**Case Report**

Small Intestinal Lymphangioma with Bleeding: A Case Report

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**Abstract**

Small intestinal lymphangioma is a rare benign tumor of the lymphatic system. Lymphangiomas account for 6% of small intestinal tumours seen in children but a smaller percentage, approximately 1.4–2.4% in adults. Here we describe the first case of small intestinal lymphangioma with bleeding in a 57-year-old woman. She presented with persistent gastrointestinal bleeding and upper and lower endoscopies did not reveal a bleeding source but lots of rufous intestinal contents. Laparotomy was performed and endoscopy revealed a small intestinal tumor. Partial resection of the small intestine was thus performed and the final pathological diagnosis was lymphangioma. However, accurate diagnosis could not usually be established preoperatively in spite of modern imaging techniques. To arrive at a definitive diagnosis and exclude malignancy, partial resection of the small intestine was considered to be the required treatment.

**ABBREVIATIONS**

OB: Stool Occult Blood

**INTRODUCTION**

Lymphangioma are rare benign tumors that appear to arise from birth defects of the lymphatic system. The formation of these tumors may be explained by obstruction of endolymphatic communication, dysembioplastic vascular tissue and systemic circulation [1-3]. Lymphangioma most commonly present as cystic or cavernous lesions. Herein, we report a case of Lymphangioma of the small intestine with bleeding.

**CASE PRESENTATION**

A 57-year-old woman suffering from dizzy for 4 mo and repeated melena for more than 3 mo of unknown origin. The complete blood count showed severe anemia, and stool occult blood (OB) was positive. Gastroscopy showed duodenal erosion. Colonoscopy showed there were a collection of rufous intestinal contents in entire colon, transverse colon and cecum were significantly; consider the possibility of large small intestinal bleeding. She underwent capsule endoscopy. This testing raised suspicions that the source of her bleeding was from the jejunum. Exploratory laparotomy was performed. However no significant bleeding lesions were found. So we performed intraoperative enteroscopy exploration. As a result, a gray mass was observed at 9cm distal to the ligament of Treitz. The enteroscopy was advanced as far as possible to examine the distal jejunum and ileum, but no other source of bleeding was evident. The mass was ill-defined, and the size was approximately 0.5 cm × 0.4 cm. Partial resection was thus performed including adjacent mesentery and small intestinal, with an end-to-end anastomosis. The small intestinal tumor was soft, and subsequent histopathological examination demonstrated that the stroma of the tumor was composed of clusters of lymphatic vessels with marked cystic dilatation, it was lined with regular endothelial cells. The cyst walls were made up of thin layers of smooth muscle, consisting of inflammatory infiltration in both the mucosal lamina propria and the submucosa (Figure 1). The pathology findings were compatible with lymphangioma with lymphangiectasia. At the annual follow-up, no recurrence was observed, and the patient is currently enjoying normal life.

**Figure 1** There are a lot of dilated lumen in the mucosa and submucosa; The lumen lined with endothelial cells, with pink lymph in it; Local small smooth muscle hyperplasia and local inflammatory cell infiltration.

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Keywords

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- Lymphangioma
- Benign tumor
- Gastrointestinal bleeding
DISCUSSION

Lymphangiomas are a heterogeneous group of lymphatic malformations that are composed of cystically dilated lymphatics. These malformations occur predominately in young patients, especially in newborns and infants, but rarely in adults. The common sites are the head, neck, and axillary regions. Other locations such as the abdominal or mediastinal cavity are rare, accounting for approximately 5% of lymphangiomas [4]. The etiology of lymphangiomas remains unclear, but a popular theory, the ‘blind sac’ theory hypothesis, postulates that a lack of lymphatic connections causes them to proliferate and dilate [5]. Lymphangiomas are classified as simple capillary, cavernous, and cystic types, depending on the size of the lymphatic spaces and the nature of the lymphatic wall [6].

Incidence of lymphangiomas varies from 1.2 to 2.8 per 1000 newborns, and both genders are equally affected [7]. The diagnosis in most cases (90%) is made before the age of two [8]. 60% of those patients display symptoms at the time of birth. Patients with small intestinal lymphangiomas have variable presentations including gastrointestinal bleeding, abdominal pain, intussusception, and protein-losing gastroenteropathy [9,10].

The clinical onset of lymphangiomas can vary from a slow growth of cyst over a period of years to an aggressively enlarging but non-invasive tumor. Their size varies based on the anatomical location and relationship to the adjacent tissues. Small tumors are usually superficial, whereas the larger ones are located in deeper layers and have a cystic texture. Usually complicated by spontaneous or traumatic hemorrhage, rupture, infection and acute intestinal obstruction. Gross examinations of the tumors are usually palpated as soft and compressible masses. Histologically, lymphangiomas consist of dilated lymphatics.

The standard management of lymphangiomas until recently has been through surgical resection, especially when the tumor increases in size and applies pressure on the surrounding tissues. Surgeons usually aim for complete removal of the tumor with surrounding organs of potential invasion, because there is a possibility of recurrence and invasion to surrounding organs [11]. The recurrence rates vary depending on the complexity of the tumors, the anatomical location and the adequacy of the excision. However, lesions that have been completely excised present 10%–27% recurrence, while 50%-100% of partially resected tumors may recur. There are reports of laparoscopic resections. Biopsy or percutaneous aspiration of the lymphangioma is not recommended, since, due to its multi-chamber character it does not bring positive therapeutic effects.

In summary, lymphangioma of the small intestinal is an uncommon lymphatic lesion, presenting with recurrent and painless gastrointestinal bleeding. Surgery is the treatment of choice for small intestinal lymphangiomas. However, with the advent of double-balloon enteroscopy, this modality may be able to treat small tumors and this indeed has been accomplished in two cases [12,13]. Despite its low frequency, this disease should be considered when gastrointestinal bleeding is observed.

REFERENCES