

# World Journal of *Clinical Cases*

*World J Clin Cases* 2021 May 26; 9(15): 3487-3795



## Contents

Thrice Monthly Volume 9 Number 15 May 26, 2021

## OPINION REVIEW

- 3487 COVID-19 combined with liver injury: Current challenges and management  
*Deng ML, Chen YJ, Yang ML, Liu YW, Chen H, Tang XQ, Yang XF*

## MINIREVIEWS

- 3498 Cholesterol gallstones: Focusing on the role of interstitial Cajal-like cells  
*Fu BB, Zhao JN, Wu SD, Fan Y*
- 3506 Association of hidradenitis suppurativa with Crohn's disease  
*Zhang M, Chen QD, Xu HX, Xu YM, Chen HJ, Yang BL*
- 3517 Surgical treatment of hepatocellular carcinoma in the era of COVID-19 pandemic: A comprehensive review of current recommendations  
*Fancellu A, Sanna V, Scognamiglio F, Feo CF, Vidili G, Nigri G, Porcu A*

## ORIGINAL ARTICLE

## Retrospective Cohort Study

- 3531 Critical prognostic value of the log odds of negative lymph nodes/tumor size in rectal cancer patients  
*Xie JB, Pang YS, Li X, Wu XT*
- 3546 Effectiveness of adjunctive corticosteroid therapy in patients with severe COVID-19: A retrospective cohort study  
*Xiong B, He LM, Qin YY, Du H, Zhan Z, Zhou YH, Chen YK, Zhang A*

## Retrospective Study

- 3559 Multifactor study of efficacy and recurrence in laparoscopic surgery for inguinal hernia  
*Chen WL, Deng QQ, Xu W, Luo M*
- 3567 Ultrasound-guided, direct suprainguinal injection for fascia iliaca block for total hip arthroplasty: A retrospective study  
*Wang YL, Liu YQ, Ni H, Zhang XL, Ding L, Tong F, Chen HY, Zhang XH, Kong MJ*
- 3576 Changes in endoscopic patterns before and during COVID-19 outbreak: Experience at a single tertiary center in Korean  
*Kim KH, Kim SB, Kim TN*

## Observational Study

- 3586 Cleansing efficacy and safety of bowel preparation protocol using sodium picosulfate/magnesium citrate considering subjective experiences: An observational study  
*Liu FX, Wang L, Yan WJ, Zou LC, Cao YA, Lin XC*

- 3597** Clinically significant endoscopic findings in patients of dyspepsia with no warning symptoms: A cross-sectional study

*Mao LQ, Wang SS, Zhou YL, Chen L, Yu LM, Li M, Lv B*

### META-ANALYSIS

- 3607** Effect of antifoaming agent on benign colorectal tumors in colonoscopy: A meta-analysis

*Zhang H, Gong J, Ma LS, Jiang T, Zhang H*

### CASE REPORT

- 3623** Subchondral bone as a novel target for regenerative therapy of osteochondritis dissecans: A case report

*Zhang SY, Xu HH, Xiao MM, Zhang JJ, Mao Q, He BJ, Tong PJ*

- 3631** Progressive familial intrahepatic cholestasis — farnesoid X receptor deficiency due to *NR1H4* mutation: A case report

*Czubkowski P, Thompson RJ, Jankowska I, Knisely AS, Finegold M, Parsons P, Cielecka-Kuszyk J, Strautnieks S, Pawłowska J, Bull LN*

- 3637** Postoperative pain due to an occult spinal infection: A case report

*Kerckhove MFV, Fiere V, Vieira TD, Bahroun S, Szadkowski M, d'Astorg H*

- 3644** Combined cesarean delivery and repair of acute aortic dissection at 34 weeks of pregnancy during COVID-19 outbreak: A case report

*Liu LW, Luo L, Li L, Li Y, Jin M, Zhu JM*

- 3649** Brucellosis of unknown origin with haemophagocytic syndrome: A case report

*Tian LH, Dong ZG, Chen XY, Huang LJ, Xiao PP*

- 3655** Recalcitrant paradoxical pustular psoriasis induced by infliximab: Two case reports

*Xia P, Li YH, Liu Z, Zhang X, Jiang Q, Zhou XY, Su W*

- 3662** Needle tract seeding of papillary thyroid carcinoma after fine-needle capillary biopsy: A case report

*Shi LH, Zhou L, Lei YJ, Xia L, Xie L*

- 3668** Metachronous pulmonary and pancreatic metastases arising from sigmoid colon cancer: A case report

*Yang J, Tang YC, Yin N, Liu W, Cao ZF, Li X, Zou X, Zhang ZX, Zhou J*

- 3675** Infiltrating ductal breast carcinoma with monoclonal gammopathy of undetermined significance: A case report

*Ma Y, Cui S, Yin YJ*

- 3680** Roxadustat as treatment for a blood transfusion-dependent maintenance hemodialysis patient: A case report and review of literature

*Fei M, Wen XQ, Yu ZL, Kang T, Wu WH, Ou ST*

- 3689** Small bowel ulcer bleeding due to suspected clopidogrel use in a patient with clopidogrel resistance: A case report

*Lee SH, Ryu DR, Lee SJ, Park SC, Cho BR, Lee SK, Choi SJ, Cho HS*

- 3696** Recurrent abdominal pain due to small bowel volvulus after transabdominal preperitoneal hernioplasty: A case report and review of literature  
*Man Y, Li BS, Zhang X, Huang H, Wang YL*
- 3704** Malignant giant cell tumor in the left upper arm soft tissue of an adolescent: A case report  
*Huang WP, Zhu LN, Li R, Li LM, Gao JB*
- 3711** Anesthetic management of bilateral pheochromocytoma resection in Von Hippel-Lindau syndrome: A case report  
*Wang L, Feng Y, Jiang LY*
- 3716** Sarcomatoid carcinoma of the pancreas — a rare tumor with an uncommon presentation and course: A case report and review of literature  
*Toledo PF, Berger Z, Carreño L, Cardenas G, Castillo J, Orellana O*
- 3726** Fulminant amebic colitis in a patient with concomitant cytomegalovirus infection after systemic steroid therapy: A case report  
*Shijubou N, Sumi T, Kamada K, Sawai T, Yamada Y, Ikeda T, Nakata H, Mori Y, Chiba H*
- 3733** Maisonneuve injury with no fibula fracture: A case report  
*Liu GP, Li JG, Gong X, Li JM*
- 3741** Alopecia treatment using minimally manipulated human umbilical cord-derived mesenchymal stem cells: Three case reports and review of literature  
*Ahn H, Lee SY, Jung WJ, Lee KH*
- 3752** Pheochromocytoma in a 49-year-old woman presenting with acute myocardial infarction: A case report  
*Wu HY, Cao YW, Gao TJ, Fu JL, Liang L*
- 3758** Lymphangiomas associated with protein losing enteropathy: A case report  
*Ding XL, Yin XY, Yu YN, Chen YQ, Fu WW, Liu H*
- 3765** *De novo* multiple primary carcinomas in a patient after liver transplantation: A case report  
*Rao W, Liu FG, Jiang YP, Xie M*
- 3773** Contralateral hemopneumothorax after penetrating thoracic trauma: A case report  
*İşcan M*
- 3779** Bilateral posterior scleritis presenting as acute primary angle closure: A case report  
*Wen C, Duan H*
- 3787** Bilateral cerebral infarction in diabetic ketoacidosis and bilateral internal carotid artery occlusion: A case report and review of literature  
*Chen YC, Tsai SJ*



**ABOUT COVER**

Editorial Board Member of *World Journal of Clinical Cases*, Wei Wang, MD, PhD, Associate Professor, Key Laboratory on Technology for Parasitic Disease Prevention and Control, Jiangsu Institute of Parasitic Diseases, Wuxi 214064, Jiangsu Province, China. wangwei@jipd.com

**AIMS AND SCOPE**

The primary aim of *World Journal of Clinical Cases* (WJCC, *World J Clin Cases*) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

**INDEXING/ABSTRACTING**

The WJCC is now indexed in Science Citation Index Expanded (also known as SciSearch®), Journal Citation Reports/Science Edition, Scopus, PubMed, and PubMed Central. The 2020 Edition of Journal Citation Reports® cites the 2019 impact factor (IF) for WJCC as 1.013; IF without journal self cites: 0.991; Ranking: 120 among 165 journals in medicine, general and internal; and Quartile category: Q3. The WJCC's CiteScore for 2019 is 0.3 and Scopus CiteScore rank 2019: General Medicine is 394/529.

**RESPONSIBLE EDITORS FOR THIS ISSUE**

Production Editor: Ji-Hong Lin; Production Department Director: Xiang Li; Editorial Office Director: Jin-Lai Wang.

**NAME OF JOURNAL**

*World Journal of Clinical Cases*

**ISSN**

ISSN 2307-8960 (online)

**LAUNCH DATE**

April 16, 2013

**FREQUENCY**

Thrice Monthly

**EDITORS-IN-CHIEF**

Dennis A Bloomfield, Sandro Vento, Bao-Gan Peng

**EDITORIAL BOARD MEMBERS**

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

**PUBLICATION DATE**

May 26, 2021

**COPYRIGHT**

© 2021 Baishideng Publishing Group Inc

**INSTRUCTIONS TO AUTHORS**

<https://www.wjgnet.com/bpg/gerinfo/204>

**GUIDELINES FOR ETHICS DOCUMENTS**

<https://www.wjgnet.com/bpg/GerInfo/287>

**GUIDELINES FOR NON-NATIVE SPEAKERS OF ENGLISH**

<https://www.wjgnet.com/bpg/gerinfo/240>

**PUBLICATION ETHICS**

<https://www.wjgnet.com/bpg/GerInfo/288>

**PUBLICATION MISCONDUCT**

<https://www.wjgnet.com/bpg/gerinfo/208>

**ARTICLE PROCESSING CHARGE**

<https://www.wjgnet.com/bpg/gerinfo/242>

**STEPS FOR SUBMITTING MANUSCRIPTS**

<https://www.wjgnet.com/bpg/GerInfo/239>

**ONLINE SUBMISSION**

<https://www.f6publishing.com>

# Lymphangiomatosis associated with protein losing enteropathy: A case report

Xue-Li Ding, Xiao-Yan Yin, Ya-Nan Yu, Yun-Qing Chen, Wei-Wei Fu, Hua Liu

**ORCID number:** Xue-Li Ding 0000-0003-4021-2246; Xiao-Yan Yin 0000-0002-8643-0786; Ya-Nan Yu 0000-0002-7530-1647; Yun-Qing Chen 0000-0001-9964-2537; Wei-Wei Fu 0000-0001-7258-5587; Hua Liu 0000-0002-0621-5358.

**Author contributions:** Ding XL reviewed the literature and contributed to manuscript drafting; Yin XY was the gastroenterologist; Yu YN revised the manuscript to improve its academic merits; Chen YQ and Fu WW contributed histopathology and immunohistochemistry expertise; Liu H revised the article and conceived the study; all authors issued final approval for the version to be submitted.

**Supported by** The National Natural Science Foundation of China, No. 81502025; China Postdoctoral Science Foundation, No. 2018M632631.

**Informed consent statement:** Written informed consent was obtained from the patient and his son (delegator) for publication of this report and any accompanying images.

**Conflict-of-interest statement:** The authors declare that they have no conflicts of interest to report.

**CARE Checklist (2016) statement:**

**Xue-Li Ding, Xiao-Yan Yin, Ya-Nan Yu, Hua Liu,** Department of Gastroenterology, The Affiliated Hospital of Qingdao University, Qingdao 266003, Shandong Province, China

**Yun-Qing Chen, Wei-Wei Fu,** Department of Pathology, The Affiliated Hospital of Qingdao University, Qingdao 266003, Shandong Province, China

**Corresponding author:** Hua Liu, MD, Associate Chief Physician, Department of Gastroenterology, The Affiliated Hospital of Qingdao University, No. 16 Jiangsu Road, Qingdao 266003, Shandong Province, China. [kaixing1407@sina.com](mailto:kaixing1407@sina.com)

## Abstract

### BACKGROUND

Lymphangiomatosis is a multisystem disorder that is rarely localized to the gastrointestinal tract. Lymphangiomatosis usually has no specific clinical presentation and is easily misdiagnosed. A case report and review of the literature on lymphangiomatosis associated with protein-losing enteropathy will help to improve the overall understanding of this disease.

### CASE SUMMARY

We report a case of lymphangiomatosis of the bowel and other solid organs. A 78-year-old man presented with recurrent bowel bleeding and protein-losing enteropathy, as well as cystic lesions in the spleen, liver, and kidney. Imaging examinations revealed many cystic lesions on the spleen, liver, kidney, and thickened wall of the ascending colon, as well as pleural effusion and ascites. Colonoscopy revealed a strawberry mucosa, variable spontaneous bleeding, and surface erosion located in the terminal ileum. Several cystic masses with a translucent and smooth surface as well as diffuse white spots were located in the colon. A laterally spreading tumor (LST) was located in the ascending colon. Pathology indicated highly differentiated adenocarcinoma (LST) and lymphangioma-toid dilation, and D2-40 was positive. The final diagnosis was lymphangioma-tosis. The patient underwent surgery for LST and then was administered thalidomide 75-150 mg/d. His condition, however, did not improve. He eventually died 6 mo after the initial diagnosis.

### CONCLUSION

Lymphangiomatosis usually occurs diffusely and can involve many organs, such as the spleen, kidney, liver, lung, mesentery, and bowel. Recurrent bowel bleeding or protein-losing enteropathy is an important indicator that should alert clinicians about the possibility of this disease when it afflicts the bowel. Doctors

The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

**Open-Access:** This article is an open-access article that was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in accordance with the Creative Commons Attribution NonCommercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited and the use is non-commercial. See: <http://creativecommons.org/licenses/by-nc/4.0/>

**Manuscript source:** Unsolicited manuscript

**Specialty type:** Gastroenterology and hepatology

**Country/Territory of origin:** China

**Peer-review report's scientific quality classification**

Grade A (Excellent): 0  
Grade B (Very good): B  
Grade C (Good): 0  
Grade D (Fair): 0  
Grade E (Poor): 0

**Received:** January 9, 2021

**Peer-review started:** January 10, 2021

**First decision:** February 12, 2021

**Revised:** February 13, 2021

**Accepted:** March 8, 2021

**Article in press:** March 8, 2021

**Published online:** May 26, 2021

**P-Reviewer:** Ozen H

**S-Editor:** Zhang H

**L-Editor:** Wang TQ

**P-Editor:** Yuan YY



should improve the medical understanding of lymphangiomatosis.

**Key Words:** Lymphangioma; Gastrointestinal hemorrhage; Protein-losing enteropathies; Colonic neoplasms; Small intestine; Case report

©The Author(s) 2021. Published by Baishideng Publishing Group Inc. All rights reserved.

**Core Tip:** Lymphangiomatosis is a multisystem disorder that is rarely localized to the gastrointestinal tract. We report a case of lymphangiomatosis of the bowel and other solid organs (spleen, liver, and kidney). The condition was misdiagnosed because it did not present specific clinical indicators, and the treating doctors were not aware of this condition. After a series of imaging and pathologic examinations, the patient was correctly diagnosed and treated with thalidomide. Follow-up indicated that he died 6 mo after the initial diagnosis. This case emphasizes the importance of improving the understanding of lymphangiomatosis.

**Citation:** Ding XL, Yin XY, Yu YN, Chen YQ, Fu WW, Liu H. Lymphangiomatosis associated with protein losing enteropathy: A case report. *World J Clin Cases* 2021; 9(15): 3758-3764

**URL:** <https://www.wjgnet.com/2307-8960/full/v9/i15/3758.htm>

**DOI:** <https://dx.doi.org/10.12998/wjcc.v9.i15.3758>

## INTRODUCTION

Lymphangiomatosis is an uncommon malformation of the lymphatic system. Lymphatic diseases can vary from small lymphangioma to generalized lymphangiomatosis, which is a rare condition and can have several clinical manifestations[1,2]. Lymphangiomatosis mainly occurs on the neck and head in children but can occur anywhere in the body, and abdominal lymphangiomatosis has also been reported[1,2]. It rarely affects the bowel[3]. Gastrointestinal bleeding, anemia, and abdominal pain are the most frequent symptoms[4], and protein-losing enteropathy (PLE) is a rare and severe complication[2,5].

In the present study, we report a rare case of lymphangiomatosis that manifested as PLE and recurrent bowel bleeding, as well as multiple organ injury to the spleen, liver, and kidney.

## CASE PRESENTATION

### Chief complaints

A 78-year-old man presented to our clinic with melena and weakness for 15 d.

### History of present illness

The patient's symptoms started 15 d before presentation but had worsened over the previous week. He presented to the gastrointestinal department with melena, weakness, and vomiting. He then presented with anemia (hemoglobin level: 38 g/L) and a positive fecal occult blood test.

### History of past illness

The patient had undergone surgery and chemotherapy for a history of lymphatic sarcoma on the left side of the neck 20 years prior. He also had a more than 10-year history of gallstones and a 2-year history of diabetes. He had no other history of disease or allergic reactions to medication.

### Personal and family history

The patient's personal and family history was typical.

### Physical examination

Following physical examination, he presented an anemic appearance and pretibial edema. No other obvious abnormalities were observed during physical examination.

### Laboratory examinations

The laboratory values following admission indicated microcytic hypochromic anemia, with a hemoglobin value of 63 g/L, lymphocyte count of  $0.33 \times 10^9/L$ , lymphocyte percentage of 9.9%, albumin value of 20.68 g/L, and globulin value of 13.96 g/L. The laboratory examination for fecal occult blood was positive. The urinary protein was negative. No significant abnormalities were recorded in other tests.

### Imaging examinations

Abdominal computed tomography (CT) displayed multiple small cystic lesions without enhancement distributed in the spleen, kidney, liver, ascites, and pleural effusion (Figure 1A-C). CT also revealed thickening of the ascending colon wall (Figure 1D).

Colonoscopy revealed a strawberry mucosa, variable spontaneous bleeding, and surface erosion located in the terminal ileum and ileocecal valve (Figure 2A). Multiple cystic masses with a translucent and smooth surface, diffuse white spots located in the colon, and a laterally spreading tumor (LST) located in the ascending colon (Figure 2B-D) were also observed.

The biopsy of the LST indicated villous tubular adenoma with low-grade intraepithelial neoplasia as well as highly differentiated adenocarcinoma after surgery. Histological findings also showed a large amount of vascular hyperplasia and dilatation located in the muscularis mucosae and submucosa (Figure 3A and B), which were immunohistochemically positive for D2-40, a specific lymphatic endothelial marker (Figure 3C and D).

---

## FINAL DIAGNOSIS

According to the history and histopathologic characteristics, the patient was diagnosed with lymphangiomatosis (intestine, colon, spleen, kidney, and liver) and colon cancer (LST).

---

## TREATMENT

The patient underwent radical right hemicolectomy after the first colonoscopy because of the large LST. However, he continued to present melena, hematochezia, and weakness after surgery. According to the history and imaging examinations, he was diagnosed with lymphangiomatosis. The patient was treated with a medium-chain triglyceride diet, thalidomide 75 mg/d, recurrent transfusion, albumin, and diuretics. His condition, however, did not improve. The dose of thalidomide was slowly increased to 150 mg/d.

---

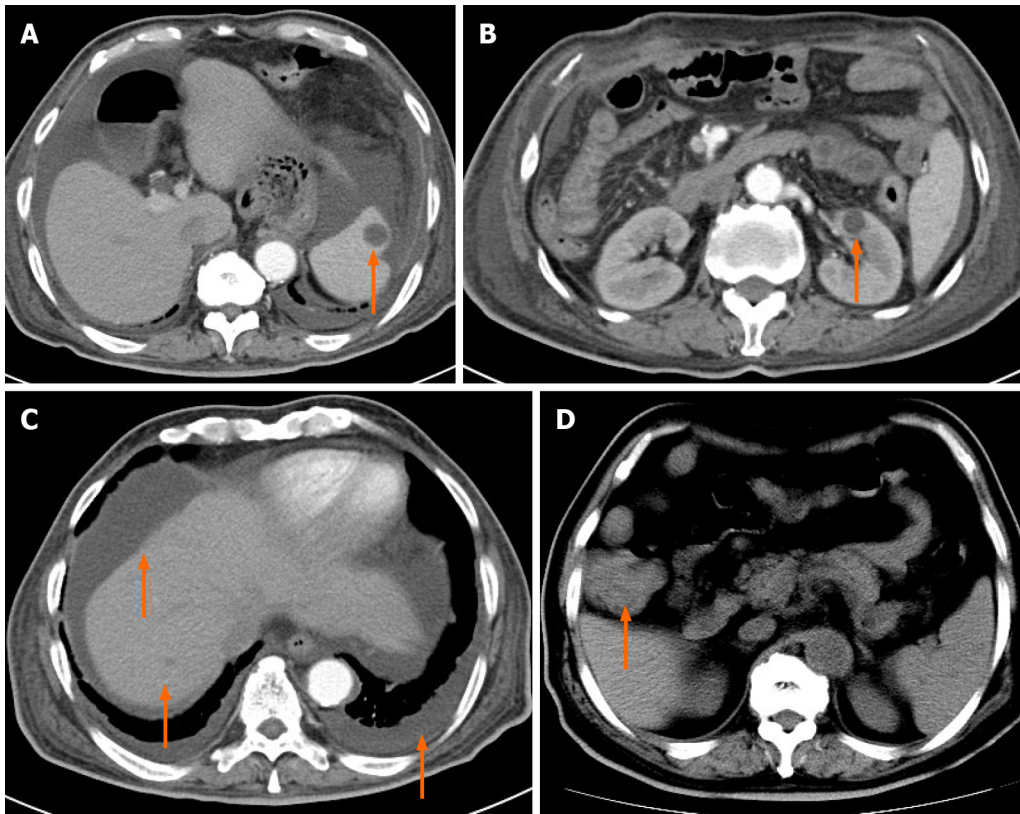
## OUTCOME AND FOLLOW-UP

Despite the patient taking thalidomide with good tolerance, he died because of recurrent bowel bleeding and consistent PLE. His hemoglobin values were 43-65 g/L despite recurrent transfusion. Similarly, his albumin values were 16.37-29.5 g/L despite recurrent albumin transfusion due to protein loss. The patient died 6 mo after the initial diagnosis.

---

## DISCUSSION

Lymphangiomatosis is a general term for excessive growth of aberrant lymphatic vessels. The acknowledged main cause is a congenital malformation of the lymphatic system, resulting in abnormal dilatation and proliferation of the lymphatic channel and leading to the formation of lymphangioma[6]. Trauma, partial lymphatic obstruction, and inflammation can also lead to secondary lymphangioma[7]. In our



**Figure 1 Abdominal computed tomography.** A: A cystic lesion in the spleen; B: A cystic lesion in the left kidney; C: A cystic lesion in the liver, ascites, and pleural effusion; D: Thickening of the ascending colon wall.

case, the patient was an elderly man with a surgical history of lymphatic sarcoma on the left side of the neck, and we speculated that the cause of his condition was secondary.

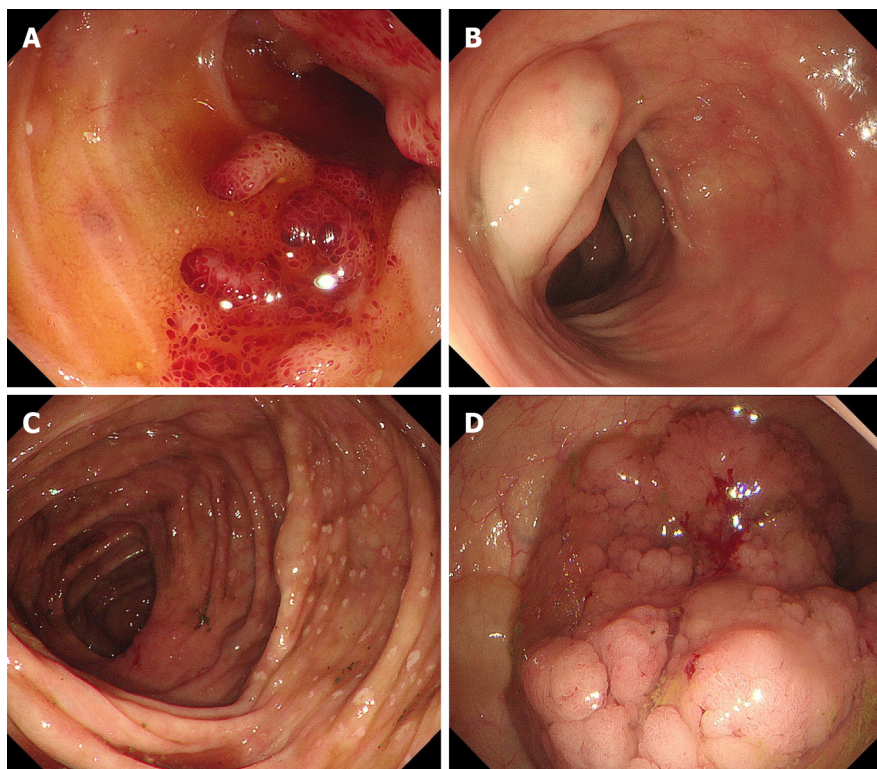
The clinical symptoms of lymphangiomatosis are complicated and atypical. They include gastrointestinal bleeding, anemia, abdominal pain, perforation, and PLE, and whether it afflicts the bowels depends on the position and size of the spread[2,4,7]. Our patient presented gastrointestinal bleeding and PLE, which is rare.

Blood examination results showed a hemoglobin decrease and positive occult blood test. Additionally, PLE caused by lymphangiomas show a decrease in the albumin, and globulin values, lymphocyte count, and lymphocyte percentage also decrease. Our patient presented these changes. CT manifestations of lymphangiomatosis include multiple hypodense cysts occupying the spleen, liver, bones, and other organs[8]. CT scans also occasionally reveal ascites and pleural effusion. Our patient had cystic lesions in the spleen, liver, and kidney, as well as ascites and pleural effusion. Endoscopy is the most important examination for gastrointestinal lymphangiomatosis[7]. Polypoid lesions, white-yellow surfaces with a strawberry mucosa, variable spontaneous bleeding and surface erosion, and cysts are the most common indicators in endoscopy[2]. Diffuse white spots are also observed in the intestine and colon. In our case, the patient had polypoid lesions, cystic lesions, and white spots in endoscopy.

The histological features of lymphangiomatosis are nonspecific, and a definitive diagnosis requires the hyperproliferation of normal lymphatic vessels with normal endothelium, predominantly in the context of submucosa, with disruption of the muscular layer and sometimes of the serosa. Regarding immunohistochemistry, D2-40 is a specific marker of lymphangioma. CD31-, CD34-, and VIII-related antigens and VEGFR3 are also helpful in the diagnosis[9].

Treatment is dictated by the nature of the symptoms, anatomic location, and the associated potential complications of treatments, including systemic therapy, endoscopic therapy, surgery, and radiation therapy[10]. Rapamycin is very effective in some cases of lymphangiomatosis[11]. In others, thalidomide may also be effective[12]. However, we only tried thalidomide because rapamycin was not available.





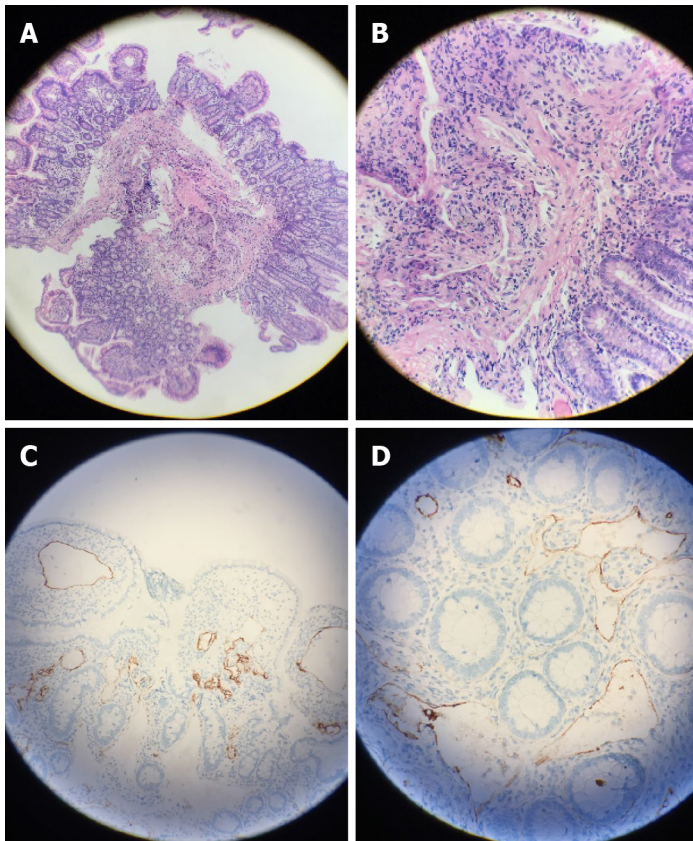
**Figure 2 Colonoscopy.** A: Colonoscopy revealed a strawberry mucosa and variable spontaneous bleeding located in the terminal ileum; B and C: Multiple cystic masses with a translucent and smooth surface, and diffuse white spots located in the colon; D: A laterally spreading tumor located in the ascending colon.

Our case required additional laboratory and imaging examinations, such as an ascites test,  $\alpha$ -antitrypsin clearance rate which is difficult to popularize in clinic because of its complicated detection method, capsule endoscopy test, and lymphangiography, to diagnose the disease. Capsule endoscopy can show lesions in or changes to the intestine. Lymphangiography is an imaging technique that shows the lymphatic system. Performing lymphangiography could perhaps have indicated the disease of our patient and revealed that the disease was primary or secondary. Unfortunately, the patient rejected undergoing complete capsule endoscopy. Additionally, our hospital did not have the capability to perform lymphangiography, and he was too weak to travel to Beijing to undergo the procedure.

Only a few lymphangiomatosis cases have been reported to be associated with PLE to date[2,5]. PLE is a severe complication of lymphangiomatosis when it affects the gastrointestinal tract and is characterized by an excessive loss of proteins in the gastrointestinal tract due to impaired integrity of the mucosa. It is characterized by edema and hypoproteinemia. In severe cases, pleural effusion and peritoneal effusion may occur. Gastrointestinal lymphangiectasia and lymphangioma are the causes of protein-losing enteropathy. Endoscopy clearly revealed pathological changes in the intestine and colon, likely explaining why our patient had PLE[2,5]. PLE is an important indicator that should alert clinicians about the possibility of this disease when it affects the bowel.

## CONCLUSION

Lymphangiomatosis usually occurs diffusely and can involve many organs, such as the spleen, kidney, liver, lung, mesentery, and bowel. Lymphangiomatosis is a rare disease that usually has no specific clinical presentation and can be easily misdiagnosed. A history of recurrent bowel bleeding or protein-losing enteropathy is an important indicator that should alert clinicians to the possibility of this disease. Doctors should improve their medical knowledge of lymphangiomatosis.



**Figure 3 Pathology.** A and B: A large amount of vascular hyperplasia and dilatation was observed in the mucosal muscular layer and submucosa [A: Hematoxylin-eosin stain (HE)  $\times 100$ ; B: HE  $\times 400$ ]; C and D: Immunohistochemically stained tumor sections showed that lymph vessels in the intestine were dilated, and D2-40 was positive (C:  $\times 40$ ; D:  $\times 100$ ).

## REFERENCES

- 1 **Valakada J**, Madhusudhan KS, Ranjan G, Garg PK, Sharma R, Gupta AK. Abdominal Lymphangiomas With Intestinal Lymphangiectasia Diagnosed by Magnetic Resonance Lymphangiography: A Case Report. *Curr Probl Diagn Radiol* 2018; **47**: 200-202 [PMID: [28554788](#) DOI: [10.1067/j.cpradiol.2017.04.006](#)]
- 2 **Lin RY**, Zou H, Chen TZ, Wu W, Wang JH, Chen XL, Han QX. Abdominal lymphangiomas in a 38-year-old female: case report and literature review. *World J Gastroenterol* 2014; **20**: 8320-8324 [PMID: [25009412](#) DOI: [10.3748/wjg.v20.i25.8320](#)]
- 3 **Ilhan M**, Oner G, Alibeyoglu A, Yeğen G, Gök AF, Akyüz F, Bicen F. Primary intestinal lymphangiomas of the ileum in an adult-the role of surgical approach. *J Surg Case Rep* 2016; **2016**: rjw133 [PMID: [27534888](#) DOI: [10.1093/jscr/rjw133](#)]
- 4 **Giuliani A**, Romano L, Coletti G, Walid A Fatayer M, Calvisi G, Maffione F, Muolo C, Vicentini V, Schietroma M, Carlei F. Lymphangiomas of the ileum with perforation: A case report and review of the literature. *Ann Med Surg (Lond)* 2019; **41**: 6-10 [PMID: [30992989](#) DOI: [10.1016/j.amsu.2019.03.010](#)]
- 5 **Glöckler M**, Severin T, Arnold R, Greiner P, Schwab KO, Uhl M, Schlensak C, Rössler J, Dittrich S. First description of three patients with multifocal lymphangiomas and protein-losing enteropathy following palliation of complex congenital heart disease with total cavo-pulmonary connection. *Pediatr Cardiol* 2008; **29**: 771-774 [PMID: [18188635](#) DOI: [10.1007/s00246-007-9194-8](#)]
- 6 **Bellini C**, Hennekam RC. Clinical disorders of primary malfunctioning of the lymphatic system. *Adv Anat Embryol Cell Biol* 2014; **214**: 187-204 [PMID: [24276895](#) DOI: [10.1007/978-3-7091-1646-3\\_14](#)]
- 7 **Ding XL**, Yu YN, Jing X, Tian ZB, Liu H, Yin XY. Diagnostic values of digestive endoscopy in small intestinal lymphangioma. *Zhonghua Weichang Neijing Dianzi Zazhi* 2017; **4**: 119-122 [DOI: [10.3877/cma.j.issn.2095-7157.2017.03.005](#)]
- 8 **Kwag E**, Shim SS, Kim Y, Chang JH, Kim KC. CT features of generalized lymphangiomas in adult patients. *Clin Imaging* 2013; **37**: 723-727 [PMID: [23391872](#) DOI: [10.1016/j.clinimag.2012.12.003](#)]
- 9 **Mehmedovic Z**, Mehmedovic M, Custovic MK, Sadikovic A, Mekic N. A rare case of giant mesenteric cystic lymphangioma of the small bowel in an adult: A case presentation and literature review. *Acta Gastroenterol Belg* 2016; **79**: 491-493 [PMID: [28209109](#)]
- 10 **Xiao NJ**, Ning SB, Li T, Li BR, Sun T. Small intestinal hemolymphangioma treated with enteroscopic injection sclerotherapy: A case report and review of literature. *World J Gastroenterol*

2020; **26**: 1540-1545 [PMID: [32308353](#) DOI: [10.3748/wjg.v26.i13.1540](#)]

- 11 **Laforgia N**, Schettini F, De Mattia D, Martinelli D, Ladisa G, Favia V. Lymphatic Malformation in Newborns as the First Sign of Diffuse Lymphangiomas: Successful Treatment with Sirolimus. *Neonatology* 2016; **109**: 52-55 [PMID: [26506225](#) DOI: [10.1159/000440939](#)]
- 12 **Pauzner R**, Mayan H, Waizman A, Rozenman J, Farfel Z. Successful thalidomide treatment of persistent chylous pleural effusion in disseminated lymphangiomas [corrected]. *Ann Intern Med* 2007; **146**: 75-76 [PMID: [17200231](#) DOI: [10.7326/0003-4819-146-1-200701020-00022](#)]





Published by **Baishideng Publishing Group Inc**  
7041 Koll Center Parkway, Suite 160, Pleasanton, CA 94566, USA

**Telephone:** +1-925-3991568

**E-mail:** [bpgoffice@wjgnet.com](mailto:bpgoffice@wjgnet.com)

**Help Desk:** <https://www.f6publishing.com/helpdesk>

<https://www.wjgnet.com>

