中文題目：以糞石表現之迴腸淋巴癌

英文題目：Ileal lymphoma presented as intestinal bezoar

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Introduction: Extranodal marginal zone lymphoma is a relatively uncommon subtype of non-Hodgkin lymphoma. It is mainly seen in adults with median age at diagnosis of 66 years old. Most gastrointestinal lymphoma emerges at stomach, followed by small intestine and produces symptoms due to localized involvement. However, the symptoms may be varied according to tumor's severity and locations. In patients with gastric lymphoma, reflux symptoms, epigastric pain or discomfort, anorexia, weight loss, or occult gastrointestinal bleeding are common presentations. In patients with intestinal lymphoma, patients might suffer from intermittent diarrhea, colicky abdominal pain, and malabsorption. Some patients may present with other rare symptoms like the patient presented with intestinal bezoar in this case. Physicians may be aware of GI lymphoma in clinical practice.

Case presentation: A 68-year-old female with history of hypertension and type 2 diabetes mellitus presented to emergency room with abdominal pain and vomiting. She had experienced an episode of influenza A pneumonia related acute respiratory failure with mechanical ventilation admitted to intensive care unit 3 months ago. Bloody stool was once noted, however, esophagogastroduodenoscopy and colonoscopy only showed mild reflux esophagitis and hemorrhoids. No recurrent gastrointestinal tract bleeding was noted after last discharge. At emergency room, mild tachycardia to 131 beats/min was noted with hypertension 152/73 mmHg. No fever or dyspnea was noted. Physical examination showed soft and ovoid abdomen with lower abdominal tenderness. Her bowel movement was decreased. She denied any fever, diarrhea, constipation, chest pain, chest tightness or dyspnea.

Laboratory examination showed leukocytosis(WBC:32900/ul) with left shift and elevated CRP level (4.84 mg/dl). KUB showed positive stack of coin sign. (Figure 1). Abdominal CT showed bezoar at terminal ileum (Figure 2.a, arrowhead), with surrounding focal wall thickening and increased enhancement (Figure 2.b, arrow). Small bowel proximal to the bezoar was dilated. No lymphadenopathy in the para-aortic retroperitoneum and pelvis. Barium small-bowel series could successfully flow through. Double balloon enteroscopy was done from anal route to distal ileum at 60 cm proximal from ileocecal valve (Figure 3). Friable mucosa and circumflex ulceration leading to stenosis was found and scope can't path through. The bezoar resolved after pre-endoscopy laxatives use. Multiple endoscopic biopsy was performed and pathology showed intestinal mucosal tissue with ulceration and inflammatory exudate. Both Cytomegalovirus PCR and acid fast stain were negative. Surgery was suggested for management of ileal stenosis, but patient hesitated. For further evaluation of the nature of stricture, MR enterography was done and showed a 7.3 cm length circumferential wall thickening at distal ileum, with high SI on T2WI and mild contrast enhancement (Figure 4). Enlarged lymph nodes at lower abdomen mesentery were also noted.

There is no peri-intestinal fat stranding or intestinal fistula. Patient received abdominal surgery after MR enterography. A 9 x 9 cm solid tumor 60 cm proximal from ileocecal valve with involvement of greater omentum and enlarged mesentery lymph nodes were found and resected. Histopathological examination showed mucosa-associated lymphoid tissue(MALT) lymphoma (Figure 5.a). Expanded follicular dendritic meshwork were seen by CD21 stain. CD20 (Figure 5.b) and BCL-2 stain were positive, while CD3, CD43, CD5, CD10, CD23 and cyclin D1 stains were negative. Patient received adjuvant chemotherapy(R-CHOP) for lymphoma management and remained tumor free until 5 years after.

Discussion: Gastrointestinal bezoars are concretions of poorly digested fibers, skins, and seeds of fruits and vegetables or foreign bodies in the alimentary tract. Most GI tract bezoars form in the stomach, but can migrate to the small bowel, causing obstruction. The main clinical presentation includes small bowel obstruction, nausea, vomiting, abdominal pain and epigastric distress. CT findings of small bezoar are an intraluminal ovoid or round mottled-appearing mass with soft-tissue density containing air in its interstices and outlined by fluid or oral contrast material in the dilated small bowel at the site of the obstruction. Small bowel bezoar accounted for 1-4% of small bowel obstruction. Surgery is often suggested for treatment while conservative treatment is sometimes considered.
Primary gastrointestinal lymphoma constituted about 1%-4% of all gastrointestinal lymphoma. The most commonly involved sites is stomach, followed by small intestine and ileocecal region. Ileal mucosa-associated lymphoid tissue (MALT) lymphoma is rare. Many patients with extranodal marginal zone lymphoma have a history of autoimmune disease (ex: Sjögren's syndrome, systemic lupus erythematosus, Hashimoto's thyroiditis) or even with coexisting severe infections. The main treatment for gastrointestinal lymphoma are surgery, chemotherapy, and radiotherapy. Treatment strategy varied on age, histological subtype, extent and burden of disease, co-morbidity and clinical scenario. Dawson’s criteria and Ann arbor staging are commonly employed on staging of gastrointestinal lymphoma for further management guidance.

Conclusion: Ileal mucosa-associated lymphoid tissue (MALT) lymphoma with bezoar presentation is rare and would easily misdiagnose if absence of biopsy. Surgical treatment was suggested for both bezoar and ileal lymphoma if clinical condition available.