

World Journal of *Gastrointestinal Surgery*

World J Gastrointest Surg 2024 April 27; 16(4): 974-1217



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Monthly Volume 16 Number 4 April 27, 2024

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ABOUT COVER

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The primary aim of *World Journal of Gastrointestinal Surgery* (WJGS, *World J Gastrointest Surg*) is to provide scholars and readers from various fields of gastrointestinal surgery with a platform to publish high-quality basic and clinical research articles and communicate their research findings online.

WJGS mainly publishes articles reporting research results and findings obtained in the field of gastrointestinal surgery and covering a wide range of topics including biliary tract surgical procedures, biliopancreatic diversion, colectomy, esophagectomy, esophagostomy, pancreas transplantation, and pancreatectomy, *etc.*

INDEXING/ABSTRACTING

The WJGS is now abstracted and indexed in Science Citation Index Expanded (SCIE, also known as SciSearch®), Current Contents/Clinical Medicine, Journal Citation Reports/Science Edition, PubMed, PubMed Central, Reference Citation Analysis, China Science and Technology Journal Database, and Superstar Journals Database. The 2023 Edition of Journal Citation Reports® cites the 2022 impact factor (IF) for WJGS as 2.0; IF without journal self cites: 1.9; 5-year IF: 2.2; Journal Citation Indicator: 0.52; Ranking: 113 among 212 journals in surgery; Quartile category: Q3; Ranking: 81 among 93 journals in gastroenterology and hepatology; and Quartile category: Q4.

RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: Zi-Hang Xu, Production Department Director: Xiang Li, Cover Editor: Jia-Ru Fan.

NAME OF JOURNAL

World Journal of Gastrointestinal Surgery

ISSN

ISSN 1948-9366 (online)

LAUNCH DATE

November 30, 2009

FREQUENCY

Monthly

EDITORS-IN-CHIEF

Peter Schemmer

EDITORIAL BOARD MEMBERS

<https://www.wjgnet.com/1948-9366/editorialboard.htm>

PUBLICATION DATE

April 27, 2024

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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>



Successful splenic artery embolization in a patient with Behçet's syndrome-associated splenic rupture: A case report

Guang-Zhao Zhu, Dong-Hua Ji

Specialty type: Immunology

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
Grade D (Fair): D
Grade E (Poor): 0

P-Reviewer: Liu P, China; Shonaka T, Japan

Received: October 23, 2023

Peer-review started: October 23, 2023

First decision: January 6, 2024

Revised: February 2, 2024

Accepted: March 6, 2024

Article in press: March 6, 2024

Published online: April 27, 2024



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Abstract

BACKGROUND

Splenic rupture associated with Behçet's syndrome (BS) is extremely rare, and there is no consensus on its management. In this case report, a patient with BS-associated splenic rupture was successfully treated with splenic artery embolization (SAE) and had a good prognosis after the intervention.

CASE SUMMARY

The patient was admitted for pain in the left upper abdominal quadrant. He was diagnosed with splenic rupture. Multiple oral and genital aphthous ulcers were observed, and acne scars were found on his back. He had a 2-year history of BS diagnosis, with symptoms of oral and genital ulcers. At that time, he was treated with oral corticosteroids for 1 month, but the symptoms did not alleviate. He underwent SAE to treat the rupture. On the first day after SAE, the patient reported a complete resolution of abdominal pain and was discharged 5 d later. Three months after the intervention, a computed tomography examination showed that the splenic hematoma had formed a stable cystic effusion, suggesting a good prognosis.

CONCLUSION

SAE might be a good choice for BS-associated splenic rupture based on good surgical practice and material selection.

Key Words: Splenic artery embolization; Behçet's syndrome; Splenic rupture; Case report

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Core Tip: This study presents a case of splenic rupture associated with Behçet's syndrome (BS). BS was confirmed using the International Criteria for Behçet's Disease. The patient opted for splenic artery embolization (SAE) over splenectomy, resulting in successful hematoma control. The patient's postoperative recovery was favorable, with no complications, suggesting the effectiveness of SAE in BS-associated splenic rupture management. These findings emphasize the significance of early BS diagnosis and the potential benefits of SAE in selected cases, contributing to the understanding and treatment of this rare but critical manifestation of BS.

Citation: Zhu GZ, Ji DH. Successful splenic artery embolization in a patient with Behçet's syndrome-associated splenic rupture: A case report. *World J Gastrointest Surg* 2024; 16(4): 1184-1188

URL: <https://www.wjgnet.com/1948-9366/full/v16/i4/1184.htm>

DOI: <https://dx.doi.org/10.4240/wjgs.v16.i4.1184>

INTRODUCTION

Behçet's syndrome (BS) is an autoimmune disease associated with numerous systemic manifestations, including skin, arthritis, ocular, and vascular lesions[1]. Vascular lesions are characterized by the involvement of vessels of all types (veins and arteries) and sizes. Arterial involvement develops in only 1%-7% of the patients with BS and is mainly observed in isolated cases. Thus, arterial involvement is significantly less frequent than venous involvement and is extremely rare in clinical practice. Nevertheless, it has a high mortality rate. Among patients with arterial involvement, arterial rupture (e.g., BS-associated splenic rupture) is the most common cause of death in patients with BS[2]. Since BS-associated splenic rupture is extremely rare, no consensus exists on its treatment.

CASE PRESENTATION

Chief complaints

A 58-year-old man presented to the emergency department in September 2021, complaining of pain in the abdominal upper left quadrant after a brief loss of consciousness during a bowel movement 24 h earlier.

History of present illness

After the admission examination, the patient consented to partial splenic artery embolization (SAE) to control subcapsular hematoma rather than splenectomy, and SAE was performed. After surgery, BS was treated with thiazolidine. There were no postoperative complications, such as recurrent splenic hemorrhage or abdominal or systemic infection. Finally, the patient was discharged from the hospital in a stable condition.

History of past illness

The patient had a 2-year history of hypertension and type 2 diabetes, both under control. He was diagnosed with BS 2 years ago with symptoms of oral and genital ulcers. At that time, he was treated with oral corticosteroids for 1 month, but the symptoms did not alleviate.

Personal and family history

The personal history of the patient was with hypertension and type 2 diabetes. The family history was not specified.

Physical examination

Multiple oral and genital aphthous ulcers were observed (Figure 1A-C). Acne scars were found on his back (Figure 1D). The patient denied any trauma or bouts of coughing.

Laboratory examinations

The low hemoglobin levels (63 g/L) supported internal bleeding. The C-reactive protein levels were elevated at 49.4 mg/L, suggesting significant systemic and/or vascular inflammation. Five days after surgery, the patient's hemoglobin levels were stable at 65 g/L without significant decline.

Imaging examinations

After admission, splenic arteriography did not show clear culprit blood vessels or lesions such as aneurysms or pseudoaneurysms (Figure 1E). Computed tomography (CT) showed splenic rupture with a subcapsular hematoma of the spleen (Figure 1F). Angiography was performed immediately after CT and during SAE and showed reduced swelling laterally to the spleen (Figure 1G).

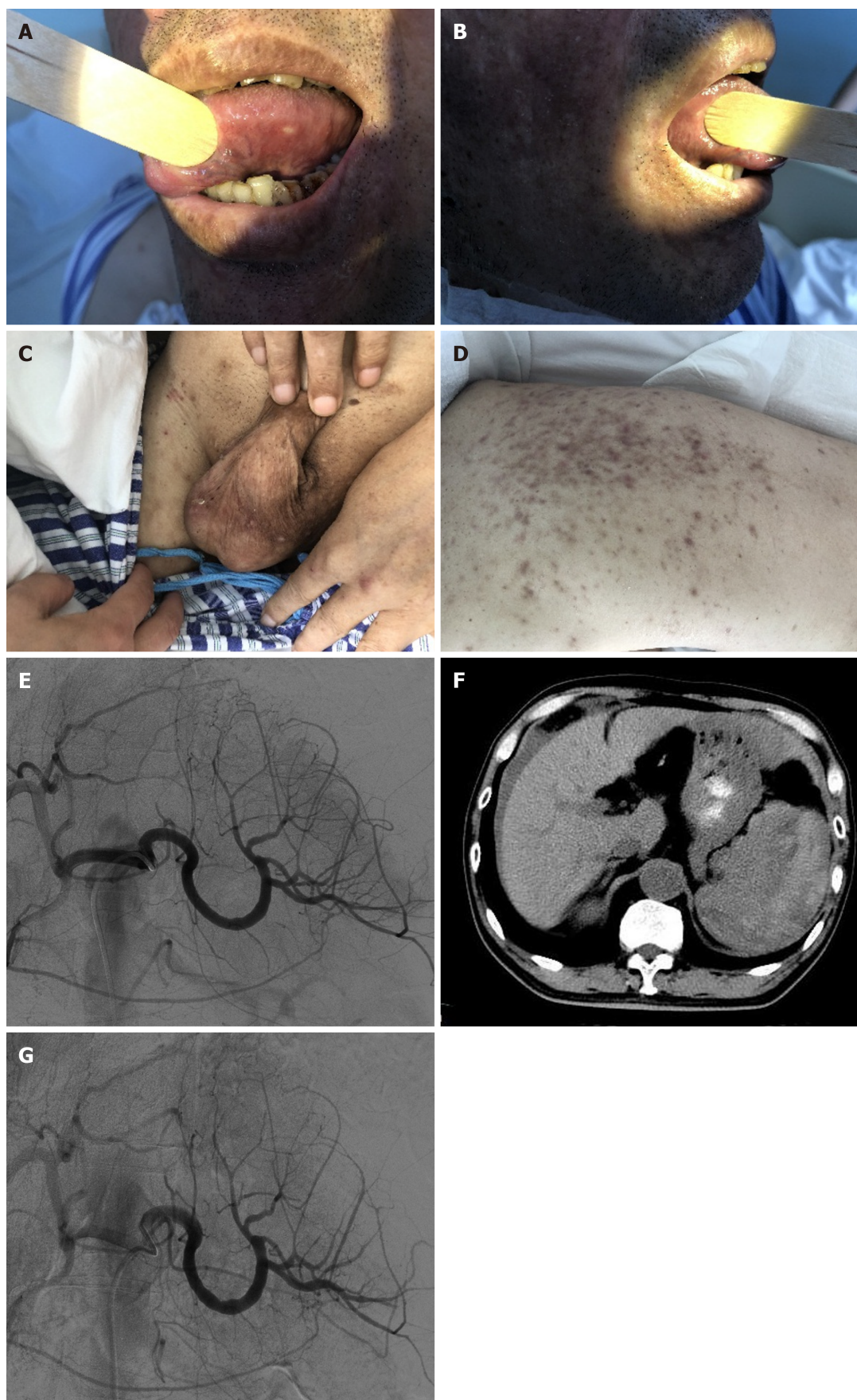


Figure 1 Physical and imaging examinations of the patient. A-D: Multiple oral and genital aphthous ulcers were observed, and acne scars were found in the index admission examination; E: The angiography before splenic artery embolization, splenic arteriography did not show clear culprit blood vessels or lesions such as aneurysms or pseudoaneurysms; F: Computed tomography showed the existence of splenic rupture with a subcapsular hematoma of the spleen; G: Postoperative angiography showed reduced swelling laterally to the spleen.

FINAL DIAGNOSIS

CT showed splenic rupture with a subcapsular hematoma of the spleen (Figure 1F).

TREATMENT

A 5F short sheath was placed in the right femoral artery for reverse puncture. An RH catheter was placed for splenic arteriography. No clear signs of contrast agent exudation, aneurysms, or false aneurysms were found in the splenic artery trunk and branches. The embolization was performed using a microcatheter (Progreat; Terumo, Tokyo, Japan) that supplied 900-1200- μ m microspheres (Embosphere; Merit Medical Systems, South Jordan, Utah, United States).

OUTCOME AND FOLLOW-UP

Angiography was performed immediately after CT and during SAE and showed reduced swelling laterally to the spleen (Figure 1G). Abdominal CT showed no hematoma hyperplasia. A follow-up CT examination 3 months later showed that the splenic hematoma had formed a stable cystic effusion, suggesting that the patient had a good prognosis after SAE.

DISCUSSION

The most commonly used surgical treatment for splenic rupture is splenectomy or partial splenectomy[3,4]. To date, there is only a single reported case of BS-associated splenic rupture with splenic artery aneurysm[5], in which the patient was treated with splenectomy, but the patient's follow-up was not mentioned. Furthermore, complications such as secondary infection can occur after splenectomy because the spleen is the largest peripheral immune organ in humans, participating in the body's immune response through the production of lymphocytes and monocytes. Hence, splenectomy will likely cause immune system malfunction, resulting in a weakened immune system.

Therefore, spleen preservation is important in maintaining proper immune and hormonal functions, especially in patients with BS. In this manner, SAE is safer for immune function than splenectomy[6]. Still, SAE can carry a risk of hemorrhage, reintervention, and conversion to splenectomy, but no such complications were observed in the patient reported here. It suggests that good surgical practice and material selection might be important factors in preventing adverse reactions. The take-home message of this case is that SAE might be a valuable treatment option for BS-associated splenic rupture, as suggested previously in non-BS cases[7,8].

It must be highlighted that there was no absolute proof that BS caused the splenic rupture in the case reported here. Still, splenic rupture was possibly caused by BS since all other possible causes of splenic rupture (*e.g.*, trauma and bouts of coughing) were excluded, but no pathological evidence supports it. On the other hand, splenic rupture has been reported in BS[5].

CONCLUSION

A patient with BS-associated splenic rupture was successfully treated with SAE. The postoperative prognosis was good. Although further clinical practice and more cases are needed to confirm the findings, it is suggested that good surgical practices, proper use of surgical materials, and standard medical therapy of SAE might be feasible for treating BS-associated splenic rupture.

FOOTNOTES

Author contributions: Zhu GZ and Ji DH carried out the studies, participated in collecting data, and drafted the manuscript. All authors read and approved the final manuscript.

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

Conflict-of-interest statement: The authors declare that they have no conflict of interest to disclose.

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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S-Editor: Zhang H

L-Editor: A

P-Editor: Xu ZH

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