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Case Report

A Rare Case of Jejunal Lymphangioma Presenting with Obscure Gastrointestinal Bleeding

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Abstract

Small bowel lymphangiomas are rare benign tumours of the lymphatic system. Most lymphangiomas are asymptomatic and do not require treatment. Resection is required for patients with bleeding, intestinal obstruction, and intussusception. Obscure gastrointestinal bleeding poses a medical challenge for physicians. We report a case of jejunal lymphangioma diagnosed using video capsule endoscopy and treated with surgical resection in a 36-year-old woman with obscure gastrointestinal bleeding.

Keywords: Lymphangioma; Small Intestine; Video Capsule Endoscopy

Introduction

A lymphangioma is a benign tumour caused by dilatation of the lymphatic channels. Lymphangiomas typically occur in the head, neck, and axillary areas, as well as in parenchymal organs such as the spleen, liver, and bone. However, they rarely occur in the gastrointestinal tract, and they are especially rare in the small intestine. Lymphangiomas may occur during childhood and are thought to represent congenital malformations of the lymphatic spaces that form during early development. They may experience secondary changes as a result of inflammation, fibrosis, or obstruction of the lymphatic spaces. Lymphangiomas represent 6% and 1.4% to 2.4% of all small bowel tumours in children [1] and adults [2], respectively. Small bowel lymphangiomas are commonly asymptomatic when they are small; however, their enlargement can cause abdominal pain secondary to intestinal irritation, followed by gastrointestinal bleeding, anemia, intestinal obstruction, intussusception, and protein-losing enteropathy [3-5]. Obscure gastrointestinal bleeding (OGIB) remains challenging for gastroenterologists. However, more cases of small bowel lymphangiomas have been observed since the advent of video capsule endoscopy (VCE) and enteroscopy. We report a rare case of jejunal lymphangioma that was diagnosed using VCE and treated with surgical resection.

Case Presentation

We observed a case of jejunal lymphangioma in a 36-year-old woman who presented with OGIB. She had a 1-year history of dyspnea on exertion and iron-deficiency anemia. Her complete blood count revealed severe anemia (hemoglobin, 5.1 g/dL), and the fecal occult blood test result was positive. Although esophagogastroduodenoscopic and colonoscopic examinations

and computed tomography evaluations were performed at another hospital, the source of bleeding was not identified. She was referred to our hospital for capsule endoscopy. Capsule endoscopy (MiroCam®; IntroMedic, Minneapolis, MN, USA) revealed an actively bleeding polypoid lesion within the jejunum (Figure 1A). Enteroscopy showed a 1.6-cm polypoid lesion with white spots and a blood clot on the surface that was oozing at the proximal jejunum (Figure 1B). Two hemoclips were applied to the opposite side of the tumour. The patient underwent exploratory laparotomy with tumour resection (Figure 1C).

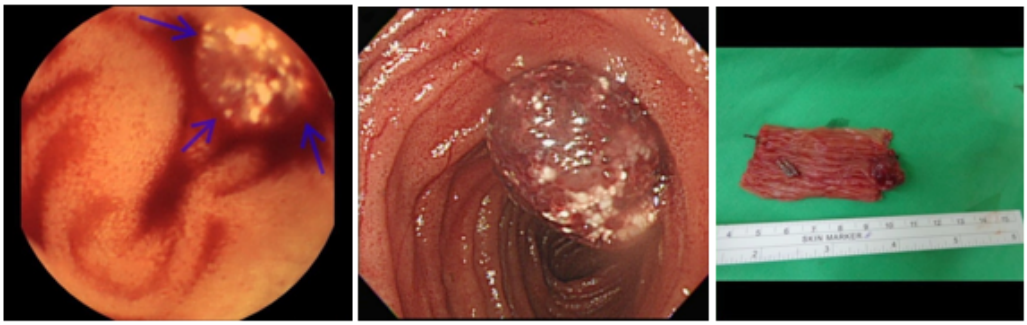

Figure 1: A: Capsule endoscopy reveals a polypoid lesion covered with white spots and active hemorrhage within the jejunum. 1B: Enteroscopy shows a 1.6-cm polypoid lesion with a broad base, white spots, and a blood clot on the surface that is oozing at the proximal jejunum. It is occupying almost half of the lumen. 1C: A tumor with hemorrhage is noted at the jejunum at 30 cm distal to the Treitz ligament.

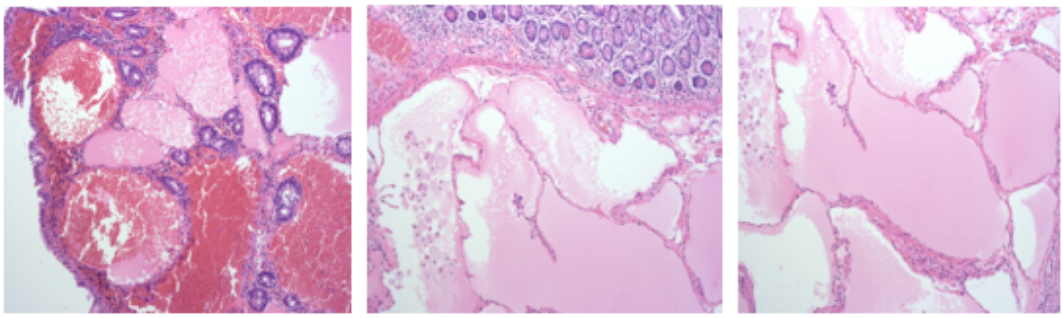

Figure 2: A: Hematoxylin and eosin staining of the small bowel specimen (×25) demonstrating a tumor involving the mucosal and submucosal layers comprising dilated lymphatic channels with surface hemorrhage. 2B: Higher magnification (×100) of the same specimen using hematoxylin and eosin staining. The tumor is mainly located at the submucosa. 2C: Higher magnification (×200) of the same specimen using hematoxylin and eosin staining. The channel wall is thin and lined with flattened cells. The channels are filled with lymphatic fluid.

Results

A histological examination revealed that the tumour comprised dilated lymphatic channels. The lesion was mainly located in the submucosa and included surface haemorrhage. The channels walls were thin and lined with flattened cells; furthermore, the channels were filled with lymphatic fluid (Figure 2A-C). The final pathological analysis results indicated small bowel lymphangioma with haemorrhage. The postoperative course was uneventful, and the anemia-related symptoms resolved after surgery.

Discussion

Lymphangiomas are benign tumors characterized by the presence of dilated lymphatic channels. These tumors are most common in the head, neck, and axillary regions. Lesions affecting the jejunum or ileum are extremely rare, accounting for less than 1% of all lymphangiomas [6]. Small bowel lymphangiomas are commonly asymptomatic and often diagnosed incidentally; however, they

sometimes present with acute or chronic gastrointestinal bleeding, abdominal pain, intestinal obstruction, volvulus, intussusception, and protein-losing gastroenteropathy [5-9]. The mechanism of gastrointestinal bleeding caused by lymphangiomas remains unclear. It has been postulated that lymphatic flow obstruction in such lesions increases pressure on the lymphatic–venous connections, thus causing retrograde blood flow into the lymphatic channel, resulting in gastrointestinal bleeding [10]. Depending on their size and clinical manifestations, small bowel lymphangiomas can be diagnosed using a barium enema, computed tomography, and endoscopic evaluation. During a review of 34 reported cases of lymphangiomas in Japan from 1967 to 1991, the clinical manifestations were divided into three groups depending on the type and location of the tumour: obstruction; bleeding; and symptoms arising from local irritability. According to previous reports, jejunal tumours most often present with melena, followed by abdominal pain and cramping; however, ileal tumours may manifest with intussusception or intestinal obstruction [2]. Another review of the English language literature from 1960 to 2009 revealed only 19 reports of 40 patients with small bowel lymphangiomas; during that review, equal sex distribution and a wide age range at presentation (5-75 years of age) were observed [11]. Since the advent of VCE and enteroscopy, the number of reported cases of lymphangioma has increased. Typical endoscopic findings include a soft submucosal mass with white spots on the surface and drainage of milky fluid after biopsy, which can be used to diagnose lymphangioma [12]. VCE can help locate small intestinal masses by allowing a direct view of the entire small intestinal mucosa, thus providing important clues indicating the need for further enteroscopic or surgical exploration of the lesions. Double-balloon enteroscopy is an effective procedure for diagnosing and treating OGIB. Several studies have demonstrated that bleeding intestinal lymphangiomas can be successfully treated by double-balloon enteroscopic therapy, such as endoscopic polypectomy, hemoclipping, and argon plasma coagulation [10,13,14]. In our case, VCE revealed a polypoid lesion with white spots on the surface and active haemorrhage at the proximal jejunum. Enteroscopy was performed; however, the lesion was not managed endoscopically because of its broad base and active haemorrhage, and because it occupied almost half of the lumen. Surgical resection is the standard treatment for symptomatic lymphangioma. However, endoscopic treatments such as coagulation and polypectomy have been applied for suitable cases [10,13,14]. This case report highlights the contributions of VCE to accurate preoperative diagnoses and strategic surgical planning.

Conclusion

Small bowel lymphangiomas are rare tumours of the small intestine with nonspecific clinical manifestations. This report describes a patient with gastrointestinal bleeding caused by a small bowel lymphangioma that was diagnosed with VCE and treated with surgical resection. Despite their rarity, small bowel lymphangiomas should be considered when patients present with OGIB.

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References

1. Fonkalsrud EW (1994) Congenital malformations of the lymphatic system. Semin Pediatr Surg. 3: 62-69.
2. Hanagiri T, Baba M, Shimabukuro T, Hashimoto M, Takemoto H, et al (1992) Lymphangioma in the small intestine: Report of a case and review of the Japanese literature. Surg Today. 22: 363-367.
3. Lawless ME, Lloyd KA, Swanson PE, Upton MP, Yeh MM (2015) Lymphangiomatous Lesions of the Gastrointestinal Tract: A Clinicopathologic Study and Comparison Between Adults and Children. American Journal of Clinical Pathology. 144: 563-569.
4. Chin CC, Shiau J, Luo CW, Hou MF (2023) Lymphangioma of small bowel in adults: A rare cause of abdominal symptoms. Asian Journal of Surgery. 46: 863-867.
5. Wang Z, Shen L, Zhou J, Tang J, Yi L, et al (2022) Clinical manifestation and treatment of small intestinal lymphangioma: A single center analysis of 15 cases. Front. Med. 9: 975698.
6. Bucciero F, Marsico M, Galli A, Tarocchi M (2015) Small bowel lymphangioma: A rare case of intestinal bleeding. Dig Liver Dis. 47: 815.
7. Wang L, Feng H, Chen B, Luan F (2022) Case Report: Endoscopic manifestations and clinical features of small intestinal lymphangioma A report of two cases. Front. Oncol. 12: 916295.
8. Kondo H, Ohki T, Ogawa S, Omori T, Onizuka H, et al (2022) Multiple intestinal lymphangiomas with episodic hemorrhage requiring partial laparoscopic resection: a case report. Surg Case Rep. 8: 56.
9. Zhao N, Fu Y, Wang Z, An Q, Jia W (2022) Case report: Submucosal cavernous lymphangioma causing jejuno-jejunal intussusception in an adult, Front. Surg. 9: 953840.
10. Wu TL, Hsu HT, Yen HH (2021) Jejunum lymphangioma: a rare case of obscure gastrointestinal bleeding with successful endoscopic therapy. Endoscopy. 53: E307-E308.
11. Morris-Stiff G, Falk GA, El-Hayek K, Vargo J, Bronner M, et al (2011) Jejunal cavernous lymphangioma. Case Reports. 2011: bcr0320114022–bcr0320114022.
12. Iwamuro M, Kawai Y, Takata K, Okada H, Yamamoto K (2013) Observation of Lymphangioma of the Duodenum by a Magnifying Endoscope with a Narrow-Band Imaging System. Case Rep Gastroenterol. 7: 229-233.
13. Kida A, Matsuda K, Hirai S, Shimatani A, Horita Y, (2012) A pedunculated polyp-shaped small-bowel lymphangioma causing gastrointestinal bleeding and treated by double-balloon enteroscopy. WJG. 18: 4798.
14. Li F, Osuoha C, Leighton JA, Harrison ME (2009) Double-balloon enteroscopy in the diagnosis and treatment of hemorrhage from small-bowel lymphangioma: a case report. Gastrointestinal Endoscopy. 70: 189-190.