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 lymph node enlargement in the inguinal area. He recommended biopsy of the enlarged lymph nodes that was unrevealing. She developed lymphorrhea for about 2 weeks after the procedure.

She acutely complained of higher fevers and more weakness about 2 months after the start of her initial symptoms. 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 increased white blood cell count (WBC: 20070/mm3, 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). 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 rifampicin (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.

What was the cause of the leg edema?

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 lymph node enlargement in the inguinal area. He recommended biopsy of the enlarged lymph nodes that was unrevealing. She developed lymphorrhea for about 2 weeks after the procedure.

She acutely complained of higher fevers and more weakness about 2 months after the start of her initial symptoms. 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 increased white blood cell count (WBC: 20070/mm3, 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). 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 rifampicin (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.

What was the cause of the leg edema?

Diagnosis
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. No findings of active infection in the retroperitoneal space were found 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). Specifically the tissue specimens obtained during surgery from the retroperitoneal space showed infiltration by a malignant lymphoproliferative disease. In addition, 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+).

Teaching points
Pyomyositis is classified in 2 main types, tropical and non-tropical pyomyositis. The disease may occur in immunocompromised patients mainly due to infection with the human immunodeficiency virus (HIV) but also due to haematological malignancies and other forms of neoplasia. The usual microbial cause of pyomyositis is Staphylococcus aureus that is responsible for about 90% of cases. 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 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 Staphylococcus aureus. However, obtaining specimens from the affected muscles for microbiological cultures may considerably help the clinician in the selection of the appropriate antimicrobial treatment. This is because, occasionally, pyomyositis is caused by bacteria other than Staphylococcus aureus and also because this pathogen may be resistant to anti-staphylococcal penicillins (mexitilin resistant Staphylococcus aureus or else MRSA).

In Table 1 we present the available evidence from publications regarding patients with pyomyositis associated with haematological malignancies. 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 immunosuppressive agents (including corticosteroids).

Relatively little attention has been paid to the association of haematological diseases with pyomyositis. Both malignant and non-malignant haematological 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 haematological neoplastic disease associated with pyomyositis: 4 patients with acute lymphoblastic leukemia, 4 with acute myelogenous leukemia, 2 with myelomonocytic leukemia (one of them with the acute form), 2 patients with Hodgkin’s disease, 4 patients with Non-Hodgkin lymphoma, 3 patients with chronic lymphocytic leukemia, 1 patient with chronic myelogenous leukemia, 7 patients with multiple myeloma, 1 patient with plasma cell leukemia and 7 patients with myelodysplastic syndromes (3 of them with refractory anemia with excess blasts in transformation), and 2 patients with myeloproliferative disease. Non-malignant diseases associated with pyomyositis are sickle cell anemia and aplastic anemia.

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 computed tomography guidance. We present below a summary of the following characteristics of these 44 patients with pyomyositis and an underlying haematological malignancy: responsible pathogen, musculature involved, drainage of the abscesses and outcome of the infection.

Staphylococcus aureus was the commonest cause of pyomyositis [grew in culture in 25 patients out of 44 patients (56,8%)] and was the causative pathogen of the pyomyositis in our patient as well. Bacteroides fragilis and Mycobacterium tuberculosis 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 haematological malignancy, each: Serratia marcescens, Salmonella sp, Klebsiella pneumoniae, Pseudomonas aeruginosa, Stenotrophomonas maltophilia, Clostridium septicum, Bacteroides fragilis, Nocardia asteroides, Acromonas hydrophila, Acremonium sp, Fusarium sp. The pathogen was recovered from cultures of the pus (drained or aspirated) in 30/44 patients [in 7 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 only, while one pathogen was found on histological examination. 2 patients had negative blood and pus cultures and no data exist for 9 patients, regarding the site of growth of the microbial pathogen. The muscles of the thigh were most commonly affected [18/44 patients (40,9%)]. Unilateral muscle involvement is more common [in 37/44 patients (84%)] in pyomyositis associated with a haematological 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 be also promptly provided. The outcome of the infection is usually successful if treated promptly. In 28/44 patients surgical drainage was performed and in 2 of these patients debridement was performed additionally. Drainage with radiological guidance was performed in 2/44 patients. Drainage was not performed in 4 patients (in one of them due to thrombocytopenia). No data exist about drainage for 11/44 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 29/44 patients. 5/44 patients with pyomyositis died. One of them due to secondary sepsis due to E. coli and one patient due to massive gastrointestinal bleeding. No data exist about the outcome of 11/44 patients.

References

Acknowledgement
This case was prepared for our website by Drs. P. Rafailidis, A. Kapaskelis, and G. Peppas (it was also submitted for consideration for publication).