Case Report

Ileal lymphangioma as a cause of intestinal obstruction in an adult patient: A rare case report

Hasmukh Babaldas Vora[1], Badri Narayan Sharma[2]

From [1]Associate Professor, [2]Second Year MCH Resident, Department of Surgical Gastro-Enterology, SMT NHL Municipal Medical College, Ahmedabad, Gujarat, India

ABSTRACT

Most abdominal lymphangiomas are diagnosed at a very early age and are rarely found in adults. Abdominal lymphangioma is mainly found in the visceral organs, the retroperitoneum, the mesentery, or in the bones. In the gastrointestinal tract, they comprise <1% and are very rarely found in the ileum. Most of the time, gastrointestinal tract lymphangiomas are asymptomatic and are diagnosed incidentally. Symptomatic lymphangioma may present with a painless, palpable mass, colicky pain, and rarely with acute abdominal symptoms which leads to obstruction either due to volvulus or extrinsic mass effect which exerts traction on the bowel. A 38-year-old male case of ileal lymphangioma is described here which presented with lump and obstructive symptoms due to mass effect. Resection and anastomosis of the involved segment were performed with histopathology suggestive of submucosal cystic lymphangioma.

Key words: Adult ileal lymphangioma, Intestine, Obstruction

Cystic lymphangioma was first described in 1913 by Gaudier and Gorse as a rare benign cystic lesion containing chylous or serous fluid and composed of lymphatic channels lined by endothelial cells. In literature, it is reported that most lesions are diagnosed in children and there is no sex predominance with the most common site being the neck followed by the axillary region and very rarely in mediastinum, visceral organs, retroperitoneum, and mesentery. Lymphangioma of the gastrointestinal tract comprises <1% [1,2]. Lymphangiomas are usually solitary and not connected to the lymphatic channels in that region resulting in stasis and proximal dilatation. Most cases are asymptomatic and incidentally diagnosed. When symptomatic, patients typically present with a painless abdominal mass. Very rarely, the mass may cause acute abdominal pain or intestinal obstruction due to luminal obstruction or, less commonly, small bowel volvulus. Diagnosing small bowel lymphangiomas pre-operatively is challenging, even with gastro-enteroscopic biopsy. On abdominal computed tomography (CT), they typically appear as well-defined submucosal cystic lesions without contrast enhancement.

Small bowel lymphangiomas are rare in adults, with only a few published case reports. Our case also presents with intermittent obstructive symptoms with abdominal mass in an adult patient and the CT scan is not conclusive of any specific diagnosis so planned for diagnostic laparoscopy. The lesion was surgically resected with 5 cm macroscopic negative margins and the specimen was sent for histopathological examination which is suggestive of ileal lymphangioma. In an adult patient, ileal lymphangioma is very rarely seen so I am presenting this case report.

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CASE REPORT

A 38-year-old male was admitted with chief complaints of on-and-off generalized abdominal pain and distension for 1 year. The pain was insidious in onset, mild to moderate colicky, non-radiating, non-referred, increased during walking, jolting, and relieved by medications. Pain and distension were associated with constipation, nausea, and vomiting with gastric contents occurring an hour after taking meals with aggravation of symptoms from the past 15 days with inability to pass motion and flatus from the past 3 days. Due to fear of abdominal pain, the patient started taking only a liquid diet. The patient had no other significant history except laparoscopic appendicectomy 5 years back.

On examination, the patient had tenderness and fullness in the right iliac with an increase in local temperature. A boggy lobulated mobile lump was palpable in the right iliac fossa which was around 5 × 5 cm in size. Peristalsis was present. On per rectal examination, tenderness is present in the right anterior side.

Correspondence to: Badri Narayan Sharma, C/O Ram Ray Sharma, Brahmin Mohalla, VPO. -Rajpura, Teh. -Bassi, Jaipur - 303305, Rajasthan, India. E-mail: badrinarayansharma913@gmail.com

Laboratory investigations were done which were within normal limits. Ultrasound abdomen was suggestive of minimally edematous wall of the base of cecum with multiple enlarged hypoechoic lymph nodes seen in the right iliac fossa, the largest conglomerated lymph node measures 29 × 37 mm.

Contrast-enhanced CT was suggestive of ill-defined nodular wall thickening involving a small bowel loop (proximal ileum) in the right lower abdomen with adjacent localized multi-lobulated area abutting terminal ileum and cecum. The size of multi-lobulated area was approximately 95 × 90 × 60 mm and wall thickness was 10–12 mm with a segment of 6.5 cm. A mildly thickened wall of the cecum, ascending colon, and terminal ileum was seen with a wall thickness of 4–5 mm. Multiple mesenteric nodes were seen in the right iliac fossa (Figs. 1 and 2).

Differential diagnoses in our case include ileocecal tuberculosis, small bowel malignancy, mesenteric lymphangioma, and carcinoid tumor. Due to its location and CT finding, we suspected for tuberculosis, small bowel malignancy, and carcinoid tumor, and these are difficult to rule out on the basis of clinical evaluation, investigations, and imaging findings.

After thorough investigations and obtaining prior consent, the patient was prepared for surgery. A diagnostic laparoscopy was done. During laparoscopy, a conglomerated cystic mass arising from the wall of ileum on the mesenteric side was seen which was around 40 cm proximal to the ileocecal junction with many mesenteric lymph nodes (Fig. 3). After inspecting the abdominal cavity laparoscopically, the abdomen was opened and resection and anastomosis were done with 5–8 cm grossly negative margin (Fig. 4a).

The resected sample (Fig. 4b) was sent for histopathological examination which showed one submucosal lesion on the mesenteric surface measuring 70 × 45 × 30 mm. The lesion was 8.0 cm away from the proximal surgical margin and 5.0 cm away from the distal surgical margin. On the cut section, the lesion was yellowish-brown with blood-filled cystic spaces and microscopic findings suggestive of submucosal cystic lymphangioma with serositis and micro-abscess formation (Fig. 5).

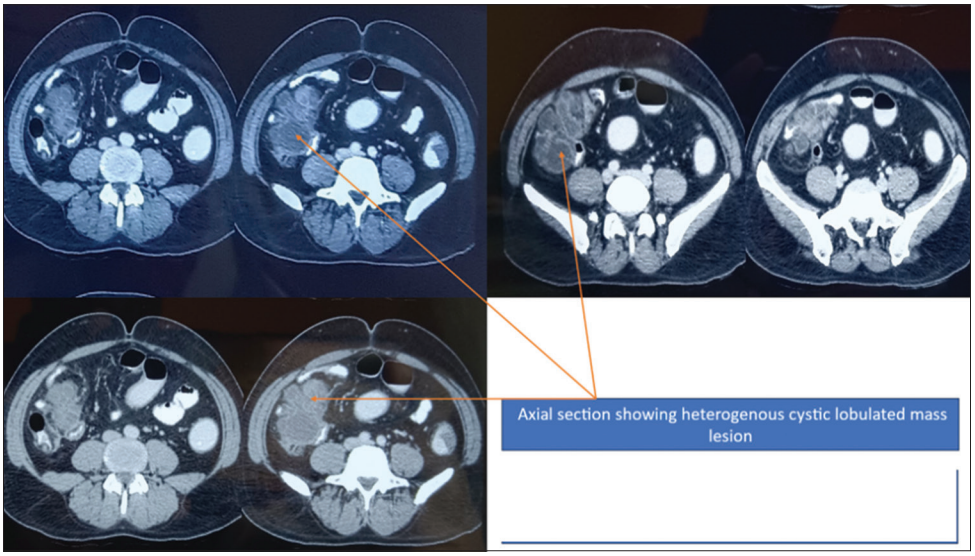

Figure 1: Axial section of contrast-enhanced computed tomography abdomen showing mass in the right iliac fossa

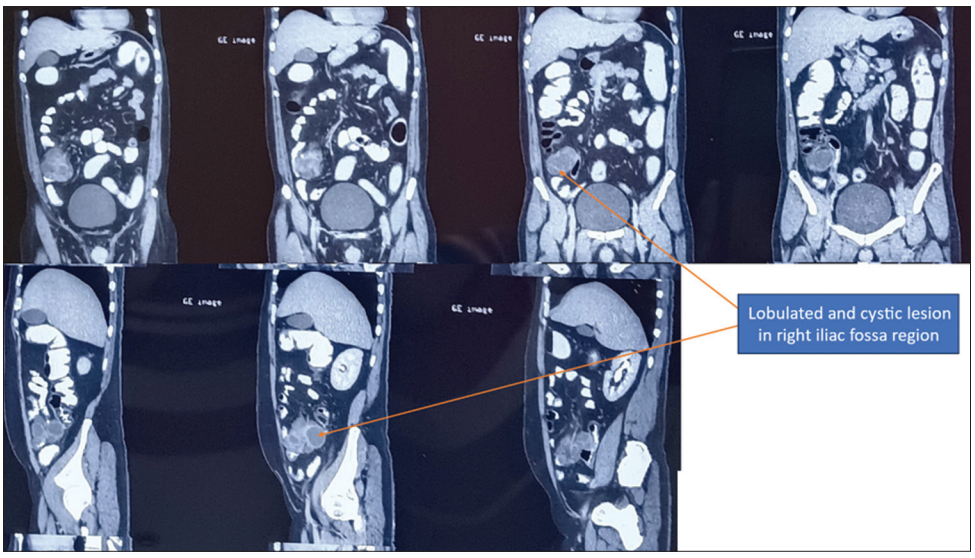

Figure 2: Contrast-enhanced computed tomography abdomen coronal and sagittal section showing cystic lobulated lesion in right iliac fossa

Table 1: Cases of lymphangioma originating from the ileum and ileocecal valve in adults

Case no.	First author	Year	Age	Sex	Main symptom	Location	Intervention
1	Sato *et al*. [5]	1981	41 years	Male	Abdominal pain	Ileocecal	Surgery (open)
2	Hanagiri *et al*. [6]	1992	53 years	Male	Abdominal pain	Ileum (250 cm from treitz ligament)	Surgery (open)
3	Uncu *et al*. [7]	1997	43 years	Male	Palpable mass	Ileum	Surgery (open)
4	Uncu *et al*. [7]	1997	19 years	Male	Vomiting	Ileum	Surgery (open)
5	Yao *et al*. [8]	2001	56 years	Female	Abdominal pain	terminal ileum	Surgery (open)
6	Konstantinidis *et al*. [9]	2005	-	Female	Abdominal pain	Ileum	Surgery (Lap)
7	Li *et al*. [10]	2009	69 years	Male	Melena	210 cm from front teeth	Endoscopic hemostasis
8	Al-Obeed and Abdulla [11]	2014	56 years	Male	Epigastric pain	Ileocecal valve	Surgery (Lap)
9	Ilhan *et al*. [12]	2016	43 years	Female	Weight loss	Ileum	Surgery (open)
10	Kohga *et al*. [13]	2017	20 years	Female	Right upper abdominal pain	Ileum	SIL-assisted ileal resection
11	Khan *et al*. [14]	2017	24 years	Female	Abdominal pain	Ileum	Surgery (open)
12	Giuliani *et al*. [15]	2019	41 years	Male	Abdominal pain	Ileum (80 cm from ileocecal valve)	Surgery (open)
13	Mohammed and Musa [16]	2019	31 years	Male	Abdominal pain	Ileum (60 cm from ileocecal valve)	Surgery (open)
14	Imamura *et al*. [17]	2020	37 years	Male	Abdominal pain	Ileum (100 cm from ileocecal valve)	Surgery (Lap)

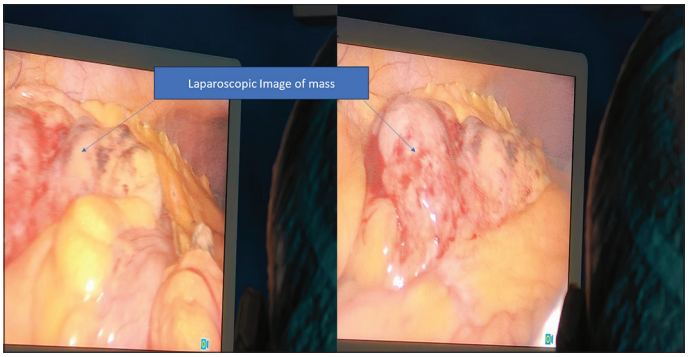

Figure 3: Laparoscopic photograph of ileal submucosal lymphangioma

Postoperatively, the patient was well and discharged uneventfully on post-operative day 7 after starting oral feeds.

DISCUSSION

Lymphangiomas are rare lymphatic system tumors with an incidence of 6% of small bowel tumors in children and 1.4–2.4% in adults. Approximately, 60% of patients with lymphangiomas are younger than 5 years; however, a significant number of abdominal lymphangiomas do not manifest until adulthood. The common sites of lymphangiomas are the head, neck, and axillary regions. Tumors at other locations, such as the abdominal or mediastinal cavity, are rare and account for approximately 5% of all lymphangiomas. Among these, lymphangiomas of the small bowel are very rare, and lymphangiomas in the jejunum or ileum are extremely rare, accounting for <1% of all lymphangiomas.

The clinical presentation of lymphangioma is unspecific and often presents with nausea, vomiting, and abdominal discomfort or pain. Other symptoms are hemorrhage, infection, perforation, intussusceptions, torsion, rupture, and protein-losing gastroenteropathy.

The majority of the lesions are diagnosed incidentally and rarely before surgery. The disease may be suspected with the

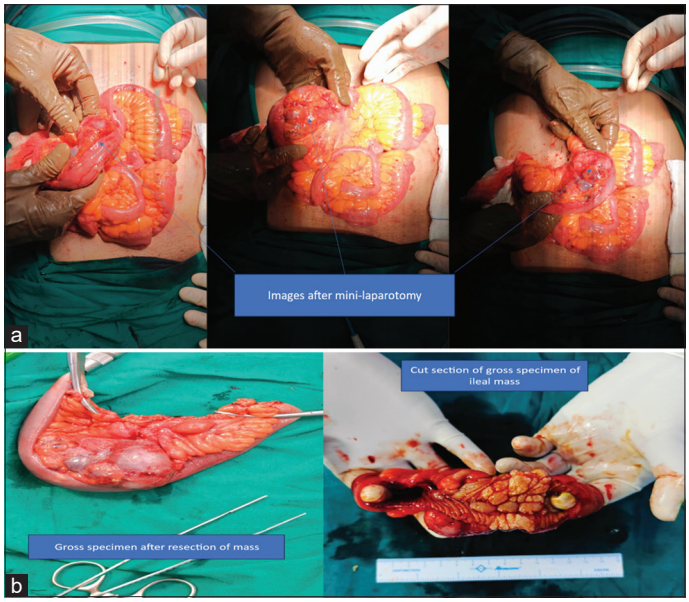

Figure 4: (a) Intraoperative photograph after mini-laparotomy; (b) Specimen after resection of lymphangioma mass segment of ileum

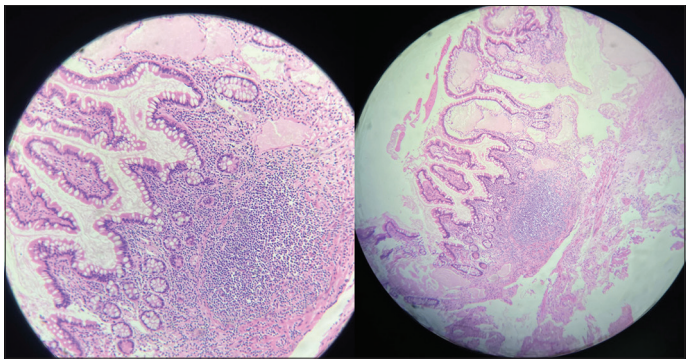

Figure 5: Microscopic images of ileal lymphangioma

typical radiological features. In endoscopy or enteroscopy, the lesions appear as elevated, yellowish, or white submucosal polypoidal lesions with intact overlying mucosa and dilated

overlying vessels. In the pediatric age group, lymphangiomas tend to be larger in size than those in adults, and most arise from the mesenteric. Clinically, most patients have emergency presentations such as acute abdomen or intestinal obstruction, while in adults they are usually discovered incidentally or present with abdominal mass.

Intra-abdominal lymphangiomas usually occur in the retroperitoneum but may affect the small bowel mesentery, liver, and pancreas. To date, we found only 20 case reports on lymphangiomas of the ileum and ileocecal valves in adult patients, and not a single case report was found in India. A case series of 18 adult patients from 1999 to 2019 with lymphangioma in duodenum were 6 cases (33.33%), 8 cases (44.44%) from the jejunum, and 2 patients (11.11%) from ileum [3]. Another case series of 15 adult patients from January 2010 to January 2021 were pathologically diagnosed with small intestinal lymphangioma. Lymphangioma was located in the ileum in 3 cases (20%) and jejunal-ileal junction in one (6.7%) [4]. Table 1 summarizes the previous case reports [5-17].

Lymphangioma may have characteristic radiological findings. Al-obeed and Abdulla reported that in CT scans it appears as homogenous, non-enhancing lesions with variable attenuation values. Attenuation depends on whether the fluid is chylous or serous.

Surgical resection is required when clinical symptoms are so severe that they cannot be relieved by other measures, or when pre-operative diagnosis is not available and there is suspicion of malignancy. Small lesions <2 cm accessible endoscopically are managed by submucosal resection. Complete surgical resection with negative margins offers a good overall prognosis. When complete resection is done, long-term follow-up is not recommended as the recurrence is very rare. In cases of incomplete resection, close follow-up is recommended as there is a chance of 10% recurrence rate in the microscopic positive resection margin. Surgical segmental resection of the bowel, including the lesion, is the optimal treatment for avoiding recurrence. Our patient underwent surgical resection of a small bowel segment including the lesion.

CONCLUSION

Ileal lymphangiomas are rare benign lesions with non-specific symptoms but sometimes present with specific symptoms due to mass effects or complications. Usually, diagnosis is suggested by imaging studies but requires histopathological confirmation for proper definitive diagnosis. Complete surgical resection is the main form of therapy whenever possible. When complete resection is performed, long-term follow-up is usually not recommended as the recurrence is very rare.

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