Surg Today (2009) 39:733–737
DOI 10.1007/s00595-008-3911-7

SURGERY TODAY

Spontaneous Rupture of the Spleen Caused by a *Bacillus* Infection: Report of a Case

SHIGEAKI AOYAGI, TOMOKAZU KOSUGA, TOSHIRO OGATA, and MASAFUMI YASUNAGA

Department of Surgery, Kurume University School of Medicine, 67 Asahi-machi, Kurume 830-0011, Japan

Abstract
We report a case of spontaneous splenic rupture (SSR) caused by a *Bacillus* species (sp.) infection. A 36-year-old man on warfarin therapy since an aortic valve replacement at the age of 13 was admitted to our hospital with a 3-week history of a high fever. He had been asymptomatic until 4 months previously, when he suffered a cerebral embolism despite adequate oral anticoagulation. Abdominal computed tomography revealed splenic infarctions, which resulted in splenic rupture 2 days later. After embolization of the splenic artery, splenectomy was successfully performed. Pathologic examination revealed splenic infarction, resulting from septic emboli, with associated rupture of the splenic capsule, but no abscess was found. *Bacillus* sp. was isolated from cultures of arterial blood preoperatively, and the excised splenic specimens, postoperatively. In addition to rupture of the suppurating intrasplenic vessels with hematoma formation, the anticoagulant therapy possibly contributed to distension of the intrasplenic hematoma.

Key words *Bacillus* species · Splenic infarction · Splenic rupture · Anticoagulant therapy

Introduction

Spontaneous splenic rupture (SSR) is a rare event, which can occur in association with many conditions, including hematologic diseases, splenic infarcts, viral or bacterial infections, pancreatitis, rheumatologic diseases, pregnancy, and occasionally, anticoagulant therapy.[1] There are also sporadic reports of SSR caused by Niemann–Pick disease, an inherited metabolic disorder.[2] On the other hand, *Bacillus* species (sp.) rarely cause serious systemic infections because *Bacillus* bacteremias are usually eradicated easily by removal of an infected intravascular device and antibiotic therapy.[3] We report a case of SSR resulting from splenic infarction caused by a *Bacillus* infection in a patient on anticoagulant therapy for a mechanical prosthetic valve.

Reprint requests to: S. Aoyagi
Received: March 27, 2008 / Accepted: July 28, 2008

Case Report

A 36-year-old man was admitted to our hospital for investigation of a high fever of 3 weeks' duration. The patient had undergone aortic valve replacement with a mechanical valve at the age of 13, after which a mild periprosthetic leak had been noted. He denied any history of intravenous drug abuse, trauma, or indwelling intravenous catheters. He was asymptomatic until 4 months before this admission, when he suffered sudden and severe headache. Computed tomography (CT) of the brain revealed a small infarct in the left occipital lobe and a cerebral embolism was diagnosed. At the time, the level of anticoagulation with warfarin was 3.04 on the International Normalized Ratio (INR) scale. On admission, a systolic ejection murmur and diastolic blowing murmur were heard over the fourth left sternal border, but he was hemodynamically stable without any symptoms or signs of heart failure. The liver and the spleen were not palpable and no neurological sequelae were found. The white blood cell count was 8100/mm^3, C-reactive protein was 13 mg/dl, and the INR was 3.38. Hematological studies, blood chemistry, liver function tests, amylase, and myocardial profiles were all within normal limits. Transthoracic echocardiography revealed normal leaflet movement of the prosthetic valve and a mild periprosthetic leak, but no vegetations or annular abscesses. Abdominal CT showed two low-attenuation areas, 1.5 × 2.0 cm and 1.0 × 1.2 cm in size, suggestive of infarctions, in the spleen (Fig. 1). Sudden epigastric

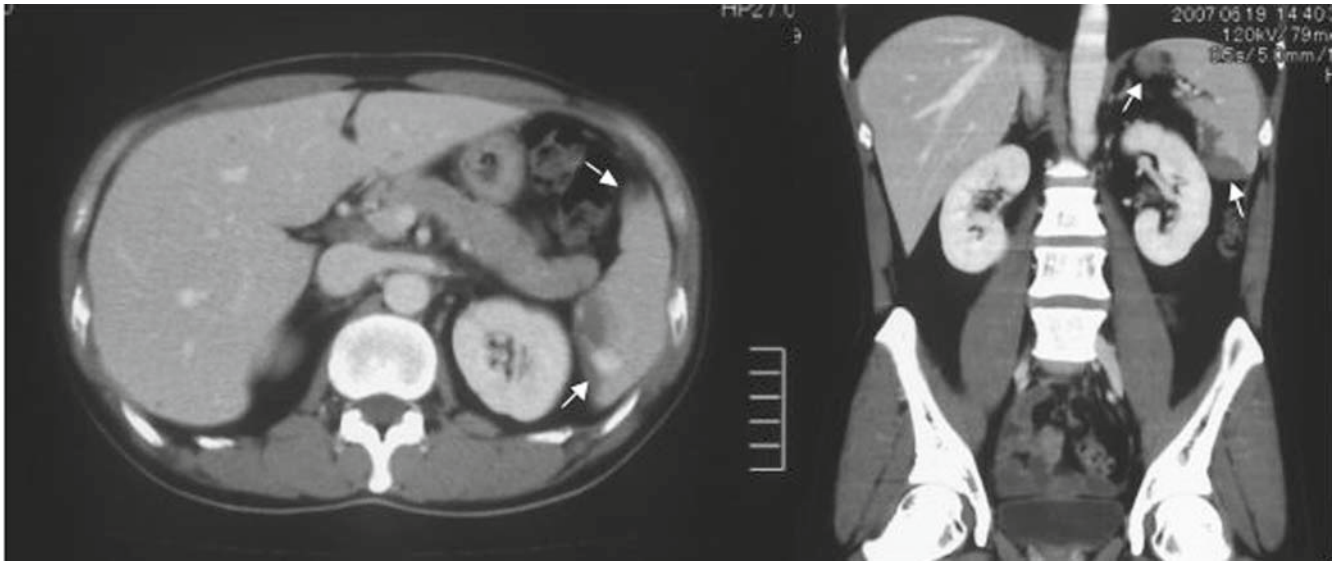

Fig. 1. Abdominal computed tomography on admission showed two low-attenuation areas, suggestive of infarctions, in the spleen (*arrows*)

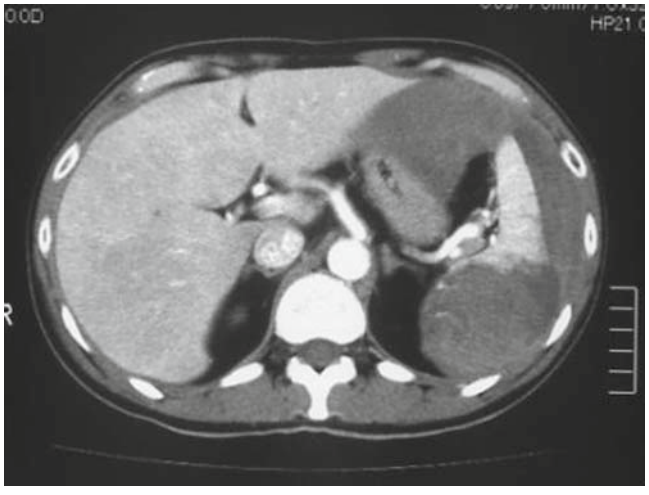

Fig. 2. Abdominal computed tomography performed emergently showed splenic infarction and massive hematoma around the spleen

pain developed 2 days later, with subsequent shock. Emergency CT showed abrupt enlargement of the low attenuation area and a massive fluid collection, consistent with a hematoma, around the spleen (Fig. 2). Selective splenic arteriography revealed the extravasation of contrast material from the spleen without aneurysms of the splenic artery (Fig. 3). We performed embolization of the splenic artery as a life-saving procedure. By that time, *Bacillus* sp. had been isolated from one of the blood cultures taken during the first 3 days after admission. Based on these findings, we diagnosed SSR resulting from the splenic infarction caused by a *Bacillus* sp. infection. Splenectomy was immediately performed because the hematoma was enlarging on subsequent CT scans and because of the necessity for permanent anticoagulant therapy.

At surgery, a massive hematoma was found around the spleen and splenectomy was carried out without any difficulties. Pathologic examination showed a normal-sized spleen containing infarctions with an intrasplenic hematoma. Microscopically, some of the infarctions were without inflammatory cellular (neutrophilic granulocytes and lymphocytes) infiltration, resulting from arterial embolization, whereas others were caused by arterial occlusion by septic emboli, and included neutrophilic granulocytes with surrounding inflammatory cellular infiltration and intrasplenic hemorrhage (Fig. 4). However, no abscess was found in the spleen. Cultures of the excised specimens were positive for *Bacillus* sp.

Intravenous antibiotic therapy with meropenem trihydrate was started at the time of surgery and continued for 4 weeks after splenectomy. The patient recovered uneventfully, and the white blood cell count and C-reactive protein had normalized by postoperative day (POD) 40. Postoperative blood cultures were sterile and echocardiography showed no deterioration of the periprosthetic valve leak. Because the patient was hemodynamically stable, surgical intervention for the aortic prosthetic valve was deemed unnecessary and he was discharged on POD 41. He is doing well without any sign of recurrence of the *Bacillus* sp. infection or heart failure 10 months after splenectomy.

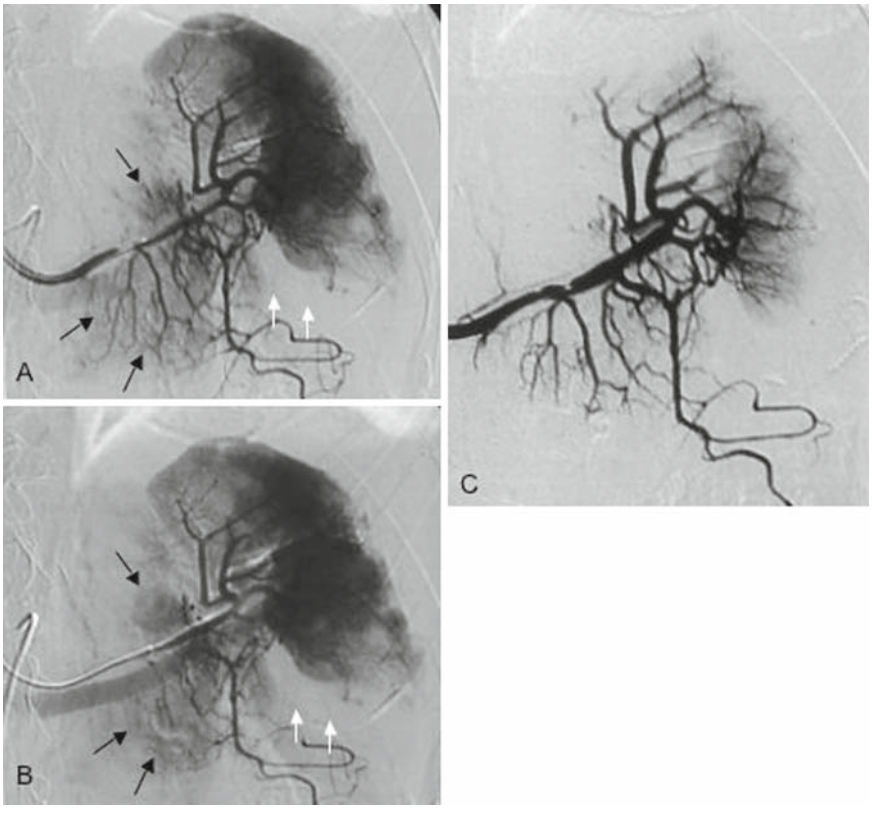

Fig. 3. Selective splenic angiograms (**A** arterial phase; **B** venous phase) showing extravasation of contrast material (*arrows*) from the spleen without aneurysms of the splenic artery, and a hypovascular area in the spleen. **C** Selective arteriogram showing disappearance of extravasation after selective embolization of the splenic artery

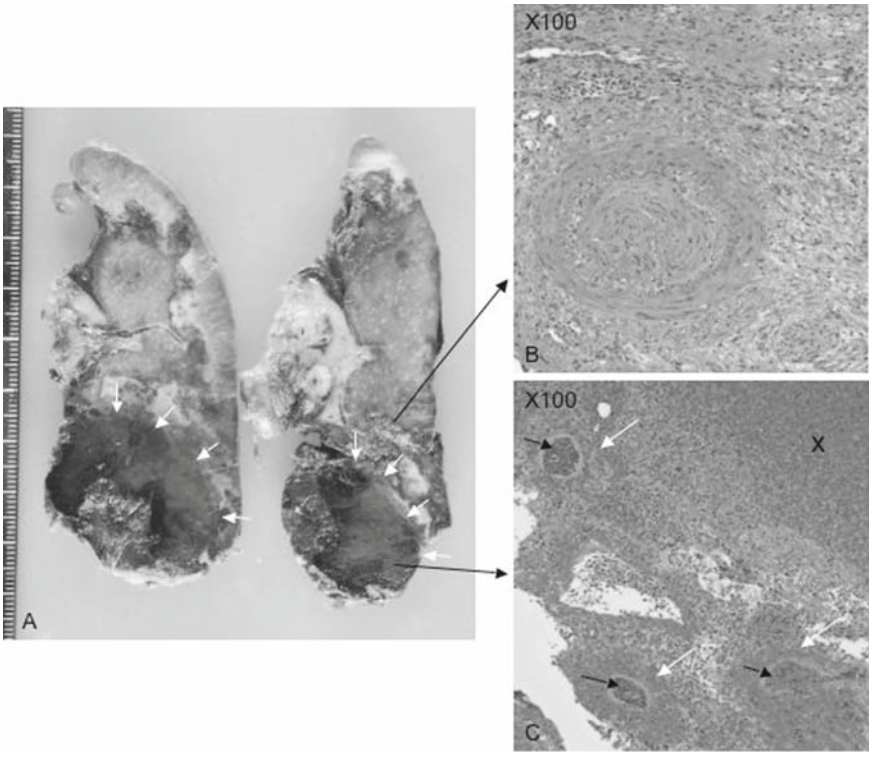

Fig. 4. A Photograph of the excised spleen showing infarctions associated with rupture of the splenic capsule and hematoma (*white arrows*). **B** Microscopic photograph showing the infarction resulting from embolization of the splenic artery, without infiltration of the inflammatory cells. **C** Microscopic photograph showing the infarction that contained arterial occlusion by septic emboli with invasion of the neutrophils (*black arrows*) and infiltration of the inflammatory cells (*white arrows*), and hemorrhage (*cross*)

Discussion

This patient had two rare pathologies of serious systemic infection: possible prosthetic valve endocarditis in the aortic position, and spontaneous splenic rupture associated with septic infarctions caused by *Bacillus* sp. Although we were not able to establish the exact primary source of *Bacillus* sp. bacteremia, infection of the aortic prosthetic valve is thought to be likely and could have caused the splenic infarction triggering the rupture, even though vegetations were not detected by echocardiography. The presence of a prosthetic valve with a periprosthetic valve leak, high fever, history of repeated systemic emboli, and positive blood culture for *Bacillus* sp. met the modified Duke criteria[4] and allowed us to diagnose possible infective endocarditis. According to reviews of 70 cases of *Bacillus* sp. infections,[3,5] endocarditis, meningitis, osteomyelitis, ophthalmitis, and pneumonia have all occurred as serious infections, but only one case of splenic abscess and two cases of endocarditis have been reported.[3,5] To our knowledge, this is the first reported case of spontaneous splenic rupture caused by *Bacillus* sp. infection. Moreover, we found only two case reports of prosthetic valve endocarditis caused by *Bacillus* sp. in the English literature[6,7] published in the last two decades.

Bacteria of the genus *Bacillus* are Gram-positive, aerobic, spore-forming rods, and the organisms are ubiquitous in nature. The prevalence of positive blood cultures for *Bacillus* sp. ranges from 0.1% to 0.9% of specimens investigated.[3] In addition to compromised hosts, intravenous drug abuse and intravascular devices, especially intravenous catheters, are the main predisposing factors to *Bacillus* infections; however, serious systemic infections are rare because *Bacillus* bacteremias are generally eradicated easily by removing an infected intravascular device or antibiotic therapy.[3] Therefore, antibiotic therapy should be initiated when a *Bacillus* sp. infection is diagnosed. No antibiotics were given to our patient preoperatively because a positive blood culture for *Bacillus* sp. was not obtained until just before the emergency surgery. Furthermore, no surgical intervention for the prosthetic valve was performed because the patient was hemodynamically stable without any symptoms or signs of heart failure, as demonstrated by a previous report.[8]

Spontaneous splenic rupture is associated with many conditions, including hematologic diseases, splenic infarcts, viral or bacterial infections, pancreatitis, rheumatologic diseases, pregnancy, and occasionally, anticoagulant therapy.[1] Our patient had splenic infarction and a *Bacillus* sp. infection, and he was on anticoagulant therapy. Splenic infarction usually occurs in association with emboli of cardiac origin or hematologic disorders and with atheromatous disease of the aorta and/or splenic artery; however, the results of the hematological examination were negative and splenic arteriography suggested an embolic rather than atheromatous origin in this patient.

Several mechanisms of splenic rupture, including rupture of a splenic abscess, rupture of intrasplenic vessels in an area of infarction, and rupture of a mycotic aneurysm of the splenic artery, have been associated with splenic infarction resulting from septic emboli of cardiac origin.[9] Our patient had no splenic abscess or aneurysm of the splenic artery, so a septic infarction may have induced suppurative changes in the intrasplenic vessels causing an intrasplenic hematoma, which may have then dissected toward an area of perisplenitis, eventually distending and rupturing the splenic capsule. Anticoagulant therapy with warfarin could also have contributed to distension of the intrasplenic hematoma because an association of SSR with anticoagulant therapy has been described in many reports.[1,10]

Since emboli to the spleen and resultant infarctions are frequently asymptomatic, we believe that every patient with conditions associated with a risk of SSR should be monitored with routine abdominal CT scanning, which is the most sensitive and specific imaging technique to diagnose splenic lesions.[11] Although selective splenic arteriography is another diagnostic tool for splenic rupture and embolization of the splenic artery is a life-saving procedure, evidence of active bleeding on CT imaging and long-term anticoagulant therapy would be indications for splenectomy. Splenectomy is indicated for abscess or persistent sepsis, large lesions over 2 cm, or peripheral lesions. Prophylactic splenectomy should be considered for such patients as splenic embolization is not enough when septic infarcts may progress to abscess formation with a risk, albeit small, of rupture.[11]

In summary, we report a rare case of SSR resulting from splenic infarction caused by a *Bacillus* infection in a patient on anticoagulant therapy. The patient was successfully treated by combined splenectomy with antibiotic therapy.

References

1. Kapan M, Kapan S, Karabicak I, Bavunogle I. Simultaneous rupture of the liver and spleen in a patient on warfarin therapy: report of a case. Surg Today 2005;35:252–5.
2. Chen E, Chen Z, Wu S, Wang X. Niemann-Pick disease resulting in spontaneous splenic rupture in an adult: report of a case. Surg Today 2008;38:473–5.
3. Sliman R, Rehm S, Shlaes DM. Serious infections caused by *Bacillus* species. Medicine 1987;66:218–23.
4. Baddour LM, Wilson WR, Bayer AS, Fowler VG Jr, Bolger AF, Levison ME, et al. Infective endocarditis. Diagnosis, antimicrobial therapy, and management of complications. A statement for healthcare professionals from the committee on rheumatic fever,

endocarditis, and Kawasaki disease, council on cardiovascular disease in the young, and the councils on clinical cardiology, stroke, and cardiovascular surgery and anesthesia, American Heart Association. Circulation 2005;111:e394–433.
5. Tuazon CU, Murray HW, Levy C, Solny MN, Curtin JA, Sheagren JN. Serious infections from *Bacillus* sp. JAMA 1979;241:1137–40.
6. Krause A, Gould FK, Forty J. Prosthetic heart valve endocarditis caused by *Bacillus circulans*. J Infect 1999;39:160–2.
7. Gatermann S, Mitusch R, Djonlagic H, Hollandt H, Marre R. Endocarditis caused by *Bacillus circulans*. Infection 1991; 19:445.
8. Truninger K, Attenhofer Jost CH, Seifert B, Vogt PR, Follath F, Schaffner A, et al. Long-term follow-up of prosthetic valve endocarditis: what characteristics identify patients who were treated successfully with antibiotics alone? Heart 1999;82:714–20.
9. Dimitrakakis G, Von Oppell U, Zilidis G, Srivastava A. Splenic rupture complicating aortic valve replacement for bacterial endocarditis. Int Cardiovasc Thorac Surg 2008;7:138–40.
10. Llanwarne N, Badic B, Delugeau V, Landen S. Spontaneous splenic rupture associated with *Listeria* endocarditis. Am J Emerg Med 2007;25:1086.e3–5.
11. Ting W, Silverman NA, Arzouman DA, Levitsky S. Splenic septic emboli in endocarditis. Circulation 1990;82(suppl IV):IV-105–9.