

World Journal of *Clinical Cases*

World J Clin Cases 2020 December 26; 8(24): 6213-6545



MINIREVIEWS

- 6213 Role of gut microbiome in regulating the effectiveness of metformin in reducing colorectal cancer in type 2 diabetes
Huang QY, Yao F, Zhou CR, Huang XY, Wang Q, Long H, Wu QM

ORIGINAL ARTICLE**Retrospective Cohort Study**

- 6229 Impact factors of lymph node retrieval on survival in locally advanced rectal cancer with neoadjuvant therapy
Mei SW, Liu Z, Wang Z, Pei W, Wei FZ, Chen JN, Wang ZJ, Shen HY, Li J, Zhao FQ, Wang XS, Liu Q

Retrospective Study

- 6243 Three-year follow-up of Coats disease treated with conbercept and 532-nm laser photocoagulation
Jiang L, Qin B, Luo XL, Cao H, Deng TM, Yang MM, Meng T, Yang HQ
- 6252 Virus load and virus shedding of SARS-CoV-2 and their impact on patient outcomes
Chen PF, Yu XX, Liu YP, Ren D, Shen M, Huang BS, Gao JL, Huang ZY, Wu M, Wang WY, Chen L, Shi X, Wang ZQ, Liu YX, Liu L, Liu Y
- 6264 Risk factors for *de novo* hepatitis B during solid cancer treatment
Sugimoto R, Furukawa M, Senju T, Aratake Y, Shimokawa M, Tanaka Y, Inada H, Noguchi T, Lee L, Miki M, Maruyama Y, Hashimoto R, Hisano T

- 6274 Cause analysis and reoperation effect of failure and recurrence after epiblepharon correction in children
Wang Y, Zhang Y, Tian N

Clinical Trials Study

- 6282 Effects of different acupuncture methods combined with routine rehabilitation on gait of stroke patients
Lou YT, Yang JJ, Ma YF, Zhen XC

Observational Study

- 6296 Application of endoscopic submucosal dissection in duodenal space-occupying lesions
Li XY, Ji KY, Qu YH, Zheng JJ, Guo YJ, Zhang CP, Zhang KP
- 6306 Early renal injury indicators can help evaluate renal injury in patients with chronic hepatitis B with long-term nucleos(t)ide therapy
Ji TT, Tan N, Lu HY, Xu XY, Yu YY

Prospective Study

- 6315** Neoadjuvant chemoradiotherapy plus surgery in the treatment of potentially resectable thoracic esophageal squamous cell carcinoma
Yan MH, Hou XB, Cai BN, Qu BL, Dai XK, Liu F

CASE REPORT

- 6322** Uterine rupture in patients with a history of multiple curettages: Two case reports
Deng MF, Zhang XD, Zhang QF, Liu J
- 6330** Pleural effusion and ascites in extrarenal lymphangiectasia caused by post-biopsy hematoma: A case report
Lin QZ, Wang HE, Wei D, Bao YF, Li H, Wang T
- 6337** Eighty-year-old man with rare chronic neutrophilic leukemia caused by CSF3R T618I mutation: A case report and review of literature
Li YP, Chen N, Ye XM, Xia YS
- 6346** Sigmoid colon duplication with ectopic immature renal tissue in an adult: A case report
Namgung H
- 6353** Paraplegia from spinal intramedullary tuberculosis: A case report
Qu LM, Wu D, Guo L, Yu JL
- 6358** Confocal laser endomicroscopy distinguishing benign and malignant gallbladder polyps during choledochoscopic gallbladder-preserving polypectomy: A case report
Tang BF, Dang T, Wang QH, Chang ZH, Han WJ
- 6364** Sclerosing stromal tumor of the ovary with masculinization, Meig's syndrome and CA125 elevation in an adolescent girl: A case report
Chen Q, Chen YH, Tang HY, Shen YM, Tan X
- 6373** Primary pulmonary malignant melanoma diagnosed with percutaneous biopsy tissue: A case report
Xi JM, Wen H, Yan XB, Huang J
- 6380** SRY-negative 45,X/46,XY adult male with complete masculinization and infertility: A case report and review of literature
Wu YH, Sun KN, Bao H, Chen YJ
- 6389** Refractory case of ulcerative colitis with idiopathic thrombocytopenic purpura successfully treated by Janus kinase inhibitor tofacitinib: A case report
Komeda Y, Sakurai T, Sakai K, Morita Y, Hashimoto A, Nagai T, Hagiwara S, Matsumura I, Nishio K, Kudo M
- 6396** Immunotherapies application in active stage of systemic lupus erythematosus in pregnancy: A case report and review of literature
Xiong ZH, Cao XS, Guan HL, Zheng HL

- 6408** Minimally invasive maxillary sinus augmentation with simultaneous implantation on an elderly patient: A case report
Yang S, Yu W, Zhang J, Zhou Z, Meng F, Wang J, Shi R, Zhou YM, Zhao J
- 6418** Congenital nephrogenic diabetes insipidus due to the mutation in *AVPR2* (c.541C>T) in a neonate: A case report
Lin FT, Li J, Xu BL, Yang XX, Wang F
- 6425** Primary gastric melanoma in a young woman: A case report
Long GJ, Ou WT, Lin L, Zhou CJ
- 6432** Extreme venous letting and cupping resulting in life-threatening anemia and acute myocardial infarction: A case report
Jang AY, Suh SY
- 6437** Novel conservative treatment for peritoneal dialysis-related hydrothorax: Two case reports
Dai BB, Lin BD, Yang LY, Wan JX, Pan YB
- 6444** Clinical characteristics of pulmonary cryptococcosis coexisting with lung adenocarcinoma: Three case reports
Zheng GX, Tang HJ, Huang ZP, Pan HL, Wei HY, Bai J
- 6450** Fracture of the scapular neck combined with rotator cuff tear: A case report
Chen L, Liu CL, Wu P
- 6456** Synchronous colonic mucosa-associated lymphoid tissue lymphoma found after surgery for adenocarcinoma: A case report and review of literature
Li JJ, Chen BC, Dong J, Chen Y, Chen YW
- 6465** Novel mutation in the *ASXL3* gene in a Chinese boy with microcephaly and speech impairment: A case report
Li JR, Huang Z, Lu Y, Ji QY, Jiang MY, Yang F
- 6473** Recurrent thrombosis in the lower extremities after thrombectomy in a patient with polycythemia vera: A case report
Jiang BP, Cheng GB, Hu Q, Wu JW, Li XY, Liao S, Wu SY, Lu W
- 6480** Status epilepticus as an initial manifestation of hepatic encephalopathy: A case report
Cui B, Wei L, Sun LY, Qu W, Zeng ZG, Liu Y, Zhu ZJ
- 6487** Delayed diagnosis of prosopagnosia following a hemorrhagic stroke in an elderly man: A case report
Yuan Y, Huang F, Gao ZH, Cai WC, Xiao JX, Yang YE, Zhu PL
- 6499** Oral myiasis after cerebral infarction in an elderly male patient from southern China: A case report
Zhang TZ, Jiang Y, Luo XT, Ling R, Wang JW
- 6504** Rare case of drain-site hernia after laparoscopic surgery and a novel strategy of prevention: A case report
Gao X, Chen Q, Wang C, Yu YY, Yang L, Zhou ZG

- 6511** Extracorporeal shock wave therapy treatment of painful hematoma in the calf: A case report
Jung JW, Kim HS, Yang JH, Lee KH, Park SB
- 6517** Takotsubo cardiomyopathy associated with bronchoscopic operation: A case report
Wu BF, Shi JR, Zheng LR
- 6524** Idiopathic adulthood ductopenia with elevated transaminase only: A case report
Zhang XC, Wang D, Li X, Hu YL, Wang C
- 6529** Successful endovascular treatment with long-term antibiotic therapy for infectious pseudoaneurysm due to *Klebsiella pneumoniae*: A case report
Wang TH, Zhao JC, Huang B, Wang JR, Yuan D
- 6537** Primary duodenal tuberculosis misdiagnosed as tumor by imaging examination: A case report
Zhang Y, Shi XJ, Zhang XC, Zhao XJ, Li JX, Wang LH, Xie CE, Liu YY, Wang YL

ABOUT COVER

Peer-Reviewer of *World Journal of Clinical Cases*, Dr. Adonis Protopapas is a gastroenterology Resident at the first Propaedeutic Department of Internal Medicine of the Aristotle University of Thessaloniki (Greece), located at the A.H.E.P.A Hospital. He earned his Bachelor's degree in 2015 from the Democritus University of Thrace, followed by three Master's of Science degrees, with specializations in clinic pharmacology, medical research methodology, and healthcare management. His research interests are mainly focused on the area of hepatology, although he also participates in various projects related to endoscopy and inflammatory bowel disease. He is particularly fascinated by research on cirrhosis and its complications. (L-Editor: Filipodia)

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, 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.

RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: *Ji-Hong Liu*; 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

Semimonthly

EDITORS-IN-CHIEF

Dennis A Bloomfield, Sandro Vento, Bao-gan Peng

EDITORIAL BOARD MEMBERS

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

PUBLICATION DATE

December 26, 2020

COPYRIGHT

© 2020 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>



Pleural effusion and ascites in extrarenal lymphangiectasia caused by post-biopsy hematoma: A case report

Qiong-Zhen Lin, Hui-En Wang, Dong Wei, Yun-Feng Bao, Hang Li, Tao Wang

ORCID number: Qiong-Zhen Lin 0000-0002-1763-5913; Hui-En Wang 0000-0001-5500-2597; Dong Wei 0000-0001-8913-2779; Yun-Feng Bao 0000-0002-7536-156X; Hang Li 0000-0001-6937-2863; Tao Wang 0000-0002-7370-7299.

Author contributions: Lin QZ reviewed the literature and contributed to manuscript drafting; Wang HE and Wei D are a thoracic surgeon and urologist, respectively, and both gave critical advice in the management of the patient and preparation of the revision; Bao YF prepared the medical imaging; Li H and Wang T revised the manuscript for important intellectual content; Wang T was the patient's nephrologist and conceived the study; All authors issued final approval for the version to be submitted.

Informed consent statement: Informed written consent was obtained from the patient for publication of this report and any accompanying images.

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

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and

Qiong-Zhen Lin, Department of Nephrology, The Third Hospital of Hebei Medical University, Shijiazhuang 050051, Hebei Province, China

Hui-En Wang, Department of Thoracic Surgery, Hebei Provincial General Hospital, Shijiazhuang 050051, Hebei Province, China

Dong Wei, Department of Urology, Hebei Provincial General Hospital, Shijiazhuang 050051, Hebei Province, China

Yun-Feng Bao, Department of Medical Imaging, Hebei General Hospital, Shijiazhuang 050051, Hebei Province, China

Hang Li, Department of Nephrology, Peking Union Medical College Hospital, Beijing 100045, China

Tao Wang, Department of Nephrology, The First Hospital of Hebei Medical University, Shijiazhuang 050000, Hebei Province, China

Corresponding author: Tao Wang, MD, PhD, Chief Physician, Professor, Department of Nephrology, The First Hospital of Hebei Medical University, No. 89 East Donggang Road, Shijiazhuang 050000, Hebei Province, China. yanzhang@hbmh.edu

Abstract

BACKGROUND

The renal system has a specific pleural effusion associated with it in the form of "urothorax", a condition where obstructive uropathy or occlusion of the lymphatic ducts leads to extravasated fluids (urine or lymph) crossing the diaphragm *via* innate perforations or lymphatic channels. As a rare disorder that may cause pleural effusion, renal lymphangiectasia is a congenital or acquired abnormality of the lymphatic system of the kidneys. As vaguely mentioned in a report from the American Journal of Kidney Diseases, this disorder can be caused by extrinsic compression of the kidney secondary to hemorrhage.

CASE SUMMARY

A 54-year-old man with biopsy-proven acute tubulointerstitial nephropathy experienced bleeding 3 d *post hoc*, which, upon clinical detection, manifested as a massive perirenal hematoma on computed tomography (CT) scan without concurrent pleural effusion. His situation was eventually stabilized by expeditious management, including selective renal arterial embolization. Despite

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: Medicine, research and experimental

Country/Territory of origin: China

Peer-review report's scientific quality classification

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

Received: June 26, 2020

Peer-review started: June 26, 2020

First decision: September 24, 2020

Revised: October 1, 2020

Accepted: November 4, 2020

Article in press: November 4, 2020

Published online: December 26, 2020

P-Reviewer: Desai DJ

S-Editor: Gao CC

L-Editor: Filipodia

P-Editor: Liu JH



good hemodialysis adequacy and stringent volume control, a CT scan 1 mo later found further enlargement of the perirenal hematoma with heterogeneous hypodense fluid, left side pleural effusion and a small amount of ascites. These fluid collections showed a CT density of 3 Hounsfield units, and drained fluid of the pleural effusion revealed a dubiously light-colored transudate with lymphocytic predominance (> 80%). Similar results were found 3 mo later, during which time the patient was free of pulmonary infection, cardiac dysfunction and overt hypoalbuminemia. After careful consideration and exclusion of other possible causative etiologies, we believed that the pleural effusion was due to the occlusion of renal lymphatic ducts by the compression of kidney parenchyma and, in the absence of typical dilation of the related ducts, considered our case as extrarenal lymphangiectasia in a broad sense.

CONCLUSION

As such, our case highlighted a morbid passage between the kidney and thorax under an extraordinarily rare condition. Given the paucity of pertinent knowledge, it may further broaden our understanding of this rare disorder.

Key Words: Urothorax; Pleural effusion; Perirenal hematoma; Renal lymphangiectasia; Lymphatic drainage; Case report

©The Author(s) 2020. Published by Baishideng Publishing Group Inc. All rights reserved.

Core Tip: It is known that obstructive uropathy or occlusion of the lymphatic ducts may lead to extravasated fluids (urine or lymph) crossing the diaphragm *via* innate perforations or lymphatic channels. Therein, this clinical phenomenon is addressed as the “urothorax”. Among the diverse etiologies, renal lymphangiectasia is a congenital or acquired abnormality of the lymphatic system of the kidneys. Under this instance, pleural effusion of lymphoid origin may develop when the renal parenchyma is tightly compressed by a perirenal hematoma. Arguably, tight compression of the renal parenchyma may keep the draining lymphatic vessels shut but not prevent the inflow from the capsular lymph plexus. Thus, our report has for the first time described this extremely rare scenario and raises clinical awareness of the underlying passage, through which upward spread of perirenal infection could result in lung abscess.

Citation: Lin QZ, Wang HE, Wei D, Bao YF, Li H, Wang T. Pleural effusion and ascites in extrarenal lymphangiectasia caused by post-biopsy hematoma: A case report. *World J Clin Cases* 2020; 8(24): 6330-6336

URL: <https://www.wjgnet.com/2307-8960/full/v8/i24/6330.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v8.i24.6330>

INTRODUCTION

The renal system has a specific pleural effusion associated with it in the form of “urothorax”, a condition where obstructive uropathy or occlusion of the lymphatic ducts leads to extravasated fluids (urine or lymph) crossing the diaphragm *via* innate perforations or lymphatic channels^[1]. In rare cases, this pleural effusion may be caused by renal lymphangiectasia, which is a congenital or acquired disorder of the lymphatic system of the kidneys^[2]. The exact etiology of this disorder remains unknown, whereas the most common hypothesis is the occlusion of draining lymphatic ducts secondary to trauma, scarring, infection, inflammation or malignant cells of the kidneys^[3]. There is only one report from the American Journal of Kidney Diseases^[4] describing extrinsic compression of the kidney by hemorrhage leading to extrarenal lymphangiectasia, and we hereby described such an extremely rare case with resultant pleural effusion. The study was approved by our institutional review board (No. 2020-22), and written informed consent was obtained from the patient.

CASE PRESENTATION

Chief complaints

A 54-year-old man was admitted for serum creatinine (Scr) elevation for 1 mo.

History of present illness

He initially visited a local clinic due to loss of appetite and was found to have an elevated Scr without dysuresia. Fluid infusion had indiscernible effect on the abnormal Scr and no evidence of secondary kidney disease was found. He was then referred to us, pending renal biopsy.

History of past illness

The patient was a capable farm hand *ex ante* without a known medical history.

Personal and family history

He denied any family history of hypertension, diabetes or kidney disease.

Physical examination

On arrival, his blood pressure was 150/90 mmHg, and he had a slightly anemic complexion. Otherwise, physical examination yielded no remarkable findings.

Laboratory examinations

Laboratory tests revealed a hemoglobin concentration of 108 g/L (reference: 130-150 g/L), platelet count of $202 \times 10^9/L$ ($100-300 \times 10^9/L$), plasma albumin of 41.3 g/L, Scr of 679.1 $\mu\text{mol/L}$ ($44.2-132.6 \mu\text{mol/L}$), normal coagulation function including D-dimer and negative results for anti-nuclear antibody, anti-neutrophil cytoplasm antibody and immunofixation electrophoresis. Screening for hepatitis B and malignancy was negative. After the routine workup, renal biopsy confirmed the diagnosis of acute tubulointerstitial nephropathy.

Imaging examinations

Plain chest X-ray was clear. Furthermore, the kidneys appeared normal on sonography without aberrant echogenicity.

FINAL DIAGNOSIS

Acute tubulointerstitial nephropathy.

TREATMENT

Three days after the biopsy, however, the patient experienced bleeding, which, upon clinical detection, manifested as a large perirenal hematoma without concurrent pleural effusion (Figure 1A). Laboratory tests *post hoc* showed a surge in the Scr level to 1094.3 $\mu\text{mol/L}$ (Figure 1, upper panel), whereas the hemoglobin concentration, platelet count and plasma albumin were 56 g/L, $166 \times 10^9/L$ and 36.7 g/L, respectively. Continuous renal replacement therapy was employed due to transient oliguria, and the patient's situation was eventually stabilized by expeditious management, including selective renal arterial embolization.

While with good hemodialysis adequacy and under stringent volume control for Page kidney, computed tomography (CT) scan 1 mo later found further enlargement of the perirenal hematoma with heterogeneous hypodense fluid, massive left-sided pleural effusion and a small amount of ascites (Figure 1B). These fluid collections had a CT density of 3 Hounsfield units (HU), and drained fluid of the pleural effusion yielded a light-colored transudate with lymphocytic predominance (> 80%). Additionally, Scr remained elevated, and plasma albumin was generally stable, with a hemoglobin concentration of 97 g/L and platelet count of $240 \times 10^9/L$. Furthermore, the pleural effusion and ascites consistently showed the same HU and lymphatic nature 4 mo later (Figure 1C), accompanied by lowering of the Scr and rise in the albumin. Of note, the patient was free of pulmonary infection and cardiac dysfunction during the whole episode.

As schematically outlined in Figure 2, tight compression of the renal parenchyma

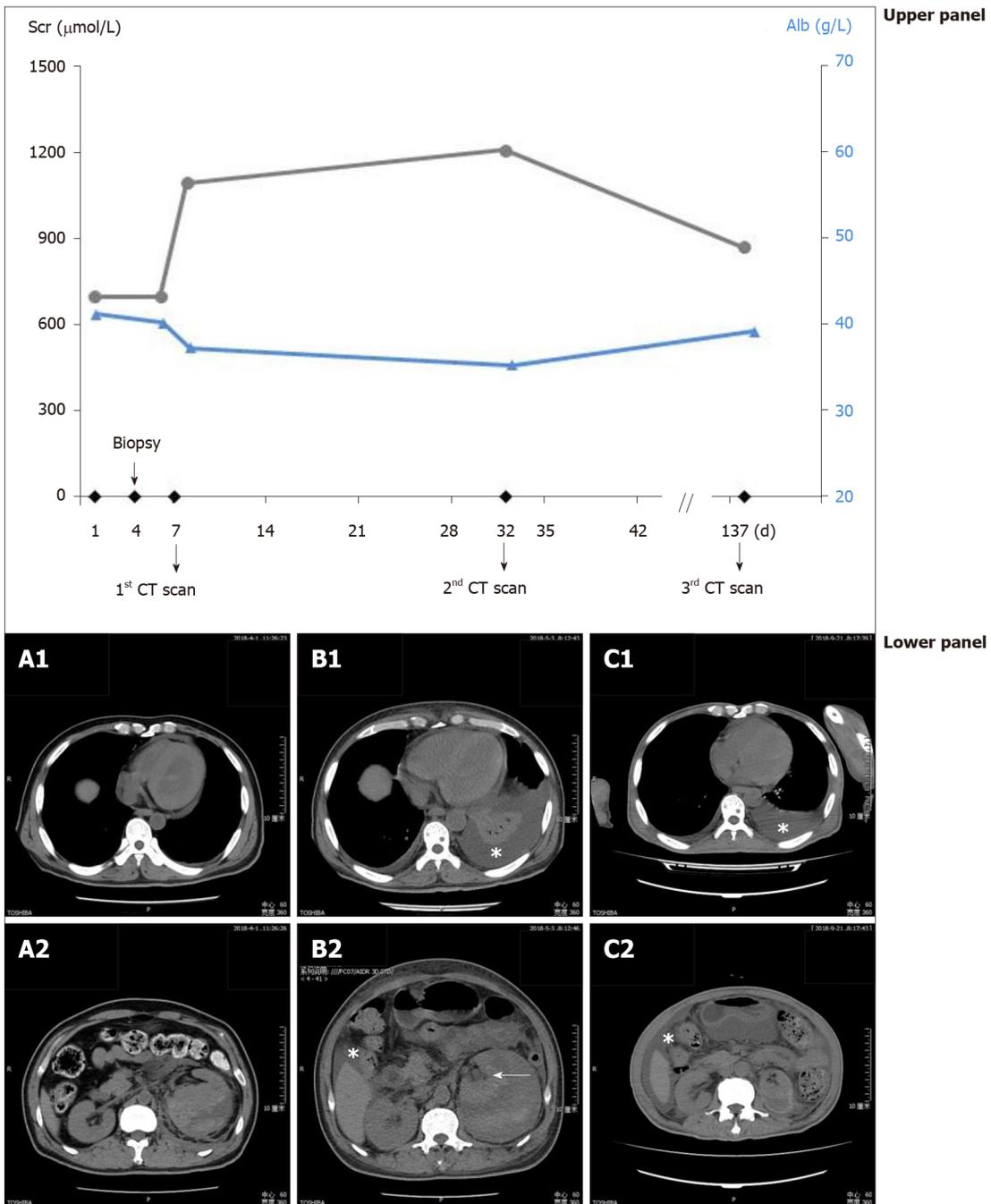


Figure 1 Evolution of the perirenal hematoma, pleural effusion and ascites in parallel with the corresponding serum creatinine and plasma albumin. Upper panel: Values of serum creatinine (circles connected by black lines) and plasma albumin (triangles connected by blue lines) at different time points; Lower panel: A: Computed tomography scan shortly after the detection of hemorrhage showing perirenal hematoma (A2), without pleural effusion (A1); B: Perirenal hematoma and small amount of ascites (B2), with pleural effusion (B1, asterisk) 1 mo after the hemorrhage, the compressed kidney is also visible (B2, arrow); C: Perirenal fluid retention and ascites (C2), with pleural effusion (C1, asterisk) 5 mo after the hemorrhage. Alb: Albumin; CT: Computed tomography; Scr: Serum creatinine.

may keep the draining lymphatic vessels shut but not prevent the inflow from the capsular lymph plexus. On the basis of clinical features, imaging findings and laboratory tests, the diagnosis of extrarenal lymphangiectasia was made, as classified previously^[4]. The patient was then kept on hemodialysis, with a special focus on dry weight and urinary volume.

OUTCOME AND FOLLOW-UP

He was eventually detached from hemodialysis 10 mo after the hemorrhage and

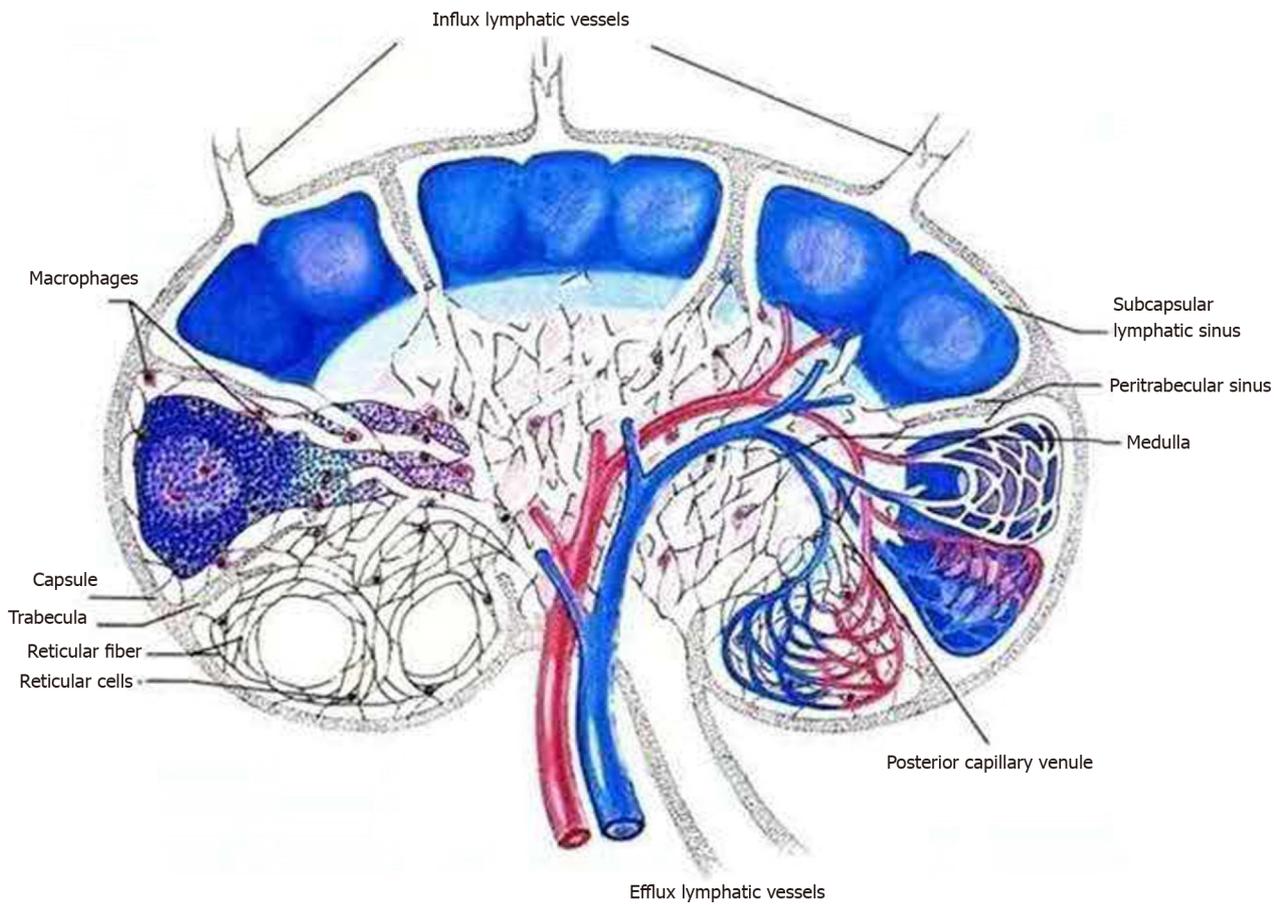


Figure 2 Schematic showing the flow of renal lymphatic fluid. Inflow from the capsular lymph plexus went through the renal parenchyma via subcapsular and peritrabecular lymphatic sinuses and pericapillary space and eventually made confluence at the efflux lymphatic vessels for out-draining.

treated for chronic kidney disease stage 5, according to the “Kidney Disease: Improving Global Outcomes” Guideline^[5]. His Scr fluctuated between 300-400 $\mu\text{mol/L}$ and had a daily urinary volume of approximately 1000 mL.

DISCUSSION

Renal lymphangiectasia is a rare benign disorder characterized by abnormal and ectatic lymphatic vessels within and around the kidneys. The abnormal dilatation of these lymphatic ducts arises from their failure to communicate with larger retroperitoneal lymph vessels. Although it is bilateral in nature in more than 90% of cases^[2], this disorder may also be unilateral or focal^[6]. The diagnosis is based mainly on imaging results, although perinephric/pleural fluid analysis and kidney biopsy are definitely helpful^[2,4]. As such, imaging findings of renal lymphangiectasia may include peripelvic cysts (intrarenal lymphangiectasia) and perinephric fluid collections (extrarenal lymphangiectasia)^[4,7]. However, locular cystic lesions within the renal sinuses may be absent in cases of perirenal compression of the kidney parenchyma or bilateral renal vein thrombosis^[8,9]. In addition to the perinephric and/or retroperitoneal fluid collection, it may also manifest ascites and, rarely, pleural effusion.

Renal lymphangiectasia may confer pleural effusion through the passage of “urothorax”. Under such circumstances, pleural drainage usually reveals chylous fluid with lymphocytic predominance ($\geq 90\%$)^[4], while occasionally, cell staining may prove colorless at gross examination^[2]. In another chance encounter supporting this finding, we recently admitted another patient on maintenance peritoneal dialysis for dyspnea on exertion. His discomfort was exacerbated after the infusion of peritoneal dialysate, and a CT scan found a large amount of right-sided pleural effusion (Supplementary Figure 1). After the addition of methylene blue to the dialysate, colored pleural drainage was observed. Indeed, urinary tract infection may spread upward and lead to lung abscess^[10]. Taken together, our case highlighted a *de novo* morbid passage

between the kidney and thorax under extraordinary conditions.

Clinically, renal lymphangiectasia is usually asymptomatic and incidentally diagnosed. When symptomatic, the most common presentations are abdominal pain (42%) and abdominal distension (21%), followed by fever, hematuria, fatigue, weight loss, hypertension and occasional deterioration in renal function^[1]. A unique entity, namely, the Page kidney, was considered in this case with the associated hypertension after subcapsular hematoma, and the management required sufficient fluid control^[12]. In this respect, our patients on maintenance hemodialysis generally manifested good Kt/V in both a cross-sectional study^[13] and 10-year follow-up^[14], and the critically ill patients receiving continuous renal replacement therapy had fine volume control^[15]. Therefore, it is highly unlikely that the observed pleural effusion was derived from fluid overload. Of essential importance, caution regarding polycythemia and the associated deep venous thrombosis has been recommended in renal lymphangiectasia^[2]. In this regard, a detailed description of the differential diagnosis^[2] and therapeutic approach^[4] is available elsewhere.

CONCLUSION

In conclusion, we reported an extremely unusual case of pleural effusion caused by extrarenal lymphangiectasia, which resulted from occlusion of the lymphatic ducts due to compression of the renal parenchyma by a hematoma. Given the paucity of pertinent knowledge, these findings may further improve our understanding of this rare disorder.

REFERENCES

- 1 **Jones GH**, Kalaher HR, Misra N, Curtis J, Parker RJ. Empyema and respiratory failure secondary to nephropleural fistula caused by chronic urinary tract infection: a case report. *Case Rep Pulmonol* 2012; **2012**: 595402 [PMID: [23198240](#) DOI: [10.1155/2012/595402](#)]
- 2 **Bazari H**, Attar EC, Dahl DM, Uppot RN, Colvin RB. Case records of the Massachusetts General Hospital. Case 23-2010. A 49-year-old man with erythrocytosis, perinephric fluid collections, and renal failure. *N Engl J Med* 2010; **363**: 463-475 [PMID: [20818867](#) DOI: [10.1056/NEJMcp1004086](#)]
- 3 **Rastogi R**, Rastogi UC, Sarikwal A, Rastogi V. Renal lymphangiectasia associated with chronic myeloid leukemia. *Saudi J Kidney Dis Transpl* 2010; **21**: 724-727 [PMID: [20587880](#)]
- 4 **Wani NA**, Kosar T, Gojwari T, Qureshi UA. Perinephric fluid collections due to renal lymphangiectasia. *Am J Kidney Dis* 2011; **57**: 347-351 [PMID: [20888101](#) DOI: [10.1053/j.ajkd.2010.06.028](#)]
- 5 **Floege J**, Barbour SJ, Catran DC, Hogan JJ, Nachman PH, Tang SCW, Wetzels JFM, Cheung M, Wheeler DC, Winkelmayer WC, Rovin BH; Conference Participants. Management and treatment of glomerular diseases (part 1): conclusions from a Kidney Disease: Improving Global Outcomes (KDIGO) Controversies Conference. *Kidney Int* 2019; **95**: 268-280 [PMID: [30665568](#) DOI: [10.1016/j.kint.2018.10.018](#)]
- 6 **Surabhi VR**, Menias C, Prasad SR, Patel AH, Nagar A, Dalrymple NC. Neoplastic and non-neoplastic proliferative disorders of the perirenal space: cross-sectional imaging findings. *Radiographics* 2008; **28**: 1005-1017 [PMID: [18635626](#) DOI: [10.1148/rg.284075157](#)]
- 7 **Varela JR**, Bargiela A, Requejo I, Fernandez R, Darriba M, Pombo F. Bilateral renal lymphangiomatosis: US and CT findings. *Eur Radiol* 1998; **8**: 230-231 [PMID: [9477271](#) DOI: [10.1007/s003300050368](#)]
- 8 **Al-Dofri SA**. Renal lymphangiectasia presented by pleural effusion and ascites. *J Radiol Case Rep* 2009; **3**: 5-10 [PMID: [22470619](#) DOI: [10.3941/jrcr.v3i10.317](#)]
- 9 **Riehl J**, Schmitt H, Schäfer L, Schneider B, Sieberth HG. Retroperitoneal lymphangiectasia associated with bilateral renal vein thrombosis. *Nephrol Dial Transplant* 1997; **12**: 1701-1703 [PMID: [9269653](#) DOI: [10.1093/ndt/12.8.1701](#)]
- 10 **O'Brien JD**, Ettinger NA. Nephrobronchial fistula and lung abscess resulting from nephrolithiasis and pyelonephritis. *Chest* 1995; **108**: 1166-1168 [PMID: [7555135](#) DOI: [10.1378/chest.108.4.1166](#)]
- 11 **Schwarz A**, Lenz T, Klaen R, Offermann G, Fiedler U, Nussberger J. Hygroma renale: pararenal lymphatic cysts associated with renin-dependent hypertension (Page kidney). Case report on bilateral cysts and successful therapy by marsupialization. *J Urol* 1993; **150**: 953-957 [PMID: [8345618](#) DOI: [10.1016/s0022-5347\(17\)35660-4](#)]
- 12 **Sterns RH**, Rabinowitz R, Segal AJ, Spitzer RM. 'Page kidney'. Hypertension caused by chronic subcapsular hematoma. *Arch Intern Med* 1985; **145**: 169-171 [PMID: [3970635](#) DOI: [10.1001/archinte.145.1.169](#)]
- 13 **Wang T**, Zhang Y, Niu K, Wang L, Shi Y, Liu B. Association of the -449GC and -1151AC polymorphisms in the DDAH2 gene with asymmetric dimethylarginine and erythropoietin resistance in Chinese patients on maintenance hemodialysis. *Clin Exp Pharmacol Physiol* 2017; **44**: 961-964

[PMID: 28590543 DOI: 10.1111/1440-1681.12793]

- 14 **Wang T**, Li Y, Wu H, Chen H, Zhang Y, Zhou H, Li H. Optimal blood pressure for the minimum all-cause mortality in Chinese ESRD patients on maintenance hemodialysis. *Biosci Rep* 2020; **40**: BSR20200858 [PMID: 32756870 DOI: 10.1042/BSR20200858]
- 15 **Wang T**, Zhang Y, Li Q, Jia S, Shi C, Niu K, Liu B. Acute kidney injury in cancer patients and impedance cardiography-assisted renal replacement therapy: Experience from the onconeurology unit of a Chinese tertiary hospital. *Exp Ther Med* 2017; **14**: 5671-5677 [PMID: 29285109 DOI: 10.3892/etm.2017.5244]



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

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

<https://www.wjgnet.com>

