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CASE REPORT | ENDOSCOPY

Therapeutic Banding for Bleeding Duodenal Lymphangiectasias: A Novel Approach

Robert Duve, MD[1], Kevin Robillard, MD[2,3], and Kazunori Kanehira, MD[4]

[1]*Department of Internal Medicine, Jacobs School of Medicine and Biomedical Sciences, University at Buffalo, Buffalo, NY*
[2]*Division of Gastroenterology, Roswell Park Cancer Institute, Buffalo, NY*
[3]*Division of Gastroenterology, Hepatology and Nutrition, Jacobs School of Medicine and Biomedical Sciences, University at Buffalo, Buffalo, NY*
[4]*Division of Pathology, Roswell Park Cancer Institute, Buffalo, NY*

ABSTRACT

With endoscopic advancements, the number of detected intestinal lymphangiectasias has been on the rise. They are generally considered benign and incidental; occasionally, these lesions carry complications, and best management options need to be established. Bleeding intestinal lymphangiectasias should be considered a rare cause in the differential diagnosis for gastrointestinal bleeding. References in the literature primarily indicate surgical treatment in these situations. In this study, we report an uncommon case of a man with esophageal adenocarcinoma who developed acute gastrointestinal bleeding from duodenal lymphangiectasias that were successfully banded.

KEYWORDS: lymphangiectasia; duodenal; banding; adenocarcinoma; acute bleed; endoscopy; theraputic

INTRODUCTION

Intestinal lymphangiectasias are dilated enteric lymphatics appearing as multiple scattered white spots, diffuse prominent villi with whitish tips, or focal small whitish macules.[1] Primary intestinal lymphangiectasia (Waldmann disease) is a rare pediatric disease with unknown incidence, and fewer than 200 cases reported from 1961 to 2014, the majority diagnosed before 3 years of age.[2] It is characterized by diffuse development of intestinal lymphangiectasias with consequential leakage of lymph into the gut lumen causing hypoalbuminemia and lymphopenia.[3] Pathophysiology is unclear but thought to be due to mutations in genes that regulate lymphogenesis such as vascular endothelial growth factor receptor 3.[2] This is contrasted from secondary causes of lymphangiectasias, which can arise because of the obstruction of downstream lymphatics, sometimes because of malignancy.[4] Secondary lymphangiectasias can be frequently found in the elderly population and are generally regarded as incidental findings.[5]

In a study evaluating 1866 consecutive esophagogastroduodenoscopy (EGD) endoscopic examinations, 3.2% noted duodenal lymphangiectasias, and histological confirmation of duodenal lymphangiectasias was evident in 1.9% of these total cases.[1] These lesions most frequently occur in the small bowel; therefore, they are sometimes seen on EGD and more commonly found on video capsule endoscopy. Diagnosis is usually made through endoscopy revealing typical white, swollen, and cystic lacteals often protruding into the gut lumen, with pathologic confirmation through biopsy possible.[6,7] It is sometimes important to rule out malignancy as a cause of lymphangiectasias in the adult population, namely, lymphoma.[3] The most often reported intestinal site is the duodenum, possibly because of easy detection with EGD.[8]

CASE REPORT

A 72-year-old man presented to the hospital for the evaluation of fatigue over the past 2 months. This was associated with 40–50 lb weight loss, early satiety, and solid food dysphagia. His medical history included hypertension, hyperlipidemia, type 2 diabetes with neuropathy, and asthma. He had never undergone prior EGD or colonoscopy. He was a former 10 pack-year smoker and drank 6–10 beers per week without a history of illicit drug use. Physical examination and vital signs were unremarkable. Laboratory test results revealed normocytic anemia with hemoglobin (Hgb) 11.0 and mild acute kidney injury, and coagulation studies were unremarkable.

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Correspondence: Kevin Robillard, MD (Kevin.robillard@roswellpark.org).

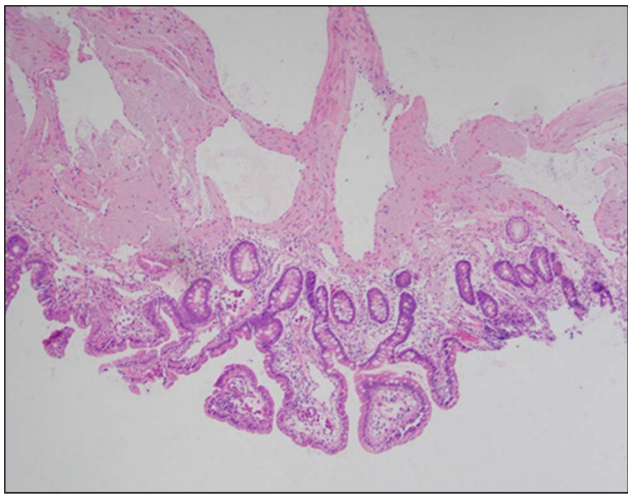

Figure 1. H&E staining characteristic of lymphangiectasias at 40×. H&E, hematoxylin and eosin.

Abdominal and pelvic computed tomography revealed a distal esophageal mass extending to the gastroesophageal junction. EGD was performed and revealed an ulcerated circumferential mass 34–40 cm in the esophagus. Pathology results revealed poorly differentiated invasive adenocarcinoma. Positron emission tomography/computed tomography revealed localized disease. Repeat EGD/EUS was performed for staging and was found to be uT3N2 and Siewert II.

He began neoadjuvant chemoradiotherapy with paclitaxel and carboplatin, with plans for esophagectomy. Four of 6 planned chemo cycles and 24 fractions of radiotherapy were completed before being stopped because of anemia, hemoglobin 7.4, thrombocytopenia, platelets 78,000, and acute kidney injury. Repeat positron emission tomography/computed tomography showed partial metabolic response to therapy without metastasis. He then experienced melena and 3 hospitalizations for upper gastrointestinal (GI) bleeding over the course of 5 weeks, separated roughly 2 weeks apart and each admission lasting 2–3 days. His Hgb remained unstable between and during admissions, reaching as low as 6.6. EGD revealed friable lymphangiectasias measuring 20–25 mm in D2 and D3 portions of the duodenum with confirmatory biopsies (Figure 1). These were treated with sucralfate, proton-pump inhibitors, intravenous iron, and as-needed blood transfusions. There was no obvious active bleeding at the primary tumor. Computed tomography angiography showed no alternative source of bleeding. Because of continued upper GI bleeding, repeat EGD was performed and again visualized lymphangiectasias, which were friable and with low-grade spontaneous bleeding (Figure 2). With continued bleeding confirmed on multiple EGDs, therapeutic endoscopy was attempted. Of available modalities for the prevention of further bleeding, endoscopic band ligation was chosen. Lymphangiectasias are dilated lymphatic channels in the lamina propria (below the basement membrane of the mucosa); therefore, more superficial options (such as argon plasma coagulation) were felt less likely to be efficacious. Four lymphangiectasias were

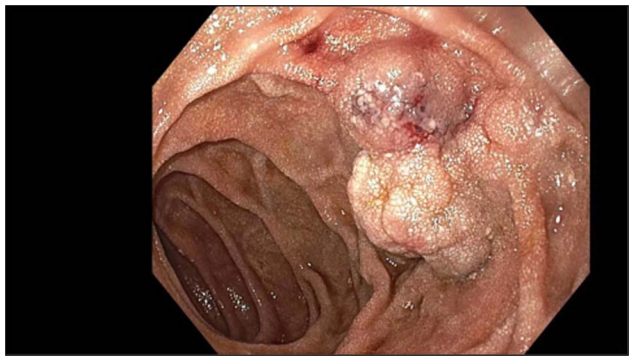

Figure 2. Lymphangiectasias with active bleed seen in the duodenum on EGD. EGD, esophagogastroduodenoscopy.

banded with effective cessation of bleeding (Figure 3). Repeat EGD 3 weeks later showed 4 nonbleeding duodenal ulcers correlating with where previous bands were placed. After banding, his blood counts stabilized and increased to his baseline of 11.0 without the need for repeat transfusion. He received a total of 13 units before banding.

The remainder of his clinical course was complicated by hydronephrosis, pulmonary embolism, and cerebrovascular accident. These combined with continual deconditioning prevented him from being considered as a surgical candidate for esophagectomy. He died of esophageal cancer.

DISCUSSION

Intestinal lymphangiectasias are typically regarded as benign incidental findings on endoscopy but occasionally can have clinical manifestations. Most commonly is lower extremity edema due to hypoalbuminemia from leakage of lymph. Other rarer consequences include intestinal obstruction, occult small intestinal bleed, and massive GI bleed.[9]

The differential diagnosis for acute upper GI bleed is extensive, and common pathologies include peptic ulcer disease, varices, esophagitis, arteriovenous malformations, gastric antral vascular ectasia, and Dieulafoy lesions. It is quite rare for bleeding lymphangiectasias to be identified as the culprit lesion and has

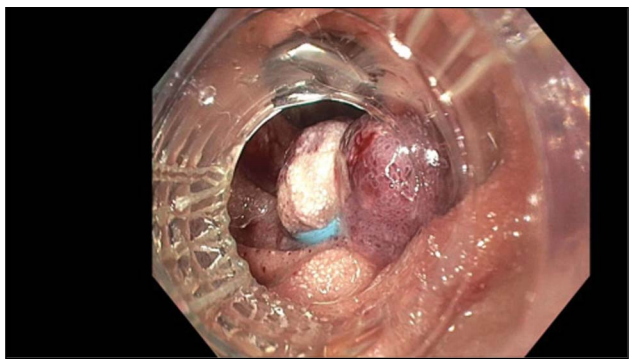

Figure 3. Duodenal lymphangiectasia status post banding.

been done so in only a few reported cases. Baichi et al described a case of persistent acute GI bleed where oozing lymphangiectasias were discovered on enteroscopy, tattooed, and successfully treated with small bowel resection.[10] Lom et al described a similar case of acute GI bleed that was again treated with partial small bowel resection, and pathology results returned as intestinal lymphangiectasia.[11] In our case, biopsy results confirmed the culprit lesion to be intestinal lymphangiectasias. Although most classically these lesions have been treated with small bowel resection, because of our patient's debilitated state, this was not a viable option. These lesions resulted in a significant GI bleed requiring multiple transfusions. Cessation of bleeding was achieved after banding of the lesions.

A handful of mechanisms have been proposed to explain the pathophysiology for bleeding lymphangiectasias. One study found a positive correlation between intestinal angiodysplasia and lymphangiectasias, finding associated lymphangiectasias in 52% of those with small bowel angiodysplasia.[12] Others' claim raised intraluminal pressure in the lymphatics opening latent connections between lacteals and venous or arterial vessels.[13,14]

In this patient's case, it was not the esophageal mass causing bleeding but the initially less suspicious group of duodenal lymphangiectasias. A literature search was unable to provide any similar endoscopic treatments. This is likely the first case described in the literature detailing successful endoscopic management of bleeding intestinal lymphangiectasias with banding.

DISCLOSURES

Author contributions: R. Duve wrote and edited the article and reviewed the literature. K. Robillard edited the article and is the article guarantor. K. Kanehira supplied and interpreted the pathology images. All authors approved the final version of the manuscript.

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