CASE REPORT

Dieulafoy's Lesion In The Oesophagus: A Rare Cause Of Upper Gastrointestinal Bleeding

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SUMMARY

A Dieulafoy's lesion is defined as a dilated, aberrant, submucosal artery that erodes overlying mucosa in the absence of an underlying ulcer, aneurysm or intrinsic mural abnormality. It is a rare cause of upper gastrointestinal (GI) bleed with a very high mortality rate if it goes unidentified. It is most commonly located in the lesser curvature of the stomach but is extremely rare in the oesophagus. We are reporting a 55-year-old man who had massive haematemesis. Emergent endoscopy showed Dieulafoy's lesion in the distal oesophagus. It was sclerosed using endoscopic therapy. Patient's symptoms of GI bleeding resolved, and he was discharged home in a stable manner. Although a Dieulafoy's lesion is exceedingly rare in the oesophagus, it is associated with a high mortality if undiscovered. Its amenability to life-saving endoscopic therapy prompts us to keep this as a possible differential diagnosis of an upper GI bleed.

BACKGROUND

A Dieulafoy's lesion is defined as a dilated, aberrant, submucosal artery that erodes overlying mucosa in the absence of an underlying ulcer, aneurysm or intrinsic mural abnormality.¹ It accounts for ~1.5% of upper gastrointestinal (GI) bleeds. Its insidious onset, tendency to cause intermittent but severe bleeding and difficulty of endoscopic diagnosis has led to a very high mortality rate of up to 80%.² In just over 100 years since Dieulafoy's original case report, there have been about 280 published case reports in the literature, most commonly located in the lesser curvature of the stomach but rarely in the

Figure 2 Endoscopic post-treatment image showing complete obliteration of Dieulafoy lesion at 10 o'clock position.

oesophagus.³ We aim to show a case of a 55-year-old man who presented with upper GI bleed caused by Dieulafoy's lesion and to illustrate the significance of endoscopic therapy in these lesions.

CASE PRESENTATION

A 55-year-old man with a history of hypertension and hyperlipidaemia presented to the emergency room after a presyncopal episode at home. He presented with 3 days of melena associated with generalised fatigue. On presentation, he had a significant amount of haematemesis followed by a syncopal episode. He reported that he never had an esophagogastroduodenoscopy (EGD) before and denied a history of GI bleeds in the past. His only medication was aspirin 81 mg daily. He denied use of non-steroidal anti-inflammatory drugs, anticoagulants or alcohol. His last colonoscopy was about 5 years ago. Vitals showed a heart rate of 120 beats per minute and blood pressure of 59/39 mm Hg. On examination, the patient was alert and oriented. The abdomen was soft, non-distended with no rebound. There was a significant amount of blood around the mouth and blood stains on his clothes. Nasogastric suctioning, with saline lavage, revealed frank red blood on return. Haemoglobin was initially 9.5 g/dL and trended down to 7.4 g/dL after 2 hours.

DIFFERENTIAL DIAGNOSIS

On initial presentation, the patient's differential diagnosis was broad. The various considerations were peptic ulcer disease, oesophageal varices, Mallory-Weiss syndrome, bleeding disorders,

Figure 1 Endoscopic image of a blood vessel protruding at seven o'clock position, without surrounding ulceration consistent with Dieulafoy's lesion.

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Figure 3 Repeat EGD which was performed 8 weeks postdischarge, negative for any evidence of recurrence or rebleeding.

angiodysplasia, a malignancy of the upper GI tract, arteriovenous malformations and gastric antral vascular ectasia. His coagulation profile was within normal limits, and endoscopy only showed visible submucosal vessel in the distal oesophagus suggestive of a Dieulafoy's lesion, thus ruling out all other differentials.

TREATMENT

The patient was emergently resuscitated with intravenous fluids and two units of packed red blood cell transfusion. Emergent endoscopy was performed in the operating room which revealed a visible submucosal vessel in the distal oesophagus suggestive of a Dieulafoy's lesion (figure 1). It was sclerosed using an injection of epinephrine, and complete obliteration of the vessel was achieved using gold probe cautery (figure 2). He was subsequently transferred to the intensive care unit for close monitoring.

OUTCOME AND FOLLOW-UP

Patient's symptoms of GI bleeding resolved. He was vitally stable and was tolerating a full oral diet. The decision was made to discharge the patient home, and he was recommended to follow-up outpatient with gastroenterology for subsequent endoscopy in 8 weeks. Repeat EGD which was performed 8 weeks post discharge was negative for any evidence of recurrence or re-bleeding (figure 3).

DISCUSSION

Dieulafoy's lesions were first reported by Gallard in 1884 and were more precisely reviewed by Georges Dieulafoy in 1898, a French surgeon who erroneously believed that these lesions were early peptic ulcers.⁴ It is an unusual cause of GI haemorrhage, accounting for less than 2% of acute GI bleeding episodes. The incidence is higher in older patients and male population. These lesions are commonly called calibre-persistent arteries due to its pathophysiology since these lesions maintain constant arterial calibre, of approximately 1–3 mm, which is up to 10-fold larger than the maximal calibre of such submucosal vessels.^{5 6} They are most commonly found in the stomach (prevalence 75%–90%) but are exceedingly rare in the oesophagus (prevalence 8%).^{3 7} Previously reported cases have been listed in table 1.^{3 7–14} Patients most commonly present with haematemesis and melena.¹⁵ Although originally thought not to be a radiologically diagnosable entity, Dieulafoy's lesions can be seen on enhanced CT of the abdomen.¹⁶ However, endoscopy has become the mainstay of diagnosis and treatment.¹⁷ The endoscopist may choose to use clipping, thermo-coagulation or epinephrine injection into the lesion as illustrated in our case. The mortality rate has dropped from 80% to 8.6% since endoscopy has been employed in the identification of causes in GI bleeds, and it has been reported to be effective in more than 90% of patients. Endoscopy has 70% sensitivity, and if it fails to diagnose the lesion, angiography can be used to embolise the lesion.³ Giles *et al* reported TC-325 also known as Hemospray can be used as a rescue therapy when haemostasis cannot be achieved using standard interventions and as the first line in cases of diffuse bleeding that cannot be controlled using the latter. In cases of spurting haemorrhage, usage of TC-325 has led to rebleeds, so it is not

Table 1 Previously reported cases of Dieulafoy's lesions of the oesophagus^{3 7–14}

Author	Country	Age/gender	Presenting symptom	Location of the Dieulafoy's lesion	Intervention	Outcome
Malliaras ³	USA	55/M	Haematemesis	Distal portion of the oesophagus	Epinephrine injection sclerosis and resolution clip placement	No recurrence
Scheider ⁷	USA	NA	NA	NA	NA	NA
Peixoto <i>et al</i> ⁸	Brazil	66/M	Haematemesis	Mid-oesophagus	metallic clip	No recurrence
Rikke Kjærgaard <i>et al</i> ¹²	USA	48/M	Haematemesis, melena, stomach pain	Distal oesophagus	None	Death
Thimmapuram <i>et al</i> ¹⁰	USA	38/M	Aspiration pneumonia, melena, haematemesis	Mid-oesophagus	Epinephrine injection followed by haemoclip application	No recurrence
Ertekin <i>et al</i> ¹¹	Turkey	25/F	Haematemesis and melena	Distal oesophagus	Endoscopic band ligation	No recurrence
Abraham <i>et al</i> ¹³	India	54/M	Painless haematemesis	NA	Epinephrine injection and argon plasma coagulation	No recurrence
Inayat <i>et al</i> ¹⁴	USA	53/M	Haematemesis and melena	Distal oesophagus	Epinephrine injection followed by haemoclip application	No recurrence
Inayat <i>et al</i> ¹⁴	USA	68/M	Haematemesis and melena	Distal oesophagus	Epinephrine injection followed by haemoclip application	No recurrence
Inayat <i>et al</i> ¹⁴	USA	80/M	Haematemesis and melena	Distal oesophagus	Epinephrine injection followed by haemoclip application	No recurrence
Divyesh <i>et al</i> (current case)	USA	55/M	Haematemesis	Distal oesophagus	Sclerosed using an injection of epinephrine, and obliteration using gold probe cautery	No recurrence

Learning points

- Although a Dieulafoy's lesion is exceedingly rare in the oesophagus, it should be kept as a possible differential diagnosis of an upper gastrointestinal bleed since it has high mortality rate up to 80% if not treated promptly.
- Endoscopy has become the mainstay of diagnosis and treatment. If minimally invasive approaches fail, surgery is reserved as a last resort.
- TC-325 also known as Hemospray can be used as a rescue therapy when standard interventions fail to achieve complete haemostasis; however, it is not effective for spurting haemorrhage.

recommended in those cases. Yau *et al* recommended its use as a temporary measure or a bridge toward more definitive therapy.^{18 19} Thirty-day rebleeding was significantly lower with Doppler-guided endoscopic haemostasis compared with standard visually guided haemostasis in patients with severe upper GI haemorrhage from ulcers or other lesions.²⁰ If minimally invasive approaches fail, surgery is reserved as a last resort.³ Although a Dieulafoy's lesion is exceedingly rare in the oesophagus, it is associated with a high mortality if undiscovered. Its amenability to life-saving endoscopic therapy prompts us to keep this as a possible differential diagnosis of an upper GI bleed.

Contributors DRN: substantial contributions to the conception or design of the work, or the acquisition, analysis or interpretation of data. DRN and MPR: drafting the work or revising it critically for important intellectual content. DRN, MPR and JY: final approval of the version published. HL-F: agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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