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CASE REPORT

Distal femur Brucella osteomyelitis in infancy: A rare case report

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ABSTRACT

Brucella disease is an infectious disease caused by Brucella bacteria. It is transmitted through the consumption of unpasteurized dairy products and undercooked meat and penetration through the skin of individuals in contact with farm animals. A detailed medical history is of utmost importance in the diagnosis. Headache, cyclical fever, sweating, vomiting, abdominal pain, and wandering arthralgia are among the main clinical symptoms. Brucella infection is usually characterized by inflammation in the musculoskeletal system, and osteomyelitis is rarely seen. In this article, we report a case of osteomyelitis after neglected brucellosis.

Keywords: Brucella, infection, osteomyelitis.

Brucella is a disease common worldwide and caused by the Brucella family of bacteria. Consumption of contaminated dairy products, contact with infected animals, and inhaling aerosolized bacteria are the main transmission routes. The characteristics of Brucella infection are not specific, and different clinical pictures are observed from person to person. Fever, arthralgia, sweating, weight loss, lymphadenopathy, or hepatosplenomegaly are the main symptoms.[1] The diagnosis is usually missed, since the physical examination and clinical picture can be more specific. The prognosis for Brucella disease is usually excellent. The possibility of complications is extremely high in patients with delayed diagnosis after skeletal system involvement and in patients who are not treated appropriately.

Osteomyelitis or septic arthritis may be seen, particularly in patients with bone involvement. patients with septic arthritis may present variable blood and synovial parameters. Making decision based on the commonly used synovial white blood cell (WBC) count cut-off value of 50,000/mm^3 may lead to misdiagnosis. To avoid misdiagnosis or delay in treatment, it is of utmost importance not to exclude the diagnosis acutely and suspicion of septic arthritis should remain even with unlikely values. Clinical follow-up of these patients should be continued and culture results should be followed.[2]

In this article, we present a case of osteomyelitis after neglected brucellosis.

CASE REPORT

An 18-month-old female patient was admitted to the emergency department with the complaint of limping. Her medical history revealed that the patient did not step on her left side for about 20 days and had a fever occasionally. The family reported that they were examined by an orthopedic surgeon two weeks ago with these complaints and discharged with antibiotics and painkillers. The patient was admitted to our clinic, when her complaints persisted. Orthopedic examination of the patient without fever revealed minimal effusion

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in the left knee. Left knee joint movements were almost complete but painful. Suprapatellar and intraarticular punctures were performed to evaluate possible pathologies, but no material was obtained. Knee radiographs revealed effusion and edema in the joint (Figure 1). The patient's general condition was good, consciousness was clear, and no pathology was found in other system examinations. Blood tests revealed a WBC of 5.75 10^3/uL and C-reactive protein (CRP) of 15 mg/L. When the anamnesis was examined in detail, it was learned that the family was engaged in animal husbandry, and the mother was being treated for Brucella arthritis. The patient was hospitalized in the ward for further investigation and treatment. Brucella agglutination test was positive during the follow-up in the ward.

Magnetic resonance imaging (MRI) of the knee was performed for detailed evaluation. It revealed a lesion medial to the distal epiphysis of the left femur, which was hypointense in T1A sequences, markedly hyperintense in T2A sequences, and showed marked contrast uptake after contrast agent administration (Figure 2). Osteomyelitis was detected in the distal femur, and the patient was operated on for debridement. Brucella bacteria were grown in the blood culture taken preoperatively and in the deep tissue culture taken during surgery.

The patient was hospitalized in the ward for Brucella osteomyelitis and treated with ampicillin and amikacin for 18 days. On Day 19 of admission, the pediatric infection department changed the antibiotic treatment to rifampicin, trimethoprim-sulfamethoxazole, and gentamicin. Treatment was completed on Day 45 of hospitalization, the clinical picture improved, and the patient was discharged with appropriate recommendations.

One year later, a control radiograph showed irregularities in the distal epiphyseal region of the femur (Figure 3). Based on MRI findings and radiographs, the findings regressed. Clinical



FIGURE 1. Two-way knee radiograph of the patient. The lateral radiograph shows edema and effusion around the knee.

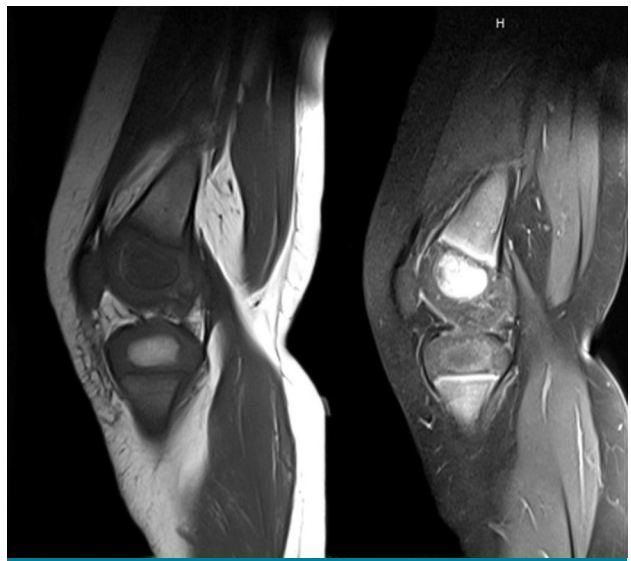


FIGURE 2. T1-T2 sagittal knee magnetic resonance images of the patient. The medial aspect of the distal epiphysis of the patient's left femur shows hypointense areas on T1A sequences and hyperintense areas on T2A sequences.

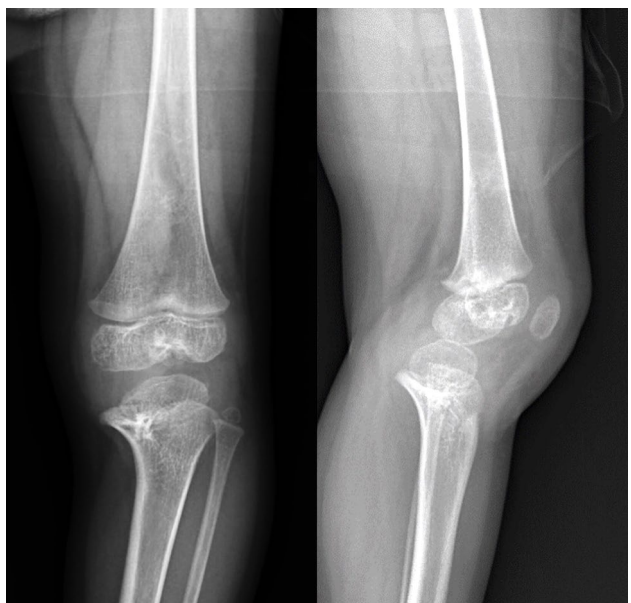


FIGURE 3. Two-way knee radiographs of the patient at postoperative 1st year. The radiograph shows irregularities in the distal epiphyseal region of the patient's femur.

follow-up was continued and, in the first postoperative year, tapering in the medial metaphyseal region of the left tibia and irregularities in the distal epiphyseal region of the femur were detected. Knee joint movements were evaluated as usual.

DISCUSSION

Brucella disease is more common in developing countries and is a significant public health problem. It is more common in children than in adults. The disease progresses insidiously, and the clinical picture can mimic many diseases. Clinical symptoms and laboratory findings vary, thereby leading to delay in the diagnosis. The disease usually presents with fever, arthralgia, and hepatosplenomegaly in children. Blood biochemistry tests may show average white blood cell count, slightly elevated sedimentation, and liver enzymes. Serological tests (Wright and Rose-Bengal tests) are used in the diagnosis. However, the definite diagnosis is made by the production of the agent in blood or bone marrow material.[3]

Radiological imaging has an essential place in the evaluation of musculoskeletal involvement. Sacroiliitis, discitis, and spondylitis are frequently seen in musculoskeletal involvement. Sacroiliac joint involvement is rarely seen in children. Brucella infection is usually characterized by inflammation in the musculoskeletal system, and osteomyelitis is seldom seen. Vertebral osteomyelitis is a vital complication, particularly in patients with psoas abscess. Calcaneus,[4] femur,[5,6] and pubis[7] osteomyelitis after Brucella have also been reported in the literature. Direct radiographs are usually normal in the first two or three weeks. In the following period, bone sclerosis and destruction may be observed. In the early period, MRI and bone scintigraphy helped make the diagnosis. On MRI, the lesion appears hypointense on T1 sequences and hyperintense on T2A sequences. Scintigraphy shows marked radioactive material uptake in the area of osteomyelitis. However, radiographic changes are not specific to Brucella osteomyelitis. Pyogenic osteomyelitis and childhood bone tumors should be considered in the differential diagnosis. The diagnosis may sometimes be delayed, or patients may be misdiagnosed due to sclerosis and destruction of the bone. Detailed anamnesis, evaluation of infection parameters, and serological tests are essential in excluding malignant bone tumors.

In the literature, there are reports that peripheral skeletal system involvement is as joint as vertebral involvement.[8-10] Arthritis, arthralgia, bursitis, or tenosynovitis may be observed in patients after Brucella due to skeletal system involvement. Brucella should be considered in the differential diagnosis after arthritis, particularly in pediatric patients living in endemic areas. In the initial period of the disease, traveling polyarthritis accompanied by fever is observed. However, some patients may have progressive monoarthritis or destructive septic arthritis. In children, monoarthritis, usually involving the hip or knee joint, is the most common type of brucellosis.[11]

After a detailed anamnesis and a detailed physical examination, serological tests are critical in diagnosing Brucella. Standard agglutination testing is the most common method of diagnosis in endemic areas. Polymerase chain reaction (PCR) tests are also used for rapid diagnosis.[12] Culture is the gold-standard diagnostic method. Samples taken from cases with suspected Brucella should be cultured within two hours; if cultivation cannot be performed within two hours, the materials taken should be stored at 4 to 10°C.[13] The main goal of treatment is to eliminate symptoms and prevent possible relapses. Treatment is classified under two main headings: medical and surgical. Antibiotics are the mainstay of medical treatment. However, surgical treatment is inevitable in endocarditis, pyogenic joint involvement, and paraspinal abscesses.[12-15]

Although controlling the symptoms after Brucella disease takes time, the prognosis is usually excellent. Recurrence is also infrequent. Complications are rare, if patients are treated timely and appropriately. However, pediatric patients, particularly those with joint involvement, are not treated appropriately and the rate of focal complications may increase in those patients with longer symptom duration.[16] In that case, gait disorders due to cartilage destruction and bone destruction may occur in the future due to deformity of the bones.[17]

Raw milk consumption and contact with animals are the main risk factors for Brucella disease in children. Sawafi et al.[18] reported that 10% of children developed osteomyelitis/septic arthritis in a study of 57 cases in Oman. Buzgan et al.[19] reported osteoarticular involvement in 25% of cases in a study of 1,028 patients. Again, Shaalan et al.[20] reported osteomyelitis in 8.5% of patients in a study of 115 cases. In addition, Tsolia et al.[21] reported femoral head subluxation due to arthritis after Brucella infection.

In conclusion, a detailed history and a thorough physical examination are crucial in diagnosing

Brucella disease. The clinical manifestations of the disease are very variable and can range from an asymptomatic infection to a severe clinical picture. There is no pathognomonic finding to make a diagnosis of Brucella disease. However, fever, malaise, peripheral arthritis, splenomegaly, and hepatomegaly are among the most common clinical signs and symptoms. Brucella disease should be considered in the differential diagnosis in patients with faint physical examination findings and serologic tests should confirm the diagnosis. In particular, in cases with bone involvement, there may be delays in diagnosis, which may lead to gait disorders in the future with cartilage and bone destruction.

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Author Contributions: Idea, study design, figures, control/supervision, analysis and/or interpretation: A.Y.; Data collection, materials, literature review, critical review: A.Y., M.C.A.

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