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Brucella pelvic tubo-ovarian abscess with a history of chronic brucellosis

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ARTICLE INFO

Article history: Received 30 September 2020 Received in revised form 9 December 2020 Accepted 13 December 2020

Keywords: Brucellosis Ovarian Tubo-ovarian abscess Serology Magnetic resonance imaging

ABSTRACT

Brucellosis is a zoonotic disease located especially in Central and South America, India, the Mediterranean and the Middle East. Human brucellosis occurs as a systemic infectious disease with various clinical manifestations. We present a case of 45-year-old female patient, nulliparous, not sexually active, with a previous medical history of a treated brucellosis, and no surgical or gynecological history. The patient presented with a history of fever for 7 days of 39 degrees Celsius, chills and acute abdominal pain. She was diagnosed with diffuse peritonitis with left tubo-ovarian abscess and was admitted for an urgent diagnostic laparoscopy. A left adnexectomy was performed. The diagnosis of genital brucellosis was made. This case report discusses an unusual complication of brucellosis represented by a tuboovarian abscess associated with acute peritonitis, treated by a laparoscopic adnexectomy and antimicrobials. Acute peritonitis associated with a tubo-ovarian abscess is an unusual complication of brucellosis.

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Introduction

Brucellosis is a zoonotic disease located especially in Central and South America, India, the Mediterranean and the Middle East [1]. Brucella species are Gram-negative unencapsulated, nonmotile, non-spore-forming, facultative intracellular bacilli. Human brucellosis occurs as a systemic infectious disease with various clinical manifestations. The generalized symptoms, consisting of fever, night sweats and malaise, are not disease specific and can mimic many diseases. This infection can be transmitted to humans through direct contact with infected animals, or animal discharge, and through consumption of infected milk, milk products, or raw meat [2]. Although frequently asymptomatic, brucellosis can cause serious complications such as acute hepatitis, liver abscess, osteomyelitis and epididymo-orchitis [3]. Meningoencephalitis, infective endocarditis and peritonitis are also some rare manifestations of brucellosis. Brucella can affect the genitourinary tract in 10 % of the cases [4].

The objective of this report is to present the case of a tuboovarian abscess and diffuse peritonitis due to brucella infection.

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Case report

A 45-year-old female patient, nulliparous, not sexually active, no previous medical, surgical or gynecological history, with frequent consumption of raw meat and unpasteurized milk. She presented in July 2019 to the Emergency Department (ED) with fever of 39.5 degrees Celsius, chills and moderate abdominal pain. Laboratory results showed Hb 12 g/dL, WBC 3700/µL, elevated liver enzymes (AST 222 U/L, ALT 240 U/L, GGT 96 U/L) and CRP 101 mg/L. She was admitted to the Infectious Disease ward and an abdominal computerized tomography (CT) scan showed a diffuse hepatosplenomegaly with diffuse periportal hypodensities and edema of the vesicular bed evoking acute hepatitis. A 4.3 cm left ovarian mass was also seen on the CT scan, for which a pelvic ultrasound was performed showing a probable endometrioma.

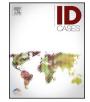
Urine culture was negative, and anti-hepatitis A IgG, anti-HBs and anti-CMV IgG were all positive. Brucella serology was positive for 1:5120. A blood culture was positive after seven days to Brucella melitensis resistant to trimethoprim-sulfamethoxazole. She was started on rifampin 600 mg per day and doxycycline 200 mg per day for 3 months and discharged from the hospital at day 14 with normal CRP and normal liver enzymes (AST 24 U/L, ALT 32 U/L, GGT 45 U/L). She presented with fever and chills two months after the end of her treatment, with normal liver enzymes and she was started again on rifampin 600 mg per day and doxycycline 200 mg once daily for 2 months. Her brucella serology decreased to 1:1280.

http://dx.doi.org/10.1016/i.idcr.2020.e01029

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Case report





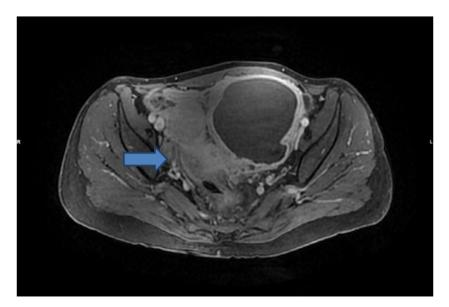


Fig. 1. Pelvic MRI showing 10.2 × 8.7 × 8.8 cm left tubo-ovarian abscess presenting an intermediate signal in T2 with a restriction of the diffusion (arrow).

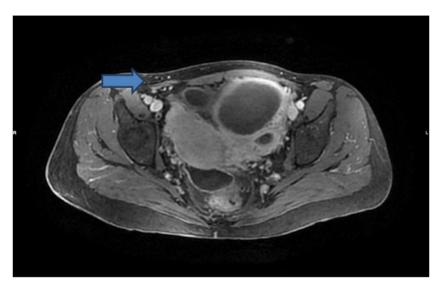


Fig. 2. Pelvic MRI showing acute Peritonitis (arrow).

In April 2020, the patient presented with a history of fever for 7 days of 39 degrees, chills and acute abdominal pain. She consulted an ID specialist who started rifampin and doxycycline 5 days before. In the ED, blood pressure was 120/60 mmHg, heart Rate 86/ min, temperature 37.8 degrees. On physical examination, her abdomen was tense and painful.

Laboratory results showed normal liver enzymes, normal creatinine, WBC 13,100/ μ L with 80.1 % PMN, CRP 175 mg/L with an elevation of her *Brucella* serology to 1:5120.

A pelvic magnetic resonance imaging (Figs. 1 and 2) was performed and showed the presence of pelvic ascites reaching the pouch of Douglas, evoking an acute peritonitis with a 10.2 cm left ovarian cystic mass, increased in size compared to the pelvic ultrasound done on July 2019.

She was admitted for urgent diagnostic laparoscopy (Figs. 3,4) that showed false membranes and diffuses abdominopelvic

peritonitis reaching the sub hepatic and sub pancreatic space, with a huge left tubo-ovarian abscess adherent to the sigmoid posteriorly. A Left adnexectomy was performed. No colonic diverticulitis was seen, the right ovary and tube had normal appearance.

She was started on piperacillin/tazobactam 4.5 g every 6 h and doxycycline 200 mg once daily. Urine culture returned negative. She had a trans-thoracic echocardiography which returned normal. *Brucella spp* was isolated from a blood culture after eleven days (Fig. 5). The final pathology report showed significant chronic and acute necrotic and suppurative inflammatory changes destroying the ovarian parenchyma with chronic and acute salpingitis. No sign of malignancy was seen. The morphological aspect raises the hypothesis of an infectious origin.

On Post-operative day 2 the patient seemed well, no fever, no abdominal pain, with a decreased level of CRP to 124 mg/L and



Fig. 3. Laparoscopy showing a tubo-ovarian abscess with peritonitis and false membranes. a: Left Pelvic Tubo-ovarian abscess, b: abdominal wall.



Fig. 4. Laparoscopy showing a tubo-ovarian abscess adherent to the sigmoid posteriorly. a: left tubo-ovarian abscess, b: sigmoid, c: false membranes and peritonitis.

WBC to $5100/\mu$ L. She is discharged 5 days later on rifampin 600 mg per day for 2 months, ciprofloxacin 500 mg BID and doxycycline 200 mg per day.

Discussion

This case report discusses an unusual complication of brucellosis represented by a tubo-ovarian abscess associated with acute peritonitis, treated by a laparoscopic adnexectomy and antibiotics.

Brucella is a facultative intracellular organism proliferating in the reticulo-endothelial system (RES), multiplying in the local lymph nodes, and then spreading by the hematogenous way [5]. Some hypothesis can explain how *brucella* can have a gonadal tropism: the high gonadal production of fructose through the Polyol Pathway and Pentose Phosphate Pathway attracts the *brucella* for tropism [6]. The Pentose phosphate Pathway produces the erythritol, a C4 alcohol sugar, the preferred carbon/energy source of *Brucella* [7].

In male patients, the scrotal area was the most frequent involved site. Epididymo-orchitis, orchitis and epididymitis are the most frequent genitourinary localizations of the disease. However, studies are limited in female patients [8]. Pyelonephritis was significantly higher in female patients with a percentage of 86.8 %, but Fallopian tube abscesses (5.3 %) and tubo-ovarian abscesses (2.6 %) were rare [8]. Pelvic abscess is a collection of infected fluid in the pelvis, caused by pelvic inflammatory disease [9], diverticulitis, appendicitis, or infectious disease. It requires an early diagnosis, hospitalization, and surgical or medical treatment. Our Patient has all the risk factors to have brucellosis. She had a history of consuming unpasteurized milk and raw meat. She lives in an endemic region: 742 documented cases of brucellosis in Lebanon between 2017 and 2018 [10].

The diagnosis is usually made by the identification of the *brucella* organism in blood cultures, or by testing *brucella* serology. Our patient presented to the ED with all the clinical symptoms of brucellosis and pelvic abscess: high grade fever, abdominal pain, leukocytosis and elevated C-reactive protein. Blood cultures show the growth of *brucella* spp after eleven days of culture (Fig. 5).

The diagnosis of human brucellosis is based on positive serological testing. In the presence of the clinical signs and

	Brucel	Brucella spp	
	Sensibilité	CMI (mg/L)	
Amoxicilline + ac. clavulanique	S		
Pipéracilline+Tazobactam	S		
Céfotaxime	S		
Ceftriaxone	S		
Amikacine	S		
Tétracycline	S		
Triméthoprime+sulfaméthoxazole	R		
Chloramphénicol	S		
Rifampicine	S		
Erythromycine	R		

Fig. 5. Antibiotic sensitivity against the Brucella isolated in the culture.

symptoms of brucellosis, the gold standard test to confirm the diagnostic of brucellosis are high agglutination titers of 1:160 for anti-*Brucella* antibodies using the standard tube agglutination (STA) test. In our case, it was very high (1:5120) [11].

However, the nucleic acid amplification assays NAATs, are more sensitive than conventional cultures and more specific than the serological tests for diagnosing human *Brucella* infection [12]. Unfortunately, most of these tests have been evaluated in only a small number of patients, and no extensive comparative studies of commercial kits and in-house PCR assays have been published [13] [14].

Pelvic inflammatory disease is low probable in our case. The patient is not sexually active, neither in the optimal range of age of PID and does not have any vaginal discharge, neither intrauterine contraceptive device.

The clinical presentation includes diverticulitis. the patient doesn't have any risk factors such as chronic constipation, age, a low fiber and high fat diet, obesity, smoking, exposure to non-steroidal anti-inflammatory drugs [15]. Furthermore, the MRI did not show any sign of diverticulosis nor diverticulitis with reported sensitivity of 86–94% and specificity of 88–92% [16,17].

This case represents a probable endometrioma sub infection, noting that the patient presented a 4.2 cm endometrioma on a previous ultrasound. To our knowledge, 8 cases of documented acute peritonitis due to *brucella* infection [18], and 2 cases of dermoid cyst subinfection were found in the literature.

Treatment of *brucella* infection is based on two-drug therapy, the most common combinations of antibiotics are: fluoroquinolones with rifampin or doxycycline with rifampin [19]. In this case, the laparoscopic surgical treatment was inevitable due to the acute peritonitis and the extreme pelvic pain in combination with the empiric antibiotics (piperacillin/tazobactam 4.5 g every 6 h, doxycycline 200 g one daily and ciprofloxacine 500 mg BID). The improvement of the clinical and biological markers with the final pathology report showing an infectious cause confirms the diagnosis of the *brucella* abscess. Due to the previous positive blood culture, cotrimoxazole was not used in the regimen.

In conclusion, acute peritonitis associated with a tubo-ovarian abscess is an unusual complication of brucellosis. The clinician should be aware especially in the endemic regions to establish a rapid diagnosis and an urgent treatment to this life-threatening situation. This approach requires adequate equipment and advanced laparoscopic surgical skills.

Authors' contributions

All authors have contributed equally in the literature search and redaction of this letter to the editor.

Financial & competing interests disclosure

The authors have no relevant affiliations or financial involvement with any organization or entity with a financial interest in or financial conflict with the subject matter or materials discussed in the manuscript. This includes employment, consultancies, honoraria, stock ownership or options, expert testimony, grants or patents received or pending, or royalties. No writing assistance was utilized in the production of this manuscript

CRediT authorship contribution statement

Bernard Najib: Conceptualization, Data curation, Formal analysis, Writing - original draft. **Wael Abdallah:** Conceptualization, Data curation, Formal analysis, Writing - original draft. **Elie Haddad:** Investigation, Methodology, Writing - review & editing. **David Atallah:** Project administration, Resources, Supervision, Validation, Visualization, Writing - review & editing.

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