



A rare cause of severe gastrointestinal bleeding in a thrombocytopenic patient with acute myeloid leukemia

Question:

A 24-year-old female who had been diagnosed with acute myeloid leukemia (AML) 1 month prior was admitted to our clinic with acute-onset substernal chest pain, hematemesis, and melena. She had completed her second cycle of chemotherapy (Cytosine arabinoside/ idarubicin.) 2 weeks ago. Apart from AML, her past medical history was unremarkable. Her physical examination was normal, except for melena in her digital rectal examination. Laboratory test results were as follows: hemoglobin level: 7.7 g/dL (11.7–15.5 g/dL), white blood cell count: $0.2 \times 10^9/L$ (4.1×10^9 – $11.2 \times 10^9/L$), platelet count: $14 \times 10^9/L$ (159×10^9 – $388 \times 10^9/L$), creatinine level: 0.62 mg/dL (0.5–0.9 mg/dL), alanine transaminase level: 19 U/L (0–33 U/L), aspartate transaminase level: 25 U/L (0–31 U/L), and international normalized ratio: 0.9 (0.8–1.2). Her hemoglobin level decreased in repeated measurements, leading to the development of tachycardia and hypotension. Upper gastrointestinal endoscopy was performed after

red blood cell and platelet transfusions. Endoscopy showed a double lumen in the middle portion of the esophagus separated by a mucosal bridge that was coated with a hematoma (Figure 1).

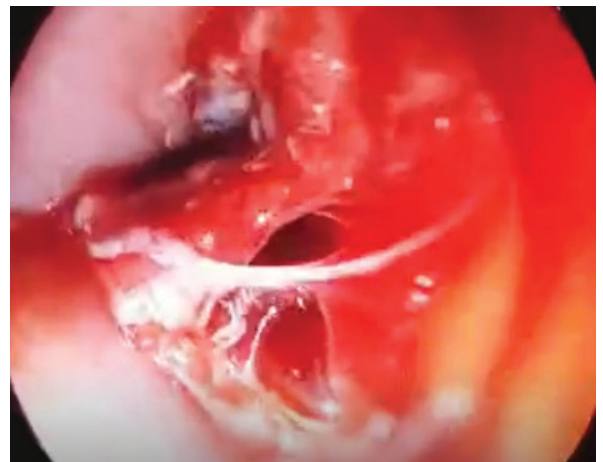


Figure 1. Endoscopic view of the patient showing a double lumen in the middle portion of the esophagus separated by a mucosal bridge that was coated with a hematoma

Answer: Spontaneous Intramural Esophageal Dissection

The patient was diagnosed with spontaneous intramural esophageal dissection (SIED). Oral intake was terminated, and the patient was conservatively managed with an intravenously administered proton pump inhibitor (pantoprasole, 8 mg/h infusion), fluid/nutritional support, and red blood cell and platelet transfusions. Four units of red blood cell suspension was administered to the patient. The patient's hemodynamic condition improved, and there were no bleeding recurrences at the follow-up. Control endoscopy revealed healing of the esophagus 21 days after initial presentation (Figure 2).

Spontaneous intramural esophageal dissection is a rare clinical entity that is generally seen secondary to bleeding disorders. There have been reports of various inherent and acquired bleeding disorders such as anticoagulant therapy, thrombolytic therapy, antiaggregant therapy, hemophilia, chronic renal failure, and liver cirrhosis causing SIED in the literature (1-3). Other causes of SIED include eosinophilic esophagitis, esophageal web/diverticula, persistent retching, and arteriovenous malformations (4). Severe thrombocytopenia was the associated risk factor in the present case. To the best of our knowledge, this is the first reported SIED case induced by severe thrombocytopenia.

Hematemesis, chest pain, dysphagia, and odynophagia are the most common symptoms of SIED. Hematemesis, which is usually clinically insignificant, spontaneously resolves in most cases. However, there have been a few patients with SIED in the literature presenting with severe gastrointestinal bleeding, similar to that seen in our patient (3).

Barium esophagogram, upper gastrointestinal endoscopy, computed tomography, and magnetic resonance imaging are useful methods for making a definitive diagnosis. Two different lumens, a true esophageal lumen and false lumen separated by a mucosal bridge, can be frequently identified by endoscopy as in our

patient. However, endoscopy should be carefully performed in these patients because forcing the endoscope into the false lumen can lead to esophageal perforation (5).

Spontaneous intramural esophageal dissection treatment is generally conservative and includes cessation of oral intake, administration of fluid and nutritional support, and close monitoring when the dissection is limited by the submucosal plane. Endoscopic interventions including incision of the septum between the true and false lumens, balloon dilatation, metal stent insertion, endoscopic clip placement, or surgical treatment may be necessary if a patient is refractory to conservative management or has a complication. Complications such as esophageal perforation, esophageal fistula, mediastinal abscess, and pneumomediastinum rarely occur (1-3).

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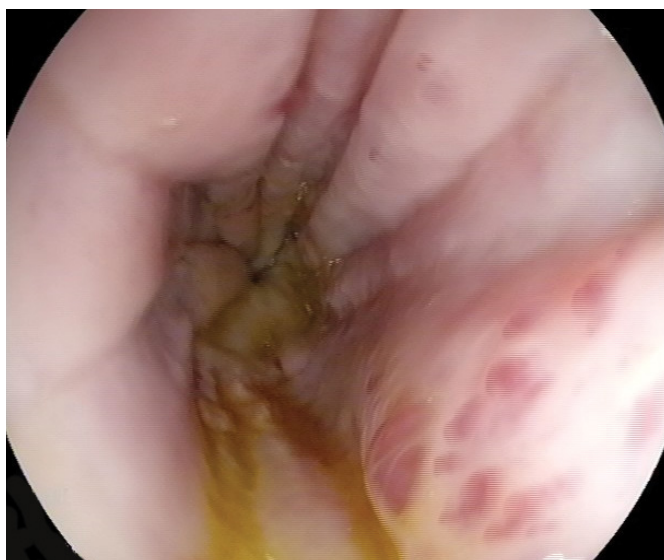


Figure 2. Endoscopic view of the patient at the follow-up showing hyperemia and erosions at the previously dissected areas and signs of dissection are no longer present