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ORIGINAL ARTICLE

Pyogenic sacroiliitis in children: don't forget the very young

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Abstract

Pyogenic sacroiliitis (PS) is rare with less than 100 pediatric cases reported in the medical literature. To better characterize PS in the pediatric population, we investigated a series of children presenting with PS. Retrospective data analysis was done at an academic tertiary center between the years of 2000 and 2017. All hospitalized children ≤ 16 years of age with PS were evaluated. Of the 894 children hospitalized with osteoarticular infections, 18 were diagnosed with PS (2%) and are included in the review. Two clinically distinct groups were identified. PS in infants ($n = 13$, 72.2%, mean age 1.1 years) had an indolent course and a faster recovery without any bacterial source identified. In contrast, the group of older children ($n = 5$, 27.8%, mean age 11.6 years) had a more complicated course and a higher rate of identified bacterial infections.

Conclusion: We describe an under-recognized entity of PS in infants with a mild clinical course and fast recovery that differ from the "classical" septic sacroiliitis. Infants with PS did not suffer from invasive complications, and pathogen characteristics of older children were not identified. Infants with fever, irritability, decreased range of motion in the pelvic area, and pain during diapering should alert the clinician to this diagnosis.

What is Known:
- *Pediatric pyogenic sacroiliitis is an extremely rare condition usually caused by Staphylococcus aureus with highest incidence in adolescents.*
- *The diagnosis of PS is challenging due to its rarity and difficulty in assessing the sacroiliac joint.*

What is New:
- *We describe an under-recognized entity of PS in infants with a mild clinical course, without invasive complications and with fast recovery that differ from "classical" septic sacroiliitis.*
- *Infants with fever, irritability, decreased range of motion in the pelvic area and pain during diapering should raise clinical suspicion of this diagnosis.*

Keywords Osteoarticular infections · Osteomyelitis · Septic arthritis · Infants · *Kingella kingae* · *Staphylococcus aureus*

Abbreviations

GPC	Gram-positive cocci
MSSA	Methicillin-sensitive *Staphylococcus aureus*
PS	Pyogenic sacroiliitis

Introduction

Septic arthritis is common in children and usually caused by blood-borne *Staphylococcus aureus*, most frequently involving the knee (40%) and the hip (22–40%) joints [4]. Pyogenic sacroiliitis (PS) is a rare condition accounting for 1–2% of all osteoarticular infections in children with less than 200 cases in all age groups and less than 100 pediatric cases reported in the English literature [14, 18]. The incidence of PS is highest in adolescents and young adults, presumably due to the enhanced blood supply to the sacroiliac joint at this age. The diagnosis of PS is challenging due to its rarity, difficulty in assessing the joint, and the low yield of diagnostic findings identified in conventional radiographs. The clinical manifestations of PS including limping and inability to bear weight are common complaints in the pediatric population and are associated with a broad differential diagnosis which is age dependent [5]. The non-specificity of these findings can lead to the wrong diagnosis. The diagnosis is even more difficult in

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younger children as it is often hard to localize the source of the pain. A delay in the diagnosis of PS may result in severe complications such as abscess formation or sepsis [12]. Conventional X-rays have low sensitivity in the early phases of the bone infection in general and even less so in the sacroiliac area. Bone scintigraphy can identify the pathological focus, but does not allow appreciation of the extent of the involvement of the bone and the joint. MRI is the best imaging modality, allowing clear visualization of bone and articular involvement and complications. However, one should first localize the area of interest. If such clinical decision is difficult, localization of the disease with bone scintigraphy, with following MRI could provide maximal imaging information [8]. In this study, we describe the manifestations and clinical course of pediatric PS diagnosed during 2000–2017 at our Center. We describe a previously under-recognized entity of very early PS with a clinical phenotype and course that differ from the septic sacroiliitis found in older children. Our findings underscore the importance of a high index of suspicion which would facilitate the diagnosis of PS in infancy.

Methods

We reviewed the database of the Hadassah-Hebrew University Medical Center (a large tertiary medical center located on two campuses with a combined ~ 30,000 emergency room admissions) between the years of 2000–2017. We searched for ICD-9 code 720.2 in children ≤ 16 years for the diagnosis of sacroiliitis. For the diagnosis of osteomyelitis and septic arthritis, we retrieved the ICD-9 codes 730 and 711 respectively. All cases of PS in hospitalized children were identified and reviewed.

PS was diagnosed by a combination of clinical signs including presence of high fever (above 38.5 °C), markers of increased systemic inflammation, and radiological findings consistent with sacroiliac joint inflammation on bone scintigraphy, CT scan or MRI. Patients with autoimmune spondyloarthropathy, spondylodiscitis, or reactive arthritis were excluded from the analysis. Patients with malignant and hematological disorders or with immunodeficiency were not excluded; however, no such patients were identified in our cohort.

Medical charts of the patients were retrospectively reviewed, and ambulatory post-discharge follow-up was performed in all patients. An analysis of incidence, age, gender composition, presenting signs and symptoms, radiologic findings, treatment, and outcome was performed. Statistical comparison between the groups was done by Student's *t* test or Mann-Whitney test. Follow-up of cases was done by phone questionnaires and included the presence of any musculoskeletal complaints or sequela of PS.

The institutional review board of the Hadassah-Hebrew University Medical Center approved the study protocol (0140-15-HMO August 2015). Oral informed consent was obtained from all patients prior to phone interview.

Results

Demographics

During the study period a total of 894 children were diagnosed with osteoarticular infection: 686 (76.7%) with osteomyelitis and 208 (23.2%) with septic arthritis. Within this group, 18 (2%) children were diagnosed with PS (13 males, 72.2%). The mean age at diagnosis was 4 ± 5.2 years (range 0.75–16 years).

Two subgroups based on age of presentation were discernable among the PS patients. The patients in one group ($n = 13$, 72.2%) were infants and toddlers (mean age 14.1 ± 5.8 months, range 7–26 months), while the remaining 5 children in the second group were school age (mean age 11.6 ± 4 years, range 6–16 years, $p = 0.002$). Patient characteristics are summarized in Table 1.

Clinical presentation

Clinical symptoms in infants were non-specific and included irritability (8/13, 61.5%), refusal to walk (5/13, 38%), sit, crawl, and pain during diapering (4/13, 31% for each). In contrast, 4/5 patients (80%) in the older group complained of lower back pain. Younger children presented on average earlier than the older ones (Table 1). On examination, the most common finding was sensitivity above the sacroiliac area (4/5, 80% of older children; 8/13, 61.5% of infants). Elevated blood inflammatory markers (e.g., white blood count, C reactive protein or erythrocyte sedimentation rate) were found in all cases with mean leukocyte count of 13×10^3 cells/μL (range 12,300–19,000), ESR of 52.8 mm/h (range 15–100), and CRP of 7.8 mg/dL (range 0.5–14.7). Most inflammatory parameters that were measured were similar between the younger and the older group of children. However, neutrophilia was present only in the older age group (mean PMN %, young (52%) vs. old (77%), $p = 0.003$) (Table 1).

Blood cultures were performed in all patients and were positive in 3 (two methicillin-sensitive *Staphylococcus aureus* (MSSA) and one *Streptococcus pneumoniae*), all of whom were in the older group (60%). Two patients from the older group, one of whom had a concomitant positive blood culture, developed local abscesses which were surgically drained and grew MSSA. In summary, in 4/5 (80%) of the older children, a pathogen was identified from blood or articular fluid, while none of the younger children had positive cultures. It should be noted that our laboratory routinely cultures for *Brucella*

Table 1 Clinical and demographic characteristics of children with PS

	All ($n = 18$)	Young* ($n = 13$)	Old* ($n = 5$)	p value
Age, year	4 (1.2)	1.1 (0.1)	11.6 (1.8)	
Females, (%)	27	36	20	1
Duration of symptoms prior to admission (days)	3.7 (0.7)	2.8 (0.5)	5.9 (1.9)	0.05
White blood cells (10^3)	13.3 (1.2)	15.2 (0.7)	13.5 (0.6)	0.199
Polymorphonuclears (%)	59.3 (4)	55 (3)	77 (5)	0.003
Pathogen isolated (%)	22.2	0	80	0.002
Intravenous treatment (day)	17.8 (3.1)	12.2	32.6 (6.8)	0.05
Total treatment (day)	33.9 (2.7)	32.8 (3.5)	36.8 (3.2)	0.53
Admission length (day)	12 (1.3)	12.4 (1.7)	10.8 (1)	0.342

Mean (SEM). *Young, children younger than 30 months; old, children 6–16 years of age

species, and none of the patients had a positive *Brucella* culture.

Imaging studies used to support the diagnosis of PS included: bone scintigraphy (Fig. 1) in 15 patients, CT in 3, and MRI (Fig. 2) in 4.

Clinical course

Almost all the patients in our study group received intravenous, followed by oral, antibiotics. In the younger group, 10 (77%) children were treated with cefuroxime. Others were treated with 1st or 3rd generation cephalosporins, penicillin-G, or methicillin. One child in the younger group did not receive antibiotic treatment and recovered spontaneously. Average duration of antibiotic treatment was 33.9 days (range 0–42) and did not differ between the young and the old groups. However, older children were treated significantly longer with intravenous antibiotics as compared to the younger ones (32.6 (7–42) days vs. 12.2 (0–21) days, $p = 0.03$) (Table 1). Complications arose only in the older group—two patients developed a local abscess requiring surgical drainage, in both with a recent history of deep skin laceration. Clinical suspicion of an abscess arose due to persistence of fever and systemic inflammation despite intravenous treatment with a subsequent CT scan confirming the clinical diagnosis. In these two patients, articular fluid was positive for MSSA.

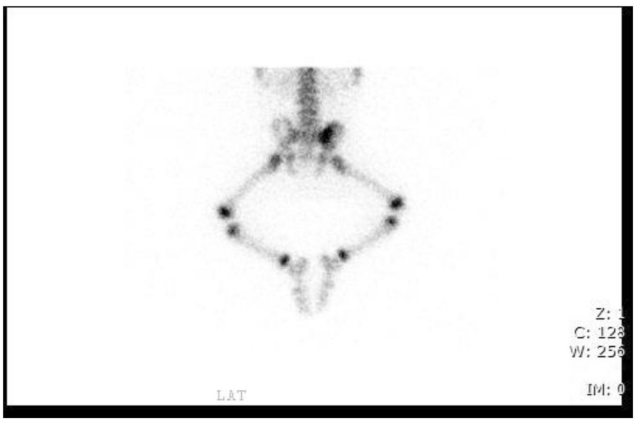

Fig. 1 Tc 99-m bone scan image (PA) of 9-month-old boy, demonstrating increased uptake in the right sacroiliac joint

Telephonic follow-up of all patients after an average of 10.6 years (range 1–18 years) from the initial diagnosis revealed that none of the patients in our study had residual musculoskeletal complaints or other manifestations that could be related to PS.

Discussion

In this study, we describe a novel under-recognized clinical entity of infantile sacroiliitis. This group of infants had mild disease without significant sacroiliac joint inflammation, negative microbiological workup, and responded to a short course of antibiotic therapy. The majority of infants had limitations in their ability to ambulate (sit, walk, or crawl) or irritability when being diapered. This disorder differed from the classical school-aged PS which had a more aggressive course, frequently complicated, with high systemic inflammation, positive cultures, and mandates prolonged antibiotic treatment.

In our series, PS represented 2% of all pediatric osteoarticular infections during the study period, which is consistent with other reports [14]. In previous reports of PS, most of the cases involved older children. Schaad et al. summarized previously reported cases and small series of a total of 77 children with PS [13]. None of the children reported were under the age of 2 years. Two more recent smaller scale studies also did not report on children with PS who were under the age of 2 years [11, 16]. Only a few cases of infantile PS have been reported [7]. In contrast, we found that among the 18 patients with PS in our series, the majority (72%) were less than 2 years old. It is also interesting that none of our patients were preschool age (2.5–6 years). Recently, Donzelli et al. reported a similarly bimodal age distribution and distinct clinical manifestations [1].

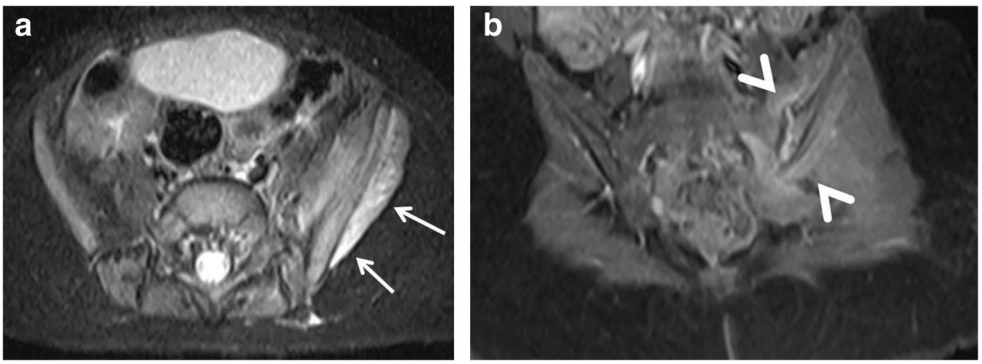

Fig. 2 Axial T2 weighted with fat suppression (**a**) and coronal T1 weighted fat suppressed contrast enhanced (**b**) MRI images of 9-month-old boy. Noted are marked bone marrow edema in the left iliac bone, sacrum, soft tissue edema, and enhancement. Mild amount of fluid is seen in the left sacroiliac joint

Older children in our cohort differed from infants and toddlers as they presented later with higher rates of bacteremia and abscess formation requiring surgical drainage suggesting a more advanced and invasive PS at presentation. Gram-positive cocci (GPC), and specifically MSSA, were the most common pathogen. MSSA is a well-recognized pathogen in PS, accounting for up to 75% of older childhood in reported cases of PS [14, 17] .

The association that we found between skin trauma and MSSA has been previously described in adult patients [15]. None of the children in our study has community-associated methicillin-resistant *S. aureus* (MRSA) infection that has recently emerged as a potential pathogen for osteoarticular infections [6, 10].

The different clinical presentation between the two age groups may be related to a less aggressive pathogen in the young age group, specifically *Kingella kingae*, an emerging pathogen in osteoarticular infections in young children, accounting for up to 53% of all osteoarticular infections in children younger than 4 years [3]. This would also explain the paucity of positive cultures in our cohort of young children, as *K. kingae* is difficult to isolate with routine culture methods, and necessitates molecular diagnoses in many cases [2], which were not routinely done at our center. Another possible explanation may be the presence of a unique non-bacterial inflammatory response in the young age group to a yet unknown stimulus. This could be analogous to transient hip synovitis, with a predisposition to the sacroiliac joint.

Cefuroxime is a reasonable empirical choice for pediatric PS, regardless of age, as it provides adequate coverage for both GPC and *K. Kingae*. The optimal duration of parenteral therapy is less clear. In our series, duration of treatment ranged from 7 to 42 days. This wide range reflects the lack of evidence-based guidelines for the duration of parenteral treatment in pediatric osteoarticular infections during the period of the study [9].

Limitations of the study are its retrospective nature and the wide range of diagnostic and therapeutic modalities utilized during the long course of the study. In addition, the infectious agent was not identified in most of the cases. Joint aspirates allowing for culture and PCR based would have most likely increased diagnostic yields for the pathogenic bacteria, but such invasive testing was not clinically justified.

In conclusion, we describe two clinically distinct age groups presenting with inflammation of the sacroiliac joint. The two PS phenotypes differ in their severity and microbiological findings. Although the diagnosis of PS is challenging in infants and toddlers, pediatricians should be alert to this diagnosis in this age group as it is more common than previously reported. Specific clinical clues such as fever, irritability, and pain upon walking, crawling, or diapering can aid the diagnosis, in conjunction with findings of systemic inflammation. When clinical suspicion arises, an imaging study, either MRI or bone scintigraphy, is recommended for definitive diagnosis. Further studies are needed to corroborate our findings and to identify the pathophysiology associated with infantile PS.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval The Institutional Review Board of Hadassah-Hebrew University Medical Center approved the study protocol (0140-15-HMO August 2015). Oral informed consent was obtained from all individual participants included in the study prior to phone interview.

Authors' Contributions EL, AGH & YB designed the study, collected and analyzed the data and drafted the manuscript. NS, DA and IW provided substantial contribution to draft the paper and reviewed the final manuscript. All authors gave their final approval of the version to be published and agree to be accountable for all aspects of the work.

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