Recurrent massive bleeding due to dissecting intramural hematoma of the esophagus: Treatment with therapeutic angiography

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Abstract

Spontaneous or traumatic intramural bleeding of the esophagus, which is often associated with overlying mucosal dissection, constitutes a rare spectrum of esophageal injury called dissecting intramural hematoma of the esophagus (DIHE). Chest pain, swallowing difficulty, and minor hematemesis are common, which resolve spontaneously in most cases. This case report describes a patient with spontaneous DIHE with recurrent massive bleeding which required critical management and highlights a potential role for therapeutic angiography as an alternative to surgery.

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INTRODUCTION

Dissecting intramural hematoma of the esophagus (DIHE) is a rare but well-known esophageal injury[1,2]. It is characterized by a concentric or eccentric intramural hematoma associated with dissection of the esophageal wall[2]. Forceful vomiting, mechanical insult, and underlying coagulopathy are common causes. It can also occur spontaneously without any evident cause[2]. DIHE is considered a benign disease. The hemorrhage from DIHE does not have clinically significant consequences, although in rare cases massive bleeding with hypovolemic shock can occur. We report a case of DIHE presenting as recurrent massive intraluminal bleeding that was treated by transarterial embolization.

CASE REPORT

A 57-year-old man visited the emergency room complaining of a sore throat and difficulty swallowing for 5 d. He had a history of alcoholic liver cirrhosis (Child-Pugh class B), but he continued drinking. Laryngoscopy and cervical computed tomography (CT) revealed severe laryngopharyngitis. Upper gastrointestinal endoscopy showed marked laryngeal edema and a small ulceration in the upper esophageal sphincter. There was no intraluminal mass, except for small varices in the distal esophagus. The gastric mucosa was moderately congested. The patient denied any history of cervical trauma or instrumentation. He was treated medically with antibiotics, and his initial symptoms subsided within 5 d.

One week later, the patient complained that he regurgitated blood from the throat. He continued to spit up small volumes of fresh blood repeatedly. His hemoglobin dropped from 13.9 to 6.5 g/dL, and 4 U of packed red cells were transfused. Emergency endoscopy revealed a small opening in the cervical esophagus and
mucosal bridging with a large mucosal defect around the esophagogastric junction (Figure 1). Active bleeding was detected from a vessel exposed on the ulcer base in the cardia. After hemostasis with endoscopic clipping, his vital signs and hemoglobin stabilized. Chest CT performed because of the esophageal lesions revealed dissection of the wall and a large circumferential intramural hematoma in the esophagus (Figure 2). Based on the endoscopy and chest CT findings, DIHE with cardiac ulcer bleeding was diagnosed. The patient was treated conservatively without oral intake.

Eight days later, the hematemesis recurred. Conservative treatment with massive transfusion of packed red cells and fresh frozen plasma was ineffective. Endoscopy showed no bleeding at the previously treated site. However, continuous oozing of blood from the distal esophagus was observed. His vital signs were unstable.

Since surgery was very risky due to underlying liver cirrhosis, less invasive celiac angiography was performed for hemostasis. A small hyperstaining pseudoaneurysmal lesion was observed at an esophageal branch of the left gastric artery (Figure 3). The branch was embolized using glue and lipiodol. Transarterial embolization using glue and coils was reattempted, and no further bleeding occurred. Follow-up chest CT 4 wk later showed a resolving hematoma and double lumens separated by the dissected mucosa. The embolization site was seen as focal lipiodol uptake in the distal esophagus (Figure 4). Endoscopic examination
was declined. Supportive care with parenteral nutrition was maintained. The patient was discharged without complications after 6 wk.

DISCUSSION

The esophagus is susceptible to various extrinsic injuries (from ingested foods, instruments, and bougienage) or intrinsic sheering forces induced by retching, vomiting, or coughing. DIHE lies in the spectrum of esophageal injuries between a mucosal tear (Mallory-Weiss syndrome) and a transmural laceration (Boerhaave’s syndrome). Although these syndromes are usually associated with severe vomiting, DIHE is not always caused by emesis. One out of five patients reports no history of trauma. However, an underlying coagulopathy is found in many patients with so-called spontaneous DIHE. Portal hypertension and endoscopic variceal sclerotherapy are also associated with DIHE in cirrhotic patients. Although the direct cause was not clear in this present case, the cervical esophageal ulcer and underlying portal hypertension may have been precipitating factors.

Acute chest pain is a common presenting symptom and should be differentiated from acute myocardial infarction and aortic dissection. Hematemesis and difficulty swallowing may ensue, and these are helpful for differentiating from other critical diseases. The typical triad of DIHE (chest pain, dysphagia, and hematemesis) is evident only in one third of cases. Therefore, the rarity of DIHE and atypical symptoms can delay correct diagnosis. In our case, repeated hematemesis with hypovolemia was the only clinical feature.

DIHE is generally benign. Most patients recover fully with conservative management. Esophageal obstruction and major bleeding constitute two major complications of DIHE. Esophageal obstruction by the hematoma may cause or aggravate difficulty swallowing. Successful endoscopic decompression or surgical treatment have been reported in such cases.

A major intraluminal hemorrhage seems to be caused by overlying mucosal rupture and evacuation of the hematoma, however, the exact mechanism remains unclear. Although half of patients are reported to experience hematemesis, the volume of bleeding is usually small, and major bleeding with hypovolemic shock is uncommon.

We reviewed 119 patients who were presented in 87 English-language reports since 1968. Major bleeding was defined as more than 500 mL hematemesis with hypovolemic shock or bleeding that required transfusion of at least 4 U. Eleven cases (9.2%), including the present case, were collected and summarized in Table 1. Seven cases were elderly patients (five females and two males). The bleeding was attributed to anticoagulation therapy in three cases. The majority were treated conservatively. However, this type of treatment was not always effective. An elderly patient who was treated conservatively died after 5 d.

Table 1  Summary of 11 patients with dissecting intramural hematoma of the esophagus presenting major hemorrhage

| Ref.         | Sex/age (yr) | Underlying diseases | Precipitating factor | Primary symptoms | Treatment | Outcome |
|--------------|--------------|---------------------|----------------------|------------------|-----------|---------|
| Atefi et al  | M/59         | Diabetes            | Retching             | Hematemesis      | Conservative | Died    |
| Kerr et al   | F/72         | Hypertension        | None                 | Epigastric pain  | Conservative | Resolved |
| Ortiz et al  | F/80         | None                | Food (rice)          | Chest pain, dysphagia | Surgical | Died    |
| Freeman et al| F/59         | None                | None                 | Chest pain        | Conservative | Resolved |
| de Vries et al| M/88        | None                | Minor head trauma    | Retrosternal pain | Conservative | Resolved |
| Folan et al  | M/58         | Alcoholism          | Vegetable (broccoli) | Dysphagia         | Surgical    | Resolved |
| Takaoka et al| F/87         | Suppurative cholangitis | Nasobiliary catheter | Hematemesis      | Conservative | Resolved |
| Cullen et al | F/74         | Hypertension        | Heparin IV           | Chest pain, dysphagia | Conservative | Resolved |
| Yamashita et al | F/67     | Cerebral aneurysm   | Heparin IV           | Hematemesis      | Conservative | Resolved |
| Bandopadhyay et al | M/86 | Hypertension | Retching             | Warfarin          | Conservative | Died    |
| Present case | M/57         | Liver cirrhosis     | None                 | Hematemesis      | Therapeutic angiography | Resolved |

Outcome

Resolved

None

Hypovolemic shock or bleeding that required transfusion of at least 4 U. Eleven cases (9.2%), including the present case, were collected and summarized in Table 1. Seven cases were elderly patients (five females and two males). The bleeding was attributed to anticoagulation therapy in three cases. The majority were treated conservatively. However, this type of treatment was not always effective. An elderly patient who was treated conservatively died after 5 d. Two patients were treated with surgery, one of them had a 5-cm longitudinal tear with a pulsatile bleeding vessel between 25 and 30 cm from the alveolar ridge on endoscopy, and the active arterial bleeder was identified and treated with an emergency thoracotomy. In our case, angiography located and treated the bleeding site at an esophageal branch of the left gastric artery. Extensive DIHE and major bleeding might be caused by a small bleeding artery. In this situation, therapeutic angiography may be effective.

Therapeutic angiography has been used to control major non-variceal gastrointestinal bleeding and has been reported to be safe, effective, and durable. It is usually considered when endoscopic therapy has failed or when emergency surgery carries a high risk of mortality. It has also been effective in the hemostasis of uncontrolled Mallory-Weiss syndrome and in the treatment of a giant gastric intramural hematoma.

In conclusion, massive intraluminal bleeding can be a complication of DIHE. Although most bleeding in DIHE can be managed medically, prompt, appropriate treatment should be attempted in hemodynamically unstable patients. In such cases, therapeutic angiography may be a useful treatment alternative to surgery.
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