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## Case Report

# Splenic abscess due to brucellosis: a case report and a review of the literature



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#### SUMMARY

Splenic abscess due to acute brucellosis is a rare event. We report a case of multiple splenic abscesses caused by *Brucella melitensis* in a 45-year-old woman and review the English language literature based on a PubMed/MEDLINE search of the last 50 years. The majority of the cases published in the literature were due to *B. melitensis* and a splenectomy was required in half of the cases. Antibiotics alone without surgical intervention can be successful in the treatment of patients with splenic brucellosis in the early stages of the disease.

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#### 1. Introduction

Brucellosis is a systemic zoonotic infectious disease caused by Gram-negative bacilli of the genus *Brucella*. Although the disease is prevalent worldwide, it is particularly endemic in many Middle Eastern countries, the Mediterranean region, and the Arabian Peninsula.<sup>1</sup> It occurs mainly because of the consumption of unpasteurized milk and contact with infected animals. Clinical manifestations vary from multisystem involvement to asymptomatic infection.<sup>2</sup> Almost every organ in the body may be involved.<sup>3</sup> Isolated splenic abscess is a very rare condition with a very high mortality unless treated adequately.<sup>4</sup>

### 2. Case report

A 45-year-old woman was admitted with a 2-month history of fever with chills, generalized malaise, night sweats, and weight loss (9 kg in 2 months). She had undergone a dilation and curettage procedure for a first-trimester abortion 45 days prior to admission and had been evaluated since then by various physicians for a fever

of unknown origin (FUO). She had no history of recent travel or contact with sick persons with a similar presentation, but reported the consumption of unpasteurized milk products.

On physical examination she was lean with a regular peripheral pulse of 100 bpm and a blood pressure of 110/70 mmHg. She was febrile with a body temperature of 39.8  $^{\circ}$ C. Her conjunctivae were pale, but there was no scleral icterus or lymphadenopathy. Cardiac and pulmonary findings were normal, and hepatosplenomegaly was not detected.

Laboratory tests revealed the following results: hemoglobin 10 g/dl, leukocyte count  $2.6\times10^9/l$ , platelet count  $90\times10^9/l$ , Creactive protein 82 mg/l, and erythrocyte sedimentation rate 100 mm/h. Biochemical workup revealed normal serum transaminases and kidney function tests. Chest X-ray and echocardiography were normal. As she was admitted with FUO, brucellosis was suspected; Brucella slide and tube agglutination tests (Wright, at 1/640) were positive. Abdominal computed tomography revealed splenomegaly and multifocal hypodense lesions of various sizes, with the largest diameter of 1.5 cm in the spleen.

Two sets of blood cultures yielded *Brucella melitensis*. The patient was thought to have multiple splenic abscesses due to brucellosis and was started on oral doxycycline (200 mg/day) and rifampin (600 mg/day) for 6 weeks. Defervescence of the fever occurred on day 3 and she was discharged on day 5 with outpatient follow-up and repeat ultrasound appointments. Her clinical condition improved gradually and all the lesions had disappeared by the end of treatment on follow-up ultrasound examinations.

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**Table 1** Clinical characteristics and outcome of the patients

Case No.	Author and reference	Year published	Patient age, years/sex	Risk factor	Localization	Brucella antibody titer	Isolate	Therapy	Outcome
1	Spink <sup>14</sup>	1964	51/M	Farmer	Liver/spleen, bone	1:320	B. suis	Surgical drainage + TET	Recovered
2	Spink <sup>14</sup>	1964	58/M	Butcher	Spleen	1:320	B. suis	Splenectomy + TET $\times$ 3 months	Recovered
3	Spink <sup>14</sup>	1964	54/M	Cattle buyer	Liver/spleen, lymph node	1:80	B. suis	NA	Died of hemorrhage from esophageal varices
4	Spink <sup>14</sup>	1964	53/M	Farmer	Spleen	1:1280	B. suis	Splenectomy + TET	Recovered
5	Spink <sup>14</sup>	1964	53/M	Farmer	Spleen	1:1280	None	Splenectomy + antibiotic (NA)	Recovered
6	Ates et al. 15	1992	30/F	NA	Liver/spleen	1:1280	B. melitensis	TET + STREP IM × 4 weeks	Recovered
7	Ates et al. <sup>15</sup>	1992	50/M	NA	Spleen	1:1280	B. melitensis	TET + STREP IM × 4 weeks then RIF 4 weeks	Recovered
8	Saadeh et al. <sup>18</sup>	1996	23/M	Farmer	Spleen, aortic valve	1:2560	B. melitensis	RIF + DOX × 6 months, splenectomy	Recovered
9	Solera et al. 19	1996	49/F	None	Spleen	1:640	None	Antibiotics (NA), splenectomy	Recovered
10	Colmenero et al. <sup>11</sup>	2002	53/M	History of brucellosis	Spleen	1:80	Serum B. melitensis DNA by PCR	2 cycles of DOX × 2 months + STREP IM × 21 days	Relapsed
11	Colmenero et al. <sup>11</sup>	2002	80/NA	History of brucellosis	Spleen	1:320	Tissue B. melitensis DNA by PCR	DOX × 2 months and STREP IM × 21 days + splenectomy followed by (DOX + RIF) × 3 months	Died of other causes
12	Colmenero et al. <sup>11</sup>	2002	72/NA	History of brucellosis	Spleen	1:40	Serum and tissue B. melitensis DNA by PCR	DOX × 2 months and STREP IM × 21 days + splenectomy followed by (DOX + RIF) × 3 months	Recovered
13	Yayli et al. <sup>20</sup>	2002	70/F	Unpasteurized milk consumption	Spleen	1:320	B. melitensis	(DOX+RIF)×6 weeks and STREP IM 21 days	Recovered
14	Yilmaz et al. <sup>21</sup>	2003	19/M	Unpasteurized milk consumption	Spleen, aortic valve	High titer	B. melitensis	Splenectomy + antibiotic (NA)	Recovered
15	Ruiz Carazo et al. <sup>22</sup>	2005	60/M	NA	Spleen	NA	NA	Splenectomy + antibiotic (NA)	Recovered
16	Del Arco et al. <sup>16</sup>	2006	39/F	Unpasteurized milk consumption	Spleen	1:1280	B. melitensis DNA by PCR	DOX × 6 weeks + STREP IM × 2 weeks, splenectomy, RIF + DOX 1 month	Recovered
17	Sayilir et al. 10	2008	61/M	NA	Spleen	1:1280	Negative	RIF + DOX and TMP-SMZ	Recovered
18	Park et al. <sup>17</sup>	2009	45/M	Livestock industry worker	Spleen, aortic valve	1:160	B. abortus	GEN and RIF+DOX+TMP-SMZ 12 months	Recovered
19	Eruz et al. <sup>9</sup>	2011	52/F	Raw meatball consumption	Spleen, respiratory system	1:640	B. melitensis	RIF + DOX $\times$ 6 weeks	Recovered
20	Deveer et al. <sup>8</sup>	2013	21/M	Farmer	Spleen	1:1280	B. melitensis	$(DOX + CIP) \times 12$ weeks + STREP $IM \times 3$ weeks	Recovered
21	Present case	2013	45/F	Unpasteurized milk consumption	Spleen	1:640	B. melitensis	$(RIF + DOX) \times 6$ weeks	Recovered

M, male; F, female; TET, tetracycline; DOX, doxycycline; STREP, streptomycin; GEN, gentamicin; RIF, rifampin; CIP, ciprofloxacin; TMP–SMZ, trimethoprim–sulfamethoxazole; NA, not available; IM, intramuscular.

The patient remained asymptomatic without any signs or symptoms of disease recurrence at 1 year after the completion of treatment.

#### 3. Discussion

Since our patient had a history of consumption of unpasteurized milk products, the mode of transmission was thought to be ingestion of contaminated food. As a multisystem disease, brucellosis can often be a diagnostic dilemma as the disease is associated with a wide variety of signs and symptoms. Our patient had a duration of symptoms of 2 months on presentation, which is typical of FUO.

Based on the initiation of her symptoms, our patient had brucellosis during her pregnancy. There is evidence that brucellosis can induce abortion in humans. Positive cultures of Brucella from human placenta, aborted fetuses, and other products of conception have been reported previously. Moreover, replication of *B. melitensis* in human trophoblasts has recently been shown. Therefore, it is speculated that brucellosis causes fewer spontaneous abortions in humans than animals due to the absence of erythritol in the human placenta and fetus. Our patient might have had the abortion due to brucellosis.

Focal forms of brucellosis may occur in up to a third of patients.<sup>3,7</sup> As they are considered true complications of the disease, they tend to have a worse prognosis compared to the nonfocal forms. Osteoarticular, neurological, genito-urinary, liver, hematological, and cardiac involvements are the most frequent focal forms. Isolated splenic abscess is an extremely rare and serious complication of brucellosis.<sup>8–10</sup> Not a single case was found in one of the largest Turkish series of 1028 cases.<sup>3</sup> Nevertheless, its incidence has been reported as less than 2% in other large case series.<sup>11,12</sup> Spontaneous splenic rupture due to brucellosis has been reported in a few rare cases.<sup>13</sup>

Our PubMed/MEDLINE search (1964–2013) yielded 20 adult cases published in the English language literature to date. Baseline demographics and clinical characteristics of all the cases are given in Table 1.8–11.14–22 The mean age of the patients was 49 years (range 19–80 years) and 68% were men. Four of the cases were due to *Brucella suis* and only one was due to *Brucella abortus*; the remaining 70% were due to *B. melitensis*. None of the patients died due to splenic abscess or any other complication of brucellosis. A splenectomy was required in 10 (47%) cases. In some cases with chronic splenic abscess where serum agglutination remained as low as 1:40, diagnosis was established using PCR of peripheral blood and splenic tissue.<sup>11</sup>

The best therapeutic approach for splenic abscess due to Brucella has not been clearly established. The treatment of splenic abscess with antibiotics alone has been reported, however this modality appears to be successful only in the early stages of the disease when there is no calcification in the lesions. <sup>4,15,20</sup> A therapeutic approach in the early stages, with the use of antibiotic therapy alone or associated with non-surgical drainage, may be an initial option, but prolonged therapy over several months may be required and careful follow-up is essential because the complete cure of the disease cannot be guaranteed. <sup>12,16</sup> The treatment of chronic lesions should involve a combination of medical and surgical therapy. <sup>16</sup> Our patient responded well to the standard duration 6 weeks of treatment.

In conclusion, splenic abscess due to brucellosis is a rare entity and prompt diagnosis is crucial for initiating therapy and preventing complications. In endemic areas, patients with acute brucellosis should be investigated for splenic abscess by abdominal ultrasonography, particularly when they have had a prolonged fever. The response to antibiotic therapy alone is favorable in the early stages, however careful follow-up is required to ensure complete cure of the disease.

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#### References

- 1. Gorgulu A, Albayrak BS, Gorgulu E, Tural O, Karaaslan T, Oyar O, et al. Spinal epidural abscess due to Brucella. Surg Neurol 2006;66:141-6. discussion 6-7.
- Zhen Q, Lu Y, Yuan X, Qiu Y, Xu J, Li W, et al. Asymptomatic brucellosis infection in humans: implications for diagnosis and prevention. Clin Microbiol Infect 2013:19:F395-7.
- 3. Buzgan T, Karahocagil MK, Irmak H, Baran Al, Karsen H, Evirgen O, et al. Clinical manifestations and complications in 1028 cases of brucellosis: a retrospective evaluation and review of the literature. *Int J Infect Dis* 2010;14:e469–78.
- Secmeer G, Ecevit Z, Gulbulak B, Ceyhan M, Kanra G, Anlar Y. Splenic abscess due to Brucella in childhood. A case report. Turk J Pediatr 1995;37:403–6.
- Al-Tawfiq JA, Memish ZA. Pregnancy associated brucellosis. Recent Pat Antiinfect Drug Discov 2013;8:47–50.
- Salcedo SP, Chevrier N, Lacerda TL, Ben Amara A, Gerart S, Gorvel VA, et al. Pathogenic brucellae replicate in human trophoblasts. J Infect Dis 2013;207:1075–83
- 7. Colmenero JD, Reguera JM, Martos F, Sanchez-De-Mora D, Delgado M, Causse M, et al. Complications associated with *Brucella melitensis* infection: a study of 530 cases. *Medicine (Baltimore)* 1996;**75**:195–211.
- Deveer M, Sozen H, Cullu N, Sivrioglu AK. Splenic abscess due to acute brucellosis. BMJ Case Rep 2013;2013.
- Eruz ED, Birengel S, Azap A, Bozkurt GY. A case of brucellosis presenting with multiple hypodense splenic lesions and bilateral pleural effusions. Case Rep Med 2011;201:614546.
- Sayilir K, Iskender G, Ogan MC, Erdil F. Splenic abscess due to brucellosis. J Infect Dev Ctries 2008;2:394–6.
- Colmenero Jde D, Queipo-Ortuno MI, Maria Reguera J, Angel Suarez-Munoz M, Martin-Carballino S, Morata P. Chronic hepatosplenic abscesses in brucellosis. Clinico-therapeutic features and molecular diagnostic approach. *Diagn Microbiol Infect Dis* 2002;42:159–67.
- 12. Ariza J, Pigrau C, Canas C, Marron A, Martinez F, Almirante B, et al. Current understanding and management of chronic hepatosplenic suppurative brucellosis. *Clin Infect Dis* 2001;32:1024–33.
- 13. Demirdal T, Okur N, Demirturk N. Spontaneous splenic rupture with hematoma in a patient with brucellosis. *Chang Gung Med J* 2011;34:52–5.
- Spink WW. Host-parasite relationship in human brucellosis with prolonged illness due to suppuration of the liver and spleen. Am J Med Sci 1964;247:129– 36.
- 15. Ates KB, Dolar ME, Karahan M, Temucin G, Onaran L. *Brucella melitensis* splenic abscess: sonographic detection and follow-up. *J Clin Ultrasound* 1992;**20**:349–
- 16. Del Arco A, De La Torre-Lima J, Prada JL, Aguilar J, Ruiz-Mesa JD, Moreno F. Splenic abscess due to Brucella infection: is the splenectomy necessary? Case report and literature review. Scand J Infect Dis 2007;39:379–81.
- Park SH, Choi YS, Choi YJ, Cho SH, Yoon HJ. Brucella endocarditis with splenic abscess: a report of the first case diagnosed in Korea. Yonsei Med J 2009;50:142–6
- Saadeh AM, Abu-Farsakh NA, Omari HZ. Infective endocarditis and occult splenic abscess caused by *Brucella melitensis* infection: a case report and review of the literature. *Acta Cardiol* 1996;51:279–85.
- Solera J, Espinosa A, Geijo P, Martinez-Alfaro E, Saez L, Sepulveda MA, et al. Treatment of human brucellosis with netilmicin and doxycycline. Clin Infect Dis 1996:22:441–5.
- Yayli G, Isler M, Oyar O. Medically treated splenic abscess due to Brucella melitensis. Scand J Infect Dis 2002;34:133–5.
- Yilmaz MB, Kisacik HL, Korkmaz S. Persisting fever in a patient with Brucella endocarditis: occult splenic abscess. Heart 2003:89:e20.
- Ruiz Carazo E, Munoz Parra F, Jimenez Villares MP, del Mar Castellano Garcia M, Moyano Calvente SL, Medina Benitez A. Hepatosplenic brucelloma: clinical presentation and imaging features in six cases. Abdom Imaging 2005;30:291–6.