Case Report

Hemolymphangioma of the transverse mesocolon: a case report and literature review

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Abstract: Hemolymphangioma is an extremely rare type of lymphatic and vascular malformation, histologically comprised of both cystic dilated veins and lymphatic vessels. They have been reported to occur in the skin, extremities, pancreas, spleen, mediastinum, as well as in the gastrointestinal tract. A 61-year-old male patient presented with a 2-week history of left lower abdominal and back pain. He had no relevant personal or family past medical history. He denied fever, trauma or weight change, but had noted early satiety with eating. On physical examination, a 10 cm soft, mobile, well-defined, minimally tender mass was palpated in the lower left abdomen. Computed tomography confirmed a large intraperitoneal cystic mass, and resection was advised. The mass was completely excised laparoscopically from the transverse mesocolon. Histopathology verified the diagnosis of hemolymphangioma. The patient recovered uneventfully, and no recurrence was identified at 3 months follow-up. Hemolymphangioma is more common in women and occurs in the fourth to fifth decades of life. The intent of this case report and literature review was to highlight the key aspects of presentation, organ involvement, imaging, histopathological characteristics, and treatment of hemolymphangioma involving the gastrointestinal tract.

Keywords: Case report; hemolymphangioma; transverse mesocolon

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Introduction

Hemolymphangioma is a congenital malformation of the vascular system, comprising both venous and lymphatic components. The interval from its prenatal origin to clinical presentation is quite variable but often the evolution of hemolymphangioma has been measured in decades. To progress from a diminutive cyst to a huge cystic tumor, it is almost never aggressive (1). Review of the English literature through January 2021 revealed only a limited number of hemolymphangioma cases involving the digestive tract organs such as the pancreas, liver, stomach, duodenum, small and large intestine. The intent of this case report and literature review was to highlight the key aspects of presentation, organ involvement, imaging, histopathological characteristics, and treatment of hemolymphangioma involving the gastrointestinal tract. We present the following article in accordance with the CARE reporting checklist (available at https://dx.doi.org/10.21037/tcr-21-176).

Case presentation

A 61-year-old male patient was admitted to the Second Hospital of Lanzhou University due to a 2-week history of intermittent left lower back pain. Subsequently, the pain was noted to include the left lower abdomen, and initially, no obvious cause was identified. The patient had noted a history of abdominal distension, but denied diarrhea, nausea, vomiting, and fever. There was no history of abdominal trauma, prior surgery or significant recent

weight change. The patient had enjoyed normal health, his father and children were alive and well. His mother died of stomach cancer, not thought to be hereditary. The patient was not taking any medications. Physical examination revealed only mild tenderness in the left lower abdomen, but with no rigidity or rebound tenderness. An approximately 10 cm soft mobile well-defined mass was palpable in the left lower abdomen. Routine blood tests, biochemical examinations, and tumor markers were unremarkable. Computed tomography (CT) with vascular enhancement confirmed a large cystic mass in the left abdominal cavity, which was highly suspected to be a hemolymphangioma. It could not be clearly differentiated from other space occupying lesions in the abdominal cavity, but required surgical excision and pathological examination to provide a definitive diagnosis (*Figure 1*). The differential diagnosis was limited to such lesions as mesenteric cyst, lymphangioma, enteric duplication cyst, omental cyst, or stromal cell tumor. The patient underwent laparoscopic exploration on October 14, 2020, during which the tumor was found to originate from the transverse mesocolon and was completely excised. Pathological diagnosis of mesenteric hemolymphangioma was confirmed (*Figure 2*). The patient's postoperative course was uneventful, and at the 3-month follow-up, the patient had returned to normal health with no sign of cyst recurrence. This study was approved by the Medical Ethics Committee of the Second Hospital of Lanzhou University. The ID/number of ethical approval is 2020A-279. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient.

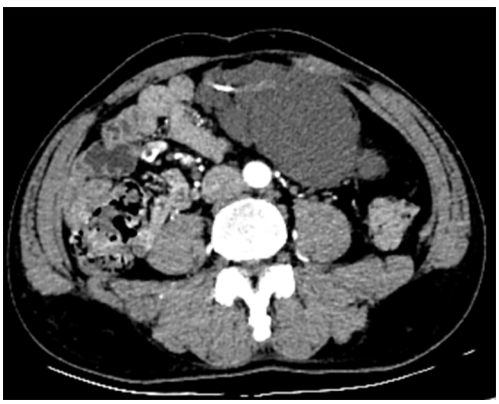

Figure 1 Computed tomography demonstrated a large, cystic, space-occupying lesion (arrow) in the left peritoneal cavity, consistent with a lymphangioma.

Discussion

We used the PubMed database to conduct a systematic review of existing medical literature, with "hemolymphangioma" as the key search word to retrieve all the studies published as of January 2021. After reviewing and summarizing each published article, we selected 25 original studies with 19 case reports describing hemolymphangioma originating from the liver, stomach, duodenum, small intestine, colon, rectum, pancreas and other digestive tract organs (*Table 1*) (2-20). To summarize, hemolymphangioma principally occurred in adult patients with an average age of 42 years (range, 3–70 years), with 84% (16/19) over the age of 20. It is more common in

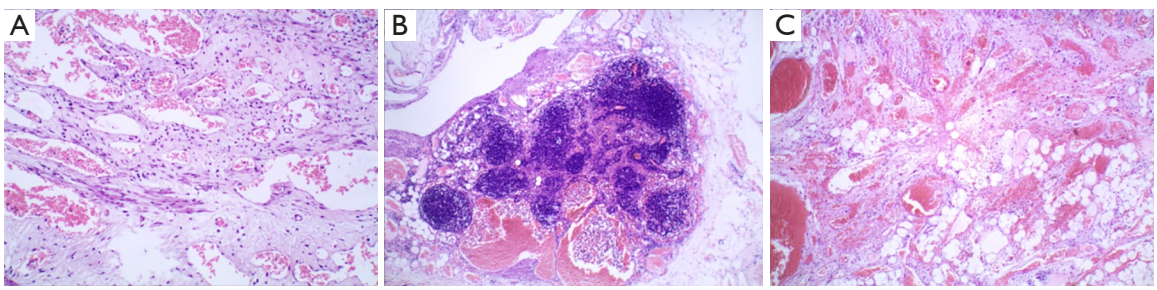

Figure 2 Hematoxylin and eosin stained slides of the cystic wall of the hemolymphangioma (H&E staining, ×40). Thin-walled lumens of different sizes were seen in the retinal tissue, which were lined with a single layer of flat epithelium. The lumens contained red blood cells or protein fluid, and lymphocytes. Proliferative lymphatic tissue was seen around some lumens. These findings were consistent with the diagnosis of hemolymphangioma (mesenteric masses).

Table 1 The clinical characteristics and management choices of 19 patients with hemolymphangioma [2–20]

Case/ number	Publication year	Age (years)/ sex	Localization	Preoperative diagnosis	Size (cm)	Chief complaint	Treatment	Physical Examination	imaging findings	Follow-up (months)	Recurrence	Evolution
2	2003	53/F	Pancreas	Space-occupying lesion of the head of pancreas	4×3	Abdominal pain and weight loss of 3 kg	Pancreatoduodenectomy with jejunostomy	Epigastric and right hypochondrium pain	Ultrasonography (US) showed a polycystic mass by the right renal pelvis. Computed tomography (CT) and magnetic resonance imaging (MRI) demonstrated a heterogeneous mass next to the head of the pancreas that partially compressed the right renal pelvis	NA	Not reported	Favourable
3	2008	53/M	Pancreas	The pancreatic neoplasm	NA	Severe anemia due to gastrointestinal bleeding	Pylorus preserving pancreatoduodenectomy was performed	Anemic appearance with distension and pain in the upper abdomen	CT revealed a heterogenous mass at the pancreatic head and suspected invasion to the duodenal wall. Ultrasonography showed a huge mass at the pancreatic head with a mixture of high and low echoic areas	12	Not reported	Favourable
4	2019	20/F	Pancreas	Large retroperitoneal tumor	18×16×12.5	A mass in abdominal cavity and epigastric discomfort about a week	Along the surface of the duodenum and pancreas, tumor (including partial transverse mesocolon and greater omentum) excision was performed.	A great abdominal mass	Abdominal computed tomography demonstrates a large tumor behind the peritoneum, possibly from the pancreas, compressing the duodenum with a polycystic structure and partial blood flow	26	Not reported	Favourable
5	2011	68/M	Stomach	Submucosal benign cystic lesion	4.5×3.1	Mild epigastric discomfort 3 months after meals	Under EUS guidance, successfully excised	Not complain of any other symptoms	The undisturbed CT scan of the upper abdomen showed a well-defined, uniform, low-attenuation mass near the lesser curvature of the posterior wall of the stomach. Contrast-enhanced CT scan showed no obvious enhancement in the arterial phase and portal vein phase, but slight enhancement in the 2-minute delay scan	18	Not reported	Favourable
6	2012	57/F	Small intestine	Small intestinal tumor	5.0×4.0	Recurrent melena more than 2 months	Partial intestinal resection	Not reported	Enteroscopy showed a gray mass with ulcers and erosion in the small intestine 30 cm distal to the flexor tendon	12	Not reported	Favourable
7	2013	37/M	The rectum	Rectal cancer	20×8×8	Rectal bleeding and tenesmus	Low anterior resection of the rectosigmoid colon with handsewn transanal colo-anal anastomosis	Mild tenderness on the left lower quadrant	Colonoscopy revealed an extensive hypervascular submucosal lesion arising from the rectosigmoid junction colon to the distal edge of the anus. Endoscopic ultrasonography demonstrated an extensive anechoic mass with clear edge. Magnetic resonance imaging (MRI) showed a significant thickness of the rectal wall, extending to the distal edge of the anus, with a narrowing lumen	12	Not reported	Favourable
8	2013	39/F	Pancreas	Mucinous cystadenoma or cystadenocarcinoma	10×7	Abdominal pain one day	Pancreatic body and tail combined with spleen resection	Slight tenderness in the left lower abdomen without rebound pain	The boundary is clear, cystic and solid, with a cable-like septum in the center of the cystic area. There is no change in enhanced CT images	NA	Not reported	Favourable
9	2014	24/F	Duodenum	Duodenal mass	4.0×1.5	Severe and undetermined anemia	A local wide excision of the tumor	Not complain of any other symptoms	Magnetic resonance demonstrated a solid, polypoid mass (40 mm ×15 mm) at the lateral wall of the second/third portion of the duodenum with mild contrast enhancement, with no evidence of ampullary obstruction or periduodenal tissue infiltration	4	Not reported	Favourable
10	2014	57/F	Pancreas	The pancreatic neoplasm	7.8×6.0	Epigastric discomfort for 10 days	A wide local resection of the tumor	Mild pain in the left hypochondrium without rebound tenderness	Abdominal computed tomography (CT) showed a cystic–solid tumor with an irregular shape, in the neck and body of the pancreas. The tumoral cystic wall and its internal division could be seen intensified on contrast-enhanced CT images compared with those on precontrast images	2	Not reported	Favourable
11	2016	3/M	Greater omentum	Intraperitoneal benign cystic lesion	20×15×6	Mild but progressively increasing abdominal pain around umbilical region for 2 days	Abdominal laparotomy followed by surgical excision	Abdominal swelling, no tenderness or rebound tenderness	Abdominal computed tomography (CT) scan revealed a large intraperitoneal mass occupying almost all of the abdominal cavity and pelvis	6	Not reported	Favourable
12	2017	57/M	Rectum	Rectal hemangioma	25 cm long lesion	Massive rectal bleeding (rectorrhagia) for 5 months	Whole of the rectum and part of the sigmoid colon were excised and sigmoid-anus anastomosis was done.	On rectal examination, fresh blood was seen around anal region and soft mass was felt on digital rectal examination	Contrast-enhanced CT showed homogeneous thickening of the intestinal wall, uneven enhancement in the venous phase, and lesions extending from the distal sigmoid colon to the entire rectum	6	Not reported	Favourable

Table 1 (*continued*)

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Case/ number	Publication year	Age (years)/ sex	Localization	Preoperative diagnosis	Size (cm)	Chief complaint	Treatment	Physical Examination	imaging findings	Follow-up (months)	Recurrence	Evolution
13	2017	42/F	Liver	Solid focal liver lesion	11.6×16.5	Right upper abdominal weakness and acute abdominal pain for 2 months	Right hemihepatectomy was conducted	a large abdominal mass and apparent conjunctival pallor	Abdominal computed tomography revealed an enormous multilocular cystic mass located at the right lobe of the liver, measuring 11.6×16.5 cm	NA	Not reported	Favourable
14	2017	45/F	Jejunum	Upper gastrointestinal hemorrhage	8-cm long	Recurrent melena for about a year	A 15-cm segment of jejunum was resected with primary anastomosis	Not reported	Video capsule endoscopy showed a zone of lymphangiectasias with red blood in the proximal jejunum	NA	Not reported	Not reported
15	2018	30/F	Pancreas	Abdominal neoplasm	12×10×7.5	Abdominal distension and an epigastric mass about 3 weeks	Body and tail pancreatectomy combined with middle colic artery and vein resection were performed	A soft mass in the upper abdomen	Computed tomography revealed a large multilocular cystic tumor in the neck and body of the pancreas	24	Not reported	Favourable
16	2018	28/M	Pancreas	A retroperitoneal mature liposarcoma or ganglioneuroma	8.0×10.0	Right upper abdominal pain for 2 days	Pylorus preserving pancreatoduodenectomy was performed	A soft abdomen, no abdominal varicose veins, and a mass approximately 8.0×10.0 cm^2 at the right upper quadrant that had an unclear border, poor activity, and tenderness (but no rebound tenderness)	Plain CT showed a cystic-solid mass of mixed density with a size of approximately 12 cm in front of the right kidney and behind the pancreatic head. Enhanced CT showed that the solidified part of the lesion was slightly strengthened, and the lesion's boundary with the surrounding adipose tissues was unclear as it partially wrapped around the duodenum. MRI showed slightly high intensity and scattered low intensity on T1-weighted imaging (T1WI) and high/low mixed intensity on T2-weighted imaging (T2WI)	NA	Not reported	Favourable
17	2018	70/M	Small intestine	The small intestine neoplasm	2.0×1.7×1.2	A 3-year history of iron deficiency anemia with occasional dark stool	Laparoscopic small bowel resection	Not complain of any other symptoms	Antegrade double-balloon enteroscopy was carried out, which demonstrated a 20 mm raised, granular lesion with white and thickened villi located 120 cm distal to the ligament of Treitz	NA	Not reported	Not reported
18	2019	20/F	Jejunum	Stomach neoplasm	6×4×3	Intermittent anemia 7 years, black stool 1 month	Laparotomy	Nemic appearance and the laboratory tests showed iron deficiency anemia (hemoglobin was 52 g/L)	Enhanced computer tomography scan showed a low-density mass sized approximately 6×3×4 cm in the middle left part of the abdomen, with partial bowel dilatation	NA	Not reported	Not reported
19	2019	55/F	Jejunum	Jejunal space-occupying lesion	3.×3；2×2	Discomfort in the right upper abdomen for 2 months	Laparotomy	mild tenderness on the right upper abdominal quadrant, with no rebound tenderness, and no abdominal mass	CT demonstrated a space-occupying lesion in proximal jejunum with calcium deposition, which had exhibited enhancement after contrast injection	6	Not reported	Favourable
20	2020	42/M	Small intestinal	Gastric ulcer and anemia	NA	Repeated episodes of melena, dizziness, fatigue, decreased athletic ability for more than 2 months	Enteroscopic injection sclerotherapy	Anemic face and upper abdominal tenderness	Capsule endoscopy revealed a prominent lesion in the jejunum about 150 cm from the distal end of the Treitz ligament	12	Not reported	Favourable

NA, the data were not available.

female patients with a male to female ratio of 1:1.38 (8/11). The anatomical locations were: pancreas 37% (7/19), small intestine 31% (6/19), rectum 11% (2/19), and stomach, duodenum, liver, and greater omentum each with 5% (1 case each). These patients typically presented with either abdominal pain or blood in the stool, but these symptoms could persist for weeks or months, complicated by repeated episodes of blood loss and significant iron-deficiency anemia. Additionally, over time, symptoms of bloating, compression, and even obstruction became evident as the mass slowly but progressively enlarged. The most common physical signs were abdominal tenderness and a palpable abdominal mass.

Hemolymphangioma, is a rare, congenital, benign malformation of the vascular system, histologically characterized by cystic dilated lymphatic and blood vessels. It originates from mesenchymal tissues, and bleeds easily with fewer clear lymphatic vessels (13). Hemolymphangiomas can be classified as primary and secondary according to the cause of the disease, with the former being considered a congenital obstruction of the venolymphatic communication between dysembryoplastic vascular tissue and the systemic circulation (11). The latter is caused by poor lymphatic drainage resulting from surgery or trauma (21,22), which is extremely rare in clinical practice. The clinical manifestations of hemolymphangioma vary depending on the size and location of the tumor. The variability of symptoms results from pressure exerted by the large cystic mass on adjacent structures, or the occurrence of complications, such as bleeding, perforation, torsion, or rupture of the tumor itself (17). In actual clinical practice, a preoperative diagnosis specifically of hemolymphangioma is virtually impossible due to its rarity, lack of characteristic imaging features or typical clinical manifestations.

Hemolymphangiomas occur predominantly in the pancreas, spleen and lower limbs, but rarely in the gastrointestinal tract. A clinical diagnosis of an intraperitoneal cystic mass is most often established on the basis of imaging. Both CT and magnetic resonance imaging (MRI) are quite valuable for determining the extent and possible invasion of tumors in general thereby facilitating preoperative surgical strategies (23). Each has its own particular advantages, and they are often complementary. A CT with vascular enhancement can identify areas of a tumor that are highly vascular or abut against important vessels. Hemolymphangiomas are a combination of dilated veins, dilated lymphatic vessels, and normal interstitial tissue. The signal for the cystic fluid by MRI examination is also related to the ratio of blood vessels to lymphatic vessels in the tumor (24). Accordingly, the T1W1 signal is mainly low or slightly low, whereas the T2W1 signal is mainly high.

As noted, abdominal hemolymphangiomas are principally manifested as space-occupying lesions in the abdomen, mainly presenting with compression symptoms, and should be differentiated from other such lesions, such as gastrointestinal stromal tumors, simple lymphangioma, and peritoneal pseudomucus. However, the final diagnosis requires pathological examination. Gastrointestinal stromal tumors are the most common stromal tumors of the gastrointestinal tract. They can also occur in the omentum, mesenteric, and retroperitoneum. They tend to grow exogenously. The histological types are mainly spindle cells or epithelioid cells (25,26). Many gastrointestinal stromal tumors show considerable cystic changes and sometimes intratumoral bleeding (27). Immunohistochemical staining is characterized by a diffuse and strong positive reaction for cd117, a positive reaction for CD34, and a negative reaction for S100 protein and desmin (28). Lymphangioma is a benign multilocular cystic mass that can occur anywhere in the abdominal cavity and is hidden between structures. Ultrasound can help distinguish intestinal duplication cysts from other mesenteric and omental cysts in the abdomen, the key sign being the double intestinal wall on the mesenteric side. Pathologically, a lymphangioma is a large, multi-cavity, thin-walled tumor, whose content is mainly chyle, but it may also be serous or hemorrhagic. Histologically, there are dilated endothelial cells arranged in the dilated lymphatic lumen (29).

Hemolymphangiomas are usually soft, have clear boundaries, exhibit polycystic changes in the cyst, and contain blood and lymph vessels in the cyst fluid. The hemolymphangioma specimen from the current case showed multi-nodular protrusions on the surface. The cut surface was grayish-yellow and covered with multiple small cysts. The maximum diameter of the cyst was approximately 12.5 cm, which was consistent with the appearance of a hemolymphangioma. The diagnosis of hemolymphangioma is established on postoperative pathology. A large number of expanded blood vessels and lymphatic vessels are characteristically observed microscopically. Moreover, hemolymphangiomas can be further distinguished through immunohistochemistry. Both vascular and lymphatic endothelial cells express CD31 and CD34, whereas D2-40 is only expressed in lymphangioma and some malignant vascular tumors. Such indicators can help solidify the diagnosis. Tumor cells in the current

case expressed CD34 (vascular and interstitial +) and D2-40 (lymphatic vessels +), thereby adding strong evidence for a pathological diagnosis of vascular lymphangioma. Complete resection provides optimal treatment, with low recurrence rates (30). Hemolymphangioma recurrence is strongly associated with the specific location of the tumor and the completeness of surgical resection. Published studies have reported that 10–27% of completely resected tumors exhibit recurrence, whereas 50–100% of only partially resected tumors recur. Non-surgical treatments, including cryotherapy, laser therapy, radiation therapy, and local injection of a sclerosing agent, have also been utilized (31). In conclusion, the present case involving an elderly male patient with hemolymphangioma provides a concise, valuable vignette to summarize this lesion. He presented with abdominal pain and a palpably soft, mobile mass, further characterized by CT scan as a cystic, noninvasive intraperitoneal benign-appearing tumor. It was completely resected, and with the combination of standard histopathology and immunohistochemistry, the diagnosis of hemolymphangioma was established. The patient recovered and had an excellent prognosis. With symptoms such as abdominal space occupation, abdominal pain and obstruction, and when common benign and malignant tumors in the abdominal cavity have been excluded, the possibility of a gastrointestinal venolymphatic cystic tumor should be considered.

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Footnote

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://dx.doi.org/10.21037/tcr-21-176). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. This study was approved by the Medical Ethics Committee of the Second Hospital of Lanzhou University (No:2020A-279). All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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