

Case Report

A Case of Brucellosis with Sternoclavicular Arthritis and Biceps Tenosynovitis

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ABSTRACT

Brucellosis is a zoonosis that affects several organs or systems. Large joint involvements are more commonly observed in brucellosis. However, small joint involvements are also seen and sternoclavicular arthritis, tendinitis, bursitis and tenosynovitis are rare complications of brucellosis. Here,

we present a case of brucellosis with sternoclavicular joint involvement and complicated by supraspinatus tendonitis and biceps tenosynovitis. Brucellosis should be included in the differential diagnosis of longstanding small joint pain in regions where brucellosis is endemic.

KEY WORDS: osteoarticular complications, small joint pain, sternoclavicular arthritis, tenosynovitis

INTRODUCTION

Brucellosis is still endemic in many of the countries in the world. Although large joint involvements are more commonly observed, peripheral joint involvements and rare complications such as sternoclavicular arthritis, tendinitis, bursitis and tenosynovitis are also seen^[1,2]. Here, we will present a case of brucellosis with sternoclavicular joint involvement and complicated by supraspinatus tendonitis and biceps tenosynovitis.

CASE REPORT

A 43-year-old male patient presented to the Infectious Diseases Outpatient Clinic with complaints of swelling on the left hand side of the neck, shoulder pain, low-grade fever and sweating. His complaints began 6 months ago. He presented to various polyclinics including orthopedics, he used non-steroidal anti-inflammatory and muscle relaxant drugs but the complaints were not resolved. In his history, habitation in a rural area, animal breeding and consumption of raw milk were noted.

On physical examination, he was afebrile and there was a non-fluctuating hyperemic edematous, mild

mass in size of 4 x 4 cm at the level of left sternoclavicular joint and there was restriction of movements in the left shoulder (Fig 1). Hepatosplenomegaly was not detected.

Laboratory values of the patient were determined as followings: white blood cell: 7500/μl, sedimentation rate: 20 mm/h, C-reactive protein: 14.7 mg/L. Rose-Bengal lam agglutination test (Seromed, Turkey) was positive and Standard tube agglutination test (Linear chemicals, S.L., Spain) for brucellosis was positive at a titer of 1:160. *Brucella melitensis* was detected in blood culture. Bact/ALERT FAN blood culture bottles (aerobic and anaerobic) for the Bact/ALERT® (bioMérieux, France) automated sensor-metric system were used, inoculated with 5–10 ml of patient's venous blood at the hospital departments. We took four blood bottles for culture. All bottles were incubated under continuous agitation and monitored for up to 7 days or until they became positive, depending on diagnosis.

The increased amounts of CO₂ produced by the bacterial growth diffuses through a semi-permeable membrane in the base of the culture bottle and reacts with water-generating hydrogen ions. The pH

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Fig 1: A non-fluctuating hyperemic edematous area with a size of 4 x 4 cm at the level of left sternoclavicular joint

decrease in the bottle results in the colour change of a built-in sensor. Reflectance values from the sensor of each culture bottle are monitored and analysed with a complex algorithm which allows differentiation of microbial from background CO₂ produced by other components in the blood.

Four of the blood culture bottles became positive. The mean detection time for *B. melitensis* was 4.5 days. A gram stain was performed with the subculture on Columbia agar medium incubated at 37 °C from the flagged positive bottle. All the procedures were carried out routinely in conventional laboratory conditions, and safety cabinets were not used. Isolated strains on Columbia agar were identified by the VITEK 2 system using gram negative cards and the strains were identified as *Brucella melitensis*.

Susceptibility testing for all isolates was done by a disk-diffusion assay on Mueller–Hinton agar in a rich CO₂ atmosphere. The following antibiotic disks (Oxoid, UK) were used: Beta lactames-amoxicillin/clavulanic acid (20 µg/10 µg), piperacillin (75 µg), piperacillin/tazobactam (75 µg/10 µg), imipenem (10 µg), ceftriaxone (30 µg), cefotaxime (30 µg), ceftazidime (30 µg), cefepime (30 µg); Aminoglycosides – gentamicin (15 µg), rifampicin (30 µg), amikacin (30 µg); Fluoroquinolones – pefloxacin (5 µg), ofloxacin (5 µg), ciprofloxacin (5 µg), rifampicin (30 µg); Tetracycline – tetracycline (30 µg); and trimethoprim/sulfamethoxazole (1.25 µg/23.75 µg).

The results from the disk diffusion method of susceptibility testing showed high level sensitivity to all examined antibiotics: amoxicillin, amoxicillin/clavulanic acid, piperacillin, piperacillin/tazobactam, imipenem, ceftriaxone, cefotaxime, ceftazidime, cefepime, gentamicin, amikacin, rifampicin, pefloxacin, ofloxacin, ciprofloxacin, trimethoprim/sulfamethoxazole and tetracycline. The VITEK 2 system was not able to show the antibiotic susceptibility of the *Brucella* strains with any of the available cards.

Cutaneous and subcutaneous edema was detected by ultrasonography. Magnetic resonance imaging (MRI) of the left shoulder revealed supraspinatus tendinitis and tenosynovitis of the biceps tendon. MRI of the neck showed thickening, signal changes and inflammatory process with contrast agent enhancement at the sternal notch of left sternoclavicular joint. The patient was administered a combination therapy of doxycycline (2x100 mg/day) + rifampicin (1x600 mg/day) with the diagnosis of brucellar sternoclavicular joint involvement and complicated by supraspinatus tendonitis and biceps tenosynovitis. Since no remission occurred in the complaints of the patient after 3 weeks of treatment, ciprofloxacin 2x500 mg/day was added to the treatment. The lesion regressed completely after one week of the combination therapy and the symptoms were markedly decreased. The combination treatment was continued for 6 weeks. Progressive improvement in the MRI appearances and final resolution was observed at the end of 3-months' follow-up without any further treatment.

DISCUSSION

Osteoarticular complications are reported in 10 - 80% of the patients in case series of brucellosis. Bone and joint lesions are arthritis, spondylitis, osteomyelitis, tenosynovitis, and bursitis. Sacroiliitis and spondylitis are the most commonly reported complications^[1,2]. While peripheral joint involvements are generally seen in weight-bearing joints like hip and knee, they can also be seen in the small joints^[3].

Sternoclavicular joint involvement is encountered as a rarely seen complication (1 - 2%)^[4-6]. When the rates reported in the literature are investigated, the number of cases is quite low. Sternoclavicular joint involvement was observed at a rate of 7% in a large-scale case series including 1729 patients^[3]. Also, when the series including 144, 452, and 511 patients were investigated, sternoclavicular joint involvement was determined at rates of 0.7%, 1.8%, and 0.8%, respectively^[3,4,7-9]. Brucellosis has a wide range of clinical forms and diagnosis may be delayed in some cases. While the clinicians suspect brucellosis more particularly in the cases with large joint involvement,

small joint involvement in brucellosis may take prolonged time until diagnosis as in our case. As in our case, the patient was admitted to various polyclinics including orthopedics after onset of complaints. Non-steroidal analgesics and anti-inflammatory drugs were given to the patient but his complaints continued. Hence the patient was admitted to infectious disease polyclinic 6 months after onset of complaints.

In treatment, we added ciprofloxacin to the combination therapy and a quick response was obtained. It was observed that the lesion regressed completely after one week of triple therapy.

In the literature, there are treatments performed with triple therapy regimens in similar cases^[10-13]. Similar to our case, streptomycin + doxycycline was started for a 26-year-old man with brucellar sternoclavicular arthritis and the patient failed to respond to the therapy. The treatment was changed to a combination therapy of doxycycline + rifampicin + ofloxacin and treatment success was obtained^[13]. Also, we preferred to add ciprofloxacin to the present antibiotherapy for better joint penetration.

CONCLUSION

Large joint involvements are more commonly observed in brucellosis. However, small joint involvements are also seen. Brucellosis has a wide range of clinical forms and diagnosis may be delayed in some cases. While the clinicians suspect brucellosis more particularly in the cases with large joint involvement, small joint involvement in brucellosis may take prolonged time until diagnosis, as in our case. Brucellosis should be included in the differential diagnosis of long-standing small joint pain in regions where brucellosis is endemic.

REFERENCES

1. Young EJ. *Brucella* species. In: Mandell GL, Bennett JE, Dolin R, editors. *Mandel, Douglas and Bennett's Principles and Practise of Infectious Disease*. Churchill Livingstone Press; 2010. p 2921-2926.
2. Colmenero JD, Reguera JM, Martos F, *et al*. Complications associated with *Brucella melitensis* infection: a study of 530 cases. *Medicine* 1996; 75:195-211.
3. Berrocal A, Gotuzzo E, Calvo A, Carrillo C, Castañeda O, Alarcón GS. Sternoclavicular brucellar arthritis: a report of 7 cases and a review of the literature. *J Rheumatol* 1993; 20:1184-1186.
4. Andriopoulos P, Tsironi M, Deftereos S, Aessopos A, Assimakopoulos G. Acute brucellosis: presentation, diagnosis, and treatment of 144 cases. *Int J Infect Dis* 2007; 11:52-57.
5. Hashemi SH, Keramat F, Ranjbar M, Mamani M, Farzan A, Jamal-Omidi S. Osteoarticular complications of brucellosis in Hamedan, an endemic area in the west of Iran. *Int J Infect Dis* 2007; 11:496-500.
6. Rahmdel K, Golsha R, Golshah E, Shirazi RR, Momtaz NS. Chest wall involvement as a manifestation of brucellosis. *J Glob Infect Dis* 2011; 3:86-88.
7. Mousa AR, Muhtaseb SA, Almudallal DS, Khodeir SM, Marafie AA. Osteoarticular complications of brucellosis: a study of 169 cases. *Rev Infect Dis* 1987; 9:531-543.
8. Ural O, Satılmış Ö, Kaya S, Kreşi D, Dikici N. Shoulder Tenosynovitis due to Brucellosis. *The Journal of Infectious Diseases and Clinical Microbiology* 2011; 16:127-130.
9. Mousa AM, Muhtaseb SA, Al-Mudallal DS, Marafie AA, Habib FM. Brucellar sternoclavicular arthritis, the forgotten complication. *Ann Trop Med Parasitol* 1988; 82:275-281.
10. Doğan M, Hakyemez IN, Akkoyunlu ME. Case of brucellar costochondritis. *Journal of Harran University Medical Faculty* 2009; 6:23-24.
11. Wong TM, Lou N, Jin W, Leung F, To M, Leung F. Septic arthritis caused by *Brucella melitensis* in urban Shenzhen, China: a case report. *J Med Case Rep* 2014; 8:367.
12. Skalsky K, Yahav D, Bishara J, Pitlik S, Leibovici L, Paul M. Treatment of human brucellosis: systematic review and meta-analysis of randomised controlled trials. *BMJ* 2008; 336:701-704.
13. Gutierrez Ruiz C, Miranda JJ, Pappas G. A 26-year-old man with sternoclavicular arthritis. *PLoS Med* 2006; 3:e293.