Case Report
J Immunol Clin Microbiol. 2020;5(4):115-122

J Immunol Clin Microbiol

A Rare Case Report in the Childhood Age Group: Salmonella Osteomyelitis of the Radius

Yaşar Dinçel[1], Abdülkadir Sarı[1], Mehmet Ümit Çetin[1], Melih Güney[1], Burak Günaydin[1], Erdem Can[1*]

1 Department of Orthopedics and Traumatology, Faculty of Medicine, Namık Kemal University, Tekirdağ, Turkey.

Abstract:

Introduction: Salmonella osteomyelitis is a rare condition predominantly seen in hemoglobinopathies such as sickle cell anemia or thalassemia. Very few cases have been reported in the literature in children without predisposing factors. Here, we present a case of Salmonella osteomyelitis developing in the radius of an immunocompetent 6-year-old child without any known predisposing conditions.

Case Report: A 6-year-old patient applied to the pediatric emergency department with complaints of pain in the left forearm and 39.5 degrees Celcius fever, which has been going on for 20 days after the fall. On radiographs, lytic lesions were detected along the Radius and an infiltrative type lesion was observed in MRI. Samples were taken for bone culture and pathological examination under general anesthesia in the operating room due to suspicion of osteomyelitis or malignant lesions. There was very intensive Salmonella species growth in culture. The patient was given 400 mg vancomycin intravenously every 8 hours, and 1 g ceftriaxone intravenously every 12 hours for 3 weeks after the pediatric consultation. The patient was followed up on an outpatient basis, with 300 mg of oral cefuroxime treatment every 12 hours for 2 weeks. MRI and X-Ray imaging was performed at 6th postoperatively. It was observed that the lytic lesion decreased significantly in the radius. On the 14th postoperative month, MRI imaging showed that the lesion disappeared.

Conclusions: Rare pathogens should be considered in the differential diagnosis of patients with prolonged insidious symptoms and appropriate bacterial cultures should be taken for specific antibiotic therapy.

Key words: *Osteomyelitis, salmonella, child, radius*

***Corresponding Author:** Erdem Can. Namık Kemal Üniversitesi Tıp Fakültesi, Ortopedi ve Travmatolji ABD, 59030, Tekirdağ, Türkiye. **Phone:** +90 282 250 00 00 **E-mail:** erdemcan@nku.edu.tr **Received:** Nov, 2020. **Accepted:** Dec, 2020.

Introduction

Osteomyelitis is rare in adolescents and children (1). The incidence in pediatric age groups is 1: 5000. Children under 5 years of age make up approximately 50% of pediatric osteomyelitis cases (2). Osteomyelitis develops mostly in hematogenously in pediatric patients and most commonly affects the metaphysis of long bones (3).

Acute bacterial osteomyelitis in children is a potential limb and life-threatening condition that requires urgent diagnosis and treatment. Delayed treatment may result in severe sepsis, chronic infection, bone deformity, or growth retardation (4).

Numerous types of bacteria can be the cause of osteomyelitis. The most common pathogen is *Staphylococcus aureus* in more than 50% of all cases (3). *Streptococcus spp, Escherichia coli, Klebsiella spp*, and *Proteus spp.* are other bacterial pathogens that can cause osteomyelitis (5).

Salmonella as an etiological agent in osteomyelitis is rare. Salmonella osteomyelitis accounts for 0.8% of all salmonella infections and only 0.45% of all types of osteomyelitis (6). Salmonella osteomyelitis predominantly occurs in patients with hemoglobinopathy such as sickle cell disease or thalassemia, and this is an important cause of morbidity and mortality in the population (7). Salmonella is predisposed for osteomyelitis, especially diabetes mellitus, systemic lupus erythematosus, lymphoma, liver and cardiovascular diseases, patients with previous surgery or trauma history, and patients receiving steroid therapy (8). In infections with high bacterial virulence, a weakening of host resistance may cause the development of Salmonella osteomyelitis in children with normal immunity.

In the literature, there are very few cases of Salmonella osteomyelitis in healthy individuals. Here, we present a case of Salmonella osteomyelitis developing in the radius of an immunocompetent 6-year-old child without any known predisposing conditions.

Case Report

A 6-year-old patient applied to the pediatric emergency department of our hospital with complaints of pain in the left forearm and 39.5℃ fever, which has been going on for 20 days after the fall. There was no bone pathological feature in X-Ray taken at another center where the patient applied with the same complaints 20 days ago (Fig. 1). It was confirmed that he received 5 days of oral amoxicillin-clavulanate and antipyretic treatment in this center due to upper respiratory tract infection. The medical history and family history of the patient were natural. Physical examination revealed swelling and tenderness in the forearm. Passive movement restriction was detected in adjacent joints. In laboratory tests; 11200 / µl (normal: <10000 / µl) serum white blood cell (WBC) count, 67 mm per hour (normal: <10 mm / s) erythrocyte sedimentation rate (ESR), C-reactive protein (CRP) 5.6 mg / L (normal: <5 mg / L), other liver function and electrolyte values were within normal limits. The blood culture taken was negative as a result. On radiographs, lytic lesions were detected along the radius (Fig.2).

Lytic lesions were also evaluated by magnetic resonance imaging (MRI). In MRI, an infiltrative type lesion was observed that was associated with the entire diaphysis of the radius and its proximal and distal metaphyses, and not associated with the epiphysis plate associated with the epiphysis line proximally, and widespread edema was also observed in all muscle groups in the parosteal region with the lesion (Fig.3).

Samples were taken for bone culture and pathological examination under general anesthesia in the operating room due to suspicion of osteomyelitis or malignant lesions.

For bone and soft tissue healing, the left upper limb was followed in a plaster splint for 3 weeks. In the pathological examination, findings compatible with osteomyelitis were observed. There was very intensive Salmonella species growth in culture. The patient was examined for hemoglobinopathies and immunodeficiency. Peripheral smear and osmotic fragility tests were evaluated as normal and immunoglobin levels were within normal limits.

The patient was given 400 mg vancomycin intravenously every 8 hours, and 1 g ceftriaxone intravenously every 12 hours for 3 weeks after the pediatric consultation. In the laboratory tests of the patient, 6700 / µl white blood cell (WBC) count, 22 mm erythrocyte sedimentation rate (ESR), and C-reactive protein (CRP) 0.6 mg / L and other biochemical values were found in the normal range. The patient was followed up on an outpatient basis, with 300 mg of oral cefuroxime treatment every 12 hours for 2 weeks. The laboratory tests of the patient, who was followed for 18 months, were repeated regularly (Table 1). MRI and X-Ray imaging was performed at 6th postoperatively. It was observed that the lytic lesion decreased significantly in the radius (Fig.4a, 4b). On the 14th postoperative month, MRI imaging showed that the lesion disappeared (Figure 5).

During the regular follow-up of the patient, no troubles were observed. Physical examination of the patient was normal, radiology, and laboratory results were evaluated as normal (Photo 1.).

Table 1. Laboratory values in postoperative follow-up.

	WBC (µl)	ESR (mm/s)	CRP (mg/dL)
Post-op 45th day	6400	6	<1
Post-op 4th month	9000	11	1,1
Post-op 6th month	5700	10	<1
Post-op 24th month	5650	10	<1

Figure 1. 6-year-old patient's left forearm AP and lateral X-Ray view. Normal X-Ray image taken at the beginning of complaints of pain and fever in the left forearm.

Figure 2. Left forearm AP and lateral X-Ray images taken 20 days after the symptoms started. Radius lytic lesion with periosteal reaction extending to the proximal epiphysis throughout the diaphysis.

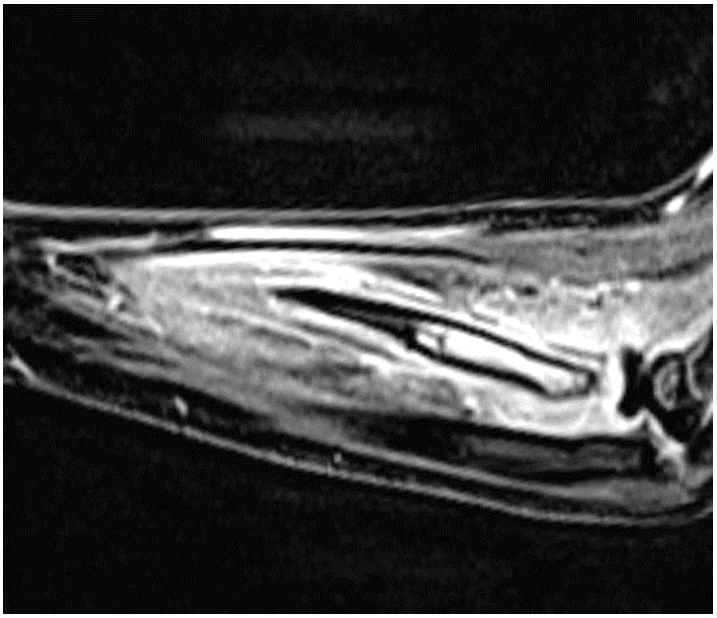

Figure 3. MRI image of the left forearm in the T1 sequence taken 20 days after the symptoms started. In the radius, diaphysis proximally infiltrative lesion associated with the pineal plate and widespread edema in the soft tissue around the lesion.

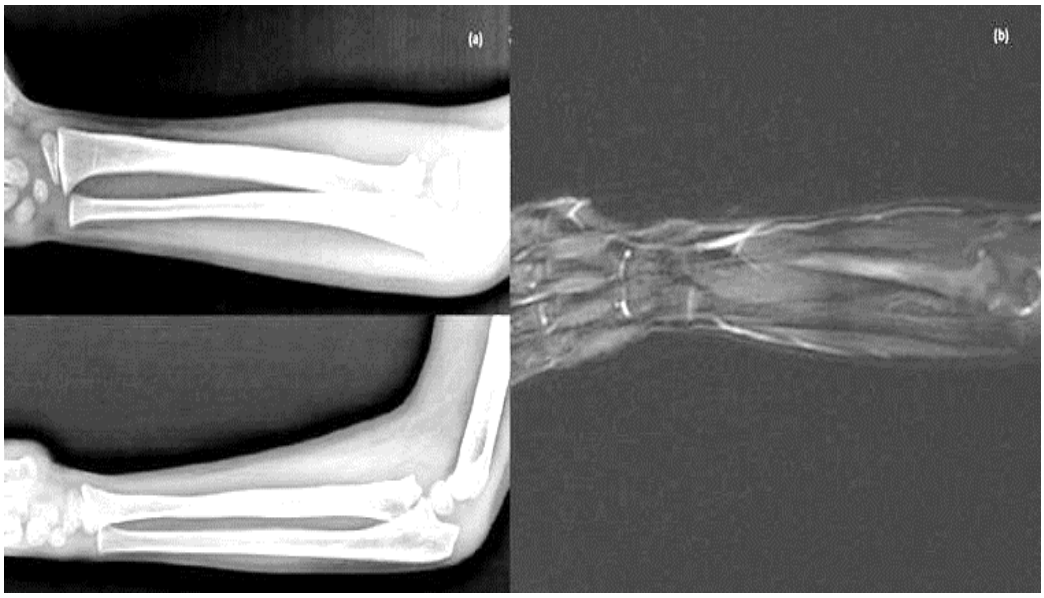

Figure 4 (a, b). (a) X-Ray image showing a significant decrease in lytic lesion taken at 6 months after surgery, (b) MRI image.

Figure 5. Normal MRI image of the patient at 14th postoperative month.

Discussion

Salmonella infection in humans can occur with different clinical symptoms. These can lead to five separate clinical conditions: gastroenteritis (food poisoning), enteric fever (typhoid), bacteremia or septicemia, focal infection (including soft tissue or bone infection), and chronic carrier (1). In only about 5-7.6% of cases, bacteremia develops in adults and children (9). Salmonella arthritis and osteomyelitis occur less than 1%, and clinical signs of gastrointestinal infection are not always seen (10). It is most often obtained by consuming contaminated foods, especially uncooked eggs, poultry, and meat products.

Salmonella osteomyelitis is usually found in the diaphyseal region of the long bones, most often in the femur and humerus (11,12). Other commonly affected bones are radius, ulna,

tibia, and vertebrae (11). Its excessive presence in the diaphysis region may be linked to increased bone marrow activity and blood destruction products (13).

Rare etiologies such as atypical osteomyelitis in immunocompromised patients may mimic an osteolytic neoplastic lesion (14). It is important to discuss tumors that show both malignancy and features of osteomyelitis. These are Leukemia and Ewing sarcoma. Most children with leukemia may complain of bone and joint pain, and often show an increase in erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP), similar to infection (15). Radiographically, leukemia shows periostitis and radiolucent bands adjacent to physics (16), the latter is not seen in osteomyelitis. Ewing sarcoma is the second most common primary bone tumor. Typically, it appears as an osteolytic lesion in the metaphyseal and metaphysodiaphyseal regions of the bone, similar to where osteomyelitis is typically found (17). Usually, a handled mass is associated with the pain site. This is not seen with typical osteomyelitis. Patients with Ewing sarcoma may also have fever and chills that mimic osteomyelitis. Due to the unique clinical and radiographic features, biopsy is very important for diagnosis. Consequently, tissue samples are needed to make a clear distinction between malignancy and osteomyelitis.

Although it has been reported that blood cultures are often positive (71%) in adults with Salmonella osteomyelitis, this does not appear to be the case in children. In Tsagris et al. (12) case series, only 1 of 4 patients was isolated from Salmonella blood cultures. In our case, the blood culture was negative.

It is considered as the best tool for the diagnosis of osteomyelitis among MRI imaging methods (18). MRI can also help diagnose earlier than plain radiography (18). Early signs of plain radiography may not be evident until two to three weeks after the onset of symptoms and signs (18).

It seems that long-term antibiotic therapy results more positively. It is thought that relapse of the disease may result from short-term treatment (12). It has been suggested by some authors that 6-week conservative treatment with IV and oral antimicrobials leads to complete recovery of acute Salmonella osteomyelitis with reduced recurrence risk. However, there is currently no consensus on the treatment of acute or chronic osteomyelitis caused by Salmonella. Successful treatment of osteoarticular Salmonella infection may require extensive and sometimes more than one debridement in addition to long-term antibiotic therapy (14,19). Antibiotic therapy should ideally be started after obtaining sufficient bacteriological samples. In these cases, third generation cephalosporins such as ampicillin, chloramphenicol, ceftriaxone and fluoroquinolones such as ciprofloxacin have been used successfully (14). Fluoroquinolones offer strong activity and good bone penetration against Salmonella; However, these drugs should be avoided in children (19).

Results

Consequently, salmonella osteomyelitis, which is rarely seen in children without underlying disease, should be kept in mind. MRI is superior in defining osteomyelitis and appropriate cultures should be taken to determine the exact effect.

Acknowledgment: The authors confirm that there are no known conflicts of interest associated with this publication and there has been no significant financial support for this work that could have influenced its outcome.

Ethics Committee Approval: Yes
Informed Consent: NA
Peer-review: Externally peer-reviewed.
Conflict of Interest: No conflict of interest was declared by the author.
Financial Disclosure: The author declared that this study has received no financial support.

References

1.Zhan C, Du J, Chen L: Salmonella osteomyelitis in a previously healthy neonate: a case report and review of the literature. Ital J Pediatr. 2018;44:28.
2.De Boeck H. Osteomyelitis and septic arthritis in children. Acta Orthop Belg 2005;71:505-15.
3.Kaplan SL. Osteomyelitis in children. Infect Dis Clin North Am 2005; 19:787-797.
4.Dartnell J, Ramachandran M, Katchburian M. Haematogenous acute and subacute pediatric osteomyelitis: a systematic review of the literature. J Bone Joint Surg Br 2012;94:584-95.
5.Dodwell ER. Osteomyelitis and septic arthritis in children: current concepts. Curr Opin Pediatr 2013;25:58-63.
6.Sanchez AA, Mazurek MT, Clapper MF. Salmonella osteomyelitis presenting as fibrous dysplasia. A case report. Clin Orthop. 1996;330:185-9.
7.Miller SI, Pegues Da. Salmonella species. In: Mandell GL, Douglas RG, Bennett JE, editors. Principles and practice of infectious diseases. Pennsylvania: Churchill Livingstone;2000;2344-60.
8.Arora A, Singh S, Aggarwal A, Aggarwal PK. Salmonella osteomyelitis in an otherwise healthy adult male: successful management with conservative treatment: a case report. J Orthop Surg. 2003;11(2): 217-20.
9.Hohmann EL (2001) Nontyphoidal salmonellosis. Clin Infect Dis 32:263-269.
10.Saphra I, Winter JW. Clinical manifestations of salmonellosis in man. An evaluation of 7779 human infections identified at the New York Salmonella Center. N Engl J Med 1957;256:1128-34.
11.McAnearney S, McCall D: Salmonella Osteomyelitis. Ulster Med J. 2015, 84:171-172.
12.Tsagris V, Vliora C, Mihelarakis I, Syridou G, Pasparakis D, Lebessi E, Tsolia M: Salmonella osteomyelitis in previously healthy children: report of 4 cases and review of the literature. Pediatr Infect Dis J. 2016, 35:116-117.
13.Rayan F, Mukundan C, Shukl DD. A case of relapsing Salmonella osteomyelitis in a thalassemia trait patient. Journal of Orthopaedics and Traumatology 2009;10:31-3.
14.Huang DB, DuPont HL. Problem pathogens: extra-intestinal com- plications of Salmonella enterica serotype Typhi infection. Lan- cet Infect Dis 2005;5:341-8.
15.Rogalsky RJ, Black GB, Reed MH. Orthopaedic manifestations of leukemia in children. The Journal of Bone & Joint Surgery. 1986;68(4):494-501.
16.Chiappini E, Mastrangelo G, Lazzeri S. A case of acute osteomyelitis: an update on diagnosis and treatment. Int J Environ Res Public Health. 2016;13(6):539.
17.Ozaki T. Diagnosis and treatment of Ewing sarcoma of the bone: a review article. J Orthop Sci. 2015;20(2):250-63.
18.Dutta A, Allen CH. Non-typhoidal Salmonella Osteomyelitis in the Midfoot of a Healthy Child and Review of the Literature. Infectious Disease Therapy 2013;1:107.
19.Bettin D, Schaphorn G, Blasius S, Becker K, Niemeyer T. A rare case of Salmonella osteomyelitis in the humerus as a differential diagnosis to a malignant bone tumor. Archives of Orthopaedic and Trauma Surgery 2002;122(9-10):544-6.

QMEL
Medicine & Publishing
Published by The QMEL®.org
Medicine & Education & Library