

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr



Case Report

Splenic artery embolization for spontaneous splenic rupture due to Babesiosis: a case report*

Behnaz Khazai, MD, MSc*, Matthew J. Homsi, DO, Cecile Diane Moliva Anendaga, MD, Eric Reiner, MD, Todd J. Everett, MD, Francis T. Flaherty, MD, Michael D. Hollander, MD

Radiology Department, Norwalk Hospital, 34 Maple St, Norwalk, CT, USA

ARTICLE INFO

Article history: Received 24 March 2022 Accepted 30 March 2022

Keywords: Babesiosis Splenic artery embolization Spontaneous splenic rupture Northeast United States Proximal vs Distal splenic artery embolization

ABSTRACT

Babesiosis incidence in the United States has been increasing with an 11% rise between 2018 and 2019 based on the latest CDC annual summary, reaching its highest ever reported incidence. This primarily tick-borne disease is particularly prevalent in New England. Despite predominantly nonspecific and at times subtle symptoms, life-threatening complications do occur. One such complication is splenic rupture which has been suggested to be more common in younger and otherwise healthy individuals. This is a report on a successful splenic artery embolization in a 65-year-old male from upstate New York who, unlike most prior studies, showed splenic rupture *after* he was discharged with negative parasitemia and general improvement following several days of targeted antibiotic therapy. Increased incidence and various presentations of Babesiosis call for an attempt to promote clinical awareness for radiologists among other specialties.

© 2022 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

Introduction

Since its first appearance in literature in 1957, human Babesiosis has been reported on all continents except for Antarctica. It became a nationally notifiable disease in the United States in January 2011 [1,2]. The latest annual summary by CDC, published in 2021, shows an 11% increase in the total number of reported cases compared with 2018; the highest annual rate ever since national data became available [2]. Out of 41 states where Babesiosis has been a reportable disease in 2019, 25 states have reported at least 1 case and the mortality rate has been 0.57%. Eighty-eight percent of these cases

* Competing Interest: None

* Corresponding author.

E-mail address: behnazkhazai@icloud.com (B. Khazai). https://doi.org/10.1016/j.radcr.2022.03.105 were residents of 7 states where tickborne transmission is well established: Connecticut, Massachusetts, Minnesota, New Jersey, New York, Rhode Island, and Wisconsin. Although transmission through transfusion, organ transplant, and placenta is possible and has been reported, history of travel to areas with established local transmission has been documented for many of the cases from those states that lack an established local transmission [2,3]. It is speculated that the thoroughness of reported statistics might have been affected by the COVID-19 pandemic and its inevitable challenges in 2019 [2]. Similar to previous years, majority of cases in 2019 were between the ages of 60-69 years (median: 64), were males (66%), and presented in spring-summer which follows ticks' activity. Fortyfour percent of cases were hospitalized for at least 1 day and the most common symptom has been the fever [2]. Symptoms are mostly nonspecific and absence of symptoms does

1930-0433/© 2022 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)



Fig. 1 - Multiple wedge-shaped hypodensities in the periphery of the spleen interpreted as infarcts.

not preclude a life-threatening complication like spontaneous splenic rupture, as in this case.

Case report

A 65-year-old Guatemalan male living in New York presented to a hospital in upstate New York, affiliated with our facility with a 4-week history of progressive vague abdominal pain, mostly localized to the left upper quadrant, associated with the onset of spiking fevers and chills for the past 3 days. He reported no exacerbating factors, but the pain was alleviated by rest. He also reported having had 2 weeks of constitutional symptoms including fever, chills, night sweats, malaise, and headache prior to his abdominal pain. He was vaccinated for COVID19 and had no significant past medical history except for appendectomy. Abdominal CT showed mild splenomegaly with multifocal wedge-shaped hypodensities consistent with acute infarcts in the absence of trauma or reported risk factors (Fig. 1). Peripheral smear showed intracellular parasites, later identified as Babesia Microti with positive Babesia PCR. He was started on Atovaquone, Azithromycin, and Doxycycline for presumed babesiosis and possible Anaplasmosis. Doxycycline was later discontinued. Lovenox was administered for DVT prophylaxis during admission. He was discharged after 7 days with improved abdominal pain, negative peripheral smear for parasites, and is afebrile for more than 48 hours. He was to continue Azithromycin and Atovaquone as an outpatient. Two days later, he presented with severe non-radiating left upper quadrant pain, dizziness, and lightheadedness. He had also noticed a trace of bright red blood on toilet paper that day. On presentation, he was hypotensive with hemoglobin of 9.5, total bilirubin of 1.1, and a positive occult blood test. He denied any trauma or recurrent fever since discharge. CT Angiogram of the abdomen was obtained which showed a significant interval increase in size and extent of splenic wedge-shaped hypodensities, the largest of which measured 4×3.3 cm with acute blood products, but without active contrast extravasation. There was an associated acute peri-splenic hematoma measuring up to 1.4 cm in thickness and mild hemoperitoneum without evidence of GI hemorrhage (Fig. 2). In summary, the patient had a splenic rupture and after stabilization including receipt of 2L of fluid, he underwent IR embolization of the splenic artery.

He had an initial temperature of 36.8°C, a heart rate of 99 bpm, respiratory rate of 18 minutes⁻¹, blood pressure of 105/72 mmHg, and O2 saturation of 98%. Lab results were remarkable for mild lymphopenia, normocytic normochromic anemia with a hemoglobin of 7.3 and hematocrit of 25.6, LDH of 359, and normal renal function. On physical exam, his abdomen was mildly distended, and tender throughout with evidence of focal peritonitis in the left upper quadrant. He received one unit of packed RBC. Surgery was consulted. Patient was transferred to the IR suite and under conscious sedation, the aorta was catheterized via a right femoral artery approach. The celiac artery was accessed followed by the splenic artery which did not show active extravasation at the time of the procedure. Proximal splenic artery was embolized with multiple 0.035" coils followed by an angiogram showing minimal splenic vascular territory filling from collaterals (Fig. 3). The catheter was withdrawn, a closure device was used at the access point and the patient was admitted to PCU. Total procedure time was 34 minutes with 6.4 minutes fluoroscopy time.

Microbiology tests over the admission showed no parasites or schistocytes on a peripheral blood smear, negative blood cultures, negative Babesia and Anaplasma PCR tests, and evidence of past EBV infection. ELISA for Lyme's was positive with subsequent negative western blot. Moreover, the patient did not report or show any rash or other evidence of Lyme's disease including articular, cardiologic, or neurologic manifestations. A negative Babesiosis test was attributed to prior antibiotic therapy. A transthoracic echocardiogram was performed to exclude an embolic etiology for the splenic infarcts and was negative. Despite hemodynamic stability and improving Hb levels, post-embolization course was complicated by a 101.5°F fever spike on day 2, and patient-reported persistent abdominal pain. Repeat abdominal CT showed a heterogeneous spleen with a normal enhancement pattern seen in a small superomedial portion of the spleen. There were new mildly rim enhancing communicating collections along the inferolateral margins of the spleen measuring up to $8.8 \times 4.8 \times 12.8$ cm with internal gas locules which were concerning for abscesses (Fig. 4). Diagnostic and potentially therapeutic CT-guided aspiration was performed which only yielded 3cc of non-purulent old blood products and precluded

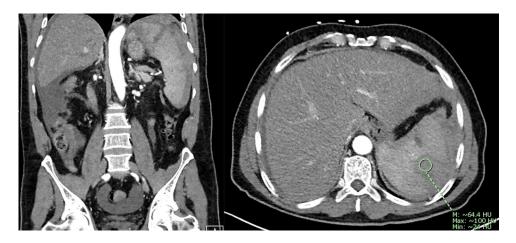


Fig. 2 – Interval increase in size and extent of splenic hypodensities with evidence of acute blood products, no active extravasation, new subcapsular hematoma, and hemoperitoneum.

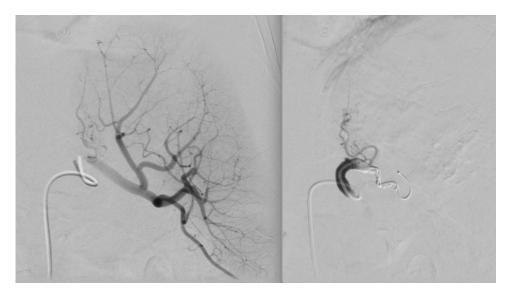


Fig. 3 - Splenic artery catheterization before and after embolization.



Fig. 4 – Status post embolization of the splenic artery with new communicating and mildly rim enhancing collections with internal gas locules.

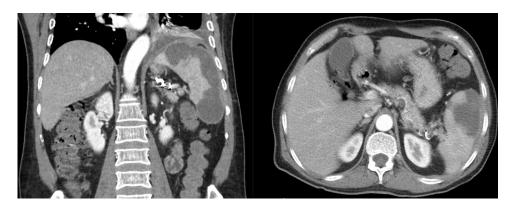


Fig. 5 - One month follow-up showing decrease in size of the collections and resolution of gas locules.

drainage catheterization. However, the patient was kept on empiric broad-spectrum antibiotics pending the culture results and because the aspiration was performed after a dose of antibiotics. With persistent negative cultures, patient's defervescence, and symptomatic improvement he was discharged on oral antibiotics. Patient continued to improve on follow-up visits and subsequent CT showed a decrease in size of the collections with a resolution of gas locules (Fig. 5).

Discussion

We presented a 65-year-old male from upstate New York who underwent successful splenic artery embolization for spontaneous splenic rupture due to Babesiosis. In the United States, Babesiosis is most common in the northeast where it has also been referred to as Nantucket fever and malaria of the northeast.

While symptoms are mostly nonspecific and could be subtle, a study on 63 confirmed cases with available imaging from New England found splenomegaly in 89%, splenic infarcts in 21%, splenic rupture in 13%, and splenic pseudoaneurysm in 5% [4]. They concluded that acute parasitemia commonly affects the spleen in spite of the prior available literature and emphasized the importance for radiologists to be aware of the entity. A case series from Rhode Island reported an incidence of 1% for splenic rupture in Babesiosis which was found to be more common in patients who were younger and healthier with a lower degree of parasitemia and less Babesiosis related end-organ damage elsewhere [5]. A case review of splenic ruptures reported that 87% of cases happened in otherwise healthy males with an average of 56 years. This is in contrast to the above-presented case which follows the general age trend of the disease [6]. Management of splenic rupture is based on patient's hemodynamic stability and evidence of contrast extravasation. While splenectomy is the gold standard for hemodynamically unstable patients that is, in hemorrhagic shock, splenic artery embolization can be successfully performed in patients with stable hemodynamics with either significant or ongoing bleeding [5,7]. Splenic artery embolization can be proximal, selective distal, or in some cases, a combination of both. Proximal embolization is favored in multifocal injuries and has a shorter procedure time as well as a lower rate of small splenic infarcts. However, there is no significant difference in splenic salvage between the proximal and distal embolization [10].

In contrast to most prior cases in the literature, the present case showed splenic rupture after the acute phase and following several days of targeted antibiotic therapy, when parasitemia was no longer detectable and patient was discharged with general improvement [5,6,8,9]. Familiarity with different possible presentations of this entity of increasing incidence is essential for clinicians including radiologists to broaden their differentials in the appropriate setting and avoid misses.

Conclusion

CDC reports an ever-increasing incidence of Babesiosis which can present with nonspecific subtle findings and yet potentially life-threatening complications like splenic rupture which could be manageable with splenic artery embolization. We call for an attempt to promote radiologists' awareness of Babesiosis and its various possible presentations especially in acute settings.

Patient Consent

An informed written consent has been obtained from the patient.

REFERENCES

- Yang Y, Christie J, Köster L, Du A, Yao C. Emerging human Babesiosis with "Ground Zero" in North America. Microorganisms 2021;9(2):440 PMID: 33672522; PMCID: PMC7923768. doi:10.3390/microorganisms9020440.
- [2] Centers for Disease Control and Prevention (CDC) Surveillance for babesiosis — United States, 2019 Annual Summary. Atlanta, Georgia: US Department of Health and Human Services, CDC; 2021.

- [3] Renard I, Ben Mamoun C. Treatment of human Babesiosis: then and now. Pathogens 2021;10(9):1120 PMID: 34578153; PMCID: PMC8469882. doi:10.3390/pathogens10091120.
- [4] Mojtahed A, Bates DDB, Hahn PF. Splenic findings in patients with acute babesiosis. Abdom Radiol (NY) 2020;45(3):710–15 PMID: 31832741. doi:10.1007/s00261-019-02362-z.
- [5] Patel KM, Johnson JE, Reece R, Mermel LA. Babesiosis-associated splenic rupture: case series from a hyperendemic region. Clin Infect Dis 2019;69(7):1212–17 PMID: 30541016. doi:10.1093/cid/ciy1060.
- [6] Li S, Goyal B, Cooper JD, Abdelbaki A, Gupta N, Kumar Y. Splenic rupture from babesiosis, an emerging concern? A systematic review of current literature. Ticks Tick Borne Dis 2018;9(6):1377–82 Epub 2018 Jun 12. PMID: 29954722. doi:10.1016/j.ttbdis.2018.06.004.
- [7] Dumic I, Patel J, Hart M, Niendorf ER, Martin S, Ramanan P. Splenic Rupture as the first manifestation of babesia Microti

infection: report of a case and review of literature. Am J Case Rep 2018;19:335–41 PMID: 29567936; PMCID: PMC5878539. doi:10.12659/ajcr.908453.

- [8] Reis SP, Maddineni S, Rozenblit G, Allen D. Spontaneous splenic rupture secondary to Babesia microti infection: treatment with splenic artery embolization. J Vasc Interv Radiol 2011;22(5):732–4 PMID: 21514529. doi:10.1016/j.jvir.2011.01.439.
- [9] El Khoury MY, Gandhi R, Dandache P, Lombardo G, Wormser GP. Non-surgical management of spontaneous splenic rupture due to Babesia microti infection. Ticks Tick Borne Dis 2011;2(4):235–8 Epub 2011 Sep 21. PMID: 22108020. doi:10.1016/j.ttbdis.2011.08.001.
- [10] Quencer KB, Smith TA. Review of proximal splenic artery embolization in blunt abdominal trauma. CVIR Endovasc 2019;2(1):11 PMID: 32026033; PMCID: PMC7224246. doi:10.1186/s42155-019-0055-3.