

A 42-year-old female with sternoclavicular arthritis and breast abscess caused by brucellosis: a case report

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Abstract

Brucellosis is a thousand-face disease and a common zoonotic infection in the endemic region. A 42-year-old female was admitted with sternoclavicular arthritis and breast abscess. After laboratory investigation and imaging, positive serological test results and positive blood culture for brucella revealed acute sternoclavicular arthritis and breast abscess due to brucellosis.

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Abstract

Brucellosis is a thousand-face disease and a common zoonotic infection in the endemic region. A 42-year-old female was admitted with sternoclavicular arthritis and breast abscess. After laboratory investigation and imaging, positive serological test results and positive blood culture for brucella revealed acute sternoclavicular arthritis and breast abscess due to brucellosis.

Keywords: Arthritis, brucellosis, Abscess, sternoclavicular

Background

Brucellosis is one of the most common zoonotic diseases worldwide, especially in Asia and Middle Eastern countries such as Iran [1,2,3], which is transmitted to humans through the consumption of unpasteurized dairy products and direct contact with infected animal tissues [4,5,6]. It has been demonstrated that brucellosis can involve any part of the body including the musculoskeletal system, genital organs, gastrointestinal system, nervous system, respiratory system, circulatory system, skin, heart, liver, spleen, bone, kidney, brain, epididymis, ovary, and gallbladder [7,8,9] and it causes a systemic infection from asymptomatic disease to fatal illness [10,11] and it can mimic other infectious and non-infectious diseases [4,12]. So it is known as a “great imitator” [10].

The onset of the disease can be insidious and when specific organ involvement is detected it is defined as focal brucellosis [2,10,11]. In addition, arthritis, spondylitis, endocarditis, meningitis, orchitis due to brucellosis are not uncommon [10,11,13]; but cutaneous and subcutaneous involvement with brucellosis are uncommon [14]. Although soft tissue locations are usually related to a penetrating injury, the hematogenous spread is considered an important pathologic route [8,13,15].

Although breast infection with granulomatous mastitis is described in animals, infection of mammary glands with brucella is extremely rare in human brucellosis [13,14,15]. Brucella-infected mammary glands have been reported in a few cases and it is often hematogenous such as a metastatic abscess in endocarditis [11]. In one case I. Gasser et al. reported the isolation of *Brucella melitensis* from a suspected breast tumor of a woman who also had uveitis [13].

Osteoarticular lesions which are the most common complications in brucellosis include peripheral arthritis, osteomyelitis, spondylitis, sacroiliitis, and bursitis. Common joints consist of the knee, hip, ankle, and sacroiliac and less frequent ones include the shoulder, sternoclavicular, and temporomandibular [10,11,16,17].

Case Presentation

A 42-year-old Middle Eastern female patient from Iran was admitted to Sina Medical Research & Training Hospital because of the acute arthritis of the left sternoclavicular joint on September 24, 2019. She was a married woman with 2 children and was a hairdresser and tattoo artist. She had no history of specific disease except a history of penicillin allergy and uterine myoma surgery seven months before admission.

She developed pain in the left sternoclavicular joint with spread to the left neck, shoulder and submental area two weeks before hospitalization, which gradually intensified and led to neck pain and limited movement of the left upper limb due to pain and then erythema and heat and swelling were added to the sternoclavicular joint. The shoulder pain was reduced by using a slightly warm compress on it. She was hospitalized with a diagnosis of acute sternoclavicular joint arthritis owing to a lack of response to outpatient treatments and topical compounds.

One and a half months ago, because of discharge from the left breast ultrasound was done. The results revealed a fibrocystic change in both breasts, reactive axillary lymph nodes on both sides and a complicated cyst in the left breast which was completely aspirated under an ultrasound guide. Then, 5 ccs of concentrated pus were drained and sent to the culture, and the result was negative.

According to the abovementioned history and examination, the patient was admitted with the diagnosis of acute left sternoclavicular arthritis.

Over one and a half months, she had been taking co-amoxiclav, dexamethasone, celecoxib, piroxicam, Gabapentin, NEUROBION, cephalixin, and vitamin E. She had no fever, chills, headache, nausea and vomiting but she occasionally complained of scant purulent discharge from the left breast and a sense of heaviness in it. Examination of the heart, lungs and abdomen were normal and she had no skin rash. Limited movement of the left upper limb was due to pain in the left sternoclavicular region and this region had slight erythema and tenderness in the palm. The right breast was normal and the left was tense.

Her vital signs at admission were: Blood pressure: 110/70, Body temperature: 36.8 axillary, Pulse rate: 82, and Respiratory rate: 18. Cardiac echocardiography, chest and neck CT scan as well as liver and spleen

ultrasound were normal.

We started to study the cause of acute sternoclavicular arthritis. MRI of the left sternoclavicular joint was performed, there was subarticular bone marrow edema at both the clavicular and sternal sides of the left sternoclavicular joint as well as some surrounding deep soft tissue edema, in favor of osteoarthritis. No obvious bony erosion was detected.

Because of the left breast discharge ultrasound was conducted and the result showed a fibrocystic change of both breasts and a hypoechoic mass in the left breast of 6.3×4.7 cm. A core needle biopsy of a left breast abscess was performed and the reported biopsy by the pathologist was as follows: Breast tissue with moderate mixed inflammation, fibrocystic change, and foci of adenosis with florid hyperplasia. For definite diagnosis and ruling out of atypical florid hyperplasia Immunohistochemistry (IHC) staining for P63, SMA, CK5/6, and ER were recommended. IHC findings did not confirm atypical changes.

Breast abscess culture revealed few colonies of coagulase-negative staphylococcus.

Despite treatment with naproxen, pain and tenderness of the left sternoclavicular joint and limited movement persisted until getting serology of brucellosis (Wright: 1/160) and positive blood culture for *Brucella*. Consequently, the patient was treated with Streptomycin and doxycycline. Four days later she felt improvement in the left sternoclavicular joint. Pain and tenderness in the joint significantly decreased and the patient was able to move the left upper limb and the discharge from the patient's left breast was stopped. After the diagnosis of brucellosis and good response to treatment, on the 14th day of hospitalization, the patient was discharged and requested for continuing treatment order with doxycycline and Streptomycin for one week followed by doxycycline and rifampin and following-up on outpatient basis.

The laboratory workup was summarized in Table 1.

30 days after discharge, she had been in good clinical condition with normal sternoclavicular joint, and normal examination of the left breast without any discharge. Also, additional ultrasound of the breasts showed a fibrocystic change in both breasts without either evidence of collection or axillary lymph nodes.

Discussion

In this article, we reported a 42-year-old woman who had no history of livestock contact, sternoclavicular joint or left breast trauma, and also there were not any risk factors of sternoclavicular arthritis such as intravenous drug use, distant site of infection, diabetes mellitus, trauma, and infected central venous line. She had experienced purulent discharge from the left breast and underwent left breast abscess drainage and antibiotic treatment on an outpatient basis one and a half months before hospitalization, but there was no improvement so occasional discharge continued following abscess evacuation. Furthermore, two weeks before admission, pain and redness, and swelling of the left sternoclavicular joint were added. So she was hospitalized with the diagnosis of acute left sternoclavicular arthritis for further examination. MRI showed signs of left sternoclavicular arthritis. In addition, laboratory tests that were performed to rule out causes of arthritis, including rheumatoid arthritis and systemic lupus erythematosus, all were negative [2]. Since brucellosis is one of the causes of arthritis in endemic areas, blood culture and serology of brucellosis were requested. Blood culture × 2 was reported positive for brucellosis and Wright agglutination test:1/160, Coombs Wright test: 1/320, 2-mercaptoethanol (2ME): 1/80 in favor of brucellosis [5,10,11]. According to the patient's complaint about purulent discharge from the left breast and history of abscess drainage one and a half months ago, at the same time as we were performing diagnostic tests for left sternoclavicular arthritis, a breasts ultrasound was performed which showed a fibrocystic change of both breasts and a mass in the left breast then she underwent left breast abscess biopsy. The results of pathology and immunohistochemistry (IHC) were not in favor of malignancy. Breast abscess culture was reported as having few colonies of coagulase-negative staphylococcus which is considered as contamination because she did not have a fever, leukocytosis and other systemic signs also colony count was low.

Breast abscess due to brucellosis is extremely rare. So to rule out endocarditis and its metastatic abscess [11] to the breast and a sternoclavicular joint, transthoracic echocardiography was performed and its result

was normal, without any vegetation or valve abnormality.

Fever, chills, sweating, fatigue, headache, splenomegaly, hepatomegaly, arthralgia and musculoskeletal pain, which are seen in most patients with generalized and localized brucellosis [1,5,11], were not present in our patient and she also had no other positive findings except left sternoclavicular arthritis and left breast abscess.

As reported in other studies, there were no specific hematological or biochemical findings in our case [2,11,12]. There was no leukocytosis in this patient and liver function tests were normal but ESR and CRP were high.

As Since the diagnosis of brucellosis arthritis is confirmed by signs and symptoms of arthritis (pain, tenderness, swelling of the joint)[17,18] in the presence of an antibody titer greater than 1:160 in the tube agglutination test or by a positive culture [10,11], brucellosis was diagnosed in our patient and considering two very rare complications of brucellosis: left sternoclavicular arthritis and left bursal abscess both simultaneously in this patient due to the lack of history of trauma and external inoculation and because she had no history of direct contact with livestock, we concluded that these complications are hematogenous. Therefore, with the diagnosis of localized brucellosis, which requires a combination of antibiotics and a prolonged course of treatment to prevent failure or relapse of brucellosis [2,10,11], the patient was treated with a standard regimen (streptomycin and doxycycline for two weeks, then rifampin and doxycycline for ten weeks). On the fourth day of treatment, she responded and the joint symptoms improved significantly the discharge from the patient's left breast was stopped after starting the treatment and she was discharged and recommended to continue the outpatient treatment and follow-up. The prognosis of brucella arthritis will be very good if appropriate and in time treatment is begun [12,19]. Unlike spondylitis, both sacroiliitis and peripheral arthritis are nondestructive and quickly curable with no sequelae [2,12].

Thirty days after discharge at the follow-up visit, the patient did not relate a feeling of stiffness or heaviness in the left breast and occasional discharge from it which she had previously experienced, and also left sternoclavicular arthritis completely was cured. She completed treatment of two localized complications of brucellosis simultaneously (sternoclavicular arthritis and left breast abscess) without any complications.

Each of the complications of sternoclavicular arthritis and Brest abscess due to brucellosis is solely a very rare complication of brucellosis. However, we found both of these rare complications simultaneously in one patient. Then, we could successfully treat her using the standard treatment of streptomycin and doxycycline for two weeks followed by rifampin and doxycycline for ten weeks.

Given that brucellosis is a thousand-face disease, especially in endemic areas, clinicians in all regions of the world, especially endemic areas should be familiar with rare complications of it so we reported this patient with very rare complications of this zoonotic disease and its successful treatment.

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Availability of data and materials

All data and materials related to this report are accessible at any time upon request.

Ethics approval and consent to participate

Ethics approval code: IR.TBZMED.REC.1399.1024

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Conflict of interest disclosure

The authors declare that they have no conflict of interest.

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Value	Parameter
9.9 × 10 ³ /μl (Neutrophils =68.2%, Lymphocytes =27.5%)	White blood cells
11.4 g/dl	Hemoglobin
250000 /mm ³	Platelets
13.8	Prothrombin time
35	partial thromboplastin time
1.2	INR
26 mg/dl	urea
0.8 mg/dl	Creatinine
86	Blood Sugar
138 mg/dl	Na

Value	Parameter
4.5 mg/dl	K
Total=8.1 mg/dl; Ionized=1 mg/dl	Calcium
3.2 mg/dl	Phosphorus
4.3 g/dl	Albumin
0.2 mg/dl	Uric acid
17 IU/L	Aspartate aminotransferase
24 IU/L	Alanine aminotransferase
232 IU/L	Alkaline phosphatase
0.34 mg/dl	Total bilirubin
Negative	HIV-Ab
37 U/L	Creatine kinase (CK)
Negative	antinuclear antibodies (ANA)
Negative	Rheumatoid factor (RF)
Negative	Anti-cyclic citrullinated (Anti CCP)
53 mm/h	Erythrocytes sedimentation rate (ESR)
36 mg/dl	C-reactive protein (CRP)
299 IU/l	Lactate dehydrogenase
1/160	Wright
1/320	Coombs Wright
1/80	2 mercaptoethanol (2ME)
Brucella was isolated on Castaneda medium after 7 days	Blood Culture × 2
Few colonies of coagulase negative staphylococcus	Breast abscess culture
Normal	Urine analysis
Negative	Urine culture

Table 1: Laboratory Tests during hospitalization; INR: international normalized ratio; HIV Ab: Human Immunodeficiency Virus-antibody.