Case Report

Esophageal Dieulafoy Lesion: A Rare and Potentially Fatal Entity

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A 35-year-old gentleman presented acutely to the emergency department with large volume haematemesis and melena. Following adequate initial resuscitation, the patient underwent emergency upper gastrointestinal endoscopy which revealed a dilated tortuous submucosal vessel which was actively bleeding at the midpoint of the esophagus. This was consistent with a Dieulafoy lesion. However, its position in the midpoint of the esophagus is rare. Our patient recovered well following intervention.

1. Introduction

Dieulafoy lesions are a rare but important cause of massive gastrointestinal (GI) bleeding. They account for less than 2% of gastrointestinal bleeds but have a significant risk of morbidity and mortality secondary to delayed diagnosis and treatment [1]. The underlying pathophysiology of these lesions is poorly understood. They are classified as dilated tortuous histologically normal submucosal arteries [2]. They classically occur in the stomach on the lesser curvature but have been reported in the colon, duodenum, and rarely in the esophagus as in our report [3]. Patients typically present with acute onset large volume haematemesis and/or melena. Unlike bleeding from a Mallory-Weiss tear, bleeding from a Dieulafoy lesion is not typically associated with preceding retching or vomiting [4]. Diagnosis usually involves endoscopy although there is a growing role for angiography particularly if suspicion remains following a normal endoscopic evaluation. Endoscopy has the added advantage of being therapeutic as well as diagnostic [5].

2. Case Presentation

A 35-year-old gentleman presented acutely to the emergency department with haematemesis and melena. He had no past medical or surgical history and was not taking any regular medications. He worked as a secondary school teacher. Of note, he had no risk factors or diagnosis of liver disease, cirrhosis, or portal hypertension. The haematemesis and melena occurred acutely with no preceding vomiting. He had been attending a choir rehearsal with his class of students when the bleeding occurred. On clinical examination, he was noted to be of normal BMI. He was hypotensive and tachycardic in the emergency department, and laboratory investigations were significant for a haemoglobin of 8 g/dl. The massive transfusion protocol was activated, and he was prepared for emergency endoscopy. At esophagogastroduodenoscopy, he was found to be actively bleeding from a dilated tortuous vessel at the midpoint of the esophagus. The bleeding was pulsatile in nature. The findings were consistent with a Dieulafoy lesion (Figure 1).

He remained stable with no further bleeds. The patient was counselled on the need for further evaluation to determine whether a definitive procedure was required to excise this vessel completely given its high risk of rebleeding. However, he refused further investigations/interventions. He was discharged home well.

3. Discussion

Dieulafoy’s lesion was initially described by Gallard in 1884 as “military aneurysms of the stomach.” However, it became synonymous with the French surgeon Dr Georges Dieulafoy in 1898 when he more accurately characterised them during his investigation of fatal upper gastrointestinal haemorrhage in three young asymptomatic men [6].
These lesions are classically found in the stomach predominantly on the lesser curvature. The precise reason for this remains controversial. Some have attributed it to the fact that the submucosal blood supply in this region of the stomach arises directly from the right and left gastric arteries which run adjacent to the lesser curvature suspended in the hepatogastric ligament and so do not have to transit a significant distance prior to reaching their end-organ [7]. However up to 1/3 of Dieulafoy lesions may occur outside the stomach, predominantly in the duodenum and colon. The aetiology for these lesions, at sites other than the lesser curvature of the stomach, remains unclear [8].

The underlying pathophysiology for the development of these lesions is poorly understood. These lesions are histologically normal, when analysed in resected specimens, but maintain a larger calibre in comparison with adjacent blood vessels. It is well documented that a normal artery of the gastrointestinal tract will narrow as it traverses the wall of the organ it is supplying [9]. This, however, is not the case in Dieulafoys lesions as these vessels maintain the same patency throughout their course. As these vessels remain larger in diameter within the submucosa, they may cause damage to the overlying mucosa secondary to forceful pulsation. Should the overlying mucosa breakdown these vessels are then exposed and subject to trauma and thus haemorrhage, often without any preceding symptoms [10].

These lesions are reported as being rare in the esophagus [11]. A literature review performed on PubMed using the following search string: ((Dieulafoy Lesion) AND (Dieulafoy's Lesion)) AND (Oesophagus OR esophagus) yielded 32 results. On review of the abstracts of these reports, 10 were found to pertain to Dieulafoy’s lesions in the oesophagus presenting with upper GI bleeding and were published in the English language with abstracts available. Table 1 reviews these previous reports. It remains unclear, however, whether this is due to under diagnosis. These vascular abnormalities can present with massive upper gastrointestinal haemorrhage and can be fatal if haemostasis
| Publication | Demographics | Presentation | Site of lesion | Treatment and outcome |
|-------------|--------------|--------------|----------------|-----------------------|
| Inayat, F., Ullah, W., Hussain, Q. and Hurairah, A., 2017. | 53 years old caucasian male. | 1 week history of melena Admitted following RTA, on day 2 of admission developed fresh rectal bleeding | Distal oesophagus | Adrenaline injection and haemoclips |
| | 68 years old Caucasian male | | | Haemoclips |
| | 80 years old Caucasian male | 2-day history of haematemesis | Distal oesophagus | All three patients recovered well and were discharged home with no further intervention |
| Nemakayala, D.R., rai, M.P., Yam, J.L. and Laird-Fick, H., 2018. | 55 years old Caucasian male. | 3-day history of melena and a presyncopal episode | Distal oesophagus | Adrenaline injection and gold probe cautery |
| | 25-year-old female | Haematemesis and melena | Distal oesophagus | Discharged home with no further intervention |
| Ertekin, C., Barbaros, U., Taviloglu, K., Guloglu, R. and Kasoglu, A., 2002. | 61-year-old male | Haematemesis | Upper oesophagus | Haemoclip |
| El Hajj, I.I., Malik, S. and McGrath, K.M., 2010. | 24-year-old male | Haematemesis | OGJ | Discharged home with no further intervention |
| Yoshida, T., Adachi, K., Tanioka, Y., Sasaki, T., Ono, S., Hanada, H., Esaki, T., Yagawa, T., Takeo, S., Saiki, Y. and Harada, T., 2004. | 74-year-old male | Haematemesis and melena | Distal oesophagus | Discharged home with no further intervention |
| Ho, K.M., 2004. Use of Sengstaken-Blakemore tube to stop massive upper gastrointestinal bleeding from Dieulafoy’s lesion in the lower oesophagus. | 71-year-old male | Haematemesis | Distal oesophagus | Adrenaline injection and bipolar diathermy |
| | | | | Further bleed controlled with Sengstaken–Blakemore tube |
| | | | | Repeat OGD did not identify a bleeding point |
| | | | | Discharged home with no further intervention |
| Malliaras, G.P., carollo, A. and Bogen, G., 2016. Esophageal Dieulafoy’s lesion: An exceedingly rare cause of massive upper GI bleeding. Journal of surgical case reports, 2016 [6], p.rjw074. | 55-year-old male | Haematemesis | Distal oesophagus | Adrenaline and haemoclip placement |
| | | | | Discharged home with no further intervention |
| Thimmapuram, J., Shah, M. and Srour, J., 2011. Esophageal Dieulafoy lesion: An unusual cause of GI bleeding. Gastrointestinal endoscopy (Print), 73 [5], pp. 1055–1056. | 55-year-old male | Haematemesis | Mid oesophagus | Adrenaline injection and haemoclip application |
| | | | | Discharged home with no further intervention |
cannot be obtained. If identified they can often be managed effectively with standard endoscopic techniques. These lesions are under recognized and should be considered in all cases of acute GI bleeding.

Data Availability

The data used to support the findings of this study are included within the article.

Conflicts of Interest

The author declares no conflicts of interest.

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| Publication | Demographics | Presentation | Site of lesion | Treatment and outcome |
|-------------|--------------|--------------|----------------|------------------------|
| Abraham, P., Mukerji, S.S., Desai, D.C. And Joshi, A.G., 2004. Dieulafoyo lesion in mid-esophagus with esophageal varices. Indian journal of gastroenterology: official journal of the Indian Society of Gastroenterology, 23 [6], pp. 220–221. | 54-year-old male | Haematemesis | Distal oesophagus | Band ligation |