Reminder of important clinical lesson

Salmonella pelvic osteomyelitis in an immunocompetent child

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Summary

A case of *Salmonella enteritidis* osteomyelitis isolated from the pelvis in a healthy adolescent is reported. Pelvis osteomyelitis is rare in childhood. Moreover, salmonella, typically responsible for bone infection in sickle cell anaemia, is unusually involved in healthy children. *Salmonella* should be considered among aetiological agents. Antibiotic treatment alone may be successful; however, sometimes, surgical drainage is required.

BACKGROUND

Acute haematogenous osteomyelitis (AHOM) of the pelvis is a rare form of childhood osteomyelitis. *Salmonella* bone infections are extremely rare in healthy children but are common in children with haemoglobinopathies. We present the case of an 11-year-old healthy boy with acute osteomyelitis of the pelvis caused by *Salmonella typhimurium*.

CASE PRESENTATION

A previously healthy 11-year-old boy presented with complaints of progressively worsening fever for 3 days (max 40°C), headache, abdominal pain and left lumbar pain associated with limp. He complained of pain at the hip, gluteus and posterior leg, but without any recent trauma or infection. Moreover, he referred to fatigue and dysuria but regular intestinal function except for diarrhoea over the past months. During his first year, the boy had had a gluteus abscess after an injection. Physical examination revealed pain and restriction of movement in the left hip particularly in external rotation and full extension. No local swelling, erythema or skin lesions were evident. Other joints had normal mobility. Laboratory investigations showed high erythrocyte sedimentation rate (ESR) (49 mm/h) and C reactive protein (CRP) (7.94 mg/dl); other lab data were normal. Pelvic, lumbar and sacral spine x-rays were normal except for an asymmetry of the pelvic bones with elevation and rotation on the left. The pelvic MRI showed dysmorphic bones and altered signal after contrast on the left suggesting low-grade inflammation. The CT scan confirmed the skeletal dysmorphism but did not show abnormalities after contrast. A technetium-labelled bone scan was negative. Suspecting a pelvic joint infection, a broad spectrum antibiotic (ceftriaxone and oxacillin) was started.

The boy continued to complain, even at night, of pain in the ischia, spreading to the thigh and the lumbar and sacral vertebral bones, with intermittent fever with shivers. Blood cultures for aerobic and anaerobic germs and fungi were repeatedly negative; *Bartonella*, *Salmonella typhi*,

Salmonella paratyphi, *Brucella* and *Rickettsia* were all negative. Molecular DNA detection for *Acinetobacter baumannii*, *Enterobacter cloacae*, *Enterococcus faecium*, *Enterococcus faecalis*, *Escherichia coli*, *Haemophilus influenzae*, *Klebsiella pneumoniae*, *Legionella pneumophila*, *Mycobacterium tuberculosis*, *Neisseria meningitidis*, *Proteus mirabilis*, *Pseudomonas aeruginosa*, *Serratia marcescens*, *Staphylococcus aureus*, *Staphylococcus epidermidis*, *Streptococcus agalactiae*, *Streptococcus pneumoniae* and *Streptococcus pyogenes*, Mantoux reaction and QuantiFERON test were all negative. Antibodies for *Streptococcus pyogenes* (ASLO and anti-DNase B) were only slightly increased. White cell count was always normal except for slight neutrophilia and microcytic anaemia (haemoglobin 11.6 g/dl, mean cell volume 75.6 fl). Liver and kidney function remained regular; lactate dehydrogenase, creatine kinase, proteins, amylase, immunoglobulins, electrolytes, enolase, peripheral blood smear, uric acid and urinary vanilmandelic acid were normal. Moreover, anti-nucleus antibodies and HLA B27 were negative. Suspecting endocarditis, a cardiologist was consulted but the clinical exam and ECG were negative.

Because of poor response, the antibiotic treatment was changed; the boy was given meropenem with teicoplanin for 10 days but again without success. Subsequently, we tried doxycycline and clarithromycin, but he still remained febrile with lumbar pain. In order to discover the agent responsible for his osteomyelitis, the lesion was biopsied in CT scan 20 days after disease onset. Bone marrow was taken too. The cultures of both bony and bone marrow specimens resulted positive for *S typhimurium*. The marrow around the bone trabecula had been replaced by inflammatory infiltrates with plasma and polymorphonuclear cells; areas with necrotic bone and fibroblasts were also found. *S typhimurium* was sensitive to levofloxacin and meropenem, although no clinical response had been obtained using the latter. Once levofloxacin was started fever and pain rapidly reduced and CRP decreased (2.45 mg/dl). The boy's immunological function was explored: lymphocyte subpopulations and granulocyte function were normal, while anti-HIV antibodies were negative. Two weeks after

biopsy, fever and pain in the iliac region started again associated with a slight CRP increase (4.44 mg/dl). A further MRI showed flogosis in the sacral and iliac bones, and gluteal and paravertebral muscles, with areas of colliquation (figure 1). A technetium bone scan revealed bone rearrangement in the same area. Linezolid was then added to the levofloxacin. Clinical conditions and blood alterations then improved. After 20 days, the MRI also showed slight improvement.

OUTCOME AND FOLLOW-UP

The boy was discharged, with levofloxacin, more than 2 months after admission. At home, he rested and had no pain or fever. Clinical exams remained negative. At the 6-month follow-up, the MRI revealed a progressive reduction of alterations of the iliac bone, the sacro-iliac joint and the muscles. Antibiotic treatment was definitively stopped 6 months after onset.

DISCUSSION

AHOM of the pelvis is a rare form (6.3–20% of cases) of childhood osteomyelitis rarely recognised primarily.¹ The ilium is the most common site, followed by ischium, pubis and acetabulum.^{1 2} Pain may be in the hip, thigh or abdomen.² Patients often limp or even refuse to walk; hip movement is often restricted. In contrast to osteomyelitis of the long bones, prior trauma is uncommon in pelvic AHOM.² Occasionally, children present local swelling

or erythema. The clinical features of salmonella infection are usually acute, with high fever, chills, severe bone pain, leucocytosis and elevated ESR; however, it may have an insidious onset. Usually the delay from onset of symptoms to start of treatment ranges from 7–13 days²; such delay has resulted in permanent disability in 3.4% of cases.² *S aureus* is the most frequent causative microorganism of pelvic AHOM, as well as for childhood AHOM overall.¹ Other causative bacteria, such as group A *Streptococcus*, *S pneumoniae*, *Fusobacterium* and *Salmonella*, have occasionally been isolated in pelvic AHOM.¹

Salmonella osteomyelitis, rare in healthy children, is common in children with haemoglobinopathies, especially with sickle cell disease. In these patients, bone infections are encouraged by the vaso-occlusive crisis.¹ *Salmonella* osteomyelitis has also been associated with immunosuppression—for example, in patients with systemic lupus erythematosus (SLE). Salmonellosis in SLE patients who were taking prednisolone is described with poor prognosis.³ Other predisposing conditions include previous surgery, connective tissue disorders and haematological neoplasms, such as lymphoma or leukaemia.¹ In patients not suffering from sickle cell disease, salmonella accounts for only 0.5% of all osteomyelitis cases, with spine involvement in approximately a quarter of these.² The few cases of non-sickle cell salmonella osteomyelitis were in women, with no immunocompromised state, but with recurring infections of the long bones.⁴ One girl, not affected by sickle cell disease, but with recurrent salmonella osteomyelitis of

Figure 1 MR image: flogistic elements in sacral and iliac bone, gluteal and paravertebral muscles with areas of colliquation.

both hands and feet from infancy until 19 years of age, was described.⁵

Tibia, distal femur and the navicular bone are other sites of salmonella bone infection in immunocompetent children.¹ Only two cases of pelvic salmonella were reported in previously healthy children.⁶ Generally, *S typhimurium* and *Salmonella enteritidis* are the common serotypes involved, which spread haematogenously.

Our case is singular because it represents one of the rare cases of pelvic salmonella osteomyelitis in a healthy child. He had no risk factors, immunodepression or prior event; although his bone deformity probably contributed to the degree of the infection. Unlike the previously reported cases, in our boy an infectious process was immediately suspected. Early diagnosis is important to prevent such potential complications as septic arthritis, chronic infection, avascular necrosis and growth abnormalities. As in the other cases, the MRI was the fundamental tool for diagnosis. The technetium-labelled bone scan was normal in the first weeks from symptom onset but became positive later.⁶ The bioptic drainage and its culture contributed highly to the aetiological diagnosis and represented the real treatment; in fact, from that moment the clinical signs and symptoms dramatically reduced. Finally, this case is unusual because of the extremely slow resolution of the infection.

In conclusion, salmonella pelvic osteomyelitis, although rare, occurs even in healthy children. Clinicians should be aware of salmonella osteomyelitis of the pelvis even in healthy children, and even when images are normal at

initial presentation, in order to start an adequate antibiotic treatment.

Learning points

- Salmonella pelvic osteomyelitis, although rare, occurs even in healthy children.
- Clinicians should be aware of salmonella osteomyelitis of the pelvis even in normal children, and even when images are normal at initial presentation, in order to start an adequate antibiotic treatment.

Competing interests None.

Patient consent Obtained.

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