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CASE REPORT | SMALL BOWEL

Duodenal-Acquired Intestinal Lymphangiectasia After Lymph Node Excision and Inferior Vena Cava Injury in an 18-Year-Old Woman

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ABSTRACT

Intestinal lymphangiectasia (IL) is a rare disorder characterized by dilation and obstruction of the small bowel lymphatic ducts, resulting in malabsorptive diarrhea and edema. While the majority of IL is congenital and presents in infancy, secondary acquired IL rarely presents as a long-term sequela after abdominal or pelvic surgeries. We report an 18-year-old woman who underwent surgical resection of ovarian cancer with subsequent endoscopic and histologic evidence of duodenal IL. This case highlights the importance of increased endoscopic awareness for iatrogenic acquired IL in clinical practice.

KEYWORDS: duodenal lymphangiectasia; iatrogenic; ovarian cancer; lymph node dissection

INTRODUCTION

Intestinal lymphangiectasia (IL) is a dilation and obstruction of gastrointestinal lymphatic vessels that occurs in <2% of the population.[1,2] Primary IL occurs due to congenital malformation of lymphatic structures and results in protein-losing enteropathy.[1] Secondary IL occurs when there is indirect acquired obstruction of the lymphatic system, such as intra-abdominal fibrosis from chronic inflammation or lymphatic stasis from increased central venous pressure, lupus, systemic sclerosis, constrictive pericarditis, lymphomas, small bowel resections, radiation therapy, and Fontan procedure.[2–8] We present a patient with duodenal-acquired IL identified on upper esophagogastroduodenoscopy (EGD) in a young woman who underwent surgical resection of ovarian cancer.

CASE REPORT

An 18-year-old woman presented to the gastroenterology outpatient clinic for evaluation of new onset frequent, nonbloody diarrhea occurring more than 5 times daily without nocturnal symptoms, accompanied by abdominal distension and nausea. She denied unintentional weight loss, history of vitamin deficiency, ascites, steatorrhea, or peripheral edema. Two months ago, she underwent laparoscopic salpingo-oophorectomy with pelvic and para-aortic lymphadenectomy for stage 1a ovarian cancer. She has a family history of celiac disease and ulcerative colitis, but not of gastrointestinal malignancy.

On physical examination, she had a well-healed midline abdominal scar, mild abdominal distension, and tenderness to deep palpation in all 4 quadrants without rebound or guarding. A systematic approach was used to rule out secondary causes of IL. Laboratory evaluation revealed normal serum protein (7.5 g/dL) and albumin (4.7 g/dL), with normal immunoglobulins, inflammatory markers, and celiac serologies. Computed tomography of the abdomen and pelvis showed no metastatic disease, lymphatic obstruction, bowel wall edema, ascites, or effusions. While transthoracic echocardiogram can be used to evaluate for cardiac causes of lymphatic congestion, this was deferred given the patient's likely surgical etiology, young age, lack of cardiovascular history, lack of peripheral edema, and absence of cardiopulmonary symptoms.

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The index diagnostic upper EGD and colonoscopy revealed numerous punctate white lesions in the second and third portions of the duodenum (Figure 1). Cold forceps biopsies of this area were obtained. On histology, the duodenal biopsies revealed benign duodenal mucosa with dilated lacteals and intact villous architecture (Figure 2). Biopsies from the duodenal bulb, stomach, and esophagus were normal. Colonoscopy showed a tortuous colon, suggestive of extrinsic compression of the sigmoid colon, possibly from surgical scar tissue.

On further review of her surgical records, the operative report commented on an inferior vena cava (IVC) tear after gentle traction of a nearby vein, with subsequent conversion of the laparoscopy to open laparotomy to complete the right salpingo-oophorectomy, left ovarian nodule biopsy, right pelvic lymph node excision, right and left para-aortic lymph node excision, and omentum excision. There were no immediate post-operative complications.

DISCUSSION

Duodenal-acquired IL can develop after abdominal or retroperitoneal surgeries that disrupt the lymphatic drainage pathways, particularly when lymph nodes are removed, or major vascular structures are injured. IL most often presents as generalized peripheral edema (83.3% of cases) and chronic diarrhea (41.7%), and less commonly with laboratory abnormalities including hypoproteinemia, hypogammaglobulinemia, hypocalcemia, and fat-soluble vitamin deficiencies, or concerning clinical manifestations such as ascites, anasarca, abdominal distension, fatigue, and abdominal pain.[3,9–11] The development of IL in this case likely resulted from both direct surgical disruption and altered lymphatic fluid dynamics. Lymphatic stasis occurs when normal flow is impeded through vessel injury or external compression from postsurgical inflammation.[12,13] In addition, changes in central venous pressure following IVC injury and repair can affect lymphatic drainage patterns, as back pressure in the venous system transmits to the lymphatics, leading to vessel dilation and lymphangiectasia.[6,14] In this case, the IVC injury during surgery likely compromised local lymphatic drainage through these combined mechanisms.

For clinical management of IL, we recommended a low-fat, high protein diet and medium-chain triglycerides (MCT) oil supplementation. At the 9-month follow-up, the patient's bowel frequency improved, her weight increased, and nutritional laboratory results remained stable. The physical examination also remained unchanged. Follow-up imaging showed no abdominal ascites, pleural or pericardial effusions, bowel wall edema, or abdominal or pelvic lymphadenopathy. Though benign in presentation, long-term monitoring was initiated, given literature evidence of potential delayed lymphatic remodeling and fibrosis developing years after initial injury.[15]

Current testing modalities for IL include direct visualization with upper EGD and biopsy sampling, alpha-1 antitrypsin clearance test, and magnetic resonance lymphangiography for evaluation of lymphatic leakage into the gut.[16] Computed tomography imaging may identify additional anatomic abnormalities contributing to IL, including fibrosis, adenopathy, and bowel wall edema.[17,18] The duodenum is particularly susceptible to due to its high density of lymphatic vessels and proximity to major lymphatic networks, making it vulnerable to pressure changes and lymphatic remodeling.[16] Video capsule endoscopy can have limited utility in diagnostic duodenal IL.[16,19] Other secondary causes of IL can be investigated by transthoracic echocardiogram to rule out right heart failure, abdominal radiographic imaging to evaluate neoplastic lymphatic compression, and infections or toxins that alter permeability of lymphatic epithelial layers.[1]

Treatments of IL primarily consists of dietary modifications, including MCT oils, low-fat diet (<2.5 g/d), and high-protein diets (2 g/kg/d). MCT oils are particularly effective as they are absorbed directly into the portal venous system, bypass the lymphatic circulation, and prevent further lymphatic vessel degradation.[1] Clinical response should be monitored through symptoms, physical examination, and nutritional markers including albumin, immunoglobulins, and fat-soluble vitamins. For refractory cases, surgical bowel resection or lymphatic embolization may be considered for focal disease.[1] While duodenal-acquired IL without significant malabsorption is generally considered benign, prolonged follow-up is essential due to potential chronic fibrotic changes and lymphangiogenesis that may

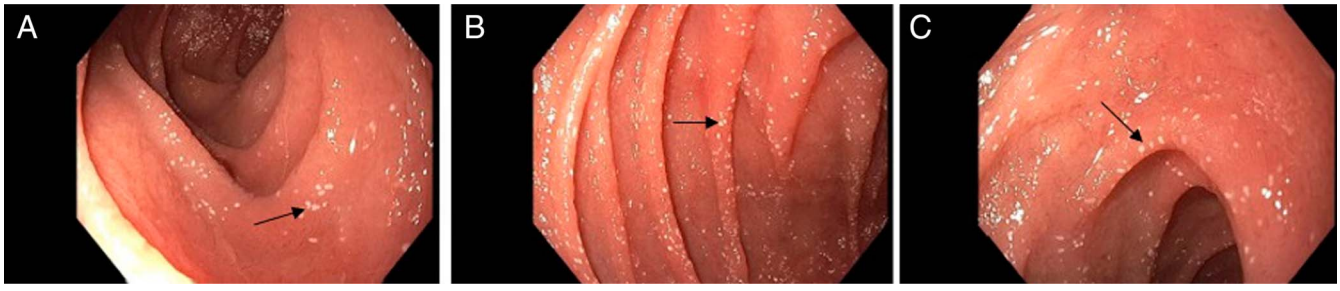

Figure 1. Images from upper endoscopy. (A) Multiple white spots with no bleeding were found in the second portion of the duodenum (black arrow). (B) Additional scattered snow-flake like areas of enlarged villi in the second portion of the duodenum (black arrow). (C) The third portion of the duodenum with additional white villi visible. The duodenal bulb appeared normal. Biopsies were taken with the cold forceps for histology (black arrow).

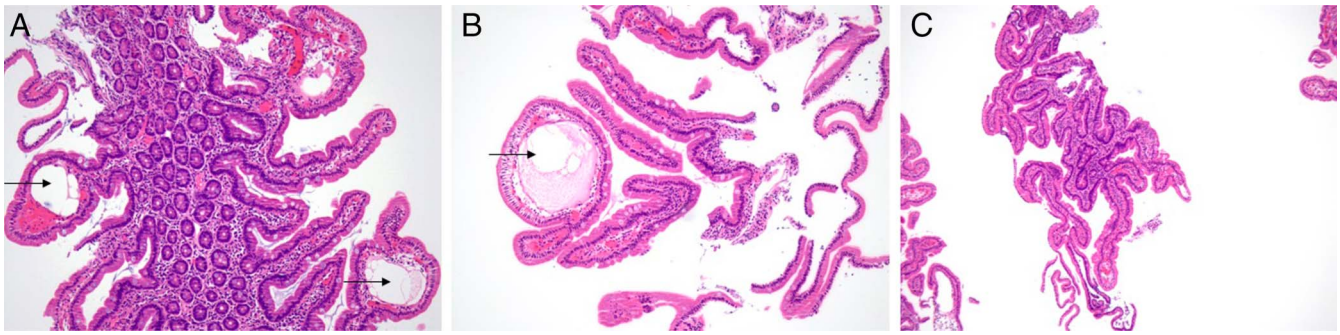

Figure 2. Duodenal biopsy pathological images. Histological examination of duodenal biopsies demonstrating dilated lymphatic vessels within intact villous architecture. (A) Low power image (100× magnification) of the descending duodenum showing multiple duodenal villi with dilated lymphatic vessels in the mucosal lamina propria (black arrows). (B) Medium power image (200× magnification) of the descending duodenum demonstrating detailed visualization of dilated lymphatic vessels and surrounding tissue architecture within the mucosal lamina propria (black arrows). (C) Scanning power image (40× magnification) of the descending duodenum highlighting preserved villous architecture and overall mucosal organization.

develop over time.[6,7,15] In our patient's case, dietary modification alone proved sufficient, with clinical improvement in symptoms and weight gain at the 9-month follow-up. However, given reports of lymphatic remodeling occurring years after the initial injury, long-term monitoring was initiated to assess for delayed complications that might require treatment adjustments.[15] This case establishes acquired IL as an important yet manageable sequela of lymph node dissection, emphasizing the value of prompt recognition and conservative intervention.

DISCLOSURES

Author contributions: D. Bickford: manuscript drafting, chart review and data abstraction, manuscript revision. C. Schmoyer: manuscript revision. CS Tse: manuscript revision and article guarantor.

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