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REVIEW

Pyomyositis associated with hematological malignancy: case report and review of the literature

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Summary Pyomyositis occurs most commonly in patients with various immunosuppressive diseases. However, the association of pyomyositis with an underlying hematological malignancy has not been reviewed. We present herein a relevant case and also review the available literature regarding the association of non-tropical pyomyositis and hematological malignancies. The case patient, a 46-year old female, had non-tropical pyomyositis of the iliopsoas and obturator muscles due to *Staphylococcus aureus* and underlying Hodgkin's disease. Forty-four patients with pyomyositis and an associated hematological malignant disease have been reported in the literature. The most common types of hematological oncology diseases found were acute lymphocytic leukemia (present in 11/44 patients (25%)) and multiple myeloma (7/44 patients (15.9%)). *Staphylococcus aureus* was the most common cause of pyomyositis (26 out of 44 patients (59.1%)). The muscles of the thigh were most commonly affected (18/44 patients (40.9%)). Medical therapy with antibiotics and surgical drainage were employed in 25/44 (56.8%) of the patients. Thirty out of 44 (68.2%) of the patients had a successful outcome. Death occurred in 5/44 (11.4%) patients. In cases of pyomyositis, the physician should consider an underlying hematological malignancy.

Introduction

Pyomyositis is classified into two main types, tropical and non-tropical. The disease may occur in immunocompromised patients mainly due to infection with the human immuno-

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deficiency virus (HIV), but also due to hematological malignancies and other forms of neoplasia. The usual microbial cause of pyomyositis is *Staphylococcus aureus*, which is responsible for 70% of the cases in HIV positive patients and in 64% of cases in non-HIV-infected individuals.[1] Other pathogens that may cause pyomyositis are various coagulase-negative *Staphylococcus* species, various streptococcal species, Gram-negative bacteria, as well as anaerobic bacteria, including *Bacteroides fragilis*, and fungi. The disease may affect practically all the skeletal muscles of the body, including muscles in the extremities and the trunk.[2] A particular form of the disease that is difficult to manage is the one that is located in the retroperitoneal space involving the muscles of the area, specifically the iliopsoas muscles and the obturator muscles.

Combined medical and surgical intervention is the preferred method of management. Antimicrobial treatment should include antibiotics with spectrum mainly against *S. aureus*. However, obtaining specimens from the affected muscles for microbiological cultures may considerably help the clinician in the selection of the appropriate antimicrobial treatment.[3] This is because, occasionally, pyomyositis is caused by bacteria other than *S. aureus* and also because this pathogen may be resistant to anti-staphylococcal penicillins (methicillin-resistant *Staphylococcus aureus* — MRSA).

Case report

A 46-year-old female patient complained of mild fever, weakness, left extremity swelling, and fullness of the left inguinal area that became progressively worse over a period of 1 month. She was seen by her primary care physician who did not find any abnormal findings in the physical examination except for lymph node enlargement in the inguinal area. He recommended biopsy of the enlarged lymph nodes, which was unrevealing. She developed lymphorrhea for about 2 weeks after the procedure.

Two months after the start of her initial symptoms, she presented at our hospital with higher fever (up to 38 °C) and more weakness. She was admitted to the hospital for further management. Physical examination showed left lower extremity swelling and lymph node enlargement in the left inguinal area (Figure 1). Routine laboratory investigation revealed an increased white blood cell count (WBC: 20.07 $\times$ 10^9/l, 81.9% neutrophils), C-reactive protein (CRP: 9.1 mg/dl (normal: 0—0.5)), and erythrocyte sedimentation rate (ESR: 87 mm 1st hour). She was found to have MRSA bacteremia. Imaging of her body with computed tomography (CT) scans revealed abscesses in the left iliopsoas and obturator muscle (pyomyositis; Figure 2) and lymphadenopathy of the inguinal, mediastinal, and preaortic areas. No other source of active infection was identified, except the findings from the retroperitoneal space. A magnetic resonance imaging (MRI) of the spine and a colonoscopy that included visualization of the terminal ileum were negative. She received intravenous antimicrobial treatment with linezolid (600 mg every 12 hours), clindamycin (600 mg every 8 hours), and rifampin (600 mg every morning and 300 mg every night) for 4 weeks that improved her condition. Specifically the fever decreased and she felt better. In addition, the laboratory indices of inflammation also improved.

A surgical consultation was obtained; the patient underwent laparoscopic surgery with incision of the peritoneum for access to the retroperitoneal space. Histological examination of the excised tissue was again unrevealing. The patient subsequently underwent an open surgery, during which more tissue was excised from the retroperitoneal space. No findings of active infection in the retroperitoneal space were found macroscopically during the operation. In addition, cultures of specimens of the excised tissue did not grow any microorganisms. Histological examination of the excised tissue revealed the diagnosis of Hodgkin's lymphoma (nodular sclerosis); Reed—Sternberg cells were visualized as well as Hodgkin monocytes surrounded by T-cell rosettes. Finally, the presence of Hodgkin lymphoma was verified by immunochemical testing (CD15+, CD30+). In addition the resected tissue contained many non-neoplastic inflammatory cells with the presence of many neutrophils, eosinophils, plasma cells, and histiocytes. Bone marrow examination was normal.

Literature review and discussion

In Table 1 we present the available evidence from publications regarding patients with pyomyositis associated with

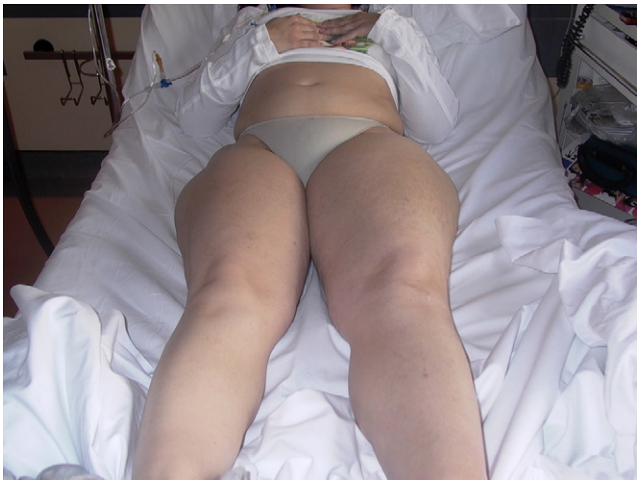

Figure 1 Edema and erythema of the left lower limb.

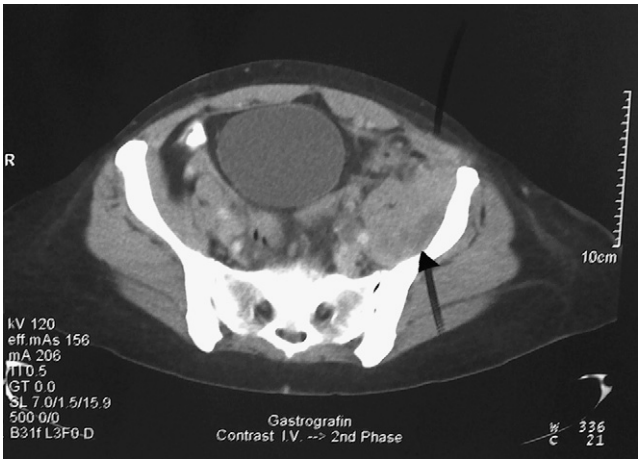

Figure 2 Abscesses in the left iliopsoas muscle on computed tomography imaging.

Table 1 Reports of patients with pyomyositis associated with hematological malignancies

First author	Year of publication [ref.]	Patient's age and sex	Hematological malignancy	Pathogen causing pyomyositis	Site of muscle involvement	Medical treatment	Surgical treatment	Outcome of infection
Kao	2006 [4]	10/F	ALL	*Staphylococcus aureus* (blood)	Thigh (R)	Vancomycin, ceftazidime	Drainage	Successful
Karmazyn	2006 [5]	NR	ALL	NR	NR	NR	NR	NR
Yu	2004 [6]	52/F	NHL	MRSA (pus)	Thigh (R)	Antibiotics (not specified)	Incision and drainage	NR
		12/F	ALL	*Staphylococcus aureus* (pus)	Thighs bilaterally	Antibiotics (not specified)	Incision and drainage	NR
		77/F	Myeloproliferative disease	*Aeromonas hydrophila* (pus)	Left lower leg	Antibiotics (not specified)	Incision and drainage	NR
		50/F	Myeloproliferative disease	Negative cultures	Thigh (R)	Antibiotics (not specified)	US guided aspiration	NR
		5/M	Diffuse large B-cell lymphoma	Negative cultures	Forearm (R)	Antibiotics (not specified)	Not performed	NR
		36/M	ALL	*Staphylococcus aureus* (pus)	Thigh (L)	Antibiotics (not specified)	US guided aspiration	NR
Chang	2005 [7]	15/F	AML (M2)	*Acremonium sp* (pus)	Calf area bilaterally	Amphotericin-B, G-CSF	Drainage	Successful
Hayashi	2003 [8]	40/M	CML	MRSA (pus)	Femoris (R)	Imipenem–cilastatin, amikacin, minocycline	Incision and drainage	Successful
		46/M	MDS (RAEB-t)	MRSA (pus)	Forearm (R)	Vancomycin, teicoplanin, amikacin	Drainage	Successful
		71/M	MDS (RAEB-t)	MRSA (sputum)	Vastus lateralis (L)	Vancomycin	Not performed due to thrombocytopenia	Death
Tsai	2003 [9]	35/M	MDS (RAEB-t)	*Stenotrophomonas maltophilia* (blood, fasciotomy wound tissue)	Soleus (L)	Ceftazidime changed to imipenem	Fasciotomy with debridement and external drainage	Successful
Torres	2001 [10]	21/M	AML	*Nocardia asteroides* (pus)	Thighs bilaterally	Trimethoprim–sulfamethoxazole	Not performed	Successful
Demir	2000 [11]	42/M	NHL	*Staphylococcus aureus* (pus)	Vastus lateralis (R), biceps femoris (L)	Ampicillin–sulbactam	Drainage and debridement bilaterally	Successful
Hossain	2000 [12]	48/M	AML	*Staphylococcus aureus*	Deltoid, quadriceps	Antibiotics (not specified)	Not performed	Successful
Matsuno	1998 [13]	68/M	MM	*Bacteroides fragilis* (pus)	Right arm and thigh	Imipenem–cilastatin	Incision and drainage	Successful
Cone	1997 [14]	22/M	Lymphoblastic lymphoma	*Staphylococcus aureus*	NR	Nafcillin	NR	Successful
		69/M	Plasma cell leukemia	*Staphylococcus aureus*	NR	Cefazolin	NR	Successful
		62/M	MDS	*Klebsiella pneumoniae*	NR	Ceftazidime	NR	Successful
		75/M	MM	*Aeromonas hydrophila*	NR	Cefazolin	NR	Successful

del Giglio	1997 [15]	27/F	ALL	*Mycobacterium tuberculosis* (biopsy)	Leg (L)	Isoniazid, rifampin, ethambutol, ofloxacin	Open surgical drainage	Successful
		74/F	AML	*Mycobacterium tuberculosis* (pus)	Gluteal and quadriceps muscles (L)	Triple antituberculous therapy	NR	Death
Corden	1996 [16]	9/M	ALL	*Staphylococcus aureus* (pus, blood)	Thighs bilaterally	Nafcillin, cephapirin, cefonicid	Incision and drainage	Successful
		10/M	ALL	*Staphylococcus aureus* (pus)	Thighs bilaterally, gluteal (L)	Cephapirin	Incision and drainage	Successful
Katagiri	1996 [17]	68/F	MM	*Bacteroides fragilis* (pus)	Femoral quadriceps (R), brachial biceps (R)	Imipenem—cilastatin, clindamycin	Incision and drainage	Successful
Gordon	1995 [18]	70/M	CLL	*Staphylococcus aureus* (blood)	Iliacus, gluteus maximus, biceps femoris (R)	Antibiotics (not specified)	Incision and drainage	Successful
		68/M	CLL	*Staphylococcus aureus* (pus)	Vastus medialis (L)	Antibiotics (not specified)	Incision and drainage	Successful
		63/M	CLL	*Staphylococcus aureus* (pus)	Gluteus maximus (L)	Antibiotics (not specified)	Drainage under CT guidance	Successful
Audran	1993 [19]	78/M	Myelomonocytic leukemia	*Salmonella sp* (blood)	Adductor	Antibiotics (not specified)	Not performed	Successful
Hoyle	1993 [20]	44/M	MM	*Staphylococcus aureus* (pus, blood, BAL)	Multiple sites in the limbs	Vancomycin, flucloxacillin	NR	Successful
Christin	1992 [21]	30/M	AMML	*Staphylococcus aureus* (pus, blood)	Multiple sites	Penicillinase-resistant synthetic penicillin, aminoglycoside	NR	Successful
		19/M	ALL	*Staphylococcus aureus* (pus)	Multiple	Penicillinase-resistant synthetic penicillin, aminoglycoside, cephalosporin 1st generation	Incision and drainage	Successful
		79/M	Hodgkin's disease	*Staphylococcus aureus* (pus)	Right paraspinal	NR	NR	NR
		NR	Hodgkin's disease	*Clostridium septicum* (pus)	Gluteus (L)	NR	NR	NR
		66/M	MDS	*Staphylococcus aureus* (pus)	Thigh (L)	Penicillinase-resistant synthetic penicillin, aminoglycoside	NR	Successful
Prallet	1992 [22]	45/F	MM	*Staphylococcus aureus* (pus, blood)	Infraspinatus, coracobrachialis, deltoid	Pefloxacin, fusidic acid	Incision and drainage	Successful
Korten	1992 [23]	64/M	MM	*Pseudomonas aeruginosa* (pus, subcutaneous nodule)	Left calf muscles	Ceftazidime, ciprofloxacin	Not performed	Successful
Bonafede	1992 [24]	66/M	Preleukemia	*Staphylococcus aureus* (pus)	Thigh	NR	NR	Successful
		70/M	MDS	*Staphylococcus aureus* (pus)	Psoas	NR	NR	Secondary *Escherichia coli* infection/death

Table 1 (*Continued*)

First author	Year of publication [ref.]	Patient's age and sex	Hematological malignancy	Pathogen causing pyomyositis	Site of muscle involvement	Medical treatment	Surgical treatment	Outcome of infection
Minor	1989 [25]	24/M	ALL	*Fusarium sp* (pus and umbilical sinus)	Calf (L)	Amphotericin B, rifampin, ketoconazole	Drainage	Death
Sarubbi	1989 [26]	67/M	MM and aplastic anemia	*Serratia marcescens* (pus, urine)	Anterior tibial compartments bilaterally	Imipenem, rifampin	Incision and drainage	Death due to massive gastrointestinal hemorrhage
Blatt	1979 [27]	19/M	ALL	*Staphylococcus aureus* (pus)	Forearm (R), calf (R)	Penicillinase-resistant synthetic penicillin, aminoglycoside, cephalosporin 1st generation	Incision and drainage	Successful
		12/M	ALL	*Staphylococcus aureus* (pus)	Thigh (L), calf (R)	Oxacillin	Incision and drainage	Successful

ALL, acute lymphocytic leukemia; NHL, non-Hodgkin's lymphoma; AML, acute myelogenous leukemia; CML, chronic myelogenous leukemia; MDS, myelodysplastic syndrome; RAEB-t, refractory anemia with excess blasts in transformation; MM, multiple myeloma; CLL, chronic lymphocytic leukemia; AMML, acute myelomonocytic leukemia; NR, not reported; MRSA, methicillin-resistant *Staphylococcus aureus*; US, ultrasound; CT, computed tomography; BAL, bronchoalveolar lavage. M, male; F, female; R, right; L, left.

hematological malignancies.[4–27] While HIV is a well-known predisposing factor for the development of pyomyositis, a number of other diseases have been reported in association with pyomyositis as well. These ailments include diabetes mellitus, trauma, rheumatologic diseases, liver cirrhosis, renal failure, respiratory diseases, organ transplantation, and conditions requiring immunosuppressive agents (including corticosteroids).[3]

Relatively little attention has been paid to the association of hematological diseases with pyomyositis. Both malignant and non-malignant hematological diseases have been reported in association with pyomyositis. Our patient proved to have Hodgkin's disease in association with pyomyositis. By reviewing the literature, we found 44 other reported patients with hematological neoplastic diseases associated with pyomyositis: 11 patients with acute lymphocytic leukemia, four with acute myelogenous leukemia, two patients with myelomonocytic leukemia (one of them with the acute form), two patients with Hodgkin's disease, four patients with non-Hodgkin lymphoma, three patients with chronic lymphocytic leukemia, one patient with chronic myelogenous leukemia, seven patients with multiple myeloma, one patient with plasma cell leukemia, seven patients with myelodysplastic syndromes (three of them with refractory anemia with excess blasts in transformation), and two patients with myeloproliferative disease.[4–27] Non-malignant hematological diseases associated with pyomyositis are sickle cell anemia and aplastic anemia.[28,29]

Diagnosis of pyomyositis is facilitated by MRI or CT scanning of the affected area and by aspiration of fluid for microbiological testing under ultrasound or CT guidance. Reviewing the 44 patients retrieved from the literature, *S. aureus* was found to be the most common cause of pyomyositis (grew in culture in 26 out of 44 patients (59.1%)) and was the causative pathogen of the pyomyositis in our patient as well. *Bacteroides fragilis*, *Mycobacterium tuberculosis*, and *Aeromonas hydrophila* were next in frequency and were present in two patients each. The following pathogens were the cause of the pyomyositis in one patient with a hematological malignancy each: *Serratia marcescens*, *Salmonella sp*, *Klebsiella pneumoniae*, *Pseudomonas aeruginosa*, *Stenotrophomonas maltophilia*, *Clostridium septicum*, *Nocardia asteroides*, *Acremonium sp*, and *Fusarium sp*. The pathogen was recovered from cultures of the pus (drained or aspirated) in 30/44 patients (in seven of these 30 patients the pathogen grew additionally in cultures taken from at least one other site). In 4/44 patients the microorganism grew in blood cultures; in one of them additionally, a fasciotomy wound tissue culture was positive. Two patients had negative blood and pus cultures and no data exist for six patients regarding the site of growth of the microbial pathogen, while one pathogen was found on histological examination. The muscles of the thigh were most commonly affected (18/44 patients (40.9%)). Unilateral muscle involvement was more common (33/44 patients (75%)) in pyomyositis associated with a hematological malignancy than bilateral involvement (in 6/44 patients (16%)).

Treatment of pyomyositis includes the appropriate antibiotics against the offending pathogen and surgical incision and drainage. Treatment of the underlying disease, if any, should also be promptly provided. The outcome of the infection is usually successful if treated promptly. In 25/44 patients surgical drainage was performed and in two of these

patients debridement was performed additionally. Drainage with radiological guidance was performed in 3/44 patients. Drainage was not performed in six patients (in one of them due to thrombocytopenia). No data exist about drainage for 13/44 patients. Our patient was treated with intravenous antibiotics for the pyomyositis with a successful outcome of the infection. The outcome of the infection was successful in 30/44 patients. Five out of the 44 patients with pyomyositis died, one of them due to secondary sepsis due to *Escherichia coli* and one patient due to massive gastrointestinal bleeding. No data exist on the outcome of 9/44 patients.

In conclusion, we have presented a case of a middle-aged woman with retroperitoneal pyomyositis and underlying Hodgkin's disease and a review of the literature regarding published cases of pyomyositis in patients with hematological malignancies. 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.

Conflict of interest: No conflict of interest to declare.

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