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Contents

Thrice Monthly Volume 10 Number 9 March 26, 2022

REVIEW

- 2660 Role of metabolites derived from gut microbiota in inflammatory bowel disease
Zheng L, Wen XL, Duan SL

MINIREVIEWS

- 2678 Roles of Wnt/ β -catenin signaling pathway related microRNAs in esophageal cancer
Chu CY, Wang R, Liu XL
- 2687 Animal models applied to acute-on-chronic liver failure: Are new models required to understand the human condition?
Gama JFG, Cardoso LMDF, Lagrota-Candido JM, Alves LA

ORIGINAL ARTICLE

Case Control Study

- 2700 Associations between coagulation factor XII, coagulation factor XI, and stability of venous thromboembolism: A case-control study
Meng Y, Li Y, Ye YJ, Ma Q, Zhang JB, Qin H, Deng YY, Tian HY

Retrospective Cohort Study

- 2710 Nomogram to predict the risk of endoscopic removal failure with forceps/baskets for treating submandibular stones
Huang Y, Liang PS, Yang YC, Cai WX, Tao Q
- 2721 Association between anesthesia technique and complications after hip surgery in the elderly population
Guo LS, Wang LN, Xiao JB, Zhong M, Zhao GF

Retrospective Study

- 2733 Perforating and nonperforating indications in repeated surgeries for Crohn's disease
Shen WS, Huang XH, Liu RQ, Li CY, Li Y, Zhu WM
- 2743 Treatment of *Pneumocystis jirovecii* pneumonia in non-human immunodeficiency virus-infected patients using a combination of trimethoprim-sulfamethoxazole and caspofungin
Wu HH, Fang SY, Chen YX, Feng LF
- 2751 Acute kidney injury in traumatic brain injury intensive care unit patients
Huang ZY, Liu Y, Huang HF, Huang SH, Wang JX, Tian JF, Zeng WX, Lv RG, Jiang S, Gao JL, Gao Y, Yu XX
- 2764 Enucleation combined with guided bone regeneration in small and medium-sized odontogenic jaw cysts
Cao YT, Gu QH, Wang YW, Jiang Q

Clinical Trials Study

- 2773 Determination of the ED₉₅ of intranasal sufentanil combined with intranasal dexmedetomidine for moderate sedation during endoscopic ultrasonography

Zou Y, Li N, Shao LJZ, Liu FK, Xue FS, Tao X

Observational Study

- 2783 Overexpression of Ubiquilin4 is associated with poor prognosis in patients with cervical cancer

Wang LN, Huang KJ, Wang L, Cheng HY

Randomized Clinical Trial

- 2792 Peplau's interpersonal relationship theory combined with bladder function training on patients with prostate cancer

Yang XH, Wu LF, Yan XY, Zhou Y, Liu X

SYSTEMATIC REVIEWS

- 2801 Efficacy of bone grafts in jaw cystic lesions: A systematic review

Wang J, Yao QY, Zhu HY

CASE REPORT

- 2811 Short stature associated with a novel mutation in the aggrecan gene: A case report and literature review

Yin LP, Zheng HX, Zhu H

- 2818 Treatment with sorafenib plus camrelizumab after splenectomy for primary splenic angiosarcoma with liver metastasis: A case report and literature review

Pan D, Li TP, Xiong JH, Wang SB, Chen YX, Li JF, Xiao Q

- 2829 Sarcomatoid intrahepatic cholangiocarcinoma with good patient prognosis after treatment with Huaier granules following hepatectomy: A case report

Feng JY, Li XP, Wu ZY, Ying LP, Xin C, Dai ZZ, Shen Y, Wu YF

- 2836 Sequential occurrence of T790M mutation and small cell lung cancer transformation in EGFR-positive lung adenocarcinoma: A case report

Hong E, Chen XE, Mao J, Zhou JJ, Chen L, Xu JY, Tao W

- 2844 Early diagnosis of Gitelman syndrome in a young child: A case report

Wu CY, Tsai MH, Chen CC, Kao CH

- 2851 Congenital intestinal malrotation with gastric wall defects causing extensive gut necrosis and short gut syndrome: A case report

Wang Y, Gu Y, Ma D, Guo WX, Zhang YF

- 2858 Delusional parasitosis as premotor symptom of parkinson's disease: A case report

Oh M, Kim JW, Lee SM

- 2864** Laninamivir-induced ischemic enterocolitis: A case report
Suzuki C, Kenzaka T
- 2871** Intramural pregnancy after *in vitro* fertilization and embryo transfer: A case report
Xie QJ, Li X, Ni DY, Ji H, Zhao C, Ling XF
- 2878** Bilateral ureteral reimplantation in a patient with an intraperitoneal ectopic bipenis: A case report
Jia YT, Shi BL, Zhang J, Li YY, Zhu J
- 2883** Lumbar disc sequestration mimicking a tumor: Report of four cases and a literature review
Li ST, Zhang T, Shi XW, Liu H, Yang CW, Zhen P, Li SK
- 2895** Parasitic leiomyoma in the trocar site after laparoscopic myomectomy: A case report
Roh CK, Kwon HJ, Jung MJ
- 2901** Giant nontraumatic myositis ossificans in a child: A case report
Xia AN, Wang JS
- 2908** Paradoxical carbon dioxide embolism during laparoscopic hepatectomy without intracardiac shunt: A case report
Jeon S, Hong JM, Lee HJ, Kim Y, Kang H, Hwang BY, Lee D, Jung YH
- 2916** Local hyperthermia combined with chemotherapy for the treatment of multiple recurrences of undifferentiated pleomorphic sarcoma: A case report
Zhou YT, Wang RY, Zhang Y, Li DY, Yu J
- 2923** Acute coronary artery stent thrombosis caused by a spasm: A case report
Meng LP, Wang P, Peng F
- 2931** Turner syndrome with primary myelofibrosis, cirrhosis and ovarian cystic mass: A case report
Xu LW, Su YZ, Tao HF
- 2938** Esophageal myoepithelial carcinoma: Four case reports
Lu H, Zhao HP, Liu YY, Yu J, Wang R, Gao JB
- 2948** Ipsilateral hemifacial microsomia with dextrocardia and pulmonary hypoplasia: A case report
Guo R, Chang SH, Wang BQ, Zhang QG
- 2954** Upper gastrointestinal bleeding from a Mallory-Weiss tear associated with transesophageal echocardiography during successful cardiopulmonary resuscitation: A case report
Tang MM, Fang DF, Liu B
- 2961** Malignant struma ovarii with papillary carcinoma combined with retroperitoneal lymph node metastasis: A case report
Xiao W, Zhou JR, Chen D

- 2969** Occult colon cancer with sepsis as the primary manifestation identified by bone marrow puncture: A case report

Wang HJ, Zhou CJ

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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.

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Treatment with sorafenib plus camrelizumab after splenectomy for primary splenic angiosarcoma with liver metastasis: A case report and literature review

Dan Pan, Tai-Ping Li, Jian-Hui Xiong, Shu-Bo Wang, Yao-Xu Chen, Jian-Feng Li, Qi Xiao

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Abstract

BACKGROUND

Primary splenic angiosarcoma (PSA) is an extremely rare and aggressive mesenchymal malignancy with high metastatic potential and a poor prognosis. There are no established treatment guidelines for PSA, even for adjuvant therapy. This rare case may provide a reliable therapeutic regime for a better prognosis.

CASE SUMMARY

A 49-year-old female who complained of right-upper quadrant abdominal pain was diagnosed as having PSA with splenic rupture and liver metastasis. After splenectomy and liver tumor resection, she received sorafenib and camrelizumab therapy. After 15 mo of follow-up, she is in good condition, without recurrence or any identified metastasis.

CONCLUSION

Immunotherapy combined with targeted therapy could be a potential option for the adjuvant therapy of PSA.

Key Words: Primary splenic angiosarcoma; Immunotherapy; Targeted therapy; Prognosis; Case report

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Core Tip: Splenectomy is the preferred treatment for primary splenic angiosarcoma (PSA); however, PSA patients may experience a good or poor prognosis after splenectomy. Although the prognosis in patients with liver metastases or rupture of the spleen is extremely poor, immunotherapy or targeted therapy has not been attempted after surgery. This case is the first report of a PSA patient with liver metastasis and splenic rupture, receiving sorafenib plus camrelizumab as adjuvant therapy. After 15 mo of follow-up, the patient is in good condition without recurrence or identified metastasis. Apart from chemotherapy and radiotherapy, targeted therapies and immunotherapy may also be an option for adjuvant therapy.

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INTRODUCTION

Primary splenic angiosarcoma (PSA), a subtype of soft tissue sarcoma (STS) with an annual incidence of 0.14-0.25 per million[1], is an extremely rare and aggressive mesenchymal malignancy with high metastatic potential and a poor prognosis. Upon histopathological examination, abnormal, pleomorphic, malignant patterns of endothelial cells, which can be rounded, polygonal, or fusiform (the hallmark of angiosarcoma or mitotic bodies), are commonly observed[2]. The typically expressed endothelial markers include von Willebrand factor, CD34, CD31, Ulex europaeus agglutinin-1, and vascular endothelial growth factor (VEGF)[2].

PSA patients with liver metastasis have a poor prognosis, especially when the spleen ruptures. Although immunotherapy and targeted therapy are widely used in other tumors, they are rarely considered in the treatment of PSA. In several cases, immunotherapy or targeted therapy is very effective in patients with angiosarcoma[3-5]. We believe that this form of therapy may have great potential in the treatment of PSA.

CASE PRESENTATION

Chief complaints

The patient developed pain in the right upper quadrant of the abdomen for one month, which exacerbated within a day, along with significant weight loss (Figure 1).

History of present illness

On 16 February 2020, a 49-year-old female with the complaint described above was brought to our emergency department. The patient did not report any abdominal trauma but complained of significant weight loss. According to the computed tomography (CT) scan on 16 February 2020 and other examinations (Figure 2), the diagnosis was non-traumatic splenic rupture and hemorrhage, with the suspected presence of liver cancer. Splenic artery embolization was suggested, which was refused by the patient and her family. Two days later, her abdominal pain was worse, after which, she and her family finally agreed to undergo splenic artery embolization (Figure 3). The patient recovered uneventfully and was discharged nine days later. One month later, she was admitted to our hospital again due to abdominal pain. According to the contrast-enhanced CT performed on 6 April 2020, the preliminary diagnosis was splenic angiosarcoma with peripheral and intrahepatic metastasis (Figure 4).

History of past illness

Surgical history included total thyroidectomy two years ago, followed by I131 therapy, hysteromyomectomy performed ten years ago, and pancreatic stone removal. The patient did not present a history of hepatitis or diabetes mellitus.

Personal and family history

There was no notable family medical history, such as cancer.

Physical examination

The patient's temperature was 36.6 °C, and blood pressure was 92/62 mmHg. Her heart rate was 101 bpm, and her respiratory rate was 20 breaths/min. Physical examination revealed a tender abdominal

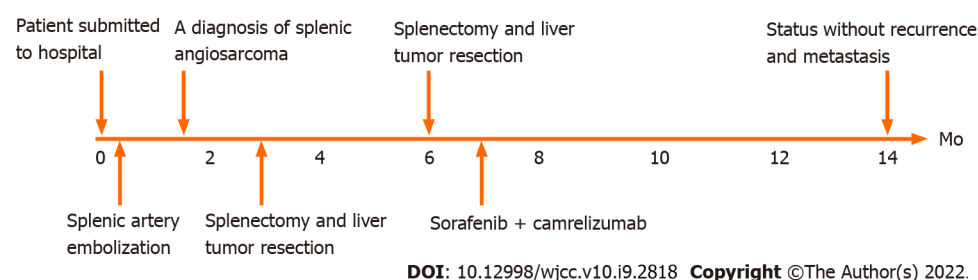


Figure 1 Treatment course.

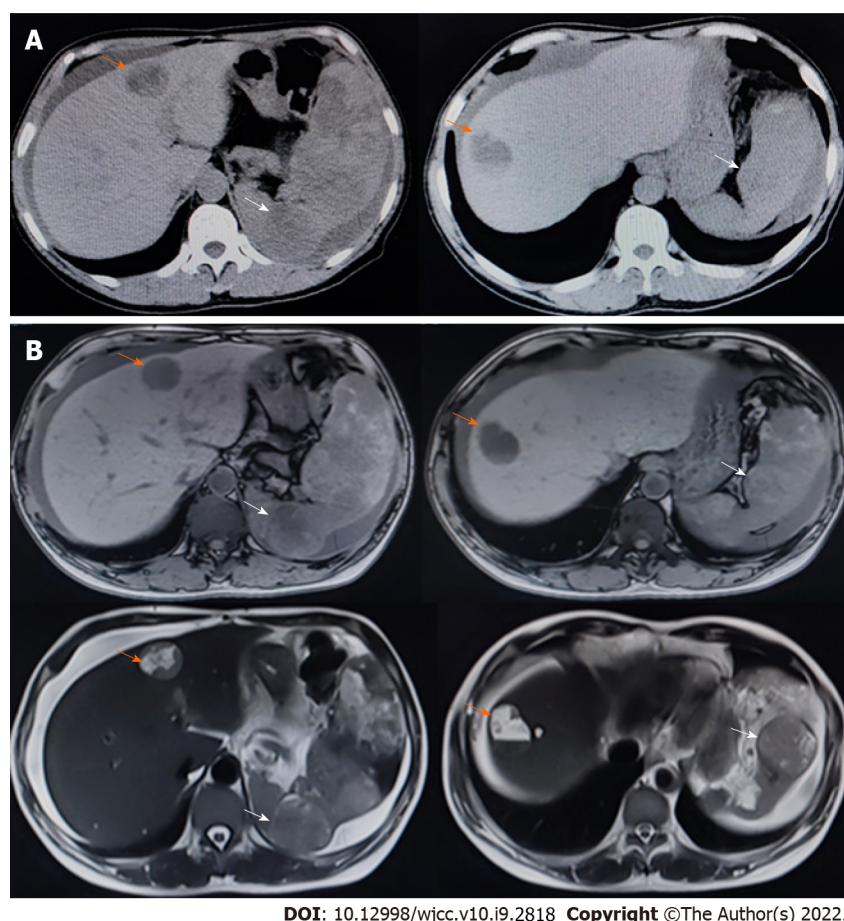


Figure 2 Computed tomography and magnetic resonance imaging performed soon after the first hospitalization. Scale bar: 10 cm. A: We observed multiple masses in the liver and spleen on computed tomography; B: We observed multiple masses in the liver and spleen on magnetic resonance imaging. Orange arrows indicate masses in the liver, while the white arrows indicate masses in the spleen.

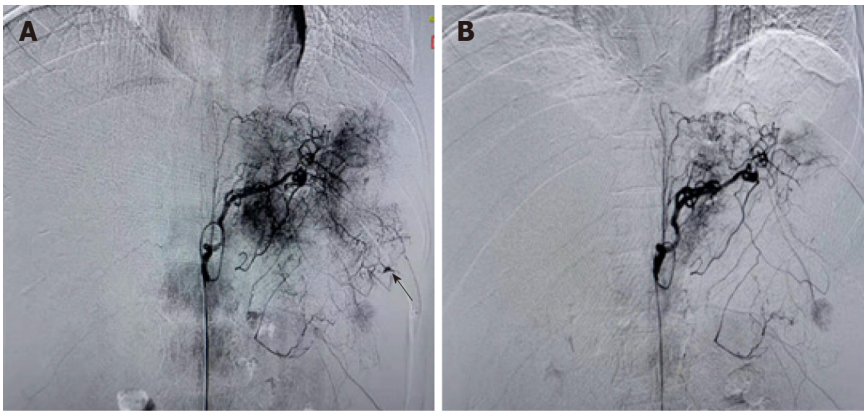
mass in the left upper abdomen and distension of the abdomen. Deep palpation showed diffuse pain, although without clinical signs of peritonitis.

Laboratory examinations

Liver function tests, hematology parameters, as well as tumor markers such as α -fetoprotein (AFP), carbohydrate antigen 199, carcinoembryonic antigen, and chromogranin A, were all normal, except for a white blood cell count of $13.54 \times 10^9/L$ [normal range: $(4.0-10.0) \times 10^9/L$] and hemoglobin concentration of 177 g/L (normal range: 120-160 g/L), which were beyond the normal range. The D-dimer level of 35.65 mg/L (normal range: 0.0-0.55 mg/L) indicated high coagulation function, while the remaining parameters were normal. Electrolyte levels, electrocardiogram, and chest X-ray were normal.

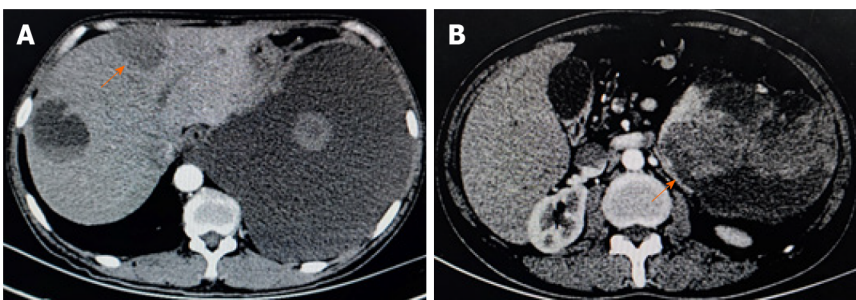
Imaging examinations

Multiple masses were observed in the liver and spleen on abdominal CT and magnetic resonance imaging on 16 February 2020 (Figure 2). Contrast-enhanced CT on 6 April 2020 revealed multiple round shadows of low-density in the spleen and liver (Figure 4).



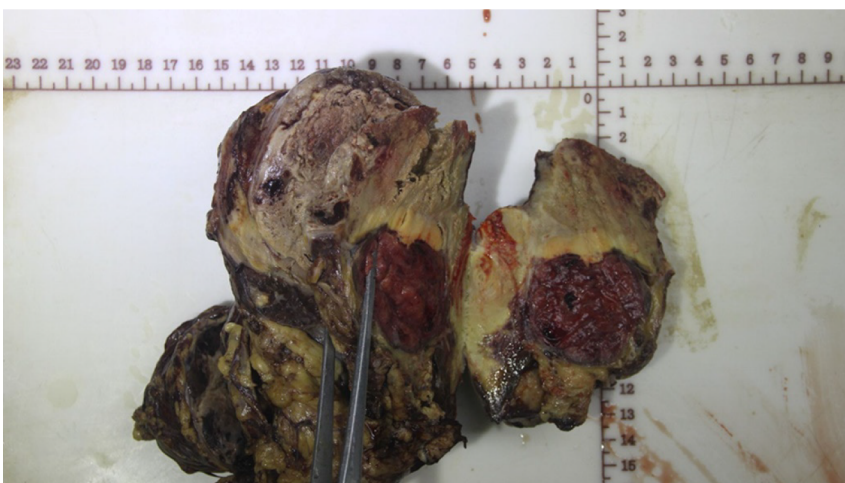
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Figure 3 Digital subtraction angiography revealed the condition of the spleen before and after artery embolization. A: Imaging before splenic artery embolization. The black arrow indicates bleeding points; B: Imaging after splenic artery embolization.



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Figure 4 Plain and enhanced computed tomography revealed multiple round shadows of low density in the spleen and liver. A: Computed tomography scan revealed that, in the left and right liver parenchyma, circular hypodensity reduction was observed with uneven density. On enhanced scan, the solid components showed slight enhancement, and no enhancement of hypodensity was observed. Massive pleural effusion; B: The spleen was enlarged, and multiple abnormal cystic solid density shadows were observed in and around the spleen. Enhanced scanning showed mild enhancement and partial fusion. The orange arrows indicate circular hypo-density regions.



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Figure 5 The resected tumor tissue. Specimen cross-sectioning revealed a demarcated yellowish-white lesion accompanied by hemorrhage and necrosis, which was 4 cm × 6 cm in size.

FINAL DIAGNOSIS

The patient was diagnosed as having PSA with a liver tumor.

TREATMENT

Splenectomy and liver tumor resection were performed not only to perform radical excision of the tumor but also for histopathological diagnosis. Intraoperatively, we observed a large scleroid tumor mass of 4 cm × 6 cm in size in the upper pole of the spleen. A cross-section of the specimen revealed a demarcated yellowish-white lesion accompanied by hemorrhage and necrosis (Figure 5). The intraoperative observation of neoplasms was in line with the imaging findings. Histopathological biopsy and next-generation sequencing (NGS) were carried out (733 gene panel genetic testing provided by 3D Medicines Inc.). The histopathology revealed that the tumor cells were arranged in sheets, fissures, or papillae, with the cytoplasm in fusiform, oval, or irregular nuclei, and mitosis was easily seen. Immunohistochemical examination revealed that the tumor cells were positive for CD31, S-100 and Ki-67 (positive rate of 60%), and negative for CD34 (Figure 6). The final pathological diagnosis was hepatic angiosarcoma and splenic angiosarcoma. From imaging, it was observed that the splenic tumors were large and numerous, while the liver tumors were relatively small and uniform in size, which was consistent with the characteristics of metastatic liver cancer. In addition, our patient had no history of hepatitis, preoperative AFP was normal, CT examination showed no obvious characteristics of primary liver cancer, and blood from the spleen returned to the liver which is the way the tumor metastasizes, and further supported the diagnosis of spleen tumor metastasized to the liver. Although the primary site was indistinguishable pathologically, the diagnosis of primary splenic malignancy with hepatic metastasis was suggested based on the characteristics of imaging and blood metastasis. The NGS revealed somatic mutations in the PDGFRA (gene amplification), KIT (gene amplification), KDR (VEGFR2) (gene amplification), and TP53 (c.376-4_380del, mutation abundance: 65.31%), while immunohistochemistry (IHC) showed the expression of programmed death ligand-1 (PD-L1) (Figure 7).

One month after the operation, the patient was given an intravenous infusion of sorafenib (400 mg) twice daily and camrelizumab (200 mg) in a 21-d cycle. After three weeks of combination therapy, she developed hand-foot syndrome and was unable to walk, after which, sorafenib was reduced to 600 mg daily.

OUTCOME AND FOLLOW-UP

Three and a half months after the operation, no metastasis was observed on single-photon emission tomography-CT (Figure 8). After 15 mo of frequent follow-up, we happily report that the patient is in good condition without any recurrence or metastasis. Also, the side effects of sorafenib and camrelizumab were tolerable. The patient believed that the treatment had little impact on her quality of life.

DISCUSSION

Since the first case of PSA reported in 1879, only about 200 cases had been reported in the literature worldwide[1]. We summarized the characteristics of PSA from case reports in the last ten years (Table 1). Unlike hepatic angiosarcoma, we observed no correlation between splenic angiosarcoma and risk factors such as thorium dioxide, vinyl chloride, or arsenic[6]. The symptoms of PSA are nonspecific, and although it may occur at any age, it is commonly observed in females aged over 60 years old[6].

Due to its low incidence, there are no recommended standardized treatments, guidelines, or even a consensus. Splenectomy is the preferred treatment for splenic angiosarcoma[2]. According to a retrospective analysis by Li *et al*[7], the mean overall survival (OS) for PSA patients was less than 8.1 mo after diagnosis. Despite the long survival of some patients, the prognosis for patients with liver metastases or splenic rupture after splenectomy was generally poor (Table 1).

Almost 30% of the patients experienced splenic rupture associated with hemoperitoneum[6]. When the spleen ruptures, the prognosis for PSA patients is poor. Kornmann *et al*[8] reported that a 69-year-old male with PSA who had splenic rupture associated with hemoperitoneum died within one month after surgery (Table 1). Abbott *et al*[9] reported that the mean OS among patients who had splenectomy after spontaneous splenic rupture was 4.4 mo. If splenectomy was performed before rupture, the mean OS increased to 14.4 mo.

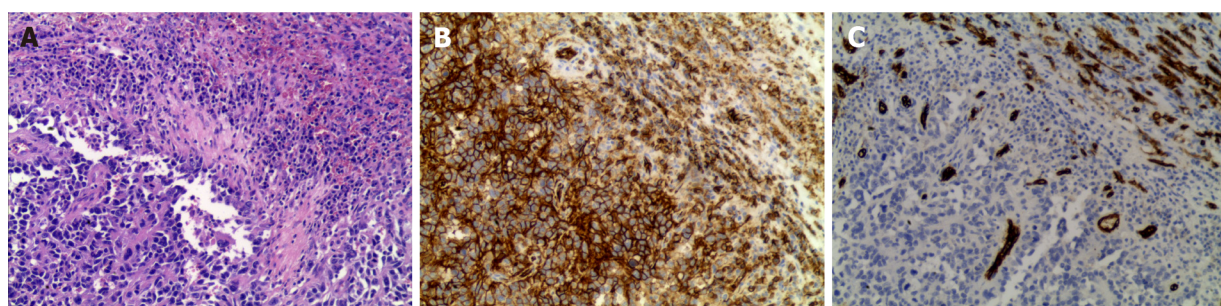
PSA is also a highly aggressive tumor, which can not only spread *via* direct invasion but also metastasize *via* the hematal and lymphatic system[2,7]. The most common site involved is the liver, followed by the lungs, bones, lymph nodes, and ovaries[7]. Metastasis in the liver, peritoneum, bone, and lung may occur in PSA patients, and approximately half of such patients died within one year after diagnosis (Table 1). The prognosis of PSA is abysmal, particularly in patients with metastases[6]. Splenic rupture and liver metastases may serve as high-risk factors for PSA. Thus, the patients with either may need more measures for prolonged survival. Adjuvant therapy may be a potential treatment strategy after surgery to reduce the likelihood of recurrence or spread to other sites.

Table 1 Review of case reports published in the last 10 years (2011 to 2021)

Ref.	Report year	Age	Sex	Etiology	Clinical manifestations	Extrasplenic metastasis	Treatment	OS
[23]	2019	80	Female	None	Progressive abdominal pain, weight loss, fatigue, and pallor	Liver and peritoneum metastases	Splenectomy	6 mo
[24]	2019	41	Male	None	Headaches, nausea, vomiting persisting	None	Splenectomy	More than 4 yr
[25]	2019	83	Female	None	Splenomegaly, acute abdominal pain	None	Splenectomy	Less than 6 mo
[26]	2018	55	Female	None	Abdominal pain and hepatosplenomegaly	Liver metastases	Palliative care	NA
[27]	2018	56	Female	None	A spleen nodule was incidentally diagnosed	None	Splenectomy	More than 18 mo
[28]	2019	42	Male	None	Severe abdominal pain	Liver and lungs metastases	Splenectomy, adjuvant chemoradiotherapy and radiation therapy	23 mo
[29]	2018	35	Female	None	Routine physical examination	Bone and abdominal cavity metastases	Laparoscopic-assisted splenectomy, radiation therapy	57 mo
[30]	2017	49	Male	None	Left upper quadrant abdominal pain, fatigue, insomnia, severe weight loss, nocturnal sweating, and impotence	Peritoneal metastases	Splenectomy and nodal sampling	11 mo
[31]	2016	55	Female	Rudimentary vascular channels	Severe tenderness in the left upper abdominal quadrant	Liver and lungs metastases	Splenectomy and adjuvant chemotherapy (paclitaxel). Second line chemotherapy (ifosfamide/ doxorubicin)	NA
[20]	2016	57	Female	None	Gastrointestinal discomfort	Distant metastases	Splenectomy and adjuvant chemotherapy (paclitaxel chemotherapy and pazopanib)	3 yr
[5]	2015	69	Male	Rupture of the spleen	Fatigue, reduced fitness, shortness of breath on exercise, anorexia, and abdominal and back pain, weight loss	Peritoneal tumor deposits	Splenectomy	Less than 1 mo
[19]	2015	45	Female	None	Left upper quadrant and left flank pain	Liver metastases	Splenectomy and adjuvant chemotherapy (paclitaxel)	5 mo
[32]	2014	38	Female	None	Upper abdominal pain and fatigue for about 1 wk	Liver metastasis	Splenectomy	Passed away 3 yr after diagnosis, that is 3 mo after recurrence and liver metastasis
[33]	2015	38	Male	None	Multiple liver masses	Liver metastases	Paclitaxel monotherapy	Less than 3 mo
[34]	2013	54	Male	None	Nonspecific complaints of abdominal pain and loss of appetite	Liver metastases	Splenectomy	3 mo
[34]	2013	77	Female	None	Hematochezia and fatigue	None	Splenectomy	1 mo
[35]	2013	80	Female	None	Syncope, hypotension, vomiting	None	Splenectomy	More than 6 mo
[36]	2013	65	Male	None	Diffuse abdominal pain and distension	Liver metastases	Splenectomy	NA
[37]	2012	70	Male	Hodgkin lymphoma treated with radiotherapy and chemotherapy 30 yr ago	Abdominal pain upper quadrants and jaundice	None	Splenectomy	More than 6 mo

[38]	2012	73	Female	None	Asymptomatic	None	Splenectomy	NA
[38]	2012	23	Female	None	Left upper quadrant pain	None	Splenectomy	NA
[21]	2011	62	Male	None	Acute onset of abdominal pain	Liver metastases	Splenectomy, palliative chemotherapy with etoposide, ifosfamide and doxorubicin	8 mo
[39]	2010	48	Male	None	Left upper quadrant pain and early satiation	Multiple pleural metastases lesions	Splenectomy	NA
[40]	2010	48	Female	None	Weight loss general fatigue, and left hypochondrial pain	Liver metastases	Splenectomy, high-dose chemotherapy with autologous peripheral blood stem cell transplantation, hepatic lobectomy, radiofrequency ablations and administration of recombinant interleukin-2	67 mo
[22]	2010	69	Female	None	Pain in the left upper quadrant of the abdomen, weight loss, progressive shortness of breath	None	Paclitaxel as neoadjuvant therapy, splenectomy and adjuvant chemoradiotherapy	More than 14 mo
[41]	2010	76	Female	None	Left upper abdominal pain	Extensive metastases	Splenectomy	NA
[42]	2010	55	Female	Radiation treatment	NA	None	Splenectomy	More than 4 yr
[43]	2009	15	Male	None	Severe right-upper quadrant abdominal and epigastric pain	Liver, bone, lymph nodes, adrenal metastasis	Splenectomy with hilar lymph node excision, chemotherapy	26.5 mo

NA: Not available; OS: Overall survival.

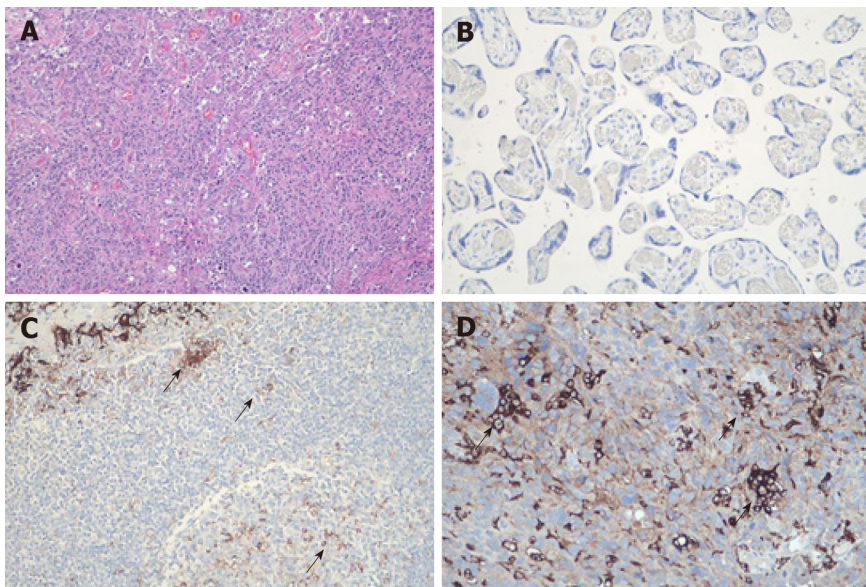


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Figure 6 Hematoxylin and eosin staining and immunohistochemistry of the specimen. A: Hematoxylin and eosin staining showed the morphology of the tumor cells; B: Immunohistochemistry (IHC) revealed that the patient was positive for CD31, which was characteristic of tumor cells derived from vascular endothelium; C: The results of IHC revealed that the patient was negative for CD34. Shown at × 100 original magnification.

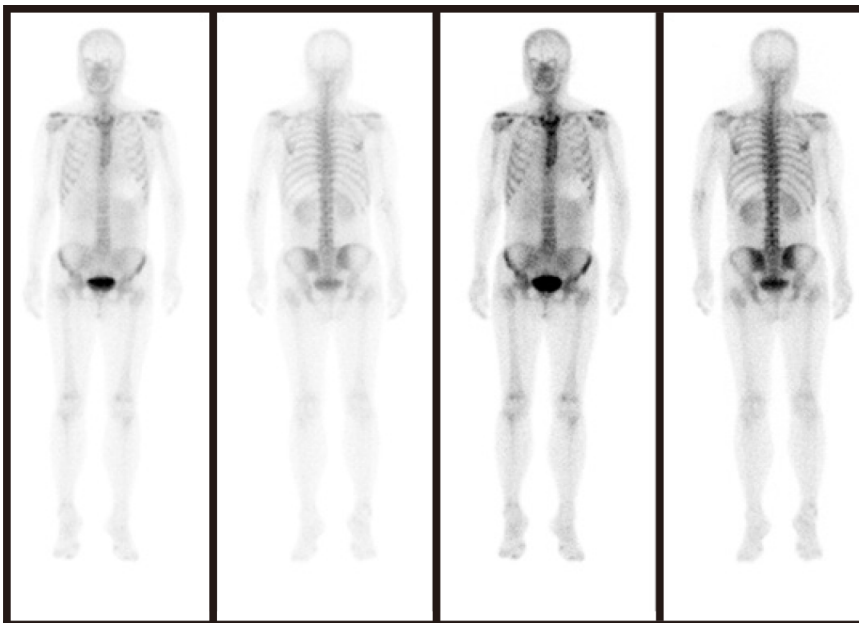
Postoperative radiotherapy and adjuvant chemotherapy provide a promising choice for sarcomas of the extremities, although the efficacy for PSA was questionable as only a few patients have been enrolled in clinical trials[10,11]. For unresectable angiosarcoma, treatment with single-agent doxorubicin or paclitaxel is recommended based on the available evidence. However, there is no precise evidence for adjuvant chemotherapy[12]. Given its extensive use in other tumors, targeted therapy and immunotherapy may provide new options for adjuvant therapy against PSA.

Immunotherapy has become a significant option for the treatment of advanced cancer[13]. A retrospective analysis reported that a cohort of patients with unresectable sarcomas, who were treated with nivolumab combined with pazopanib, presented clinical benefit in 50% of the patients after at least four cycles[14]. This may be due to increased lymphocytic infiltration and PD-L1 expression in patients with sarcomas[15,16]. Hence, it is reasonable to perform immunotherapy. Studies have demonstrated that the activity of anti-PD-1 therapy in all types of STS is consistent[17]. Targeted therapies such as bevacizumab, sorafenib, and pazopanib have shown great efficacy in the treatment of advanced STS [13]. A retrospective analysis reported that sorafenib had activity against angiosarcoma[4]. In a phase-III trial for metastatic STS, 372 patients with advanced STS whose disease had progressed despite at least one line of chemotherapy were randomly assigned to either the pazopanib arm or a placebo arm. The



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Figure 7 The level of programmed death ligand-1 protein was detected using immunohistochemistry by the Dako programmed death ligand-1 immunohistochemistry 22c3 PharmDx kit. A: Hematoxylin and eosin staining of the specimen, shown at $\times 100$ original magnification; B: Negative control for the test, shown at $\times 200$ original magnification; C: Positive control for the test, shown at $\times 200$ original magnification; D: Immunohistochemistry revealed that this patient was positive for programmed death ligand-1 (PD-L1) in the cytomembrane of tumor cells (trehalose-6-phosphate synthase = 20%, cervical pedicle screws = 22), shown at $\times 200$ original magnification. The black arrows indicate cells with high PD-L1 expression.



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Figure 8 Single-photon emission tomography combined with computed tomography showed no signs of metastasis.

difference in progression-free survival (PFS) between the two arms was statistically significant, with a median PFS of 4.6 mo for pazopanib, compared to just 1.6 mo for placebo[18]. Pazopanib has received approval for the treatment of certain STSs in 2012[14]. In an open-label, multicenter, phase-II study, bevacizumab showed great potential in the treatment of angiosarcoma in 30 patients, whose disease was deemed surgically unresectable, with a mean survival of 26 wk. Four patients showed partial response, and half of the patients had stable disease[3]. Notably, all the reported targeted drugs were anti-angiogenic agents, possibly because angiogenesis or VEGF plays an important role in carcinosarcoma [4]. Moreover, a previous study suggested that combining immunotherapy and targeted therapies may have complementary roles in cancer treatment and that combinatorial therapy might be synergistic[5]. That study gave us great inspiration and confidence.

In the present case, the patient was treated with sorafenib combined with camrelizumab as adjuvant therapy, instead of chemotherapy, because we found that the prognoses of patients on chemotherapy were stratified, that is, chemotherapy worked well in some patients and presented unsatisfactory clinical outcomes in others[19-22]. Therefore, we performed NGS and IHC for PD-L1. The patient may be sensitive to sorafenib (targets BRAF, CRAF, KIT, FLT3, RET, VEGFR-1/-2/-3, and PDGFR α/β) and PD-L1 inhibitor, as deduced from the mechanism of these drugs and thus received periodic treatment. This was a meaningful attempt of off label use. Now, after 15 mo of follow-up, there is no progress or recurrence of the disease, and the prognosis is good compared to most PSA patients with rupture of the spleen or liver metastases without adjuvant therapy among cases published in the last 10 years (Table 1).

CONCLUSION

Our data provide excellent evidence that adjuvant targeted therapy and immunotherapy may improve the prognosis in patients with PSA. Meanwhile, more evidence-based medical data and the specific cycle of targeted immunotherapy need to be further explored. NGS can be considered for patients suffering from angiosarcoma, which may suggest possible clinical treatment.

FOOTNOTES

Author contributions: Xiao Q was the physician in charge, confirmed the diagnosis and contributed the details on the histologic diagnosis; Pan D contributed to the case report design and manuscript drafting, and provided the histologic image; Wang SB and Chen YX were responsible for NGS testing; Li TP and Xiong JH reviewed the manuscript; Xiao Q and Li JF were the principal authors of the paper, had full access to all data, and are the guarantors; and all authors read and confirmed the final version of this article.

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