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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

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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***

Orak hücre hastalığı olanların Salmonella enfeksiyonlarına duyarlılıkları artmıştır. Burada ateş, öksürük ve göğüs ağrısı ile gelen S-β⁺-talasemili bir çocuk sunuldu. Hasta pnömoni, akciğer apsesi ve kaburga osteomiyeliti tanılarını aldı. Kan ve göğüs duvarı apsesinin aspirasyon materyalinin kültüründen *Salmonella irumu* izole edildi. Hasta seftriakson ve ardından siprofloksasin ile başarıyla tedavi edildi.

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

Patients with sickle cell disorders have an increased susceptibility to Salmonella infections. The mechanisms for this increased susceptibility appear to be multifactorial and include several proposed immunologic mechanisms such as inadequate opsonisation due to abnormalities in the serum complement pathway, functional autosplenectomy with loss of particulate clearing and defective neutrophil antibacterial function (1,2). Bone necrosis, which may be seen in patients with sickle cell disorders, predisposes to Salmonella osteomyelitis (3, 4). Here, we report on a child with S-β⁺-thalassemia who developed lung abscess and osteomyelitis of the ribs caused by *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 39ºC; 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/mm^3. Erythrocyte sedimentation rate was 107 mm/h and C-reactive protein level was 12.4 mg/dL.

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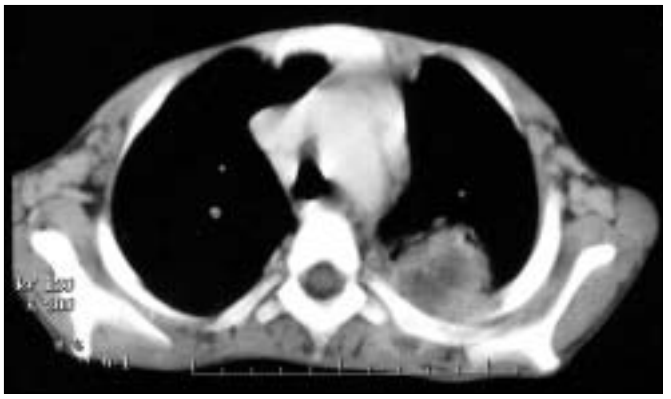

Figure 1A. Axial CT scan shows left upper lobe abscess and costal osteomyelitis. Also note the extension of the inflammatory process to the posterior chest wall, and periaortic and axillary lymphadenopaties.

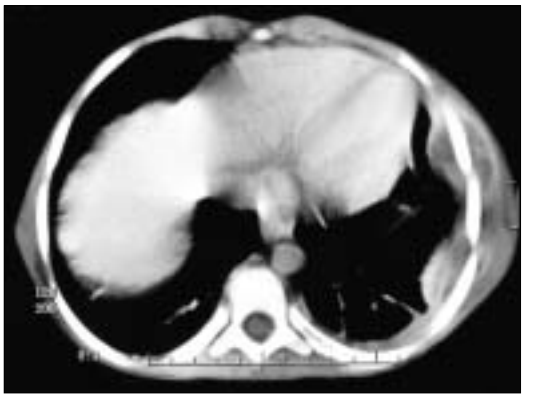

Figure 1B. CT scan of lung base demonstrates left chest wall abscesses due to costal osteomyelitis and associated pleural effusion.

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^0-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 confirmed 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-

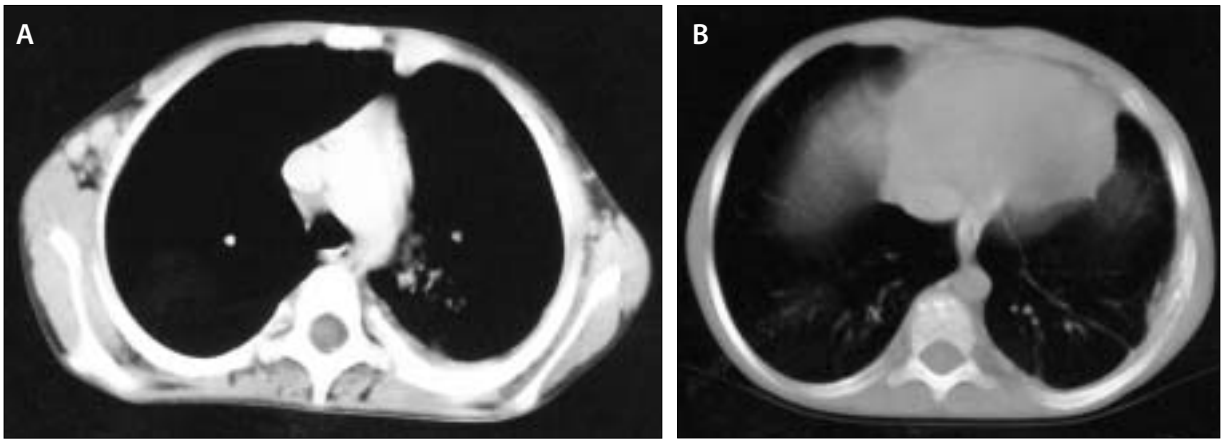

Figure 2. A ve B. Follow up CT scans of the same levels reveal nearly complete regression of the soft tissue lesions of the chest wall **(a)**. The residual costal changes due to osteomyelitis are more pronounced at bone window **(b)**.

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

1. Barret-Connor E. Bacterial infection and sickle cell anemia. Medicine 1971; 50: 97-112.
2. Gardner RV. Salmonella vertebral osteomyelitis and epidural abscess in a child with sickle-cell anemia. Pediatr Emerg Care 1985; 1: 87-89.
3. Keeley K, Buchannan GR. Acute infarction of long bones in children with sickle cell anemia. J Pediatr 1982; 101: 217-227.
4. Adeyokunnu A, Hendricks RG. Salmonella osteomyelitis in childhood. Arch Dis Child 1980; 55: 175-184.
5. Honig GR. Hemoglobin Disorders. In: Behrman RE, Kliegman RM, Jenson HB, eds. Nelson Textbook of Pediatrics. WB Saunders Company; 2000: 1478-1488.
6. Saphra I, Winter JW. Clinical manifestations of salmonellosis in man: an evaluation of 7779 human infections identified at the New York Salmonella Center. New Eng J Med 1957; 256: 1128-1134.
7. Berkeley D, Mangels J. Salmonella pneumonia in a patient with carcinoma of the lung. Am J Clin Pathol 1980; 74: 476-478.
8. Murdoch MB, Peterson LR. Nontyphoidal Salmonella pleuropulmonary infections. Arch Intern Med 1991; 151: 196.
9. Neva FA. Pulmonary involvement in typhoid and paratyphoid fevers. Ann Intern Med 1950; 33: 83-99.
10. Buscaglia AJ. Empyema due to splenic abscess with Salmonella newport. JAMA 1978; 240: 1990.
11. Kate P, Osei K, Chiemchanya S et al. Empyema due to Salmonella typhimurium with underlying alveolar cell carcinoma. South Med J 1984; 77: 234-236.
12. Fonollosa V, Bosch JA, Garcia-Bragado F et al. Hemolytic anemia, splenic abscess, and pleural effusion caused by Salmonella typhi. J Infect Dis 1980; 142: 945.
13. Harvill T. Typhoid pulmonary abscess. JAMA 1942; 119: 494-496.

14. Saphra I, Wasserman M. Salmonella choleraesuis: a clinical and epidemiological evaluation of 329 infections. Am J Med Sci 1954; 228: 525-533.
15. Hahne O. Lung abscess due to Salmonella typhi. Am Rev Respir Dis 1964; 89: 566-571.
16. Besznyak J, Pinter E, Turbok E. Throracic empyema and lung abscess due to Salmonella stanley. Arch Surg 1965; 91: 1023-1025.
17. Cohen JI, Bartlett JA, Corey R. Extra-intestinal manifestations of Salmonella infections. Medicine 1987; 66: 349-388.
18. Aguado J, Obeso G, Cabanillas JJ et al. Pleuropulmonary infections due to nontyphoid strains of Salmonella. Arch Intern Med 1990; 150: 54-56.
19. Bansal P, Harrison P. Salmonella lung abscess causing rupture of the thoracic aorta. Br J Hosp Med 1990; 43: 296.
20. Ankobiah WA, Salehi F. Salmonella lung abscess in a patient with acquired immunodeficiency syndrome. Chest 1991; 100: 591.
21. Chan JC, Raffin TA. Salmonella lung abscess complicating Wegener's granülomatosis. Respir Med 1991; 85: 339-341.
22. Albrecht H, Stellbrink HJ, Fenske S et al. Salmonella typhimurium lung abscess in an HIV-infected patient successful treatment with oral ciprofloxacin. AIDS 1992; 6: 1400-1401.
23. Satué JA, Aguado JM, Costa JM et al. Pulmonary abscess due to non-typhi Salmonella in a patient with AIDS. Clin Infect Dis 1994; 19: 555-557.
24. Riantawan P, Subhannachart P. Salmonella lung abscess and bacteraemia in an AIDS patient. J Med Assoc Thai 1996; 79: 333-336.
25. Ridha AG, Malbrain MLNG, Mareels J et al. Lung abscess due to nontyphoid Salmonella in an immunocompromised host. Case report with review of the literature. Acta Clin Belg 1996; 51: 175-183.
26. Hodges FJ, Holt JF, Jacox HW et al. Editorial comments. In: Yearbook of Radiology, Chicago: Year Book Medical Publishers, Inc., 1957: 89.
27. Hook EW, Campbell CG, Weens HS et al. Salmonella osteomyelitis inpatients with sickle cell anemia. N Eng J Med 1957; 257: 403-407.
28. Diggs LW. Bone and joint lesions in sickle cell disease. Clin Orthop 1967; 52: 119-143.
29. Hanahel JI, Hulbert TV, Larsen RA. Case report: recurrent Salmonella typhi chest wall abscess associated with a pituitary macroadenoma. Am J Med Sci 1992; 304: 43-44.
30. Lalitha MK, John R. Unusual manifestations of salmonellosis-a surgical problem. Q J Med 1994; 87: 301-309.