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HIDING BEHIND A FORESHORTENED CHROMOSOME – CML WITH PYOMYOSITIS – A CASE REPORT AND LITERATURE REVIEW

Haematology

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ABSTRACT

Occurrence of pyomyositis in CML is very rare with only three documented cases in literature so far. This is attributed mainly to the neutrophil functional defect associated with poorly controlled CML as most of these cases are diagnosed at CML presentation or in situations of resistance. Here we report one such case of CML, who presented with fever & pain in medial side of left thigh with elevated total counts and platelets after 4 years of fairly compliant therapy with Imatinib and was diagnosed as CML with Pyomyositis This report highlights many important facts - the significance of TKI Domain Mutational Analysis, the less common morbidities faced by these patients in the TKI era and the need to address the gap in the access to molecular investigations.

KEYWORDS

CML, Pyomyositis

BACKGROUND

Pyomyositis is a rare, subacute, deep pyogenic infection of the muscle tissue.[1] Pyomyositis often occurs in immunocompromised patients with underlying hematologic malignancy. Most frequently , it presents as a local abscess, but it can also manifest as a diffuse inflammatory or rapidly progressing myonecrotic process. In the occasion of failure to initiate early and appropriate treatment, pyomyositis can cause significant morbidity and mortality in immunocompromised patients. In our patient, it might be attributed to the poor neutrophil function associated with CML and it's treatment failure with Imatinib.

CASE VIGNETTE

A gentleman in his 30's came limping to the medical oncology OPD who is a diagnosed case of CML for nearly 4 years and was on a fairly compliant therapy with Imatinib since December 2019. His only drug free period was from July 2021 to November 2021 during the COVID pandemic. He presented to us with worries about his persistently elevated CBC parameters and also complained of high grade fever and pain in the left hip region for the past 20 days. Physical examination showed pallor, hepatomegaly of 6cm below the Right costal margin and splenomegaly of 12 cm below the Left costal margin reaching the umbilicus. On examining his hip region, the left lower limb was flexed and externally rotated at hip joint and flexed at knee joint. Extension at the hip joint was restricted due to the pain. A hard mass of 10x8 cm was found along the medial side of left thigh extending from left hip to mid thigh. His past medical records in 2021 and 2022, showed that CHR was never attained

Table 1: Peripheral Smear Reports From The Past

Peripheral smear	July 2021	November 2021	March 2022	September 2022
WBC	Increased	Increased	Increased	Increased
Blasts	2%	-	2%	4%
Promyelocytes	2%	1%	5%	15%
Myelocytes	24%	9%	32%	12%
Metamyelocytes	6%	22%	6%	10%
Band forms	12%	30%	12%	4%
Neutrophils	48%	25%	37%	43%
Lymphocytes	4%	6%	4%	2%
Basophils	2%	3%	2%	8%
Platelets	Normal	Normal	Normal	Normal

Table 2: Bone Marrow Reports From The Past

Bone marrow	July 2021	November 2021	March 2022	September 2022
M:E	3:1	7:1	8:1	12:1
Blasts	<3%	<5%	<5%	4%
Erythropoiesis	Normal	Suppressed	Suppressed	Suppressed

So, we proceeded with theTKD mutational analysis (Table3).

No TKD Mutations was found & he was started on cytoreductive therapy with oral Hydroxyurea and Nilotinib. Total counts started to come down but the platelets remained high. MRI of the left hip region demonstrated a T1 hypointense and T2 hyperintense abscess of size 7.1x7x7.6 cm with surrounding edema in the intra and intermuscular plane in the left obtruator group of muscles.

Table 3: ABL Kinase Domain Mutations Analysed

ABL1 kinase domain subunits	Mutations analysed
P loop	E255K/V, Y253H, G250E
C helix	V299L
C loop	F359V/C/I
Drug contact site	T315I/A, F317L/V/I/C
Myristate pocket	-

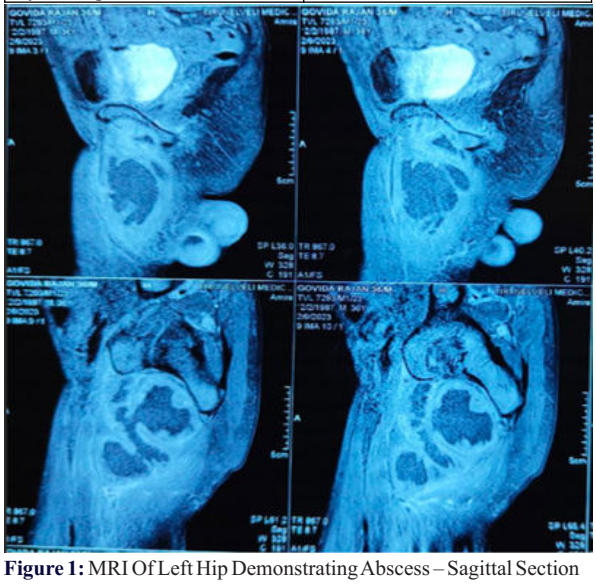

Figure 1: MRI Of Left Hip Demonstrating Abscess – Sagittal Section

I&D of the abscess was done with broad spectrum IV antibiotics and the platelets normalised in a few days and the patient was discharged home with Nilotinib. The trends of his CBC in the hospital stay is shown below.

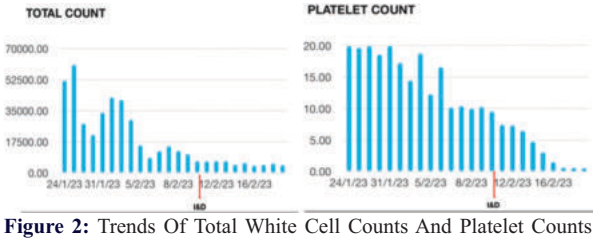

Figure 2: Trends Of Total White Cell Counts And Platelet Counts During The Hospital Stay

At his one month follow up visit, all of his CBC parameters were within normal limits with no palpable spleen or liver suggesting good response to therapy.

DISCUSSION

CML & PYOMYOSITIS

The occurrence of pyomyositis is most frequent in patients with immunosuppressive diseases. Pyomyositis is of two main types, tropical and non-tropical. Pyomyositis in immunocompromised patients mainly due to HIV infection, but also includes a myriad of causes inclusive of but not limited to hematological malignancies and other forms of neoplasia.

The organism frequently responsible for pyomyositis is Staphylococcus aureus which amounts to 70% of cases in HIV positive patients and upto 64% in non-HIV infected individuals. [1] Other organisms such as coagulase - negative Staphylococcus species, various streptococcal species, Gram-negative bacteria, as well as anaerobic bacteria, including Bacteroides fragilis, and fungi are also responsible for the disease. In reality, Pyomyositis can occur in all the skeletal muscles of the body, including muscles in the extremities and the trunk.[2] But the yield rates of cultures of pus or blood has continued to remain low.

Pyomyositis occurs most frequently in males. The male:female ratio ranges from 2:1 to 3:1.[3] The diagnosis of Pyomyositis is most often delayed due to the low level of awareness of the disease. Another explanation is that if the involved muscles are deeply placed, the symptoms can be masked and hence leading to a late diagnosis. Pyomyositis involving the retroperitoneal space – iliopsoas and obturator group of muscles can be very hard to treat.

Hip pain with clinically prominent signs of infection and a negative aspiration or arthrogram should raise the alarm of a possible pyomyositis and they should be advised to undergo MRI to exclude osteomyelitis and pyomyositis.[4-7] as MRI is the best imaging modality for the early diagnosis of pyomyositis. [3,4,5,6] It is essential to note that patients with hematological malignancies having elevated counts at baseline, have a higher probability of Pyomyositis being overlooked. The underlying hyperleukocytosis adds a significant threat to the lives of these immunocompromised patients. The hyperleukocytosis, the frequent usage of analgesics, such as acetaminophen and nonsteroidal anti-inflammatory drugs for the constitutional symptoms, can mask fever episodes and inflammatory signs normally induced by infection leading to a delay in the diagnosis and treatment delivery. This delay in initiation of antimicrobial therapy in immunocompromised patients can lead to severe systemic involvement with significant morbidity and mortality.[7,8] The choice of antibiotic therapy is to be guided by the culture and sensitivity testing of the pus aspirated under USG or CT guidance.

The differential diagnosis of hip tenderness in pediatric patients with hematological malignancies includes fracture of femur, slipped capital femoral epiphysis, avascular necrosis of head of femur, septic arthritis, osteomyelitis, leukemia, lymphoma, and metastatic invasion of solid malignancies. Anteroposterior and lateral radiographs of both hips exclude femoral fracture and slipped capital femoral epiphysis. MRI with MR angiography, demonstrate signal changes in bone and vascular structures revealing intraosseous and extraosseous changes of the joint and surrounding soft tissue in hematologic patients.

Pyomyositis is classified to three stages.[3-4]

Stage 1	Progressive muscle dull pain with low-grade fever
Stage 2	Muscle abscess formation with a local tender, swollen, erythematous soft tissue mass is occasionally associated with septicemia
Stage 3	Severe local infection and sepsis

It is likely that our patient had stage 2 disease which was cured by the combined modality therapy offered to him.

The most preferred management of pyomyositis is the combined medical and surgical approach. The antimicrobial therapy with antibiotics with coverage against Staphylococcus aureus is the initial therapy and later, it can be directed as per the sensitivity testing. This is due to the possibility of the alternative causative organism and due to the emergence of resistant strains of Staphylococcus aureus. Apart from appropriate antimicrobial therapy and drainage of the pus, the resolution of pyomyositis also depends on the effective neutrophil function recovery by the management of underlying hematological malignancy or HIV.

In our case, impairment of neutrophil function may have caused progression of pyomyositis even with antibiotic usage. Culture of the aspirated pus did not grow any organisms and that could be explained by the prior initiation of antibiotic therapy.

From the review of literature, only three cases of pyomyositis have been reported in CML till date and all of them were treated with both Incision & Drainage and antibiotics.

Table 4: Reports Of Cases With Pyomyositis & CML

First author	Year of publication	Patient's age and sex	Pathogen	Site	Medical treatment	Surgical treatment	Outcome
Asou et al.[10]	2003	40/M	MRSA (pus)	Femoris (R)	Imipenam-cilastatin, amikacin, minocycline	I&D	Successful
Yoshimoto et al.[11]	2016	68/M	Sterile	Iliopsoas (R)	Linezolid (oral)	I&D	Successful
Chen et al.[12]	2011	17/M	Sterile	B/L adductor muscles	Oxacillin	I&D	Successful

Consistent with the literature of pyomyositis, these patients were all males and two of them had sterile cultures, similar to our patient. Hence, our experience adds to the relevant literature and suggests that once a diagnosis of non-tropical pyomyositis is established one has to consider an underlying hematological malignancy and also the possibility of muscle pain being a pyomyositis in hematological malignancy.

CONCLUSION:

Pyomyositis should be considered in the differential diagnosis in those with malignancy who present with hip joint or groin muscle pain to diagnose the disease early and prevent morbidity and mortality caused by it.

Conflict Of Interest: The authors declare no conflict of interest.

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