A 10-year-old boy with limping complaint

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Case

A 10-year old boy who had a history of diarrhea and fever approximately 4 weeks before presentation and whose complaints improved with symptomatic treatment in that period without use of antibiotics presented to our outpatient clinic with pain, swelling, erythema and limping in the left foot. On physical examination, erythema, increased temperature and tenderness on the dorsum of the left foot and a purulent bulla with a size of 3 x 3 cm on the dorsolateral part of the 5th metatarsophalengeal joint were noted (Figure 1). The patient and his family reported that pain and limping in the left foot started two days after diarrhea stopped and increased gradually for the last three weeks. Swelling, erythema and increased temperature started 5 days before presentation and reached the status at the time of presentation. At presentation, the white blood cell (WBC) count in the peripheral blood was found to be 11 200/mm^3, C-reactive protein (CRP) was found to be 5,94 mg/dL and erythrocyte sedimentation rate (ESR) was found to be 40 mm/h. Foot radiographs were found to be normal. On magnetic resonance imaging, Brodie abscess starting from the distal metaphysis of the 5th metatarsal bone of the left foot and extending to the subcutaneous tissue and soft tissue edema in the periarticular area were observed (Figure 2). No growth occured in urine, stool and blood culture samples obtained at presentation.

Surgical drainage was planned for the abscess in the fifth metatarsal bone in the patient who was started on intravenous ceftriaxone (2 x 1.5 g) ampirically. During the operation, the abscess was drained with debridment of the surrounding soft tissues, decompression and curettage was performed in the distal metaphysis of the fifth metatarsal bone, the abscess content and drainage material were sent to the laboratory and cultured.

Figure 1. Swelling and eryhtema on the dorsum of the foot. Purulent bulla is observed in the dorsolateral part of the fifth metatarsophalyngeal joint

Figure 2. Bone edema, Brodie abscess (normal arrow) and soft tissue abscess extending to the subcutaneous area (dashed arrow) are observed in cross-sections on fat-saturated and contrast-enhanced T1-weighted magnetic resonance images

The left lower extremity of the patient was followed up in cast brace for three weeks to enable recovery of the bone and soft tissue.

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Diagnosis: Salmonella osteomyelitis

When group B salmonella was grown in the cultures, the patient was evaluated in terms of hemoglobinopahties and immune deficiency. Peripheral blood smear, hemoglobin electrophoresis and immunoglobulin levels were found to be normal and immune deficiency was not considered in the patient.

After surgery the patient received intravenous ceftriaxone for 2 weeks more. It was observed that swelling, eryhtema and increased temperature in the foot disappeared in the postoperative period and his WBC, CRP and ESR values regressed to 5 700/mm^3, 0.3 mg/dL and 12 mm/h, respectively. The antibiotherapy was completed to 6 weeks with oral cefixim (2 x 400 mg) after discharge. At the follow-up visit on the 2nd month, it was observed that the patient's complaints improved and he walked easily. On the follow-up visit after one year, he was found to be completely recovered.

Discussion

Hematogenous osteomyelitis is a clinical picture which occurs most frequently in the metaphyses of the long bones caused most commonly by Staphylococcus aureus in all age groups (1). In hematogenous osteomyelitis, Salmonella species are rarely isolated as the causative agent. However, complaints related with the skeletal system are observed frequently following salmonellosis. These findings may frequently be related with development of post-infectious reactive arthritis and rarely with osteomyelitis caused by Salmonella species or suppurative infections developing in the form of septic arthritis. The rate of the cases caused by Salmonella species has been reported to be 0.45% among all osteomyelitis cases (2). Individuals with hemoglobinopathy and especially sickle cell anemia, chronic liver disease and diabetes and individuals who have immunosupression because of malignancy have a higher predisposition to Salmonella osteomyelitis.

In children, regional suppurative infections including osteomyelitis caused by Salmonella are rare and generally accompany some underlying diseases. Salmonellosis is observed as four clinical pictures which occur seperately, consecutively or simultaneously including gastroenteritis, thyphoid fever or enteric fever, septicemia associated or not associated with regional suppurative infections and carrier state. Among these four clinical pictures, gastroenteritis is observed most commonly with a rate of 70%, carrier state is observed with a rate of 15%, thyphoid fever or enteric fever is observed with a rate of 8% and regional suppurative infections and septicemia are the disease form observed most rarely with a rate of 7.4% (3). In infections with high bacterial virulance, weakening of host defense may lead to development of Salmonella osteomyelitis even in children with normal immunity. It has been shown that 60% of the patients with Salmonella osteomyelitis have diarrhea or a positive stool culture at the time of diagnosis (4). In Salmonella osteomyelitis, the most common specific subgroup is group B. This is followed by group D and group C_1 (5).

The incidence of Salmonella osteomyelitis is inversely proportional to age. Locomotor system infections caused by Salmonella are observed more frequently in infants and young children. Salmonella osteomyelitis is observed at early ages and most commonly in the short tubular bones in the hands and feet. In older children, it is observed especially in long bones including mainly the femur and humerus (6). Short tubular bone involvement in our 10-year old patient was notable.

Osteomyelitis cases with mild symptoms for longer than 2 weeks in the absence of systemic findings including fever and malaise and with no growht in blood culture are considered subacute osteomyelitis whether or not supressed with antibiotics. Whatever the causative bacterium, the diagnosis of primary subacute hematogenous osteomyelitis (PSHO) is considerably difficult. While Salmonella species generally lead to acute osteomyelitis in young children, they cause to subacute osteomyelitis in older children because of variation in the relationship between the pathogen and host. Another reason for the disease to progress to the subacute form is inadequate antibiotic usage. Treatment of primary subacute hematogenous osteomyelitis is generally difficult because of absence of growth in cultures. Rasool (7) reported culture positivity with a rate of 29% in 21 PSHO cases. Although the most common causative agent in cases of primary subacute hematogenous osteomyelitis is *S. aureus*, cases of PSHO caused by extraordinary agents including *S. pneumonia* and Klebsiella species have also been reported in the literature (8). In our case presentation, the clinical picture of the patient mentioned is compatible with PSHO.

In Salmonella osteomyelitis, the bacterium is localized in the bone following septicemia. In individuals with sickle cell anemia, hepatic failure, malginancy, connec-

tive tissue disease or immune deficieny, group B Salmonella tends to form abscess in the bone by growing in the bone trabeculae due to thrombosis in the venous sinuses in the metaphyseal areas (1). This is observed considerably rarely in healthy individuals under normal conditions. In the literature, there are studies related with locomotor system infections caused by Salmonella in healthy individuals (9-10). In all of these studies, there is a history of diarrhea before development of locomotor system symptoms. Çelik et al. (9) published a case of Brodie abscess caused by Salmonella in the navicular bone in a 4-year old boy who did not have hemoglobinopathy or immune deficiency.

It should be kept in mind that Salmonella species may rarely lead to septic arthritis and osteomyelitis also in healthy children. In healthy children, severe abdominal pain, diarrhea and locomotor system findings following Salmonellosis should be cared and evaluated in terms of Salmonella osteomyelitis or septic arthritis. Conclusively, delayed diagnosis and treatment in healthy children as in the case presented may lead to a picture of PSHO and this may cause to diagnostic and therapeutic difficulties.

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