A case of brucellosis presenting with mastoiditis and hearing loss

Yeşim Alpay1, Arzu İrvem2, Haşmet Yazıcı3

1Department of Infectious Disease and Clinical Microbiology, Balıkesir University School of Medicine, Balıkesir,
2Department of Microbiology and Clinical Microbiology, Umraniye Training and Research Hospital, İstanbul,
3Department of Ear-Nose-Throat Diseases, Balıkesir University School of Medicine, Balıkesir, Turkey

Abstract
Brucellosis is a zoonotic, systemic infection which can involve any organ or system of the body. It is necessary to investigate in etiology of fever unknown. The signs and symptoms of Brucella infections can mimic those of many other diseases. Atypical findings may be seen in chronic disease and diagnosis and treatment may be delayed. In this case report, we presented a case of brucellosis with an atypical course with lymphadenitis, mastoiditis and conductive hearing loss. It is emphasized that brucellosis may have a course with atypical clinical presentations. Early diagnosis and requirements of a change in treatment are important. Furthermore, although neurosensory hearing loss can be seen in brucellosis, in our case, hearing loss was determined to be a conductive type and temporary. Therefore it is different from cases in the literature.

Keywords
Brucellosis; Mastoiditis; Hearing Loss; Atypical Presentation
Introduction

Brucellae are Gram-negative non-spore-forming and non-motile coccobacilli. *Brucella melitensis* is the most common strain cause of disease in humans. It is primarily the disease of plant-eating animals [1]. The reservoirs of brucellosis are various wild and domestic mammals such as cattle, goat, sheep, pig, deer, wolf, and dog. The pathogen can be transmitted through respiratory tract from these animals to humans, by ingestion of contaminated animal products, by contact with damaged skin or mucosa. Transmission is most commonly seen by ingestion of not boiled, unpasteurized milk and dairy products. Half a million new human cases are determined worldwide annually, and it is an important infectious disease observed as hyperendemic in the Mediterranean basin, Middle East, and South America. The incubation period of *Brucella* infection is 1-8 weeks. Bacteria are inactivated within 10 minutes at 60º C. It is sensitive to heat inactivation and pasteurization. Brucellosis is a zoonotic, systemic infection which can involve any organ or system of the body. It is necessary to investigate in etiology of fever unknown. The signs and symptoms of *Brucella* infections can mimic those of many other diseases. Atypical findings may be seen in the chronic disease. Diagnosis and treatment may be delayed [2]. Here, we presented a case of brucellosis with an atypical course with lymphadenitis, mastoiditis, and conductive hearing loss.

Case Report

A 33-year old male patient was admitted to infectious disease clinic with complaints of subfebrile body temperature, lymphadenitis and pain in the neck and temporal region, myalgia, arthralgia, loss of appetite, weight loss and hearing loss in the right ear for the previous 4 months. The patient applied to the hospital more than once; antibiotics were prescribed, but they did not treat the patient. It was learned that he consumed unpasteurized dairy products.

On physical examination, the signs were determined; fever of 37.8ºC, right cervical lymphadenitis (in size of 4x5cm) and severe sensitiveness. There were no symptoms of the central nervous system. In the otorhinolaryngologic examination performed for hearing loss; otitis media with effusion of the right ear and mastoiditis were diagnosed in computed tomography (CT) of the temporal bone (figures 1,2).

The routine laboratory tests were determined as follows; CRP: 18.4 mg/L (0-5), erythrocyte sedimentation ratio:25 (1.h:0-15), WBC:6.9x10³ K/ul, amylase:34 U/L (28-100), lipase:27 U/L (0-67), ALT: 96 U/L (0-50), AST:69U/L (0-50), anti-rubella IgG: positive, anti-rubella IgM: negative, toxoplasma antibody IgG and IgM: negative, salmonella gruber widal tests: negative, tularemia tube agglutination test: negative, PPD test: negative, brucella standard tube agglutination test:1/320 positive. No bacterial growth in blood culture was seen. Doxycycline (100 mg 2x1 po) and rifampicin (300 mg 1x2 po) treatment was initiated for the patient. During follow-ups of the patient, it was learned that the pain and swelling described in the region of the mastoid bone didn't recover completely. Hearing loss recovered completely after 2 weeks of brucellosis treatment. It has been considered that mastoiditis findings of the case were associated with brucellosis and temporary hearing loss in the same region of the lesion was associated with edema due to infection/inflammation and these recovered with present antibiotic treatment. Treatment was continued by adding ciprofloxacin (750 mg 2x1 po) treatment to combination treatment with doxycycline (100 mg 2x1 po) and rifampicin (300 mg 1x2 po). Reduction in sedimentation rate (3/1.h) and CRP (3.28 mg/L) values were observed. A regression of mastoiditis was determined in control CT. The auditory test resulted in a normal hearing. Treatment continued for 3 months.

Discussion

Brucellosis has various symptoms and findings, it may be confused with many diseases. The most important symptoms are fever, arthralgia/arthritis, and hepatosplenomegaly and these are accompanied by a headache, abdominal pain, weight loss, fatigue, and sweating [3]. Disease presentation of our case was subfebrile body temperature, pain, and lymphadenitis in the neck and temporal region, myalgia, arthralgia, loss of appetite, weight loss, hearing loss in the right ear and failure of the antibiotics administered before diagnosis. In the further evaluation, the case was determined to be complicated with mastoiditis.
and otitis media with effusion and hearing loss. Osteoarticular symptoms are most frequently seen among brucellosis complications. Sacroilitis, spondylitis, arthritis, osteomyelitis, bursitis, and tenosynovitis are among them. In this case; brucella standard tube agglutination test was 1/320 positive and sedimentation rate, and CRP levels were elevated. No bacterial growth in blood culture of our case. Blood cultures might be negative. Diagnosis of brucellosis can be difficult, especially in non-endemic regions. Serological diagnosis usually with standard agglutination tests (SAT) is important in endemic regions. The sensitivity of blood cultures ranges are between 53% and 90%, and it requires prolonged periods of incubation [10]. Therefore, serology is important in the diagnosis. During follow-ups of the patient, the pain and swelling described in the region of the mastoid bone didn’t recover completely, but hearing loss recovered completely within the 2. week of treatment. Otitis media with effusion and hearing loss have been attributed to mastoiditis findings due to brucellosis. There wasn’t any symptoms and findings of neurobrucellosis. Temporary conductive hearing loss was determined in our case and it was associated with inflammation and edema due to infection. Neurobrucellosis, peritonitis, pericarditis and pancytopenia are rarely seen clinical manifestations of brucellosis [4]. Neurobrucellosis may develop in any stage of the course of disease and variable clinical manifestations such as encephalitis, meningoencephalitis, radiculitis, myelitis, peripheral and cranial neuropathies, and psychiatric findings can be seen [5]. Generally, hearing loss is seen in neurobrucellosis [6-8]. In our case, without findings of the central nervous system, hearing loss associated with mastoiditis alone was observed and recovered within the 2. week of treatment. External otitis, myringitis, acute suppurrative otitis media, otitis media with effusion, meningitis, mumps, toxoplasmosis, brucellosis, syphilis can cause acute onset hearing loss. Relapse of brucellosis is commonly seen with monotherapy and therapeutic failure is well known. In a meta-analysis performed, triple treatment for longer than 12 weeks is recommended in complicated brucellosis. In the treatment for less than 6 weeks, therapeutic failure and relapse of brucellosis were seen to be higher. Especially in osteoarticular involvement, the combination of aminoglycoside/doxycycline has lower relapse rate compared to the combination of rifampicin/doxycycline. In a meta-analysis performed, therapeutic failure or relapse of brucellosis was seen at a rate of 5-7% for doxycycline-streptomycin regimens and 11-17% for doxycycline-rifampicin regimens [9, 10]. In our case, after diagnosis of mastoiditis, ciprofloxacin 750 mg 2x1 po has been added to the doxycycline 100 mg 2x1 po + rifampicin 300 mg 1x2 po treatment. During follow-ups, therapeutic response was obtained. Regression in mastoiditis findings were determined in control CT, and a normal hearing test result were observed on the 8th week of treatment. Treatment was given for 6 weeks. Brucellosis is an infectious disease that has a good prognosis with early diagnosis and treatment, but the delay of diagnosis and treatment can occur in cases with atypical presentation. Since the cases may have an atypical course, they should be evaluated regarding complications in the presence of localized findings. Here it is emphasized that brucellosis may have a course with atypical clinical presentations such as lenfadenitis, mastoiditis and hearing loss. Early diagnosis and requirements of a change in treatment are important. Furthermore, although neurosensorial hearing loss can be seen in brucellosis, in our case, hearing loss was determined to be a conductive type and temporary. Therefore, it is different from cases in the literature.

Scientific Responsibility Statement
The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

Animal and human rights statement
All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. No animal or human studies were carried out by the authors for this article.

Conflict of interest
None of the authors received any type of financial support that could be considered potential conflict of interest regarding the manuscript or its submission.

References

How to cite this article: