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SUPPLEMENTATION TO THE ENCYCLOPEDIA

Small Bowel Lymphangioma ^{☆ ☆}



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KEYWORDS

Small bowel; Lymphangioma; Endoscopy; Gastrointestinal bleeding; Balloon enteroscopy; Video

Abstract

Background: Lymphangiomas are uncommon benign tumors found mainly in children. Intraabdominal lymphangiomas are rare, mostly located in the mesentery. Small bowel lymphangiomas are very rare.

Patient and methods: A 38-year-old woman presented with recurrent melena and anemia. Bidirectional endoscopy was non-diagnostic. Balloon enteroscopy revealed a 1 cm actively oozing, soft, friable, polypoid lesion in the proximal-mid small bowel. The lesion was white-yellow with "strawberry" mucosal patterns. Endoscopic tattooing was performed and she underwent subsequent laparoscopic segmental small bowel resection.

Results: Histopathologic features were consistent with a cavernous lymphangioma. At last follow-up, the patient's gastrointestinal (GI) bleeding and anemia had resolved.

Conclusions: Small bowel lymphangiomas can cause gross or occult GI bleeding, anemia, abdominal pain, and/or obstruction. Endoscopists should be aware of this rare tumor and its unique endoscopic features. The optimal treatment is radical excision, since incomplete resection may lead to recurrence. Argon plasma coagulation or polypectomy have been used to achieve endoscopic ablation and palliation of GI bleeding.

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Video Related to this Article

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Background

 A 38 year old woman was referred for recurrent melena and transfusion-dependent anemia.

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^{**}The terms of this license also apply to the corresponding video.

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- Previous upper endoscopy and colonoscopy were non-diagnostic.
- Small bowel capsule endoscopy revealed fresh blood in the proximal-mid small bowel.
- The patient had no significant past medical or surgical history.

2. Materials

- Single balloon enteroscope (Olympus SIF-Q180, Olympus America, Center Valley, PA).
- Injection needle (Carr-Locke needle, US Endoscopy, Mentor, OH, USA).

3. Endoscopic procedure

- Single balloon enteroscopy was performed following abnormal capsule endoscopy findings.
- Endoscopic findings:
 - A 1 cm actively oozing, soft, friable, polypoid lesion in the proximal-mid small bowel.
 - The lesion was white-yellow with "strawberry" mucosal patterns.
 - The surrounding mucosa was not indurated.
- Endoscopic intervention:

Endoscopic tattooing was performed at the normal appearing mucosa 1-2 cm proximal and distal to the bleeding lesion.

- The patient underwent laparoscopic segmental small bowel resection.
- Surgical pathology: $1.9 \times 0.6 \times 0.5 \text{ cm}^3$ cavernous lymphangioma.

Marked dilatation of the lymphatics in the mucosa and submucosa.

• Outcome: at last follow-up, the patient's gastrointestinal (GI) bleeding and anemia had resolved.

4. Key learning points and tips and tricks

- Lymphangiomas are uncommon benign tumors found mainly in children.
 - The most common sites are the head, neck, and axillary region.
 - Intra-abdominal lymphangiomas are rare (about 9%), mostly found in the mesentery [1-3].
- Histopathologically, lymphangiomas are classified into 3 types:
 - Capillary lymphangioma: small thin-walled lymphatics, usually in the superficial dermis.
 - Cavernous lymphangioma: larger dilated lymphatic channels, often connected to normal-caliber adjacent lymphatic spaces.
 - Cystic lymphangioma: lymphatics of various sizes that contain serous, chylous, or bloody fluid, with no connection to adjacent lymphatics.

- Small bowel lymphangiomas are very rare [4-14].
 - Cavernous or cystic lymphangiomas.

Cavernous lymphangioma involves the mucosa and submucosa.

• Presenting symptoms:

GI bleeding

Abdominal pain

Obstruction

Intussusception or volvulus

• Endoscopic findings (cavernous type):

Polypoid tumor

White-yellow surface with strawberry mucosa.

Variable spontaneous bleeding and surface erosion.

• Treatment options:

The optimal treatment is radical excision. Incomplete resection may lead to recurrence.

Endoscopic ablation and palliation have been reported using argon plasma coagulation or polypectomy for GI bleeding.

6. Scripted voiceover

Voiceover Text

A 38-year-old woman is undergoing single balloon enteroscopy for abnormal capsule endoscopic findings in the small bowel.

1cm actively oozing, soft, friable, polypoid lesion is noticed in the proximal-mid small bowel.

The lesion is white-yellow with "strawberry" mucosal patterns. The surrounding mucosa is not indurated.

Due to its size, unknown etiology, and active bleeding status, we decided not to proceed with endoscopic hemostasis.

Endoscopic tattooing is being performed at the normal appearing mucosa 1-2cm proximal and distal to the bleeding lesion.

The patient underwent laparoscopic segmental small bowel resection. Surgical specimen shows a 2cm tumor with marked lymphatic dilatation within the mucosa and submucosa.

These findings are consistent with a cavernous lymphangioma.

Small bowel cavernous lymphangiomas are very rare and they involve the mucosa and submucosa.

The endoscopic findings of cavernous lymphangiomas include polypoid tumor, white-yellow surface with strawberry mucosa, variable spontaneous bleeding and surface erosion.

Endoscopists should be aware of this rare tumor and its unique endoscopic features.

The optimal treatment is radical excision, since incomplete resection may lead to recurrence.

Argon plasma coagulation or polypectomy have been used to achieve endoscopic ablation and palliation of GI bleeding.

Conflict of interest

Shou Tang and Feriyl Bhaijee have nothing to declare and have no conflict of interests.

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References

- [1] Allen JG, Riall TS, Cameron JL, Askin FB, Hruban RH, Campbell KA. Abdominal lymphangiomas in adults. J Gastrointest Surg 2006;10(5):746-51.
- [2] Hwang SS, Choi HJ, Park SY. Cavernous mesenteric lymphangiomatosis mimicking metastasis in a patient with rectal cancer: a case report. World J Gastroenterol 2009;15 (31):3947-9.
- [3] Okamoto D, Ishigami K, Yoshimitsu K, Irie H, Tajima T, Nishie A, et al. Hemorrhagic mesenteric cystic lymphangioma presenting with acute lower abdominal pain: the diagnostic clues on MR imaging. Emerg Radiol 2009;16(4):327-30.
- [4] Kida A, Matsuda K, Hirai S, Shimatani A, Horita Y, Hiramatsu K, et al. A pedunculated polyp-shaped small-bowel lymphangioma causing gastrointestinal bleeding and treated by double-balloon enteroscopy. World J Gastroenterol 2012;18 (34):4798-800.
- [5] Gerosa Y, Bernard B, Lagneau M, You K, Hoang C, Brasseur JL, et al. [Cystic lymphangioma of the duodenum revealed by digestive hemorrhage and associated with exudative enteropathy]. Gastroenterol Clin Biol 1993;17:591-3.

- [6] Onozato W, Nakamura T, Hatate K, Ozawa H, Satou T, Kokuba Y, et al. A case of lymphangioma of the ileum presenting with intussusception. Jpn J Gastroenterol Surg 2007;40:1531-5.
- [7] Mavrogenis G, Coumaros D, Lakhrib N, Renard C, Bellocq JP, Leroy J. Mixed cavernous hemangioma-lymphangioma of the jejunum: detection by wireless capsule endoscopy. Endoscopy 2011;43(Suppl 2 UCTN):E217-8.
- [8] Li F, Osuoha C, Leighton JA, Harrison ME. Double-balloon enteroscopy in the diagnosis and treatment of hemorrhage from small-bowel lymphangioma: a case report. Gastrointest Endosc 2009;70:189-90.
- [9] Hsu SJ, Chang YT, Chang MC, Yu SC, Lee JC, Shun CT, et al. Bleeding jejunal lymphangioma diagnosed by double-balloon enteroscopy. Endoscopy 2007;39(Suppl 1):E5-6.
- [10] Griffa B, Basilico V, Feltri M, Griffa A. [Submucosal jejunal lymphangioma: an unusual case with obscure gastrointestinal bleeding in an adult, detected by video-capsule endoscopy and treated by laparoscopy]. Minerva Chir 2006;61:529-32.
- [11] Ugwonali O, Coady M, Saxena R, Robert M, Horowitz N, Topazian M. Intraoperative enteroscopy for diagnosis of a bleeding jejunal lymphangioma. J Clin Gastroenterol 2000;31:333-5.
- [12] Barquist ES, Apple SK, Jensen DM, Ashley SW. Jejunal lymphangioma. An unusual cause of chronic gastrointestinal bleeding. Dig Dis Sci 1997;42:1179-83.
- [13] Day W, Kan DM. A small bowel lymphangioma presenting as a volvulus. Hong Kong Med J 2010;16(3):233-4.
- [14] Hanagiri T, Baba M, Shimabukuro T, Hashimoto M, Takemoto H, Inoue A, et al. Lymphangioma in the small intestine: report of a case and review of the Japanese literature. Surg Today 1992;22(4):363-7.