

World Journal of *Clinical Cases*

World J Clin Cases 2022 August 16; 10(23): 8057-8431



OPINION REVIEW

- 8057** Invasive intervention timing for infected necrotizing pancreatitis: Late invasive intervention is not late for collection
Xiao NJ, Cui TT, Liu F, Li W
- 8063** Clinical utility of left atrial strain in predicting atrial fibrillation recurrence after catheter ablation: An up-to-date review
Yu ZX, Yang W, Yin WS, Peng KX, Pan YL, Chen WW, Du BB, He YQ, Yang P

MINIREVIEWS

- 8076** Gut microbiota and COVID-19: An intriguing pediatric perspective
Valentino MS, Esposito C, Colosimo S, Caprio AM, Puzone S, Guarino S, Marzuillo P, Miraglia del Giudice E, Di Sessa A
- 8088** Beta receptor blocker therapy for the elderly in the COVID-19 era
Santillo E, Migale M

ORIGINAL ARTICLE**Retrospective Cohort Study**

- 8097** Nonselective beta-blocker use is associated with increased hepatic encephalopathy-related readmissions in cirrhosis
Fallahzadeh MA, Asrani SK, Tapper EB, Saracino G, Rahimi RS

Retrospective Study

- 8107** Different squatting positions after total knee arthroplasty: A retrospective study
Li TJ, Sun JY, Du YQ, Shen JM, Zhang BH, Zhou YG
- 8115** Outcomes of seromuscular bladder augmentation compared with standard bladder augmentation in the treatment of children with neurogenic bladder
Sun XG, Li YX, Ji LF, Xu JL, Chen WX, Wang RY
- 8124** Distinctive clinical features of spontaneous pneumoperitoneum in neonates: A retrospective analysis
Kim SH, Cho YH, Kim HY
- 8133** Cognitive training for elderly patients with early Alzheimer's disease in the Qinghai-Tibet Plateau: A pilot study
Wang XH, Luo MQ
- 8141** Diagnostic value of elevated serum carbohydrate antigen 125 level in sarcoidosis
Zhang Q, Jing XY, Yang XY, Xu ZJ

- 8152** Evaluation of progressive early rehabilitation training mode in intensive care unit patients with mechanical ventilation

Qie XJ, Liu ZH, Guo LM

- 8161** Comparison of demographic features and laboratory parameters between COVID-19 deceased patients and surviving severe and critically ill cases

Wang L, Gao Y, Zhang ZJ, Pan CK, Wang Y, Zhu YC, Qi YP, Xie FJ, Du X, Li NN, Chen PF, Yue CS, Wu JH, Wang XT, Tang YJ, Lai QQ, Kang K

Clinical Trials Study

- 8170** Role of H₂receptor blocker famotidine over the clinical recovery of COVID-19 patients: A randomized controlled trial

Mohiuddin Chowdhury ATM, Kamal A, Abbas MKU, Karim MR, Ali MA, Talukder S, Hamidullah Mehedi H, Hassan H, Shahin AH, Li Y, He S

Observational Study

- 8186** Short-term prognostic factors for hepatitis B virus-related acute-on-chronic liver failure

Ye QX, Huang JF, Xu ZJ, Yan YY, Yan Y, Liu LG

- 8196** Three-dimensional psychological guidance combined with evidence-based health intervention in patients with liver abscess treated with ultrasound

Shan YN, Yu Y, Zhao YH, Tang LL, Chen XM

- 8205** Role of serum β 2-microglobulin, glycosylated hemoglobin, and vascular endothelial growth factor levels in diabetic nephropathy

Yang B, Zhao XH, Ma GB

SYSTEMATIC REVIEWS

- 8212** Gallbladder neuroendocrine carcinoma diagnosis, treatment and prognosis based on the SEER database: A literature review

Cai XC, Wu SD

CASE REPORT

- 8224** Sepsis complicated with secondary hemophagocytic syndrome induced by giant gouty tophi rupture: A case report

Lai B, Pang ZH

- 8232** Spontaneous remission of autoimmune pancreatitis: Four case reports

Zhang BB, Huo JW, Yang ZH, Wang ZC, Jin EH

- 8242** Epstein-Barr-virus-associated hepatitis with aplastic anemia: A case report

Zhang WJ, Wu LQ, Wang J, Lin SY, Wang B

- 8249** Aspiration as the first-choice procedure for airway management in an infant with large epiglottic cysts: A case report

Zheng JQ, Du L, Zhang WY

- 8255** Sequential multidisciplinary minimally invasive therapeutic strategy for heart failure caused by four diseases: A case report
Zhao CZ, Yan Y, Cui Y, Zhu N, Ding XY
- 8262** Primary ascending colon cancer accompanying skip metastases in left shoulder skin and left neck lymph node: A case report
Zhou JC, Wang JJ, Liu T, Tong Q, Fang YJ, Wu ZQ, Hong Q
- 8271** Clinical and genetic study of ataxia with vitamin E deficiency: A case report
Zhang LW, Liu B, Peng DT
- 8277** Complete resection of large-cell neuroendocrine and hepatocellular carcinoma of the liver: A case report
Noh BG, Seo HI, Park YM, Kim S, Hong SB, Lee SJ
- 8284** Immunotherapy combined with antiangiogenic agents in patients with advanced malignant pleural mesothelioma: A case report
Xuan TT, Li GY, Meng SB, Wang ZM, Qu LL
- 8291** Bladder malacoplakia: A case report
Wang HK, Hang G, Wang YY, Wen Q, Chen B
- 8298** Delayed inflammatory response evoked in nasal alloplastic implants after COVID-19 vaccination: A case report
Seo MG, Choi EK, Chung KJ
- 8304** Phosphoglyceride crystal deposition disease requiring differential diagnosis from malignant tumors and confirmed by Raman spectroscopy: A case report
Ohkura Y, Uruga H, Shiiba M, Ito S, Shimoyama H, Ishihara M, Ueno M, Udagawa H
- 8312** Vulvovaginal myeloid sarcoma with massive pelvic floor infiltration: A case report and review of literature
Wang JX, Zhang H, Ning G, Bao L
- 8323** Femoral neck stress fracture and medial tibial stress syndrome following high intensity interval training: A case report and review of literature
Tan DS, Cheung FM, Ng D, Cheung TLA
- 8330** Periosteal chondroma of the rib: A case report
Gao Y, Wang JG, Liu H, Gao CP
- 8336** Papillary thyroid carcinoma occurring with undifferentiated pleomorphic sarcoma: A case report
Lee YL, Cheng YQ, Zhu CF, Huo HZ
- 8344** Laparoscopic treatment of bilateral duplex kidney and ectopic ureter: A case report
Wang SB, Wan L, Wang Y, Yi ZJ, Xiao C, Cao JZ, Liu XY, Tang RP, Luo Y
- 8352** Incontinentia pigmenti with intracranial arachnoid cyst: A case report
Li WC, Li ML, Ding JW, Wang L, Wang SR, Wang YY, Xiao LF, Sun T

- 8360** Relapsing polychondritis causing breathlessness: Two case reports
Zhai SY, Zhang YH, Guo RY, Hao JW, Wen SX
- 8367** Endodontic management of a fused left maxillary second molar and two paramolars using cone beam computed tomography: A case report
Mei XH, Liu J, Wang W, Zhang QX, Hong T, Bai SZ, Cheng XG, Tian Y, Jiang WK
- 8375** Infant biliary cirrhosis secondary to a biliary inflammatory myofibroblastic tumor: A case report and review of literature
Huang Y, Shu SN, Zhou H, Liu LL, Fang F
- 8384** Metastatic low-grade endometrial stromal sarcoma with variable morphologies in the ovaries and mesentery: A case report
Yu HY, Jin YL
- 8392** Bronchogenic cysts with infection in the chest wall skin of a 64-year-old asymptomatic patient: A case report
Ma B, Fu KW, Xie XD, Cheng Y, Wang SQ
- 8400** Incidental accumulation of Technetium-99m pertechnetate in subacute cerebral infarction: A case report
Han YH, Jeong HJ, Kang HG, Lim ST
- 8406** Metal stent combined with ileus drainage tube for the treatment of delayed rectal perforation: A case report
Cheng SL, Xie L, Wu HW, Zhang XF, Lou LL, Shen HZ
- 8417** Using ketamine in a patient with a near-occlusion tracheal tumor undergoing tracheal resection and reconstruction: A case report
Xu XH, Gao H, Chen XM, Ma HB, Huang YG

LETTER TO THE EDITOR

- 8422** Reflections on the prevalence of human leukocyte antigen-B27 and human leukocyte antigen-B51 co-occurrence in patients with spondylarthritis
Gonçalves Júnior J, Sampaio-Barros PD, Shinjo SK
- 8425** Comment on "Disease exacerbation is common in inflammatory bowel disease patients treated with immune checkpoint inhibitors for malignancy"
Argyriou K, Kotsakis A
- 8428** Intranasal sufentanil combined with intranasal dexmedetomidine: A promising method for non-anesthesiologist sedation during endoscopic ultrasonography
Wang Y, Ge ZJ, Han C

ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Peng Liang, MD, Associate Professor, Day Surgery Center, Department of Anesthesiology, West China Hospital of Sichuan University, Chengdu 610041, Sichuan Province, China. 39485572@qq.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 abstracted and indexed in Science Citation Index Expanded (SCIE, also known as SciSearch®), Journal Citation Reports/Science Edition, Current Contents®/Clinical Medicine, PubMed, PubMed Central, Scopus, Reference Citation Analysis, China National Knowledge Infrastructure, China Science and Technology Journal Database, and Superstar Journals Database. The 2022 Edition of Journal Citation Reports® cites the 2021 impact factor (IF) for *WJCC* as 1.534; IF without journal self cites: 1.491; 5-year IF: 1.599; Journal Citation Indicator: 0.28; Ranking: 135 among 172 journals in medicine, general and internal; and Quartile category: Q4. The *WJCC*'s CiteScore for 2021 is 1.2 and Scopus CiteScore rank 2021: General Medicine is 443/826.

RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: *Hua-Ge Yu*; Production Department Director: *Xiang Li*; Editorial Office Director: *Jin-Lei 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

Bao-Gan Peng, Jerzy Tadeusz Chudek, George Kontogeorgos, Maurizio Serati, Ja Hyeon Ku

EDITORIAL BOARD MEMBERS

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

PUBLICATION DATE

August 16, 2022

COPYRIGHT

© 2022 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>

Sepsis complicated with secondary hemophagocytic syndrome induced by giant gouty tophi rupture: A case report

Bin Lai, Zhi-Hui Pang

Specialty type: Rheumatology

Provenance and peer review:

Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's scientific quality classification

Grade A (Excellent): 0

Grade B (Very good): 0

Grade C (Good): C, C

Grade D (Fair): 0

Grade E (Poor): E

P-Reviewer: Deshwal H, United States; Rodrigues AT, Brazil; Romanelli A, Italy

Received: June 8, 2021

Peer-review started: June 8, 2021

First decision: July 15, 2021

Revised: July 27, 2021

Accepted: July 8, 2022

Article in press: July 8, 2022

Published online: August 16, 2022



Bin Lai, The First Clinical Medical College, Guangzhou University of Chinese Medicine, Guangzhou 510405, Guangdong Province, China

Zhi-Hui Pang, Department of Orthopedics, The First Affiliated Hospital of Guangzhou University of Chinese Medicine, Guangzhou 510405, Guangdong Province, China

Corresponding author: Zhi-Hui Pang, MD, Chief Physician, Department of Orthopedics, The First Affiliated Hospital of Guangzhou University of Chinese Medicine, No. 12 Airport Road, Baiyun District, Guangzhou 510405, Guangdong Province, China. adi_007@126.com

Abstract

BACKGROUND

Gouty tophi are a chronic granulomatous caused by a deposition of monosodium urate crystal deposition in the body. Once broken, it may easily induce severe infection. Sepsis complicated with secondary hemophagocytic syndrome induced by gouty tophi rupture is extremely rare in the clinical setting, and no such serious complications have been reported in literature.

CASE SUMMARY

This is a 52-year-old Chinese male patient with a 20-year history of gouty arthritis. At admission, the gout stone in the patient's right ankle was broken and it secreted a white mucoid substance. During the course of treatment, the patient suffered from systemic inflammatory response syndrome multiple times. His condition gradually deteriorated, further complicated by hemophagocytic syndrome. After thorough removal of gout lesions and active anti-infection treatment and control of blood uric acid level, combined with multidisciplinary cooperation, the patient was finally cured.

CONCLUSION

Sepsis complicated with secondary hemophagocytic syndrome induced by gouty tophi rupture is extremely rare in the clinical setting. Timely and accurate diagnosis is very important to save patients' lives.

Key Words: Gouty tophi; Sepsis; Secondary hemophagocytic syndrome; Multidisciplinary cooperation; Case report

©The Author(s) 2022. Published by Baishideng Publishing Group Inc. All rights reserved.

Core Tip: Sepsis and secondary hemophagocytic syndrome induced by gouty tophi rupture are very rare in a clinical setting. Since the early symptoms are similar to gouty arthritis, it is easy to ignore septic infection and immune system damage. Sepsis and hemophagocytic syndrome develop rapidly. Often, when detected by clinicians, patients have life-threatening symptoms. This case emphasizes that clinicians should screen patients with gouty tophi rupture for early complications of sepsis and immune system damage. It is, therefore, important to improve the relevant examination as soon as possible and initiate early intervention.

Citation: Lai B, Pang ZH. Sepsis complicated with secondary hemophagocytic syndrome induced by giant gouty tophi rupture: A case report. *World J Clin Cases* 2022; 10(23): 8224-8231

URL: <https://www.wjgnet.com/2307-8960/full/v10/i23/8224.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v10.i23.8224>

INTRODUCTION

Gouty tophi are a chronic granulomatous caused by the deposition of monosodium urate crystals in and around the joint[1]. The main clinical manifestations are joint fever, swelling, and severe pain. When the gouty tophi are large, they may be palpated as a subcutaneous induration. In severe cases, joint bone destruction occurs, leading to joint dysfunction or even disability[2,3]. There have been increased reports of unusual concomitant gout and infections, such as septic arthritis and necrotizing fasciitis, in some coastal areas[4,5]. Gouty and septic arthritis can cause joint fever, swelling, and local erythema, making them difficult to distinguish from each other based on clinical symptoms alone[6]. As such, bacterial culture and Gram staining are necessary to exclude septic arthritis[7]. Although this phenomenon is not common clinically, a 15-year case study has confirmed this[4]. The hemophagocytic syndrome (HPS) is a rare immune-activated disease characterized by excessive systemic inflammation. It can be divided into two types: familial and secondary[8]. Secondary HPS usually occurs in the setting of infection, malignant lesions, rheumatism, and metabolic diseases[8,9]. The main clinical symptoms include high fever, hepatosplenomegaly, and hemocytopenia, accompanied by pulmonary dysfunction and neurological symptoms (including epilepsy, meningitis, and decreased consciousness)[10]. It is not an independent disease, but a group of clinical syndromes involving multiple organs. HPS is associated with a variety of potential diseases and can occur as a genetic or acquired disease[10,11]. At present, HPS following immune system damage caused by gouty tophi is very rare and has not been reported in the relevant literature. Herein, we report a rare case caused by the rupture of huge gouty tophi, resulting in infection at the site of the rupture, rapid development of sepsis, and eventually, destruction of the immune system in the later stage of the disease, leading to secondary HPS.

CASE PRESENTATION

Chief complaints

A 52-year-old Chinese male patient complained of recurrent right ankle pain and swelling for 20 years. He presented to our out-patient department on August 8, 2019.

History of present illness

The pain in the right ankle was repeated and aggravated at night. The maximum visual analogue scale was 9 points.

History of past illness

The patient's height was 169 cm and weight was 65 kg. He had a history of hyperuricemia and alcohol drinking for 20 years, but he did not take drugs regularly.

Personal and family history

He had no trauma history, no other special diseases, or family genetic history.

Physical examination

His right ankle joint was red, swollen, and painful, the local skin temperature was increased as evident by touch, and a gouty tophi with a size of about 6 cm × 6 cm could be seen in the right ankle joint. The gouty tophi had broken and secreted white sticky secretions accompanied by scattered odor and severe edema of the right lower limb (Figure 1).



DOI: 10.12998/wjcc.v10.i23.8224 Copyright ©The Author(s) 2022.

Figure 1 Condition of the patient's wound on admission.

Laboratory examinations

Laboratory examinations were conducted for blood, blood analysis, biochemical tests, coagulation, and levels of serum C-reactive protein, serum procalcitonin, erythrocyte sedimentation rate, brain natriuretic peptide, and ferritin. In addition, we also conducted secretion bacterial culture and bone marrow biopsy. Laboratory data at admission showed a white blood cell count of 26.45×10^9 cells/L, a neutrophil count of 24.57×10^9 cells/L, and serum uric acid level of $535 \mu\text{mol/L}$ (Table 1). After the fourth operation, laboratory data showed that a white blood cell count of 0.73×10^9 cells/L, a neutrophil count of 0.02×10^9 cells/L, a lymphocyte count of 0.63×10^9 cells/L, hemoglobin levels of 58 g/L , a platelet count of 93×10^9 cells/L, and a ferritin count of 2576.81 ng/mL (Table 1). Local palpation found that the patient's liver and spleen had varying degrees of swelling.

Imaging examinations

Echocardiography showed decreased left ventricular systolic function (ejection fraction = 30%). The results of bacterial culture of four secretions after operation all indicated that methicillin-resistant *staphylococcus aureus* (MRSA) was present. In the late stage of the disease, *Candida albicans* infection was also found in the secretion culture. Bone marrow puncture results showed that the proliferation ability of bone marrow was decreased, hemophagocytic cells was increased, and granulocytes was decreased (Figure 2).

MULTIDISCIPLINARY EXPERT CONSULTATION

Qing-Ye Zhang, Associate Chief Pharmacist, Department of pharmacy, The First Affiliated Hospital of Guangzhou University of Traditional Chinese Medicine

Consultation opinion on August 10, 2019: At present, the number of leukocytes and neutrophils is abnormally high. This finding and the clinical manifestations suggest the possibility of sepsis. It is suggested that bacterial culture and gram-staining be performed to make a definite diagnosis. Piperacillin sodium and sulbactam sodium for injection (4.5 g q8h iv) was administered for temporary anti-infection treatment. The dosage can be adjusted based on the etiological results and drug sensitivity results.

Consultation opinion on August 13, 2019: The results of the bacterial culture suggest that *Staphylococcus* methoxycycline-resistant infection is the cause. Vancomycin (0.5 g q12h iv) is suggested as an anti-infection treatment. Attention should be paid to the liver and kidney function of patients, and blood analysis and bacterial culture should be reviewed in time.

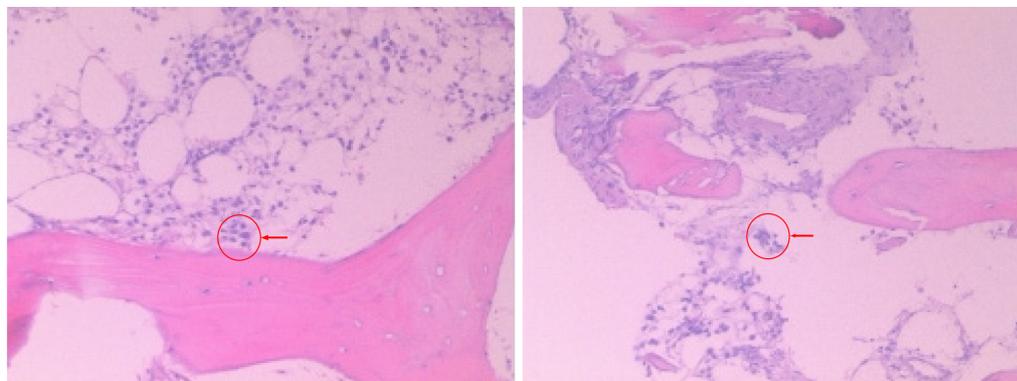
Consultation opinion on August 27, 2019: At present, the patient is still feverish (the temperature $> 38 \text{ }^\circ\text{C}$); and the highest recorded temperature was $39.4 \text{ }^\circ\text{C}$. The leukocyte and glomerular filtration rates have improved. Vancomycin (1 g qd iv) combined with piperacillin and zolbactam sodium for injection (4.5 g q8h iv) continues to be used for anti-infection treatment. Currently, the antibiotic program cover common hospital bacteria. It is suggested to use methylprednisolone tablets (12 mg qd po) for symptomatic treatment in patients with fever.

Consultation opinion on September 4, 2019: At present, the wound has healed well, the total number

Table 1 Laboratory examination during hospitalization

	On admission	After 4 th operation	Before discharge
RBC ($\times 10^{12}/L$)	4.37	2.17	2.14
WBC ($\times 10^9/L$)	26.45	0.73	4
NEU ($\times 10^9/L$)	24.57	0.02	2.02
LYM ($\times 10^9/L$)	0.66	0.63	1.50
HGB (g/L)	124	58	60
Platelet ($\times 10^9/L$)	360	93	112
UA ($\mu\text{mol}/L$)	535	380	416
Albumin (g/L)	24.3	32	34.8
AST (U/L)	18	13	6
ALT (U/L)	15	13	3
PCT (ng/L)	2.84	1.57	0.08
ESR (mm/h)	64	67	48
CRP (mg/L)	205	168	16.8
BNP (pg/L)	1470.9	474.2	643.2
D-dimer (mg/L)	5.54	2.93	
Fibrinogen (g/L)		8.05	
Ferritin (ng/mL)		2576.81	

ALT: Alanine aminotransferase; AST: Aspartate aminotransferase; BNP: Brain natriuretic peptide; CRP: C-reactive protein; ESR: Erythrocyte sedimentation rate; HGB: Hemoglobin; LYM: Lymphocyte; NEU: Neutrophil; PCT: Procalcitonin; RBC: Red blood cell; UA: Uric acid; WBC: White blood cell.



DOI: 10.12998/wjcc.v10.i23.8224 Copyright ©The Author(s) 2022.

Figure 2 Bone marrow biopsy results. The proliferation ability of bone marrow decreased, hemophagocytic cells increased (where indicated by arrow), and granulocytes decreased. Hematoxylin-eosin-stained sections, $\times 40$ magnification.

of white blood cells and neutrophils decreased, and erythrocyte sedimentation rate and C-reactive protein are increased slightly. Considering that the infection is under control, vancomycin has been used for 1 mo, it can be discontinued and replaced with levofloxacin (0.6 g qd iv) for maintenance treatment.

Consultation opinion on September 9, 2019: The patient presents with fever, sore throat, and a body temperature of $> 39\text{ }^{\circ}\text{C}$, laboratory examination indicated a drastic decrease in granulocytes and neutrophil levels, and an increase in infection indexes, such as procalcitonin level, blood sedimentation rate, and C-reactive protein level. It is recommended that imipenem and cilastatin sodium injection (0.5 g q8h iv), as well as caspofungin acetate (80 mg qd iv) are used for treatment.

Li-Wen Hu, Associate Chief Physician, Department of Hematology, The First Affiliated Hospital of Guangzhou University of Traditional Chinese Medicine

Consultation opinion on September 2, 2019: At present, the patient has high fever and fuzzy consciousness. Blood analysis indicates cytopenias (affecting at least two lineages in the peripheral blood). Local palpation revealed varying degrees of swelling in the patient's liver and spleen. Bone marrow biopsy is suggested to make a definite diagnosis. In case of infection, the use of cytokines to improve white blood cell count can be considered.

Consultation opinion on September 6, 2019: Bone marrow biopsy findings indicate that the proliferation ability of the bone marrow has decreased, hemophilic cells have increased, and granulocytes have decreased. These findings, clinical signs, and the blood test findings, considered together, indicate HPS. It is advised that the patient be transferred to the hematology department for further treatment.

FINAL DIAGNOSIS

Combined with the results of laboratory examination, bacterial culture, bone marrow biopsy, and consultation of Hematology Department and Pharmacy Department, the patient was diagnosed as: (1) Gouty arthritis; (2) Sepsis; and (3) Hemophagocytic syndrome.

TREATMENT

For the treatment of gouty tophi rupture, we conducted five surgical operations for thorough debridement combined with vacuum sealing drainage. The main cause of the disease was infection induced by the rupture of gouty tophi. Therefore, active measures should be taken to control serum uric acid levels throughout the treatment of the disease. We administered febuxostat tablets (40 mg qd po) to inhibit the formation of uric acid, and methylprednisolone tablets (40 mg qd po) to inhibit the inflammatory reaction when the symptoms are serious. For patients with repeated infections, we consulted the pharmacy department to develop a personalized antibiotic use plan for patients. On admission, we collected samples of wound secretion for bacterial culture. Considering the possibility of infection, we empirically used piper acillin sodium and sulbactam sodium for infection (4.5 g q8h iv) for 4 d. Based on the bacterial culture findings of MRSA infection, we replaced the antibiotic with vancomycin (0.5 g q12h iv) for 1 d and performed the first operation on the same day. The patient was given symptomatic treatment for uric acid reduction, detumescence, gastric protection, anti-infection, and nutritional support. The use of vancomycin (1 g q12h iv) was continued for 5 d for anti-infection treatment until the second operation. After the second operation, the patient was given oxygen inhalation, blood volume supplement, anti-thrombotic, anti-infection, and other symptomatic treatment. Additionally, vancomycin (1 g q12h iv) was administered for 4 d to prevent infections. During this period, the patient had a high fever and disturbance of consciousness again, and the laboratory examination indicated that the inflammatory index was elevated. We changed the antibiotics to vancomycin (1 g q12h iv) combined with piperacillin sodium and sulbactam sodium (4.5 g q8h iv) for 5 d. After the third operation, the secretion culture still showed MRSA infection. As such, we continued the vancomycin for injection (1 g q12h iv) combined with piperacillin sodium and sulbactam sodium (4.5 g q8h iv) for 8 d. When the patient's general condition had improved, we performed the fourth operation. After the operation, we maintained the use of vancomycin for 1 d. After the consultation and evaluation of the pharmacy department, we changed the antibiotic to levofloxacin hydrochloride injection (0.6 g qd iv) for 5d and discontinued vancomycin since the patient's infection had been controlled. During this period, the patient suddenly developed agranulocytosis, which may have been caused by drugs, according to the department of hematology. Therefore, we used recombinant human granulocyte colony-stimulating factor (100 µg qd iv) for 6 d to improve leukocyte levels. Later, the patient was diagnosed with HPS and was transferred to the department of hematology for treatment. On the 1st day, we administered imipenem cilastatin sodium (1 g q8h iv) combined with moxifloxacin hydrochloride and sodium chloride (0.1 g qd iv) for 1 d. On the 2nd day, we changed the antibiotic regimen to voriconazole (200 mg q12h iv) for 2 d and linezolid (300 mL q12h iv) for 6 d. After treatment, the patient's vital signs gradually stabilized, followed by the performance of the fifth operation. After the operation, the patient was given symptomatic treatment, such as oxygen inhalation, albumin supplement, gastric protection, and nutritional support. No bacterial infection was found in the secretions of patients after the operation. Considering that the related inflammatory indexes were still high, we administered cefoperazone sodium and sulbactam sodium (3 g bid iv) for 9 d.

OUTCOME AND FOLLOW-UP

After 2 mo of treatment, his laboratory indexes finally returned to normal (Table 1), and the right ankle wound healed well. Under the guidance of the physical therapist, the right ankle function gradually recovered. At his 5-mo follow-up in the outpatient department, his right ankle was completely healed, and the joint function was good (Figure 3).

DISCUSSION

Gouty arthritis, caused by the deposition of monosodium urate crystals in the joints, tendons, and surrounding soft tissues, is characterized by joint swelling, pain, and dysfunction[12,13]. According to a study in Taiwan, 43.6% of patients with gouty arthritis have symptoms of sepsis. Because the early symptoms are joint swelling and severe pain, these are often ignored by clinicians. As such, when these are finally detected, the patients are already experiencing life-threatening symptoms[7]. The main reason for this is that patients with a long history of gout or multiple subcutaneous gouty tophi deposits are prone to local wound ulcers and *staphylococcus aureus* infection[14]. Staphylococemia leads to the metastatic infection of inflammatory joints and the deposition of joint crystals, and the local setting of inflammation and effusion induced by monosodium urate provides an environment for the accumulation of blood-borne microorganisms and the growth of bacteria[7]. This explains why our patients repeatedly developed sepsis and septic shock during hospitalization. Sepsis progresses rapidly. Once it occurs, severe anti-inflammatory reactions may easily occur, causing the patients to undergo immune cell apoptosis and eventually become complicated with various diseases[15,16]. This may be the main reason why our patient suddenly developed HPS in the later stages of the disease. Therefore, early prevention, detection, and intervention are key in delaying the disease process. The main basis for the diagnosis of sepsis includes a clear infectious disease, body temperature changes, and associated inflammatory markers[17]. Given these, the selection of the appropriate antibiotics is the main link in preventing the further progression of sepsis. Second, we should actively remove infected tissues with obvious lesions and cut off the source of infection[18]. Our patient had an infected - local gout stone ulceration on admission. After admission, the wound appeared red and swollen and was warm to the touch. Laboratory examination revealed that the related inflammatory indices increased sharply. The results of the bacterial culture suggested an MRSA infection. We took the measures quickly. We conducted multidisciplinary consultations to develop an effective antibiotic treatment plan; however, we effectively treated the MRSA infection through surgical removal combined with local negative pressure drainage to remove the infection focus. Although the patient still had an infection and even experienced shock several times during the treatment, the infection was controlled and the patient's life was saved through active anti-infection treatment, which also proved that our treatment measures were effective. This case provides valuable experience. To improve the prognosis of patients with giant gouty tophi rupture, it is essential to carry out an early bacterial culture of the secretions, cooperate with a multidisciplinary team, and formulate the corresponding anti-infection treatment plan.

HPS is a rare and destructive immune-activated disease. The excessive inflammation in secondary HPS is mainly associated with infectious, autoimmune, or neoplastic diseases. Furthermore, *Pseudomonas aeruginosa* and MRSA have been reported to be associated with bacterial HPS[11,19,20]. Although there is no relevant study regarding secondary HPS induced by gout stone rupture, we believe that this mainly occurs due to infection with MRSA, causing septic shock, destruction of immune activation, and induction of HPS. Because of the rarity of this situation, it can be easily ignored by most clinicians. Our case provides a good example of this. Although sepsis occurred many times during the treatment period and HPS developed due to immune system damage in the later stage, which induces HPS, our timely review of relevant indicators and multidisciplinary consultation allowed for the active provision of effective treatment measures, ultimately helping the patient recover. Therefore, we believe that for patients with gouty tophi rupture, sepsis, and immune system damage should be considered in the early stage, and to avoid the occurrence of HPS. Moreover, a timely review of relevant laboratory indicators, combined with multidisciplinary consultation is the key to the successful treatment of the disease.

CONCLUSION

Sepsis complicated with secondary hemophagocytic syndrome induced by gouty tophi rupture is extremely rare in the clinical setting. Timely and accurate diagnosis is very important to save patients' lives.



DOI: 10.12998/wjcc.v10.i23.8224 Copyright ©The Author(s) 2022.

Figure 3 Wound healing of affected limb during outpatient follow-up 5 mo after discharge.

ACKNOWLEDGEMENTS

Thanks to Qing-Ye Zhang, Associate Chief Pharmacist, for guiding the formulation of antibiotic regimen; Li-Wen Hu, Associate Chief Physician, for helping diagnose hemophagocytic syndrome and guiding treatment.

FOOTNOTES

Author contributions: Both authors were involved in the treatment and case of the patient; Lai B collected the patient's data and wrote the initial draft of the manuscript; Pang ZH critically appraised and revised the overall content of the manuscript; All authors read and approved the final manuscript.

Supported by the National Natural Science Foundation of China, No. 81774336 (to Pang ZH).

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 conflict of interest.

CARE Checklist (2016) statement: 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: <https://creativecommons.org/licenses/by-nc/4.0/>

Country/Territory of origin: China

ORCID number: Bin Lai 0000-0003-2921-6844; Zhi-Hui Pang 0000-0003-2507-2564.

S-Editor: Gong ZM

L-Editor: Filipodia

P-Editor: Gong ZM

REFERENCES

- 1 Wilson L, Saseen JJ. Gouty Arthritis: A Review of Acute Management and Prevention. *Pharmacotherapy* 2016; **36**: 906-922 [PMID: 27318031 DOI: 10.1002/phar.1788]
- 2 Goel N, Khanna V, Jain DK, Gupta V. Gouty tophi presenting as multinodular lateral inguinal swelling: A case report. *Diagn Cytopathol* 2018; **46**: 801-803 [PMID: 29882294 DOI: 10.1002/dc.23972]
- 3 Qaseem A, McLean RM, Starkey M, Forciea MA; Clinical Guidelines Committee of the American College of Physicians,

- Denberg TD, Barry MJ, Boyd C, Chow RD, Fitterman N, Humphrey LL, Kansagara D, Manaker S, Vijan S, Wilt TJ. Diagnosis of Acute Gout: A Clinical Practice Guideline From the American College of Physicians. *Ann Intern Med* 2017; **166**: 52-57 [PMID: 27802479 DOI: 10.7326/M16-0569]
- 4 **Yu KH**, Luo SF, Liou LB, Wu YJ, Tsai WP, Chen JY, Ho HH. Concomitant septic and gouty arthritis--an analysis of 30 cases. *Rheumatology (Oxford)* 2003; **42**: 1062-1066 [PMID: 12730521 DOI: 10.1093/rheumatology/keg297]
 - 5 **Yu KH**, Ho HH, Chen JY, Luo SF. Gout complicated with necrotizing fasciitis--report of 15 cases. *Rheumatology (Oxford)* 2004; **43**: 518-521 [PMID: 14722350 DOI: 10.1093/rheumatology/keh097]
 - 6 **Delle Sedie A**, Riente L, Iagnocco A, Filippucci E, Meenagh G, Grassi W, Valesini G, Bombardieri S. Ultrasound imaging for the rheumatologist X. Ultrasound imaging in crystal-related arthropathies. *Clin Exp Rheumatol* 2007; **25**: 513-517 [PMID: 17888204]
 - 7 **Weng CT**, Liu MF, Lin LH, Weng MY, Lee NY, Wu AB, Huang KY, Lee JW, Wang CR. Rare coexistence of gouty and septic arthritis: a report of 14 cases. *Clin Exp Rheumatol* 2009; **27**: 902-906 [PMID: 20149303]
 - 8 **Schram AM**, Berliner N. How I treat hemophagocytic lymphohistiocytosis in the adult patient. *Blood* 2015; **125**: 2908-2914 [PMID: 25758828 DOI: 10.1182/blood-2015-01-551622]
 - 9 **Ramos-Casals M**, Brito-Zerón P, López-Guillermo A, Khamashta MA, Bosch X. Adult haemophagocytic syndrome. *Lancet* 2014; **383**: 1503-1516 [PMID: 24290661 DOI: 10.1016/S0140-6736(13)61048-X]
 - 10 **Henter JI**, Horne A, Aricó M, Egeler RM, Filipovich AH, Imashuku S, Ladisch S, McClain K, Webb D, Winiarski J, Janka G. HLH-2004: Diagnostic and therapeutic guidelines for hemophagocytic lymphohistiocytosis. *Pediatr Blood Cancer* 2007; **48**: 124-131 [PMID: 16937360 DOI: 10.1002/pbc.21039]
 - 11 **Xu P**, Zeng H, Zhou M, Ouyang J, Chen B, Zhang Q. Successful management of a complicated clinical crisis: A patient with left-sided endocarditis and secondary hemophagocytic lymphohistiocytosis: a rare case report and literature review. *Medicine (Baltimore)* 2017; **96**: e9451 [PMID: 29390580 DOI: 10.1097/MD.00000000000009451]
 - 12 **Tausche AK**, Aringer M. [Gouty arthritis]. *Z Rheumatol* 2016; **75**: 885-898 [PMID: 27730304 DOI: 10.1007/s00393-016-0206-z]
 - 13 **Dehlin M**, Jacobsson L, Roddy E. Global epidemiology of gout: prevalence, incidence, treatment patterns and risk factors. *Nat Rev Rheumatol* 2020; **16**: 380-390 [PMID: 32541923 DOI: 10.1038/s41584-020-0441-1]
 - 14 **Hess RJ**, Martin JH. Pyarthrosis complicating gout. *JAMA* 1971; **218**: 592-593 [PMID: 5171014]
 - 15 **Bone RC**. Sir Isaac Newton, sepsis, SIRS, and CARS. *Crit Care Med* 1996; **24**: 1125-1128 [PMID: 8674323 DOI: 10.1097/00003246-199607000-00010]
 - 16 **Hotchkiss RS**, Karl IE. The pathophysiology and treatment of sepsis. *N Engl J Med* 2003; **348**: 138-150 [PMID: 12519925 DOI: 10.1056/NEJMra021333]
 - 17 **Singer M**, Deutschman CS, Seymour CW, Shankar-Hari M, Annane D, Bauer M, Bellomo R, Bernard GR, Chiche JD, Cooper-Smith CM, Hotchkiss RS, Levy MM, Marshall JC, Martin GS, Opal SM, Rubenfeld GD, van der Poll T, Vincent JL, Angus DC. The Third International Consensus Definitions for Sepsis and Septic Shock (Sepsis-3). *JAMA* 2016; **315**: 801-810 [PMID: 26903338 DOI: 10.1001/jama.2016.0287]
 - 18 **Emergency Medicine Branch Of Chinese Medical Care International Exchange Promotion Association**; Emergency Medical Branch Of Chinese Medical Association; Chinese Medical Doctor Association Emergency Medical Branch; Chinese People's Liberation Army Emergency Medicine Professional Committee. [Consensus of Chinese experts on early prevention and blocking of sepsis]. *Zhonghua Wei Zhong Bing Ji Jiu Yi Xue* 2020; **32**: 518-530 [PMID: 32576341 DOI: 10.3760/cma.j.cn121430-20200514-00414]
 - 19 **Ito S**, Takada N, Ozasa A, Hanada M, Sugiyama M, Suzuki K, Nagae Y, Inagaki T, Suzuki Y, Komatsu H. Secondary hemophagocytic syndrome in a patient with methicillin-sensitive Staphylococcus Aureus bacteremia due to severe decubitus ulcer. *Intern Med* 2006; **45**: 303-307 [PMID: 16595999 DOI: 10.2169/internalmedicine.45.1535]
 - 20 **Hoshino C**, Satoh N, Sugawara S, Kuriyama C, Kikuchi A, Ohta M. Community-acquired Staphylococcus aureus pneumonia accompanied by rapidly progressive glomerulonephritis and hemophagocytic syndrome. *Intern Med* 2007; **46**: 1047-1053 [PMID: 17603250 DOI: 10.2169/internalmedicine.46.6378]



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>

