

Case Report

Focal intestinal lymphangiectasia: An unusual cause of acute overt obscure gastrointestinal bleeding

Ashish Kumar Jha, Ragesh Babu Thandassery, Mahesh Kumar Goenka

Institute of Gastroscience, Apollo Gleneagles Hospital, Kolkata, West Bengal, India

Abstract

Detection of bleeding lesion in a patient of acute overt obscure gastrointestinal bleeding is a real challenge. Recently, authors have showed superiority of urgent capsule endoscopy (CE) over angiography in patients with acute overt obscure gastrointestinal bleeding. Focal type of intestinal lymphangiectasia is a rare cause of acute gastrointestinal bleeding. Here, we describe a case of focal lymphangiectasia who presented to us with acute overt obscure gastrointestinal bleeding and diagnosed by urgent CE.

Key words

Acute overt obscure gastrointestinal bleeding, capsule endoscopy, focal lymphangiectasia, intestinal lymphangiectasia

Introduction

Detection of bleeding lesion in a patient of acute overt obscure gastrointestinal bleeding is a real challenge. Recently, authors have showed higher diagnostic yield of urgent capsule endoscopy (CE) as compared to angiography in patients with acute overt obscure gastrointestinal bleeding.^[1] In another study, emergency CE was able to identify bleeding lesions in 67% of patients with severe overt obscure gastrointestinal bleeding.^[2] Focal type of intestinal lymphangiectasia (IL) is a rare cause of acute overt obscure gastrointestinal bleeding. Here, we describe a case of focal lymphangiectasia who presented to us with acute overt obscure gastrointestinal bleeding and diagnosed by urgent CE.

Case Report


A 42-year-old male presented with 2 days history of melena. Examination revealed stable vital signs and pallor. Investigations showed hemoglobin and hematocrit of 8.6 g/dl and 24.5% respectively. Esophagogastroduodenoscopy and

colonoscopy did not reveal the source of the bleeding. Abdominal computerized tomography (CT) scan with angiography was normal. Urgent CE was done, which revealed a white plaque-like lesion in mid-jejunum with oozing of blood [Figure 1]. Mucosal biopsy was taken using a spiral enteroscope, which revealed mucosa with presence of dilated lymphatic channels. Although he received three units of packed red cell transfusion, his hemoglobin fell to 7.2 g/dl (hematocrit 21.1%). In view of the ongoing bleeding, the affected jejunal segment was resected [Figure 2]. Histopathology of the resected specimen showed mucosal and submucosal lymphatic spaces filled with lymph, consistent with lymphangiectasia.

Discussion

Intestinal lymphangiectasia is a rare disease characterized by dilated lymphatic vessels of the gastrointestinal tract. This can occur as a primary congenital disorder (Milroy's disease) or may be secondary to lymphatic obstruction. Distribution of lesion may be unifocal, multifocal or diffuse. Focal or localized IL is secondary to localized developmental abnormality of lymphatic vessels. IL may be asymptomatic or presents with malabsorption and protein-losing enteropathy.^[3] Bleeding from IL is uncommon and ranges from chronic blood loss to massive bleeding.^[4-6] Acute gastrointestinal bleeding from a unifocal type of IL is rare.^[4,6] Obstruction of the normal efferent flow of the lymph and subsequent opening of latent lymphatic-venous or abnormal lymphatic-arterial connections explains the hemorrhage in IL.^[6]

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Address for correspondence:

Dr. Ashish Kumar Jha, Institute of Gastroscience, Apollo Gleneagles Hospital, CC Road, Kolkata, West Bengal, India. E-mail: ashishjhabn@yahoo.co.in

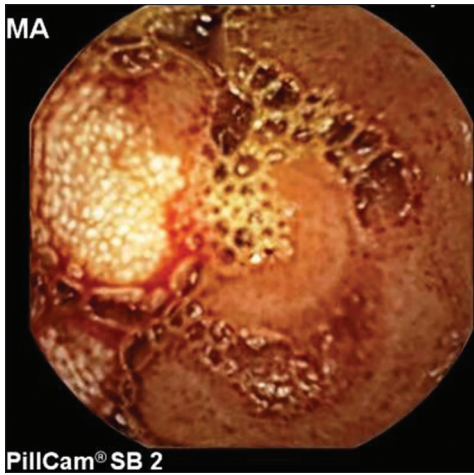


Figure 1: Capsule endoscopy image showing a white plaque-like lesion

The diagnosis of jejunoileal IL is difficult. The role of CT scan with angiography is limited in IL. Recently, small bowel evaluation by CE, double-balloon enteroscope, and spiral enteroscope have emerged as a novel tool for diagnosis of the jejunoileal lymphangiectasia. The endoscopic findings include white-tipped villi, scattered white spots or nodules, plaque-like white lesions, and submucosal elevations. Histopathological features consist of dilated intramucosal and submucosal lacteals. Treatment of IL mainly includes low-fat, high-protein diet supplemented with medium-chain triglyceride.^[3] Localized IL may be treated with surgical resection.^[7]

Conclusion

In conclusion, focal type of IL may rarely present with acute overt obscure gastrointestinal bleeding. Management of such a clinical condition warrants an urgent CE and surgical resection.

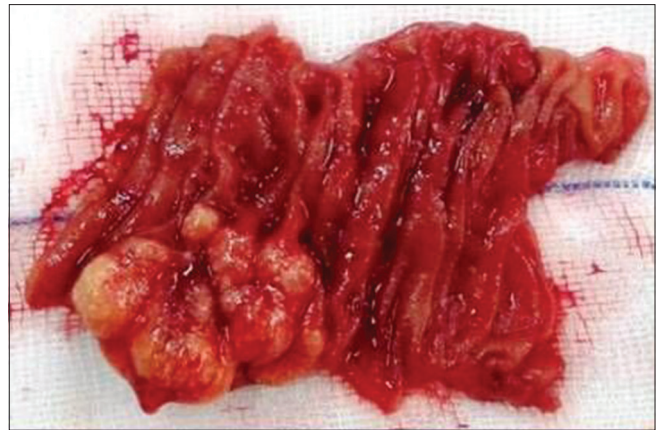


Figure 2: Resected surgical specimen

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