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Case Report

Paediatrics Section

Eosinophilia in an Acutely Limping Child: An Easy Guess of Rare Systemic Aetiology!

HARSHITA SHARMA[1], SANTOSH VENKETRAMAN KONDEKAR[2], MAAZ AHMED[3], SURBHI RATHI[4]

ABSTRACT

Painful limping child is often considered due to septic arthritis. Iliopsoas abscess (IPA) is rare in adults and children. The aetiology is often related to tuberculosis of spine. Hereby we report a case of staphylococcal IPA where incidental evaluations led to obvious diagnosis of a rare primary immunodeficiency syndrome called Job syndrome or hyperimmunoglobulin E and Eosinophilia Syndrome (HIES). This was the first case of IPA in a case of immunodeficiency syndrome including HIES.

Keywords: Iliopsoas abscess, Job Syndrome, Septic arthritis

CASE REPORT

A five-year-old boy presented with 3 weeks history of afebrile limping of left leg with difficulty in walking with mild pain across back to groin and difficulty in squatting. The child had maculopapular rash since birth; had no dysmorphism and had normal milestones. There was no history of fever, trauma or hospitalization in past. The rash was not itchy papular with spread mainly around head and neck. There was no family history of rash or any other disease including tuberculosis.

On examination, there was no fever but restriction of flexion and extension of left hip movements beyond 45 degrees was noted along with tenderness over left illaic fossa. Septic arthritis being the commonest cause of non-traumatic acute limping in pediatric age group; a blood count and ultrasound hip was asked for. The other differential diagnoses for painful hip are trauma, osteomyelitis, tuberculosis, psoas abscess and Perthes' Disease.

Blood total cell count was 27000/cumm with polymorphs 78%, lymphocytes 16% and eosinophils 5%; ESR was 24 mm/h with Hb of 11 gm%. Ultrasound abdomen confirmed an iliopsoas abscess (IPA) with approximately 50ml pus collection. Abscess was drained under ultrasound guidance. It grew *Staphylococcus aureus*. A diagnosis of staphylococcal IPA was confirmed. Repeated blood counts were normal except absolute eosinophil count 3000. A suspicion of primary immunodeficiency with hypereosinophilia was suspected and after serum IgE level came as 15000 IU/ml, a diagnosis of hyperimmunoglobulin E and Eosinophilia syndrome (HIES) was made. Evaluation for tuberculosis in the form of X-ray chest, Mantoux test and pus smear and culture for acid fast bacillus was negative.

Patient was effectively treated with vancomycin 45mg/kg/day for 14 days in divided doses and sent home with antibiotic prophylaxis with cotrimoxazole 8mg/kg/day for 2 months. Child had no pain or restriction of hip movements on discharge.

DISCUSSION

Mynter et al., described Psoas abscess first in 1881, called it psoitis [1]. Even in adults the entity is not common, and often the aetiology is tuberculous [2]. Even the latest case in a child was secondary to Potts spine [3]

IPA is usually not considered evaluating the limping child on priority [4]. Most causes of limping are orthopaedic conditions related to trauma, dysplasia and infections [4]. Amongst infections, proximally only septic arthritis, avascular necrosis of femur head and TB of hip are commonly considered. Diagnosis of IPA has always been delayed in a limping child [4].

A 15 year radiological study from France reported only 16 such cases [5]. Even in adults IPA is considered as rare declining entity, with vague presentations and diagnostic delays [2]. Commonest reported aetiology is related to tuberculosis i.e. Potts spine [2]. Neonatal idiopathic IPA is reported even as bilateral IPA with MRSA [6,7]. Bilateral IPA was also reported in a child following tuberculosis [8]. A Turkish study recently reported case series of five cases of IPA of which four were staphylococcal and one was tuberculosis. Even streptococcal and *Enterococcus faecalis* aetiology is known in children [9,10]. IPA is also rarely known in Crohn's Disease, Sickle cell disease and following appendisectomy [11,12]. It is also seen as a complication of appendicitis and chicken pox [13,14]. This case also grew Staphylococcal aspiration on pus culture, the commoner organism in children [4]. The very reason to discuss all these cases being, there is no single case reported in a case of immunodeficiency syndrome yet.

Immunodeficiency syndromes make a child prone for invasive bacterial infection even from neonatal age group. Hyper IgE syndrome or Job Syndrome was first described in 1966 as a child with multiple skin abscesses [15]. Buckley et al., had described similar cases [16]. Later they were combined as Job Syndrome which later amalgamated as Hyperimmunoglobulin E with eosinophilia syndrome (HIES). Depending on aetiology and inheritance HIES is classified as AD and AR. AD is known to be associated with STAT 3 mutations [17]. Immunoglobulin E levels range from 400 IU/ml to 25000 IU/ml and eosinophilia may range from 400 IU/ml to 20000 IU/ml [18]. Some children have dysmorphism since birth in the form of facial asymmetry, deep seated eyes, large skin pore size and prominent forehead [17,18]. As in our case, cases with maculopapular rash since birth are reported in HIES. Superficial scalp and skin abscesses are common and deep seated skin abscesses and repeated staphylococcal pneumonias and pneumatoceles are frequent in a case.

Despite being rare, series of 43 patients is on record studying linkage of HIES to chromosome 4 [19]. Another series of 50 hyper IgE cases is reported by Freeman et al., studying brain abnormalities [17]. Renner et al., in a series of 14 patients described AR as distinct entity [20]. During the Indian literature review, we noted that boys with HIES had abscesses at earlier age (less than age 5 years) compared to girls, and we could not find a case report of HIES with dysmorphism in a boy [21-29]. There are loose case reports of associations of HIES with dysmorphism, neonatal teeth, cervical rib, BCG associated tuberculosis, gingival hyperplasia, osteomyelitis, meningitis and molluscum [21-29]. HIES and other immunodeficiency syndromes are known to cause serious abscesses at various sites, but IPA in a case of HIES or Job syndrome is not yet reported.

Though, there were diagnostic hurdles to consider septic arthritis as first diagnosis in this case, an ultrasound for abdominal pain associated with same helped us zero down to the IPA and hence child was treated effectively in time. Incidental eosinophilia helped us investigate for serum IgE. The diagnosis would have probably missed if eosinophilia was overlooked.

CONCLUSION

Iliopsoas abscess should always be kept in mind while evaluating a limping child; and a CBC and ultrasound hip should be asked for if there is acute restriction of joint movements. An eagle's eye is needed to look out for eosinophilia in any case of abscess that may unmask an overlooked hyper IgE syndrome.

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PARTICULARS OF CONTRIBUTORS:
1. Intern, Department of Pediatrics, TN Medical College and BYL Nair Charitable Hospital, Mumbai, India.
2. Associate Professor, Department of Pediatrics, TN Medical College and BYL Nair Charitable Hospital, Mumbai, India.
3. Speciality Medical Officer, Department of Pediatrics, TN Medical College and BYL Nair Charitable Hospital, Mumbai, India.
4. Professor, Department of Pediatrics, TN Medical College and BYL Nair Charitable Hospital, Mumbai, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Santosh Kondekar,
Associate Professor, Department of Pediatrics, First Floor, TN Medical College Mumbai-400008, India.
E-mail: drkondekar@gmail.com

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