

Case Report

Rare Case Report of Unusual Cause of Spondylodiscitis in an Immunocompetent Lebanese Child

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Article History: | Received: 25.01.2020 | Accepted: 24.02.2020 | Published: 28.02.2020 |

Abstract: Salmonella typhi is still considered a health burden in low income countries despite the implementation of vaccination strategies since 1896. The incidence of typhoid fever in some parts of Asia accounts for 10 to 100 cases per 100,000 person-years (1). Typhoid fever typically presents with fever, malaise, constipation and abdominal pain. (2 and 3) Extra intestinal manifestations through hematogenous dissemination (including spondylodiscitis, osteomyelitis, meningitis, etc...) are very rare with a median prevalence of 5-10 % of all salmonella infections (4). Vertebral column invasion secondary to salmonella bacteremia is common in immunocompromised patients, patients with sickle cell disease and very rarely reported in the immunocompetent pediatric patient (5). In this article, we report a case of a 2 years 6 months old immunocompetent Lebanese child, who presented with fever, diffuse abdominal pain and a remarkable abdominal distension of 2 weeks duration, who was found to have salmonella spondylodiscitis.

Keywords: Salmonella Typhi (S. Typhi), Salmonella Paratyphi (S. Paratyphi), Spondylodiscitis.

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CASE PRESENTATION

Our patient is a 2 years 6 months old Lebanese female, previously healthy, with a promptly followed vaccination schedule, who presented to the Emergency Department for high grade fever and diffuse abdominal pain. History goes back to 4 weeks prior to presentation when she had fever and diarrhea treated at home with probiotics and oral rehydration solution and improved within 3 days. 2 weeks later, the patient's fever spiked again, reaching 40°C, and partially responded to antipyretics; the patient displayed again diffuse abdominal pain with vomiting, poor oral intake and constipation. The patient was admitted to another Hospital for 6 days where full laboratory workup and imaging were done and revealed: Entamoeba Histolytica cysts in stool analysis. During her stay, the patient received Ceftriaxone for 4 days and Metronidazole for 3 days. As her symptoms persisted, Widal and Wright tests were ordered and the patient

requested transfer to our care. Upon admission, the patient was febrile but hemodynamically stable, on physical examination, she had a moderately distended non rigid and diffusely tender abdomen with no costovertebral tenderness, Skin inspection didn't reveal any rash or skin lesions, no cervical or inguinal lymph nodes were noted and genitourinary examination didn't show any abnormal or remarkable finding. Initial laboratory investigations showed no leukocytosis nor left shift (WBC10500 with neutrophils 59,6 %), no thrombocytopenia was noted (platelets 335000), but normocytic anemia was noted (Hb 9 and MCV77), elevated acute phase reactant markers (CRP 120 mg/dl and ESR 44) with slight electrolytes disturbances (Na 140, K 3.74, Cl 100 and CO₂=16.6). KUB standing showed multiple small air fluid levels in the lower abdomen with fecal loading in the colon (ileus) (**Figure 1**).



Figure 1: Abdomen (KUB): multiple small air fluid levels in the lower abdomen with fecal loading in the colon (ileus)

Two days later, the patient’s abdominal pain and distension increased in severity, she began complaining of lower back pain extending to the hips bilaterally and refused to stand on her feet or to bear weight. CT-scan of the abdomen and pelvis showed: epidural extramedullary soft tissue mass within the

spinal canal at L2-L3 level compressing the thecal sac. Lumbosacral MRI discerned an anterior epidural mass with a lateral compression of the dural sac associated with an infiltration of the psoas muscles; findings were in favor of an infectious process more precisely a spondylodiscitis with an epidural extension (**Figure 2**).



Figure 2: MRI Lumbosacral: sagittal T1 after gadolinium injection showing abnormal signal intensity involving L2 L3 intervertebral disc with erosion of endplate and abnormal enhancement extending to the spinal cord and medullary canal denoting spondylodiscitis.

Widal titer result came out and was highly elevated (1/5120), blood and stool cultures taken within the second week of illness were negative. All the clinical, biological and radiological findings were highly suggestive of salmonella Spondylodiscitis. An intravenous regimen of Ceftriaxone, Vancomycin were started and continued for a total course of 4 weeks and Metronidazole was discontinued after 10 days. The patient didn’t undergo any surgical intervention for biopsy or for symptoms control as she was improving

on IV antibiotics. After 2 weeks of antibiotherapy, the patient’s clinical condition improved; and her lower back pain extending to the hips bilaterally started to relief, her refusal to stand on her feet or to bear weight was also improved with regression of the acute phase reactants markers: (WBC 9300 with neutrophils 54% and CRP 0.38). The repeated MRI of the lumbosacral tract showed a slight improvement of the previously reported findings. The clinical and radiological improvements were our guide to continue with the

ongoing therapy without proceeding to CT-guided biopsy. Follow up was arranged in 4 months for re-evaluation of disease relapse as an organized outpatient visit and the patient revealed a normal musculoskeletal exam with no trace of sequela.

DISCUSSION

Salmonella species is a facultative anaerobe gram negative bacillus (6) usually acquired by ingesting contaminated food or water, consuming raw or undercooked eggs and meat, handling or consuming reptiles like turtles, lizards and snake (7). Infections commonly present with gastroenteritis as typhoid fever and it is an uncommon cause of bacteremia in hospital (6). Studies have shown that salmonella infection is eradicated through both cellular and antibody mediated responses. Since Salmonella infections are associated with serious neurological and multiorgan complications, vaccination against salmonella species has helped reducing the major global health problem imposed by the emergence of these infections in the endemic areas. Currently, two vaccines have been established to provide protection against invasive Salmonella disease. Both are not licensed for younger subjects and provide no lifelong protection against the bacterium. Ty21a, given as a single oral dose, is the only approved live attenuated vaccine. Thus, it's able to mediate cellular and humoral responses but it is only recommended for adults and children older than 5 years of age. Vi CPS, given in multiple doses, is derived from salmonella polysaccharides and generates a cellular-mediated immune response (8). Despite its poor immunogenicity, this vaccine is prohibited for individuals younger than 2 years of age (8). Our patient was 2 year and 6 month old and had received her first dose of Vi CPS 4 months prior to admission. Her presenting condition is a clear example of the vaccine's poor efficiency. Bone and joint infection associated with salmonella account for less than 1% of all salmonella infections and as a cause of osteomyelitis, salmonella is rare, accounting for just 0.45 % of osteomyelitis cases (6). Spondylodiscitis (vertebral osteomyelitis), a combination of discitis (inflammation of one or more intervertebral disc spaces) and spondylitis (inflammation of one or more vertebrae), and these are uncommon disorders accounting for nearly 2% of all bony infections. The presentation could be sub-acute or even chronic that the patient wouldn't be able to recall the inciting event (9). Pyogenic spondylodiscitis is mostly due to staphylococcus aureus, both in adults and children, gram negative species are found in 15% of cases (5). Non pyogenic spondylodiscitis is mainly due to Mycobacterium tuberculosis infections in endemic areas (3). Spondylodiscitis caused by salmonella is encountered very rarely. While the literature demonstrates that these infections occur as a result of the hematogenous dissemination or bacterial spreading through the lymphatic system (10), Sickle cell associated vertebral column infections could show a good example. In fact, sickle cell crisis triggers the

formation of intestinal micro infarcts leading salmonella gut flora into the bloodstream and further on to the vertebral column (4). Our patient was an otherwise healthy child, with no apparent hematological or immunologic disorder according to prompt clinical evaluation and family history. Confirmation with hemoglobin electrophoresis and serologic testing for HIV and autoimmune diseases is still required. The lumbar region is mostly involved, as is the case in our patient (4, 6, 9 and 11) followed by thoracic vertebrae (4) with a male predominance (9). In most vertebral column infections, the disease is confined to vertebral bodies sparing the disc spaces due to its poor vasculature. However, in young subjects, the vertebral endplate is richly vascularized, facilitating direct hematogenous access to the disc space (2 and 5) and favoring the inflammatory process. Although enteric fever usually manifests as fever, vomiting, diarrhea and bacteremia, the typical clinical presentation of spondylodiscitis consisting of fever and back pain may not be preceded by a similar prodrome (10 and 11). Furthermore, only a few cases with neurological compromise caused by Salmonella spondylodiscitis have been reported in the English Medical literature (8 and 10). Our patient developed lower back pain radiating to the hips bilaterally and refused to stand on her feet or to bear weight. Her symptoms followed a classical typhoid fever. In the absence of any consensus about gold-standard diagnostic tools, the diagnosis of salmonella spondylodiscitis depends on the age of the patient, presence of neurological symptoms, and accessibility of tissue biopsy. In one series of 11 patients, Amritand et al found 9 positive blood cultures growing either *S. Typhi* or *S. Paratyphi*. The diagnosis of Chang's 8 patients was made by either blood and stool or tissue cultures. In both cases, Widal test was positive. The diagnosis of our patient was confirmed by a positive serology and suggestive findings on MRI. Biopsy was not obtained to prevent invasive procedure and as the clinical improvement on empirical treatment was achieved within the first two weeks. Treatment of Salmonella spondylodiscitis depends on the need for surgical intervention and bacterium species in terms of treatment length, route and type of administered antimicrobial agents (11). As aforementioned, positive cultures allowed clinicians to isolate the bacterium, and was the mainstay of the treatment decision in all studies we have searched. Thus, in the absence of any neurological manifestation, the regimen used was intravenous Ceftriaxone or Ciprofloxacin for at least 4 weeks in young patients (2). Eight to twelve week courses were more commonly followed (11, 12 and 13). In contrary, a surgical intervention- regardless of its indication- will extend the course of antibiotics for 12 to 20 weeks. Antibiotics are first given intravenously until switching to oral route becomes possible (11). Our patient received 4 weeks of empirical parenteral antibiotic therapy (Ceftriaxone and Vancomycin), a tissue biopsy was not performed and blood, urine and stool cultures turned out to be negative. In the absence

of complications or comorbidities, sequelae are rare. Patients are followed on 16 to 22 months basis, according to complications and comorbidities, and imaging is repeated to detect relapses or to follow on the outcome of procedures.

CONCLUSION

Salmonella Typhi is the most isolated pathogen among patients presenting with enteric fever. Though bone and joint infections due to salmonellosis are still extremely rare in the pediatric population, investment in the development of better vaccines is of tremendous importance given the serious complications and morbidity that can be caused by these types of infections. Innovation in vaccination strategies and planning, especially in the endemic areas and developing countries is the mainstay for the prevention from elevated morbidity and mortality resulting from salmonella infections.

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