

# World Journal of *Clinical Cases*

*World J Clin Cases* 2021 June 26; 9(18): 4460-4880



**OPINION REVIEW**

- 4460 Surgery for pancreatic tumors in the midst of COVID-19 pandemic

*Kato H, Asano Y, Arakawa S, Ito M, Kawabe N, Shimura M, Hayashi C, Ochi T, Yasuoka H, Higashiguchi T, Kondo Y, Nagata H, Horiguchi A*

**REVIEW**

- 4467 Roles of exosomes in diagnosis and treatment of colorectal cancer

*Umwali Y, Yue CB, Gabriel ANA, Zhang Y, Zhang X*

**MINIREVIEWS**

- 4480 Dynamics of host immune responses to SARS-CoV-2

*Taherkhani R, Taherkhani S, Farshadpour F*

- 4491 Current treatment for hepatitis C virus/human immunodeficiency virus coinfection in adults

*Laiwatthanapaisan R, Sirinawasatien A*

- 4500 Anti-tumor effect of statin on pancreatic adenocarcinoma: From concept to precision medicine

*Huang CT, Liang YJ*

- 4506 Roles of vitamin A in the regulation of fatty acid synthesis

*Yang FC, Xu F, Wang TN, Chen GX*

**ORIGINAL ARTICLE****Basic Study**

- 4520 Identification of the circRNA-miRNA-mRNA regulatory network and its prognostic effect in colorectal cancer

*Yin TF, Zhao DY, Zhou YC, Wang QQ, Yao SK*

- 4542 Tetramethylpyrazine inhibits proliferation of colon cancer cells *in vitro*

*Li H, Hou YX, Yang Y, He QQ, Gao TH, Zhao XF, Huo ZB, Chen SB, Liu DX*

**Case Control Study**

- 4553 Significance of highly phosphorylated insulin-like growth factor binding protein-1 and cervical length for prediction of preterm delivery in twin pregnancies

*Lan RH, Song J, Gong HM, Yang Y, Yang H, Zheng LM*

**Retrospective Cohort Study**

- 4559** Expected outcomes and patients' selection before chemoembolization—"Six-and-Twelve or Pre-TACE-Predict" scores may help clinicians: Real-life French cohorts results  
*Adhoute X, Larrey E, Anty R, Chevallier P, Penaranda G, Tran A, Bronowicki JP, Raoul JL, Castellani P, Perrier H, Bayle O, Monnet O, Pol B, Bourliere M*

**Retrospective Study**

- 4573** Application of intelligent algorithms in Down syndrome screening during second trimester pregnancy  
*Zhang HG, Jiang YT, Dai SD, Li L, Hu XN, Liu RZ*
- 4585** Evaluation of a five-gene signature associated with stromal infiltration for diffuse large B-cell lymphoma  
*Nan YY, Zhang WJ, Huang DH, Li QY, Shi Y, Yang T, Liang XP, Xiao CY, Guo BL, Xiang Y*
- 4599** Efficacy of combination of localized closure, ethacridine lactate dressing, and phototherapy in treatment of severe extravasation injuries: A case series  
*Lu YX, Wu Y, Liang PF, Wu RC, Tian LY, Mo HY*
- 4607** Observation and measurement of applied anatomical features for thoracic intervertebral foramen puncture on computed tomography images  
*Wang R, Sun WW, Han Y, Fan XX, Pan XQ, Wang SC, Lu LJ*
- 4617** Histological transformation of non-small cell lung cancer: Clinical analysis of nine cases  
*Jin CB, Yang L*
- 4627** Diagnostic value of amygdala volume on structural magnetic resonance imaging in Alzheimer's disease  
*Wang DW, Ding SL, Bian XL, Zhou SY, Yang H, Wang P*
- 4637** Comparison of ocular axis and corneal diameter between entropion and non-entropion eyes in children with congenital glaucoma  
*Wang Y, Hou ZJ, Wang HZ, Hu M, Li YX, Zhang Z*

**Observational Study**

- 4644** Risk factors for postoperative delayed gastric emptying in ovarian cancer treated with cytoreductive surgery and hyperthermic intraperitoneal chemotherapy  
*Cui GX, Wang ZJ, Zhao J, Gong P, Zhao SH, Wang XX, Bai WP, Li Y*
- 4654** Clinical characteristics, gastrointestinal manifestations and outcomes of COVID-19 patients in Iran; does the location matters?  
*Mokarram P, Dalivand MM, Pizuorno A, Aligolighasemabadi F, Sadeghdoust M, Sadeghdoust E, Aduli F, Oskrochi G, Brim H, Ashktorab H*
- 4668** AWGS2019 vs EWGSOP2 for diagnosing sarcopenia to predict long-term prognosis in Chinese patients with gastric cancer after radical gastrectomy  
*Wu WY, Dong JJ, Huang XC, Chen ZJ, Chen XL, Dong QT, Bai YY*

**Prospective Study**

- 4681** Clinical outcomes and 5-year follow-up results of keratosis pilaris treated by a high concentration of glycolic acid

*Tian Y, Li XX, Zhang JJ, Yun Q, Zhang S, Yu JY, Feng XJ, Xia AT, Kang Y, Huang F, Wan F*

**Randomized Controlled Trial**

- 4690** Tenofovir disoproxil fumarate in Chinese chronic hepatitis B patients: Results of a multicenter, double-blind, double-dummy, clinical trial at 96 weeks

*Chen XF, Fan YN, Si CW, Yu YY, Shang J, Yu ZJ, Mao Q, Xie Q, Zhao W, Li J, Gao ZL, Wu SM, Tang H, Cheng J, Chen XY, Zhang WH, Wang H, Xu ZN, Wang L, Dai J, Xu JH*

**SYSTEMATIC REVIEWS**

- 4700** Mesenteric ischemia in COVID-19 patients: A review of current literature

*Kerawala AA, Das B, Solangi A*

- 4709** Role of theories in school-based diabetes care interventions: A critical review

*An RP, Li DY, Xiang XL*

**CASE REPORT**

- 4721** Alport syndrome combined with lupus nephritis in a Chinese family: A case report

*Liu HF, Li Q, Peng YQ*

- 4728** Botulinum toxin injection for Cockayne syndrome with muscle spasticity over bilateral lower limbs: A case report

*Hsu LC, Chiang PY, Lin WP, Guo YH, Hsieh PC, Kuan TS, Lien WC, Lin YC*

- 4734** Meigs' syndrome caused by granulosa cell tumor accompanied with intrathoracic lesions: A case report

*Wu XJ, Xia HB, Jia BL, Yan GW, Luo W, Zhao Y, Luo XB*

- 4741** Primary mesonephric adenocarcinoma of the fallopian tube: A case report

*Xie C, Shen YM, Chen QH, Bian C*

- 4748** Pancreas-preserving duodenectomy for treatment of a duodenal papillary tumor: A case report

*Wu B, Chen SY, Li Y, He Y, Wang XX, Yang XJ*

- 4754** Pheochromocytoma with abdominal aortic aneurysm presenting as recurrent dyspnea, hemoptysis, and hypotension: A case report

*Zhao HY, Zhao YZ, Jia YM, Mei X, Guo SB*

- 4760** Minimally invasive removal of a deep-positioned cannulated screw from the femoral neck: A case report

*Yang ZH, Hou FS, Yin YS, Zhao L, Liang X*

- 4765** Splenic Kaposi's sarcoma in a human immunodeficiency virus-negative patient: A case report

*Zhao CJ, Ma GZ, Wang YJ, Wang JH*

- 4772 Neonatal syringocystadenoma papilliferum: A case report  
*Jiang HJ, Zhang Z, Zhang L, Pu YJ, Zhou N, Shu H*
- 4778 Disappeared intralenticular foreign body: A case report  
*Xue C, Chen Y, Gao YL, Zhang N, Wang Y*
- 4783 Femoral neck stress fractures after trampoline exercise: A case report  
*Nam DC, Hwang SC, Lee EC, Song MG, Yoo JI*
- 4789 Collision carcinoma of the rectum involving neuroendocrine carcinoma and adenocarcinoma: A case report  
*Zhao X, Zhang G, Li CH*
- 4797 Therapeutic effect of autologous concentrated growth factor on lower-extremity chronic refractory wounds: A case report  
*Liu P, Liu Y, Ke CN, Li WS, Liu YM, Xu S*
- 4803 Cutaneous myiasis with eosinophilic pleural effusion: A case report  
*Fan T, Zhang Y, Lv Y, Chang J, Bauer BA, Yang J, Wang CW*
- 4810 Severe hematuria due to vesical varices in a patient with portal hypertension: A case report  
*Wei ZJ, Zhu X, Yu HT, Liang ZJ, Gou X, Chen Y*
- 4817 Rare coexistence of multiple manifestations secondary to thalamic hemorrhage: A case report  
*Yu QW, Ye TF, Qian WJ*
- 4823 Anderson-Fabry disease presenting with atrial fibrillation as earlier sign in a young patient: A case report  
*Kim H, Kang MG, Park HW, Park JR, Hwang JY, Kim K*
- 4829 Long-term response to avelumab and management of oligoprogression in Merkel cell carcinoma: A case report  
*Leão I, Marinho J, Costa T*
- 4837 Central pontine myelinolysis mimicking glioma in diabetes: A case report  
*Shi XY, Cai MT, Shen H, Zhang JX*
- 4844 Microscopic transduodenal excision of an ampullary adenoma: A case report and review of the literature  
*Zheng X, Sun QJ, Zhou B, Jin M, Yan S*
- 4852 Growth hormone cocktail improves hepatopulmonary syndrome secondary to hypopituitarism: A case report  
*Ji W, Nie M, Mao JF, Zhang HB, Wang X, Wu XY*
- 4859 Low symptomatic COVID-19 in an elderly patient with follicular lymphoma treated with rituximab-based immunotherapy: A case report  
*Łącki S, Wyżgolik K, Nicze M, Georgiew-Nadziakiewicz S, Chudek J, Wdowiak K*

- 4866** Adult rhabdomyosarcoma originating in the temporal muscle, invading the skull and meninges: A case report  
*Wang GH, Shen HP, Chu ZM, Shen J*
- 4873** *Listeria monocytogenes* bacteremia in a centenarian and pathogen traceability: A case report  
*Zhang ZY, Zhang XA, Chen Q, Wang JY, Li Y, Wei ZY, Wang ZC*

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**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**

June 26, 2021

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**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>

## Splenic Kaposi's sarcoma in a human immunodeficiency virus-negative patient: A case report

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**Author contributions:** Zhao CJ and Wang JH contributed to the study conception; Wang JH contributed to the design and supervision; Zhao CJ, Ma GZ and Wang YJ contributed resources; Zhao CJ and Ma GZ contributed materials; Zhao CJ, Ma GZ and Wang YJ contributed to the data collection and/or processing; Zhao CJ and Wang JH contributed to the analysis and/or interpretation; Wang JH contributed to the literature search; Wang JH wrote manuscript and critically revised it for important intellectual content.

**Informed consent statement:**

Informed written consent was obtained from the patient and legal guardian 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).

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### Abstract

#### BACKGROUND

Kaposi's sarcoma (KS) is a malignancy that usually affects the skin of the lower extremities, and may involve internal organs. It originates from the vascular endothelium. It is well known that the development of KS is associated with human herpes virus 8 (*i.e.* HHV8) infections. Sporadic KS cases have mainly been found in Africa. Isolated splenic KS in Asia has rarely been reported. We present here a case of KS primarily involving the spleen in a human immunodeficiency virus (HIV)-negative Chinese patient.

#### CASE SUMMARY

A 50-year-old male patient was admitted to hospital due to abdominal distension and discomfort, reduced food intake and weight loss. Medical examination revealed that the patient had moderate anemia, a low platelet count, slight fatty liver and a huge mass in the spleen. Spleen lymphoma was considered. An anti-HIV test was negative. The whole spleen was surgically excised. The final pathological diagnosis was nodular stage spleen KS, and the patient underwent total splenectomy. He recovered well and was discharged from hospital 12 d after surgery. Two weeks later, the patient developed liver metastasis and died within 1 mo after surgery.

#### CONCLUSION

KS is difficult to diagnose and pathological examination is necessary. KS has a

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**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:** December 22, 2020

**Peer-review started:** December 22, 2020

**First decision:** March 11, 2021

**Revised:** March 22, 2021

**Accepted:** April 23, 2021

**Article in press:** April 23, 2021

**Published online:** June 26, 2021

**P-Reviewer:** Yildirim M

**S-Editor:** Gao CC

**L-Editor:** Filipodia

**P-Editor:** Wang LL



poor prognosis and should be diagnosed and treated early to improve survival.

**Key Words:** Kaposi's sarcoma; Spleen; Splenectomy; Human immunodeficiency virus negative; Human herpes virus 8; Case report

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**Core Tip:** This case describes a rare Splenic Kaposi's sarcoma (KS) in a human immunodeficiency virus-negative patient. The preoperative diagnosis was lymphoma, but the postoperative pathological diagnosis was KS. The operation and lack of proper postoperative treatments for KS may promote liver metastasis and death, as in this case. Improving the overall understanding of KS is paramount. Therefore, we report this case to provide KS experiences and lessons for future clinical works.

**Citation:** Zhao CJ, Ma GZ, Wang YJ, Wang JH. Splenic Kaposi's sarcoma in a human immunodeficiency virus-negative patient: A case report. *World J Clin Cases* 2021; 9(18): 4765-4771

**URL:** <https://www.wjgnet.com/2307-8960/full/v9/i18/4765.htm>

**DOI:** <https://dx.doi.org/10.12998/wjcc.v9.i18.4765>

## INTRODUCTION

Kaposi's sarcoma (KS) is a rare angioproliferative disease of the vascular endothelium [1]. KS usually affects the skin of extremities but can also involve internal organs. The development of KS involves human herpes virus 8 (HHV8) infection, regardless of any association with acquired immunodeficiency syndrome (*i.e.* AIDS)[2]. KS has been classified into four types: classic KS, endemic African KS, iatrogenic KS, and epidemic KS[3]. Sporadic KS cases have mainly been reported in the Mediterranean/Eastern Europe, and isolated splenic KS is rare. Cutaneous KS lesions have three forms, based on histological appearance: patch lesions, plaque stage lesions, and nodular stage lesions. Clinical features of KS vary from asymptomatic to aggressive. Pain, edema, and ulceration may occur in cutaneous or mucous lesions[4]. Visceral KS may involve the liver, gastrointestinal tract, adrenal glands and the spleen[5-9], which can be life-threatening. In splenic KS, abdominal discomfort (distension or pain), and anemia were important clinical symptoms of a previously reported case and in our patient as well [9]. Differential diagnosis from other hematological diseases is required.

We present herein a rare case of KS that primarily involved the spleen in a human immunodeficiency virus (HIV)-negative elderly male patient. We also review the literature in PubMed using the search terms "Kaposi's sarcoma", "spleen", "HIV negative", and "case report".

## CASE PRESENTATION

### Chief complaints

A 50-year-old man presented with a 2-wk history of abdominal distension, reduced food intake and weight loss.

### History of present illness

The patient began to have symptoms more than 10 d prior. He had previously visited a local clinic and received anti-inflammatory treatments to alleviate his symptoms. However, the symptoms relapsed after discontinuation of treatment. The patient was admitted to our hospital for further diagnosis and treatment. He underwent thorough laboratory examinations and was primarily diagnosed with thrombocytopenia, anemia, fatty liver and splenic space-occupying lesions. The patient was transferred from the hematology department into our ward for surgery.

**History of past illness**

The patient had a free previous medical history.

**Personal and family history**

The patient had no relevant personal and family history.

**Physical examination**

On admission, the patient's temperature was 36.5 °C, heart rate was 96 beats/min, respiratory rate was 19 breaths/min, blood pressure was 117/89 mmHg, and he had an anemic appearance. The patient had no rash or KS lesions on the skin. There were no lumps in the abdomen, the liver was not palpable, and the spleen could be touched at three transverse fingers under the ribs. There was no edema in the lower extremities.

**Laboratory examinations**

A routine blood test showed moderate anemia with a hemoglobin level of 7.9 g/dL, thrombocytopenia with a platelet count of 35000/ $\mu$ L, and an albumin level of 30.5 g/L. Bone marrow biopsy showed active myeloproliferation (80%); a decrease in the ratio of granulocytes to erythrocytes was observed and immunohistochemical investigation showed CD 34(+), MPO(+), CD20(+), and CD3(T cell +). Tests for hepatitis B virus (hepatitis B surface antigen, hepatitis B surface antibody, hepatitis B e antigen, hepatitis B e antibody, hepatitis B core antibody) were negative. Tests for HIV antigen and antibody were negative. Detection of nucleic acid for COVID-19 (coronavirus disease 2019) was negative.

**Imaging examinations**

A computed tomography (CT) scan of the chest showed an inflammatory lesion in the right upper lobe of the lung and effusion in the left pleura. Abdominal CT suggested splenomegaly, with multiple low density shadows and mixed density shadows (Figure 1A). Abdominal magnetic resonance imaging (MRI) revealed space-occupying lesions in the spleen, which were considered to be lymphoma (Figure 1B).

**Initial treatment**

During hospitalization, the patient had moderate anemia and thrombocytopenia, and was initially treated with thymosin and caffeic acid tablets to stimulate the production of blood in the bone marrow. He also received omeprazole to protect the stomach and glutathione to protect the liver. The day before surgery, the patient was administered a platelet transfusion to increase his platelet count to 50000/ $\mu$ L.

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**FINAL DIAGNOSIS**

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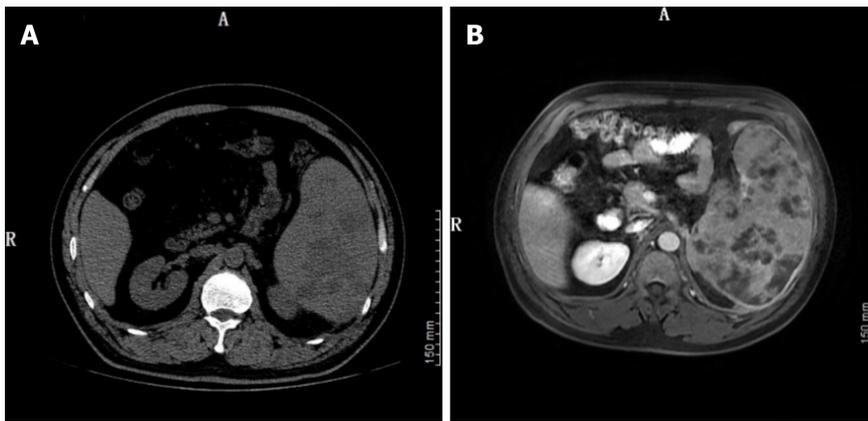
The patient underwent an elective splenectomy. Macroscopic examination revealed that the spleen was 22 cm  $\times$  18 cm  $\times$  12 cm in size, part of the spleen surface membrane was missing, and the surface was dark red with many gray and white foci. Tumor tissues were harvested and subjected to histopathological examination (Figure 2), which revealed nodular stage splenic KS. Proliferative spindle cells were crisscrossed and divided by slit-like spaces containing blood cells, which were sieve-like or beehive in shape (Figure 3A and B). Immunohistochemical examination revealed that the tumor cells were immune-positive for CD31, CD34, FIL-1, ERG, Ki-67 (10%+), CD8 (individual cells+), and D2-40 (focal+), and were immune-negative for CK, EMA, HHV8 and EBER (Figure 3C-F). The patient was readmitted to our hospital and was diagnosed with multiple liver metastases of KS based on MRI findings (Figure 4).

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**TREATMENT**

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Based on the laboratory examinations and MRI findings, spleen lymphoma was suspected. When the patient's condition was clinically stable, he underwent total splenectomy laparoscopically. During the operation, the liver was explored and no abnormal nodules were found. After surgery, the patient was given ceftriaxone for anti-infection and other medication to protect his liver and stomach. The patient recovered well and was discharged from hospital 12 d after surgery.



**Figure 1 Abdominal computed tomography scan and magnetic resonance imaging before surgery.** A: The abdominal computed tomography performed on July 20, 2020 revealed splenomegaly, with multiple low density shadows and mixed density shadows; B: The magnetic resonance imaging on July 25, 2020 revealed multiple space-occupying lesions in the spleen, which was considered to be lymphoma.



**Figure 2 Abdominal magnetic resonance imaging images of the liver pre- and post-operation.** A: The magnetic resonance imaging (MRI) performed before surgery on July 25, 2020 showed no Kaposi's sarcoma (KS) lesions in the liver; B: The MRI on September 5, 2020, when the patient was readmitted to the hospital after surgery, showed multiple KS metastatic lesions in the liver (black spots).

## OUTCOME AND FOLLOW-UP

After surgery, the patient's platelet count increased from 35000/ $\mu$ L to 114000/ $\mu$ L and then decreased 1 wk later. The patient's anemia did not resolve. Two weeks after discharge, the patient was readmitted to the hospital due to severe jaundice. MRI showed that the patient had developed multiple liver metastases of KS, and laboratory examinations revealed impaired liver function. Unfortunately, the patient did not undergo further chemotherapy and died 1 wk later.

## DISCUSSION

KS lesions are mainly localized on the skin of distal extremities but can also involve lymph nodes and visceral organs without skin involvement[3,10]. KS primarily located in the spleen in non-HIV patients has rarely been reported. Sarode *et al*[9] reported the first case of splenic KS in 1991. Mikami *et al*[11] reported a case of Kaposi-like variant of splenic angiosarcoma lacking HHV8 DNA in an HIV-negative patient in 2002. To our knowledge, we report the third case of KS located in the spleen in an HIV-negative patient. These cases have similar clinical features and malignant behavior. KS lesions have three stages: patch stage, plaque stage and nodular stage[4]. Based on low-magnification findings, our patient had nodular stage KS.

The pathogenesis of KS has still not been clearly elucidated. It is uniformly associated with HHV8 infection, irrespective of HIV infection history[12]. Szajerka *et al* [3] reviewed the multifactorial etiopathogenesis of KS, including the HIV, iron and

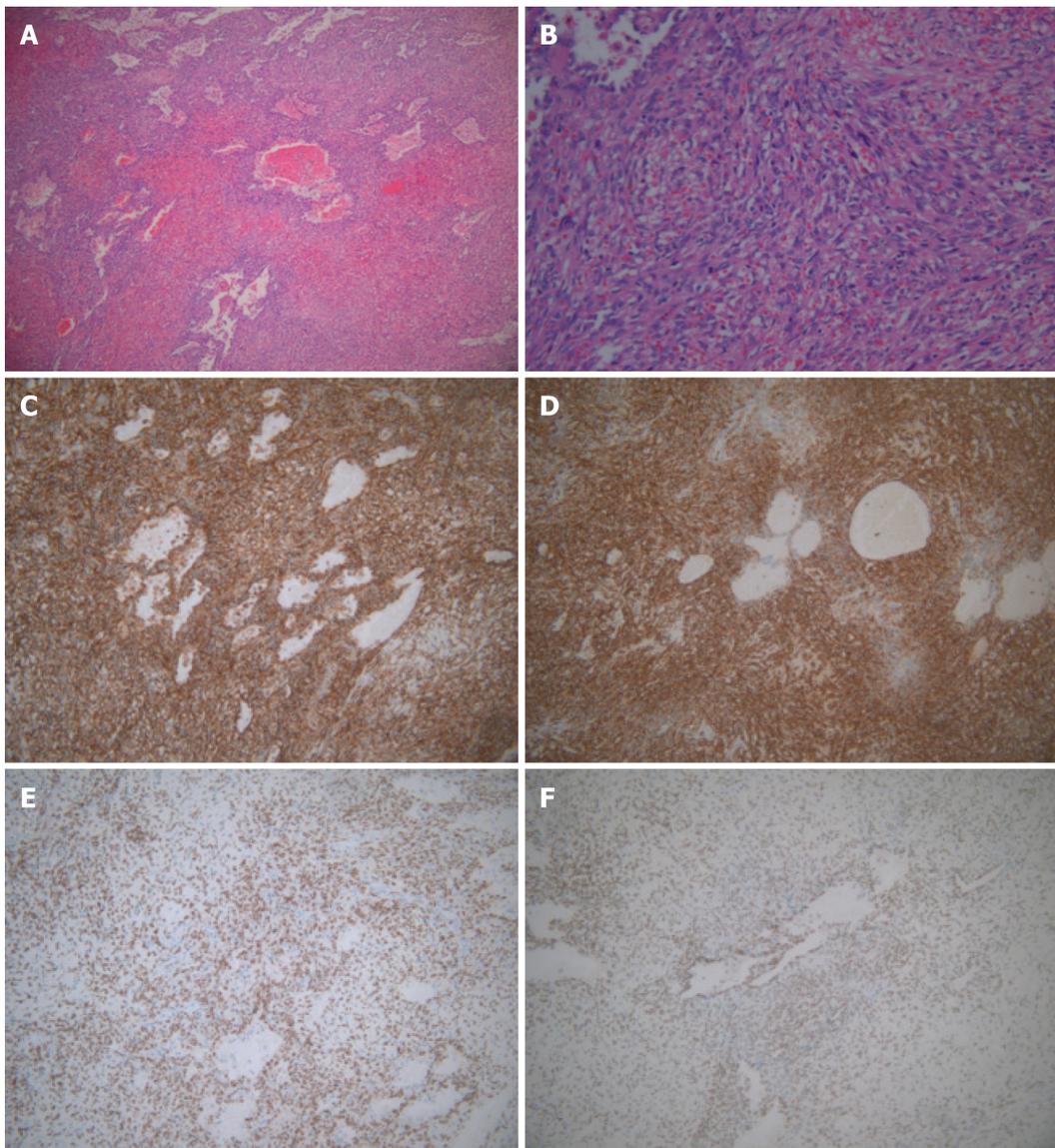
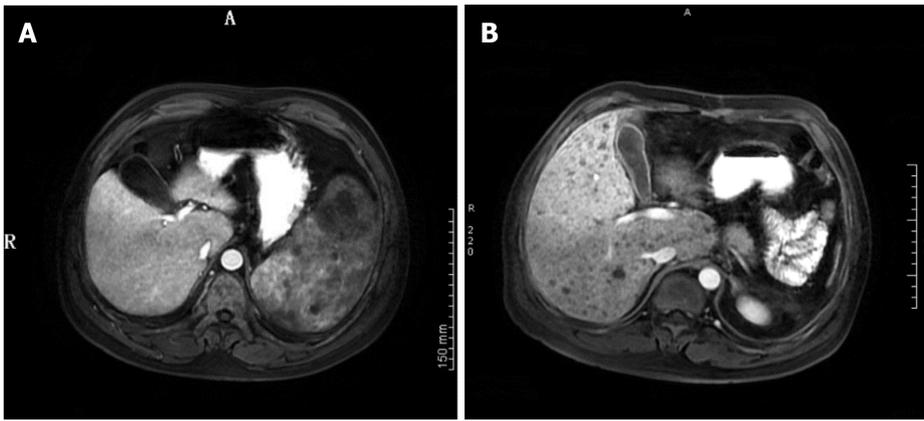


Figure 3 The excised spleen.

other minerals, insects and saliva, and HHV8 infection. Ensoli *et al*[13] indicated that KS is the result of the complex interplay between HHV8 and immunologic, genetic and environmental factors. In our case, unfortunately, tumor cells were immunonegative for HHV8 and we were unable to test for HHV8 DNA in the blood sample. However, this does not indicate that the present case did not have HHV8 infection. In addition to HHV8, cyclin D1 is also an important molecular marker to distinguish KS from angiosarcoma[14].

The clinical manifestations of splenic KS vary, and mainly include anemia, abdominal discomfort (abdominal pain or distension), and metastasis. In two reported cases, the patients had abdominal pain, and the chief complaint in our case was abdominal distension. The first reported case developed metastasis in the liver, mesentery, pleura, and lymph nodes[9]. The second case developed liver and bone marrow metastases[11]. Four weeks after surgery, the present patient was found to have liver metastasis when he was readmitted to the hospital due to jaundice. We speculate that surgery promoted tumor metastasis. It is possible that if the patient receives chemotherapy after surgery, tumor metastasis may be delayed.

Selection of the management strategy for KS depends on the location and variant of KS[15]. Epidemic KS often regresses with highly active antiretroviral therapy[16,17] and potent antiretroviral medication can dramatically reduce the incidence of KS in HIV patients[18]. For visceral KS, surgery, radiotherapy and systemic chemotherapy are necessary. The patients with splenic KS underwent splenectomy and chemotherapy according to the reports[9,11]. In our case, the whole spleen was successfully excised, but failed to improve the patient's condition and accelerated liver



**Figure 4 Hematoxylin and eosin staining and immunohistochemistry.** A: Multinodular tumors with a clear boundary and composed of proliferative spindle cells with interstitial hemorrhage ( $\times 10$ ); B: Proliferating spindle cells divided by slit-like spaces containing red blood cells, which were sieve-like or beehive in shape, and eosinophilic hyaline bodies scattered in the cytoplasm or extracellular of spindle cells ( $\times 200$ ); C: Immunohistochemical staining was positive for CD31, revealing splenic Kaposi's sarcoma (KS) ( $\times 200$ ); D: Immunohistochemical staining was positive for CD34, revealing splenic KS ( $\times 200$ ); E: Immunohistochemical staining was positive for ERG ( $\times 200$ ); F: Immunohistochemical staining was positive for FLI-1 ( $\times 200$ ).

metastasis. Although biopsy can provide correct diagnosis, considering that splenic biopsy may increase the risk of tumor metastasis, we did not perform biopsy before surgery. The prognosis of splenic KS is very poor. Patients with widespread visceral involvement are poorly responsive to treatment[4]. The present patient only lived 3 mo from the time he felt sick. Up to date, we have insufficient knowledge about KS. A splenic tumor does not explain megaloblastic anemia; therefore, the diagnosis of KS should be established until the etiology is explained. Our patient should have received early appropriate treatment for splenic KS consisting of surgery, chemotherapy and radiotherapy.

## CONCLUSION

We report a rare splenic KS in an HIV-negative patient with atypical clinical manifestations. MRI suggested splenic lymphoma and splenectomy was performed. The pathologic diagnosis was KS.

## ACKNOWLEDGEMENTS

We are grateful to the patient and his family for providing a signed informed consent. We acknowledge the contribution by our study team members. We would like to thank Professor Gengyin Zhou from the Department of Pathology of Qilu Hospital and Qianfeshan Hospital for assistance in the pathological diagnosis.

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