ACG CASE REPORTS JOURNAL

CASE REPORT | ESOPHAGUS

Large Esophageal Intramural Hematoma After Solid Food Ingestion in a Patient Without Identifiable Inherited or Acquired Coagulopathy

Michael Ladna, MD[1], and John George, MD[2]

[1]*Department of Hospital Medicine, University of Florida, Gainesville, FL*
[2]*Department of Gastroenterology, Hepatology, and Nutrition, University of Florida, Gainesville, FL*

ABSTRACT

We present the case of an elderly man without any significant medical history and not on any anticoagulant or antiplatelet therapy who presented with severe epigastric abdominal and substernal chest pain shortly after eating a baguette. He was found to have a large 15 cm dissecting intramural hematoma of the esophagus. He was managed conservatively with proton pump inhibitors. He remained stable throughout his hospitalization without evidence of an acute blood loss anemia and was discharged home. Repeat esophagogastroduodenoscopy 8 weeks after discharge showed a 5 mm scar with complete resolution of the dissecting intramural hematoma of the esophagus.

KEYWORDS: intramural hematoma of esophagus; Mallory-Weiss tear; esophageal trauma

INTRODUCTION

Dissecting intramural hematoma of the esophagus (DIHE) is a rare pathology that can be misdiagnosed as cardiac-related chest pain. This can lead to delay in diagnosis or worse if therapeutic anticoagulation with heparin is initiated should a misdiagnosis of acute coronary syndrome be made. Although this condition tends to occur in patients with some form of inherited or acquired coagulopathy, it can also occur without an identifiable coagulopathy such as in our case. Diagnosis of DIHE should prompt an immediate workup for an underlying coagulopathy such as hemophilia.

CASE REPORT

A 70-year-old man with no significant medical history presented to the emergency department with chief concern of 1 day of progressively worsening epigastric abdominal pain and substernal chest pain along with dysphagia and odynophagia that started suddenly shortly after eating a baguette. He was hemodynamically stable on initial evaluation. Physical examination was significant for mild epigastric tenderness without rigidity, guarding, or rebound tenderness. Symptomatic supportive medical management with antiemetics and analgesics was administered. Initial laboratory testing revealed unremarkable white blood cell count, hemoglobin, basic metabolic panel, and liver function tests. Chest computed tomography showed a fluid filled distal esophagus with focal thickening. He underwent an esophagogastroduodenoscopy (EGD) for further evaluation.

EGD revealed a large DIHE with a total length of 15 cm (extending 25–40 cm from incisors). No food particles were visualized in the esophagus. A 20 mm nonbleeding Mallory-Weiss tear with stigmata of recent bleeding was identified at the gastroesophageal junction. Hematin and old blood were noted in the mid esophagus and stomach. The duodenum was not examined. Prothrombin time, international normalized ratio, and activated partial thromboplastin time were all within normal limits. He was not on any anticoagulant or antiplatelet medications. He denied any family or personal history of hemophilia. The patient was kept with nothing by mouth for 12 hours, and intravenous pantoprazole twice daily was continued for 24 hours. Thoracic surgery and interventional radiology were consulted. Surgical or angiographic interventions were not required. His diet was advanced to full liquid the following

ACG Case Rep J 2023;10:e01067. doi:10.14309/crj.0000000000001067. Published online: June 8, 2023

Correspondence: Michael Ladna, MD (michael.ladna@medicine.ufl.edu).

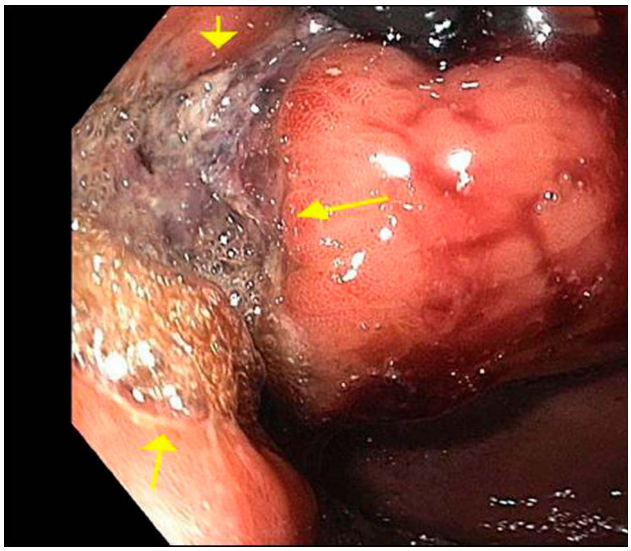

Figure 1. Endoscopy showing Mallory-Weiss tear.

day and then mechanical soft the day after that, both of which he tolerated well. His hemoglobin remained stable throughout the hospitalization. Pantoprazole 40 mg twice daily was transitioned from intravenous to oral, and he was discharged after 3 days of hospitalization with a plan for outpatient endoscopy to document healing (Figure 1).

He underwent repeat EGD 8 weeks after discharge to assess healing. Repeat EGD showed a 5 mm scar in the middle third of the esophagus, 25–30 cm from the incisors. The scar tissue was healthy in appearance. Diffuse mild mucosal changes characterized by congestion were found in the entire esophagus. The Z-line was regular and was found 40 cm from the incisors. The entire examined stomach was normal (Figure 2).

DISCUSSION

DIHE is an uncommon condition characterized by collection of blood in the wall of the esophagus. It is part of a spectrum of esophageal injuries which include local mucosal tears (Mallory-Weiss syndrome) and full-thickness rupture (Boerhaave syndrome). It tends to present with sudden-onset chest pain, abdominal pain, dysphagia, odynophagia, and hematemesis. A systemic analysis of all DIHEs in 2000 showed that 80% of patients had 2 of 3 typical symptoms of chest pain, hematemesis, and dysphagia or odynophagia, with 50% of cases being associated with sudden pressure changes in the esophagus or direct trauma to the esophagus.[1] DIHE can be spontaneous[2] or secondary to sudden change in intrathoracic and intraesophageal pressure such as during forceful vomiting, retching, or coughing or as a result of direct trauma or foreign body/toxic substance ingestion. There tends to be a coagulopathy present whether that be acquired by using an anticoagulant or be inherited. The presence of DIHE could be a clue for an underlying coagulopathy such as hemophilia.

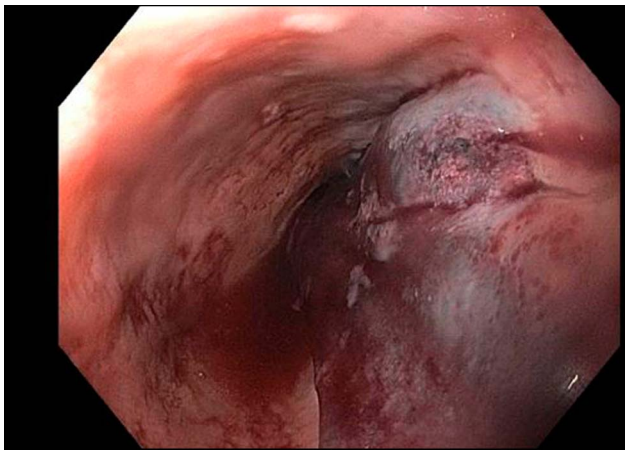

Figure 2. Endoscopy showing large 15cm intramucosal hematoma.

DIHE has been reported in the setting of warfarin use with a therapeutic international normalized ratio of 2.5,[3] chronic idiopathic thrombocytopenia,[4] hemophilia A,[5] herpes esophagitis,[6] trauma from food ingestion,[7] trauma from cardioversion,[8] acquired factor XI deficiency in a patient with relapsed T-cell prolymphocytic leukemia after allogenic hematopoietic-cell transplantation,[9] esophageal duplication cyst,[10] after surgical delivery of preterm twins in a preeclamptic woman on therapeutic enoxaparin due to multiple venous thromboembolism risk factors,[11] and after injection sclerotherapy.[12]

Most cases are benign and are effectively treated with conservative measures with resolution of symptoms typically within 1–2 weeks.[13] Conservative measures include nothing by mouth, intravenous fluid resuscitation, acid suppression with proton pump inhibitors, and correction of any coagulopathies if present. A minority of cases may require intervention by using a surgical approach or embolization with interventional radiology because of severe bleeding. A review from 2009 which included 119 patients found that major bleeding occurred in 11 cases (9.2%) and was defined as either more than 500 mL of hematemesis with hypovolemic shock or bleeding that required at least 4 units of pRBCs.[14]

Because most DIHEs resolve with conservative measures, use of surgery or embolization is especially rare and limited to individual case reports. One case report detailed the successful use of emergent thoracotomy for the treatment of massive bleeding from DIHE due to an active arterial hemorrhage.[15] Another case report of spontaneous DIHE with recurrent massive bleeding in the setting of coagulopathy from decompensated liver cirrhosis refractory to correction with fresh frozen plasma achieved hemostasis by embolization with glue and lipoidal of the esophageal branch of the left gastric artery.[14] There was also a case of achalasia and DIHE, with both pathologies being successfully treated with peroral endoscopic myotomy. There were no adverse events reported. On 2-month follow-up, dysphagia was markedly improved with complete epithelialization of the

DIHE on EGD. At 1-year follow-up, the patient did not have recurrence of DIHE.[16]

It is important to be aware of this pathology because it is often mistaken for cardiac chest pain leading to a delay in diagnosis. The outcomes for these patients could be potentially disastrous if misdiagnosed as unstable angina because the use of therapeutic heparin could rapidly worsen the hematoma and lead to massive blood loss.

DISCLOSURES

Author contributions: The case report section was written by J. George with contributions and edits by M. Ladna. The discussion section was written by M. Ladna with contributions and edits by J. George. M. Ladna is the article guarantor.

Financial disclosure: None to report.

Informed consent was obtained for this case report.

Received November 11, 2022; Accepted May 5, 2023

REFERENCES

1. Cullen SN, McIntyre AS. Dissecting intramural haematoma of the oesophagus. *Eur J Gastroenterol Hepatol*. 2000;12(10):1151–62.
2. Syed TA, Salem G, Fazili J. Spontaneous intramural esophageal hematoma. *Clin Gastroenterol Hepatol*. 2018;16(2):e19–20.
3. Alirezaei T, Irilouzadian R, Pirsalehi A, Nekooghadam SM. Intramural esophageal hematoma mimicking acute coronary syndrome: Complication of warfarin use in a mechanical heart valve patient. *Case Rep Med*. 2022; 2022:5636989.
4. Sharma K, Wang Y. Submucosal esophageal hematoma precipitated by chronic idiopathic thrombocytopenic purpura. *Radiol Case Rep*. 2017; 12(2):278–80.
5. Sugimura K, Ishii N. Esophageal hematoma mimicking a large esophageal polyp: A diagnostic clue of acquired hemophilia A. *Mayo Clin Proc*. 2019; 94(10):2142–3.
6. Takemoto K, Atagi K. Intramural esophageal hematoma with herpes esophagitis. *Intern Med*. 2022;61(6):923.
7. Sharma B, Lowe D, Antoine M, Shah M, Szyjkowski R. Intramural esophageal hematoma secondary to food ingestion. *Cureus*. 2019;11(9):e5623.
8. Hess U, Brücher BL, Cavallaro A, Hannig C, Stein HJ, Ott R. Intramural esophageal hematoma after cardioversion. *Dis Esophagus*. 1997;10(3): 225–8.
9. Harada N, Makuuchi Y, Kuno M, et al. Intramural esophageal hematoma precipitated by acquired factor XI deficiency in a patient with relapsed T cell prolymphocytic leukemia after allogeneic hematopoietic cell transplantation. *Ann Hematol*. 2022;101(11):2567–9.
10. Lee HS, Jeon HJ, Song CW, et al. Esophageal duplication cyst complicated with intramural hematoma: Case report. *J Korean Med Sci*. 1994;9(2): 188–96.
11. Garzon S, Zanconato G, Zatti N, Chiarioni G, Franchi M. Intramural hematoma of the esophagus complicating severe preeclampsia. *Case Rep Obstet Gynecol*. 2017;2017:6304194.
12. Lukashok HP, Robles-Medranda C, Santana MdA, Costa MHM, Borges AdA, Zaltmani C. Intramural esophageal hematoma after elective injection sclerotherapy. *Arq Gastroenterol*. 2009;46(4):279–83.
13. Steadman C, Kerlin P, Crimmins F, et al. Spontaneous intramural rupture of the oesophagus. *Gut*. 1990;31(8):845–9.
14. Shim J, Jang JY, Hwangbo Y, et al. Recurrent massive bleeding due to dissecting intramural hematoma of the esophagus: Treatment with therapeutic angiography. *World J Gastroenterol*. 2009;15(41):5232–5.
15. Folan RD, Smith RE, Head JM. Esophageal hematoma and tear requiring emergency surgical intervention. A case report and literature review. *Dig Dis Sci*. 1992;37(12):1918–21.
16. Fujiyoshi Y, Fujiyoshi MRA, Kimura R, et al. Achalasia with esophageal intramural hematoma treated by per-oral endoscopic myotomy (POEM). *DEN open*. 2022;2(1):e70.