervention remains the preferred treatment for lesions of the distal gastrointestinal tract. There are recent encouraging reports on the successful use of laparoscopic surgery in managing symptomatic Dieulafoy's disease (Fig. 5 and Table 4).

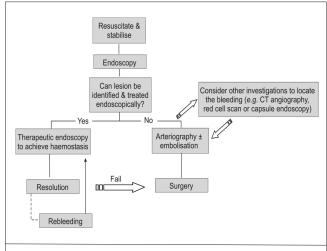


Figure 5 Treatment algorithm for Dieulafoy's lesion.

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