

PELVIC OSTEOMYELITIS DUE TO *HAEMOPHILUS INFLUENZAE* TYPE B IN A DOWN SYNDROME CHILD

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Pelvic osteomyelitis in children is uncommon, and *Staphylococcus aureus* is the most commonly encountered organism. The diagnosis of pelvic osteomyelitis is usually delayed. With an early diagnosis and commencement of appropriate anti-microbial therapy, the outcome is usually favorable. We present a 14-month-old girl with Down syndrome and right iliac bone osteomyelitis secondary to *Haemophilus influenzae* type B. To our knowledge, the association of the elements of pelvic osteomyelitis, Down syndrome and *H. influenzae* has not been previously reported.

Case Report

A 14-month-old Saudi girl, a known case of Down syndrome, and a product of full-term uneventful pregnancy, was fully vaccinated except for the *H. influenzae* conjugate vaccine. The conjugate vaccine is not a part of the national infant immunization program in the Kingdom of Saudi Arabia. The patient was not known to have any congenital heart disease. She presented to the Emergency department of King Khalid University Hospital with a history of fever associated with signs of upper respiratory tract infection (URTI) for a few days. She did not appear toxic. Blood culture was taken and she was discharged home. She was called back on the second day because the blood culture grew gram-negative coccobacilli. The coccobacilli reacted positive with *H. influenzae* B anti-sera. The organism as well grew in chocolate agar incubated at 37°C with CO₂ overnight and grew around the XV factor, but not around the X or V factors individually in the nutrient agar. The growth characteristics were consistent with *H. influenzae* B. It was sensitive to ampicillin, chloramphenicol, and second- and third-generation cephalosporins. The patient was diagnosed as "bacteremia" and treated with intravenous cefuroxime 100 mg/kg/day in three divided doses. After two days, the cefuroxime was changed to the oral route as

FIGURE 1. Three-phase bone scan (Technitium Tc99 pyrophosphate) shows a focal area of increased uptake in the right iliac bone consistent with osteomyelitis.

her general condition improved and the fever subsided. Five days after she completed a 10-day course of antibiotics, she presented with fever and apparent discomfort on nappy changing. She appeared unwell with an axillary temperature of 38°C. She was hypotonic with no meningeal signs. There was tenderness on the right iliac bone with no swelling, redness or fluctuation. The rest of the physical examination was normal. WBC count was 11.2x10⁹/L with 34% polymorphs and 60% lymphocytes. The erythrocyte sedimentation rate (ESR) was 72 mm in the 1st hour. The patient was admitted and started on IV cefuroxime 100 mg/kg/day after repeating the blood culture, which turned to be negative. Considering the anatomy of the pelvis and the previous antimicrobial therapy, culturing the pelvic bone was not tried. Pelvic x-ray was normal. Three-phase bone scan (Technitium Tc99 pyrophosphate) showed a focal area of increased uptake, with matched increase in gallium uptake in the right iliac bone consistent with osteomyelitis (Figure 1). The antibiotic was shifted to ceftriaxone 100 mg/kg once daily. The fever subsided on the second day after admission. The right iliac bone tenderness gradually improved and the ESR normalized after four weeks. She completed four weeks of ceftriaxone. At four months' follow-up, she was well and with normal WBC and ESR.

Discussion

Down syndrome patients are susceptible to frequent and chronic respiratory and gastrointestinal infections.¹ However, a review of the literature shows that they are not particularly susceptible to bone infections, especially the pelvic bones. Williams et al. described a Down syndrome neonate with osteomyelitis due to a non-encapsulated *H. influenzae*, which was not previously reported.² In general, acute hematogenous pelvic osteomyelitis (AHPO) is an uncommon lesion.³⁻⁶ It constitutes about 6% of the total number of cases of acute hematogenous osteomyelitis (AHO). From a review of 93 cases of AHPO in children published in the literature, the following features seem to characterize this particular entity:³⁻⁷ it mostly affects older

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children and adolescents, with a mean age of 8 years, both sexes are equally affected, the triad of pain at the pelvis, the presence of fever and gait disturbance (limping) is seen in 75% of the cases, in infants, discomfort during nappy change replaces the gait disturbance, as was the case in our patient, and pain on hip movement with or without limitation and point tenderness are common presenting signs. Other less frequent, but important signs include soft tissue swelling and abnormal rectal examination with rectal tenderness and rectal wall edema in 25% of the cases.⁶ A history of significant trauma was documented in almost a third of the cases which could play a role in the pathogenesis.⁶ Patients often present to the hospital within the initial 10 days of the onset of symptoms. The correct diagnosis usually takes another 1-3 weeks in most of the cases. Only 16% of the cases had a correct initial provisional diagnosis.⁶

Many reports describe the deceptive nature of this illness, the diversity of clinical presentations and the difficulties with which the diagnosis is made.³⁻⁶ In our case, the initial presentation of the child consisted of fever, irritability and discomfort during nappy changing. The latter is an important sign to suspect pelvic osteomyelitis, but it was overlooked initially, and despite the early commencement of cefuroxime for the presumed bacteremia, the dose and the duration were suboptimal to treat the pelvic osteomyelitis. The discomfort persisted and the fever relapsed and the diagnosis was made only at the third week from the initial presentation.

Laboratory findings in AHPO includes polymorphonuclear leukocytosis in 60% of the cases. So, a normal WBC count is not uncommon at presentation. ESR is uniformly elevated by the fourth day of illness. Normal ESR after three days of presentation almost precludes the diagnosis of osteomyelitis.^{5,6} Blood culture is positive in almost half of the patients.⁶ Plain x-ray is usually normal at admission.

Pelvic ultrasound usually reveals a deep soft tissue swelling, which could be very helpful in the early diagnosis of AHPO.⁸ Nuclear scanning is often positive in the initial phase, however, it could be negative.⁶ In our patient, nuclear scanning was done two weeks after the initial presentation when the diagnosis of osteomyelitis was suspected, and was found to be positive. CT scan and MRI helped greatly in the diagnosis.

The most common bones involved are the ischium and ilium followed by the pubis. Some series show a predilection for the right side,³ as was the case in our patient. In 95% of the cases, only a single bone was involved.

The causative organism is isolated in 70% of the cases. *Staphylococcus aureus* is by far the most common (80%), followed by *Salmonella* (10%) in which half of the patients have sickle cell disease. *Streptococcus pneumoniae*, *H. influenzae B* and *Streptococcus pyogenes* each are isolated in about 3%.³⁻⁷ In our patient, *H. influenzae B* was isolated from the first blood culture. The negative blood culture at

the time of diagnosis was explained by the previous 10 days of antimicrobial therapy. The isolation of *H. influenzae B* in our patient who had Down syndrome is unique and had not been previously reported. It is not clear, so far, why bones are not involved in infection as frequently as respiratory or gastrointestinal systems in this immunodeficient group of population.

Empirical therapy of AHPO in children should be directed mainly towards *S. aureus*, the principal causative organism. If the child is known to have sickle cell disease, *Salmonella* should be considered. *H. influenzae B* was an important pathogen in osteoarticular infections in infants and young children in the pre-*influenzae B* vaccine era—with potentially serious associations mainly meningitis, which could be seen in 10% of cases.⁹ With the advent of widespread *H. influenzae B* vaccination, its incidence of bone and joint infections has decreased to near zero and hence, the coverage of *H. influenzae B* as part of the empiric therapy may no longer be needed if the child is fully immunized.¹⁰ As the *H. influenzae B* conjugate vaccine is not yet part of the national infant immunization program in Saudi Arabia, high doses of cefuroxime (150-200 mg/kg/day) or a combination of cloxacillin and ceftriaxone or cefotaxime is usually used in the empirical therapy. This can then be modified according to the microbiological data.

In the past, antibiotic therapy was recommended through the IV route for 4 weeks and then orally for the same duration.^{3,4} More recently, there is a trend for shorter antibiotic courses and an earlier shift to oral therapy.⁶ Antimicrobial therapy alone is usually adequate to achieve recovery in uncomplicated cases, as seen in our patient despite being immunocompromised. The duration of treatment we decided on—4 weeks—was based on her quick clinical recovery and normalization of the ESR. Surgery is reserved for complicated cases, which could be the result of either a failure of response to antibiotics alone or the development of an extra-osseous abscess.⁴⁻⁶ If diagnosed early and appropriate treatment is commenced, more than 95% of patients achieve complete recovery without significant long-term sequelae. The abundant blood supply to the pelvis may partially explain why antibiotic treatment alone is adequate in most patients.⁶

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