Lung abscess and osteomyelitis of the ribs caused by *Salmonella irumu* in a child with S-β⁺-thalassemia

*S*-β⁺-talasemili bir çocukta salmonella irumu'ya bağlı akciğer apsesi ve kaburgalarda osteomiyelit

Erdal İnce¹, Ergin Çiftçi¹, Tanıl Kendirli¹, Mehmet Ertem¹, Haluk Güriz¹, Ahmet Derya Aysev¹, Suat Fitöz², Birsel Erdem³, Ülker Doğru¹

¹Department of Pediatrics, Ankara University School of Medicine, Ankara Turkey
²Department Of Radiology, Ankara University School of Medicine, Ankara, Turkey
³Department Of Microbiology, Ankara University School of Medicine, Ankara, Turkey

Patients with sickle cell disorders have an increased susceptibility to *Salmonella* infections. We report a child with S-β⁺-thalassemia who presented with fever, cough, and chest pain. The child diagnosed with pneumonia, lung abscess, and osteomyelitis of the ribs. *Salmonella irumu* was isolated from blood and an aspiration material of the chest wall abscess. The patient was treated successfully with ceftriaxone and subsequently ciprofloxacin.

**Key words:** S-β⁺-thalassemia, lung abscess, osteomyelitis, *Salmonella irumu*


Anahtar kelimeler: S-β⁺-talasemi, akciğer apsesi, osteomiyelit, *Salmonella irumu*

**Case report**

A 10-year-old boy was admitted to our hospital with a 7-day history of fever, cough, and chest pain. He was diagnosed with S-β⁺-thalassemia at 7 months of age, and underwent splenectomy and cholecystectomy at eight years of age. He had taken multiple blood transfusions and had hemochromatosis in his liver demonstrated by liver biopsy two years ago. His mother had sickle cell trait, and his father had β-thalassemia trait.

His body temperature was 39°C; there was a painful fluctuating swelling over the left 6th rib. His breathing sounds were decreased in the left hemithorax. Cracking rales were also noted in the same hemithorax. Laboratory examinations included hemoglobin level of 6.8 g/dL, leukocyte count of 20.800/mm³ with a shift to the left, and platelet count of 646.000/mm³. Erythrocyte sedimentation rate was 107 mm/h and C-reactive protein level was 12.4 mg/dL.
Chest x-ray examination showed pneumonic infiltration in the left upper and middle zone and laterally located dense appearance resembling left pleural effusion. A computed tomography scan revealed lung abscess (Figure 1A), costal osteomyelitis and abscesses surrounding the 4th, 5th, and 6th ribs (Figure 1B). There was not any bone involvement in bone radionuclide scans.

The abscess on chest wall was drained and purulent material was obtained. Gram staining obtained from the abscess showed Gram-negative rods and *S. irumu* was isolated from the culture. *S. irumu* was also isolated from blood specimen. Culture of stool was negative for Salmonella.

The patient, who was diagnosed having pneumonia, lung abscess, and rib osteomyelitis with chest wall abscesses, was treated with intravenous ceftriaxone 100 mg/kg per day. At the end of three-treatment weeks, follow up CT scans revealed nearly complete regression of lung abscess and costal osteomyelitis (Figure 2A and 2B).

After one-month treatment with ceftriaxone, oral ciprofloxacin was started for another 1 month and the patient was discharged from the hospital. At the end of two-treatment months the patient had no evidence of pulmonary abscess or rib osteomyelitis. At one-year follow-up he was in good condition.

**Discussion**

Sickling disorders of various degrees of severity result from hemoglobin S existing in combination with other abnormal hemoglobins or thalassemias. Several of these syndromes, including S-β⁺-thalassemia, present a clinical picture virtually indistinguishable from that of sickle cell anemia. Most of the others, including S-β⁺-thalassemia, produce less severe manifestations (5).

Although it has been well known that patients with sickle cell disorders have an increased susceptibility to Salmonella infections, we encountered two very rare manifestations of salmonellosis simultaneously in our patient. Pulmonary manifestations are uncommon in salmonellosis. Saphra and Winter reported that pulmonary involvement occurs in 1% of patients with salmonellosis (6). Lobar and bronchopneumonia are relatively frequent manifestations (7,9). Empyema thoracis is rare (10-12) and lung abscess due to salmonellae is exceptional. Approximately only seventeen cases with lung abscess caused by salmonellae have been reported previously (13-25). According to Cohen et al. only one patient with salmonella pneumonia and empyema had sickle cell anemia as underlying disease (17).

The association between sickle cell anemia and salmonella osteomyelitis was recognized by Hodges et al (26). In 1957, Hook et al., who reviewed 33 cases of sickle cell anemia and salmonella infections, reported that 94% of patients had osteomyelitis (27). Adeyokunnu and Hendrickse reviewed 63 cases of salmonella osteomyelitis and found that 90% of patients had sickle cell anemia (4). Diggs reviewed 62 cases of osteomyelitis in patients with sickle cell anemia and found that 89% of patients had salmonella osteomyelitis (28). Cohen et al. noted that out of the 150 patients with salmonella osteomyelitis 48 patients had sickle cell anemia, 7 had sickle-C disease, 2 had sickle-thalassemia, and 1 had sickle cell trait (17). Ribs, spine and long bones are most frequently affected bones in salmonella osteomyelitis (6). Roentgenographic findings of osteomyelitis appear in later stages of infection. Although we strongly suspected from osteomyelitis in our patient, we could not confirm this diagnosis until the fifteenth day of the therapy. Although salmonella associated chest wall abscess without osteomyelitis has been reported (29), possible diagnosis of osteomyelitis must be taken into consideration in patients with sickle cell disorders even if there are no osteomyelitis findings.

We could not explain whether pneumonia and lung abscess or osteomyelitis of the ribs was initial focus. Salmonel-
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la bacteremia might occur and then Salmonella inoculate in scar tissues in lung and ribs. Cohen et al. noted that 53 per cent of patients with salmonella pneumonia and empyema had positive stool culture (17). The high incidence of stool cultures from patients suggests a gastrointestinal source in the pathogenesis of their pulmonary infections. After the gastrointestinal tract has become colonized or infected, hematogenous dissemination to the lungs may occur. Alternatively, aspiration of infected gastric secretions may occur in patients with gastrointestinal infection or colonization. Rarely, salmonella pulmonary infections may occur by extension of infection from a nearby site (10). We could not determine the origin of *S. irumu* in our patient. Child did not have a history of gastroenteritis, and stool culture was negative. Because our patient had undergone cholecystectomy, Salmonella carriage state is less possible. However, it has been reported that cholecystectomy fails to eliminate the carrier state in 15% of patients. In such situations, previously damaged liver may serve as a carrier site (30). Our patient had hemochromatosis induced liver injury demonstrated by liver biopsy obtained during cholecystectomy and splenectomy. Our patient might carry *S. irumu* in his damaged liver, although we could not demonstrate this strain in his stool.

*S. irumu* is a very rare strain of Salmonella in Turkey. *S. typhmurium*, *S. typhi* and *S. cholerasuis* were reported to be the most common serotypes isolated from cases with salmonella pneumonia and empyema (17). *S. typhmurium*, *S. typhi*, *S. enteritidis* and *S. cholerasuis* were the most commonly isolated serotypes from cases with salmonella osteomyelitis (17). We did not encounter publication reporting neither osteomyelitis nor lung abscess caused by *S. irumu* in a Medline search. In addition, we did not encounter any case with sickle cell disorders who had osteomyelitis and lung abscess simultaneously. Although it is known that Salmonella infections are relatively frequent in patients with sickle cell disorders, to our knowledge, this is the first report of osteomyelitis and lung abscess caused by *S. irumu* in a patient with S-β+-thalassemia.

References