



CASE REPORT

Colonic lymphangiomatosis associated with anemia

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Abstract

Lymphangioma is an uncommon malformation of lymphatic system. Multiple colonic lymphangioma named as lymphangiomatosis is considered an extremely rare disease. Although lymphangioma is a benign tumor and most colonic lymphangiomatosis do not cause symptoms and do not require treatment, resection of lymphangioma is necessary in the presence of symptoms such as abdominal pain, bleeding, intussusceptions. We report a case of colonic lymphangiomatosis in a man who presented with abdominal discomfort and anemia, which was diagnosed and treated with endoscopic snare polypectomy.

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Key words: Colonic lymphangiomatosis; Anemia; Snare polypectomy

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INTRODUCTION

Lymphangioma is a lymphatic malformation which shows benign proliferation of lymph vessels, with the characteristics of submucosal tumors covered with normal mucosa^[1-5]. It may occur anywhere but rarely in the colon. However, it has been reported more frequently with the increasing prevalence of endoscopy. Multiple colonic lymphangiomatosis reported as "colonic lymphangiomatosis", are even rarer. We report a case of colonic lymphangiomatosis suspicious of submucosal tumor in a man who presented with anemia and abdominal discomfort, which was treated with endoscopic snare polypectomy.

CASE REPORT

A 48-year-old man with chief complaints of abdominal discomfort and anemia was admitted to our hospital. He had no significant past medical history. Physical examination revealed no remarkable abnormality in the abdomen. Laboratory tests showed hypochromic microcytic anemia, 6.3 g/dL hemoglobin, 24.2% hematocrit, 66.5 fL mean corpuscular volume (normal = 85-99), 17.3 pg mean corpuscular hemoglobin (normal = 26-34), 26.0% mean corpuscular hemoglobin concentration (normal = 32%-36%), 12 ug/dL iron (normal = 65-157), and 3.59 ng/mL ferritin (normal = 16.4-323). Tumor marker levels (CEA, CA19-9), urinalysis and blood chemistry tests were within normal ranges.

Colonoscopy revealed several protruding mucosal lesions covered with normal mucosa ranging 10-20 mm in diameter at the proximal transverse colon. These lesions were semi-transparent, and the cushion sign was positive. Some indigested food materials were got in between most protruding lesion and colonic wall (Figure 1A). The most prominent lesion underwent snare polypectomy (Figure 1B), and inner surface of the lesion was filled with seroanguinous fluid and yellowish fibroadipose tissue-like materials (Figure 1C). Abdomen computer tomography (CT) was unremarkable. Histopathologic examination showed cystic lumen covered with a single layer of flat endothelial cells (Figure 2). Based on the clinical and histopathologic findings, a diagnosis of colonic lymphangiomatosis was made. Considering that the patient did not show any bleeding signs and symptoms after the mucosectomy, we decided to follow up on an out-patient

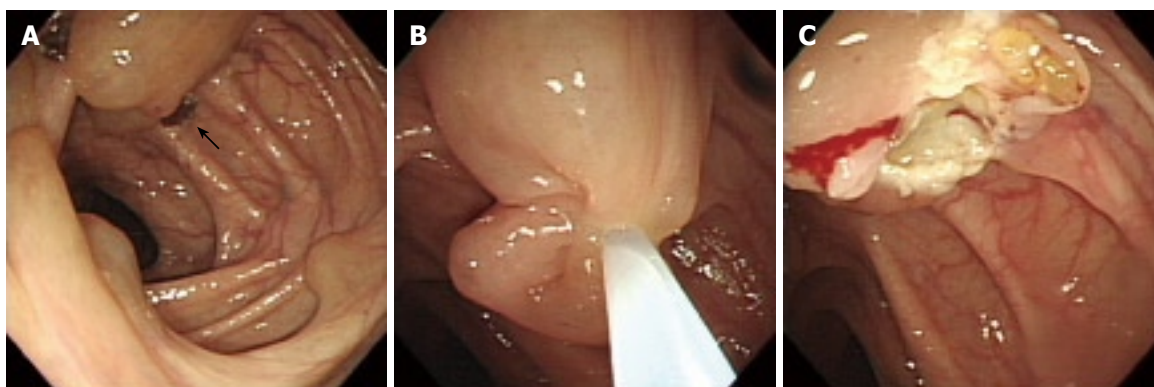


Figure 1 Colonoscopy revealing a semiparent, protruding mucosal lesion covered with normal mucosa with indigested food materials between the lesion and colonic wall (A), which underwent snare polypectomy (B), and inner surface of the lesion filled with seroanguinous fluid and yellowish fibroadipose tissue-like materials (C) after the procedure.

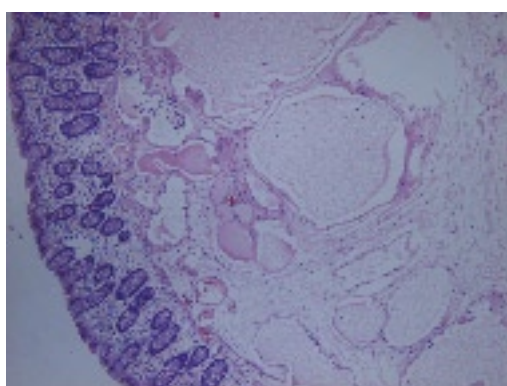


Figure 2 Histopathology showing a cystic lumen covered with a single layer of flat endothelial cells (HE, × 100).

basis. The patient had abdominal pain and anemia when he was followed up 3 mo after mucosectomy.

DISCUSSION

Lymphangioma, a benign tumor of lymphatics, rarely occurs in the abdomen, and most of them arise in the mesentery, omentum, mesocolon and retroperitoneum^[6-9]. The incidence of lymphangioma in the intestinal wall is very low, and few cases of multiple colonic lymphangioma have been reported^[1-4]. Barium enema, colonoscopy, abdomen CT, endoscopic ultrasonography are useful in its diagnosis^[10-12]. Colonoscopic findings of lymphangioma are characterized by a steep rising margin and a somewhat narrow base, the presence of properties of submucosal tumors covered with normal colon mucosa, as well as a smoother, glassier, and more translucent surface than that of adenoma, and soft lesions with their shape changed on postural alterations or compression^[1]. Our case also had such characteristics. Imaging studies may be needed to evaluate the wall structure and coexisting extracolonic lesions. Our case did not have any extracolonic lesions.

Because lymphangioma is a benign tumor and most colonic lymphangiomas are asymptomatic, and do not need treatment^[2]. However, symptoms such as abdominal pain, bleeding, intussusception or protein losing enteropathy can occur^[1,2,13-16]. The cardinal sign of our

case was anemia, which may be due to the seroanguinous cystic contents of lymphangiomatosis from the internal bleeding. Endoscopic resection has been recently performed for the diagnosis and treatment of colonic lymphangiomas^[1]. Endoscopic mucosectomy has been recommended for pedunculated tumors less than 2 cm in diameter^[1,5]. We report a colonic lymphangiomatosis associated with anemia and abdominal discomfort, which was treated with endoscopic mucosal resection.

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