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Recurrence of brucellosis on breast implants

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Keywords: Brucellosis; zoonosis; breast-implant; prosthesis infection; fever.

Background

Brucellosis is an endemic zoonosis, notably in the Middle East, commonly acquired through ingestion of unpasteurized contaminated milk or from direct contact with infected animals. Occurrence of a focal infection, including that of prosthetic devices, aggravates the disease prognosis. Here, we report the case of a 32-year-old woman with recurrence of brucellosis on breast implants two months after initial well-conducted treatment.

Case report

A 32-year-old woman, with history of bilateral breast implants 10 years prior to hospitalization and no other medical history or daily medication, developed daily fever, chills, myalgia and headache two months after travelling from France to Israel. While staying there for a 3-month trip, she reported eating raw dairy products. She consulted upon return to France with persisting symptoms and had a physical examination strictly normal. Initial laboratory tests showed serum aspartate aminotransferase level at 141 U/L and alanine aminotransferase level at 292 U/L (laboratory standards <32 IU/L) without cholestasis nor elevation of bilirubin and hepatic insufficiency. Blood cultures were positive for *Brucella melitensis* (identification using matrix-assisted laser desorption/ionization-time of flight mass spectrometry [MALDI-TOF]) and confirmed by the French reference center for brucellosis. The patient's sister, who had developed similar symptoms, was also diagnosed with brucellosis and treated in Israel.

Initial work-up did not reveal signs of endocarditis on transthoracic echocardiography (TTE), and contrast-enhanced body computed tomography imaging (CT-scan) did not show other infectious localizations (Figure 1A).

Fever resolved within three days and liver disorders quickly improved after introduction of doxycycline 200 mg/day and gentamicin 3 mg/kg/day for 7 days, then switched to doxycycline and rifampin 15 mg/kg/day for five weeks. Blood cultures, controlled at one week of treatment, remained

negative. The patient was symptom-free at the end of treatment.

Two months after the end of the antibiotic course, the patient complained of asthenia, anorexia, myalgia and left mastodynia followed by recurrence of fever and chills. On examination, she had bilateral axillary adenopathies without left breast abscess nor pus discharge. Blood analysis revealed liver enzymes in the normal range and blood cultures were positive for *B. melitensis*.

The CT-scan revealed bilateral infra- and supra-centimetric axillary adenopathies and an infiltration around the left breast implant (Figure 1B). Positron emission tomography (PET)/CT-scan revealed a diffuse and intense hypermetabolism (Standardized Uptake Value max 13.2) around breast implants with focal pseudonodular infiltrated areas and bilateral axillary adenopathies (Figure 1C). No sign of endocarditis was observed on TTE.

The patient received doxycycline and gentamicin for 10 days before she underwent a bilateral capsulectomy and removal of both implants. Next Generation Sequencing (NGS) and culture performed on the removed breast implants were negative. The pathology report revealed an inflammatory cell infiltrate without malignancy nor granuloma. Antibiotics were switched to doxycycline and rifampin, then modified to cotrimoxazole because of nausea, vomiting and dizziness, and were continued for 3 months.

Blood culture remained negative, and the patient was free of symptoms 10 months after the end of treatment. The patient did not consider undergoing new breast implant surgeries.

Discussion

Brucellosis affects half a million people annually [1]. After a phase of bacteremia revealed by fever, sweat, arthralgia, myalgia, headaches and lymphadenopathies, the disease can reach a chronic stage that may involve various organs, including breasts [1,2].

Diagnosis can be difficult. Sensitivity of blood cultures reaches 70-80% at the early stage of the disease but decreases to 20-45% in focal diseases [3]. In latter cases, the most commonly used

diagnostic methods are culture of the infected site (e.g. cerebrospinal fluid, joint fluid), even though often negative, and serological tests, even though lacking specificity [3]. Of note, molecular methods such as targeted Polymerase Chain Reaction (PCR) and NGS have also proved to yield a high sensitivity and specificity [3,4].

In our case, diagnosis of brucellosis recurrence was obtained on positive blood cultures. NGS on fixed breast tissue (transferred from the pathology laboratory) and breast implants culture remained negative probably because of the use of a 10 day-course of antibiotics prior to surgical procedure.

Although rare, *Brucella* prosthetic device infection can occur through hematogenous or lymphatic dissemination. Infections of prosthetic joints, prosthetic heart valves, ventricular defect patches, implantable cardioverter-defibrillators and breast implants have already been reported [5–9].

The first reported case of brucellosis breast implant infection was in a 48-year-old female laboratory technician living in Saudi Arabia who had relapsing symptoms with a right breast abscess after 2 months of a well conducted 6 week-treatment for acute brucellosis with doxycycline and rifampin [8]. Culture of blood and of the abscess were positive for *B. melitensis*. She underwent bilateral capsulectomy and a prolonged antibiotic therapy for 10 months.

The second case was of a 71-year-old female living in the United States of America who had developed recurring fever. Diagnosis was made upon removal of both breast implants, performed for hypotension and inflammation around her right breast implant on the second episode of fever [9].

Treatment of acute brucellosis relies on two regimens, both using doxycycline for 6 weeks, in combination with either streptomycin 1g/day for 15 days (streptomycin can be substituted by gentamicin 5 mg/kg/day for 7-10 days) or rifampin (600-900 mg/day) for 6 weeks. In case of a focal disease, the combination of doxycycline with rifampin (whether in association with gentamicin or not) is preferred and the duration of antibiotics is prolonged for a minimum of 3 months [5,10].

Treatment of device-related brucellosis is controversial but most centers practice a combination of medical and surgical therapy, whether on prosthetic valve endocarditis [5] or prosthetic joint

infections [10]. Conservative therapeutic strategies without removal of the devices have also been reported [5,10].

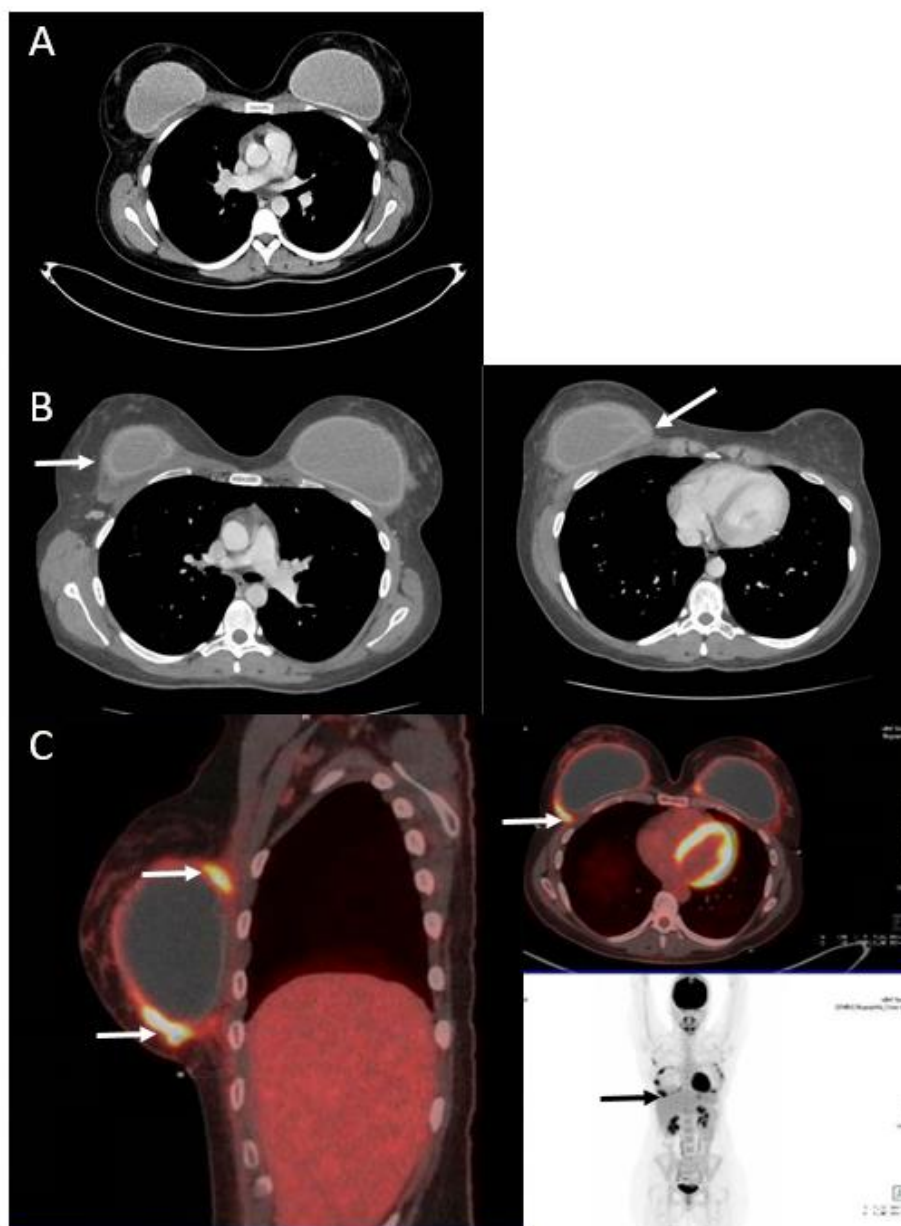
In our case, we considered this relapse of brucellosis as a breast-implant infection given the local clinical signs, the compared CT-scans showing the appearance of an infiltration around the left breast implant and a PET revealing focal pseudonodular intense hypermetabolism. No other localization, including endovascular graft, was diagnosed on PET. We opted for both a medical and surgical treatment as described for similar infections (prosthesis joint infections, endocarditis and breast implants).

To conclude, we reported a rare case of breast implant infection by *Brucella melitensis* in a young woman returning from a region of high prevalence of brucellosis. Performing an upfront PET/CT scan at the initial phase of diagnosis in patients with prosthesis and implants should be discussed to adjust the duration of treatment and to consider surgery. In any case, clinical follow-up and blood cultures drawn after the end of initial treatment can help physicians guide the therapeutic strategy, whereas follow-up serologies are not useful.

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Figure 1. A. Contrast-enhanced computed tomography imaging (CT-scan) on first *Brucella melitensis* bacteremia occurrence, with a normal aspect of the breast implants. B. Contrast-enhanced CT-scan performed during recurrence of *B. melitensis* bacteremia revealing infiltration around the left breast implant (white arrows). C. Positron emission tomography (PET) performed on *B. melitensis* bacteremia recurrence revealing a diffuse and intense hypermetabolism (Standardized Uptake Value max 13.2) around breast implants with focal pseudonodular infiltrated areas (black and white arrows).



Ethical Statement

The patient approved the report of her case.

Response to reviewers

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