Severe thoracic pyomyositis in a patient with systemic lupus erythematosus

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SUMMARY
Pyomyositis may mimic deep vein thrombosis and be misdiagnosed in patients with systemic lupus erythematosus (SLE). We report here on patient with SLE with severe thoracic pyomyositis presented with right upper arm swelling and fever. The patient fully recovered after a serial surgical debridement and antibiotic therapy. Pyomyositis, as well as deep vein thrombosis, should be considered during the differential diagnosis of patients with SLE experiencing fever and unilateral limb oedema. CT and identification of causal pathogens are crucial in the diagnosis of pyomyositis. Early effective antibiotic treatment as well as surgical intervention can together bring about a better outcome.

BACKGROUND
Pyomyositis is a pyogenic infection of skeletal muscle with the typical presentation of a localised abscess, which is mostly observed in tropical countries. It commonly affects patients with existing conditions such as diabetes mellitus, haematological malignancy, chronic renal failure, asplenia, HIV infection, autoimmune diseases and those receiving chemotherapy or immunosuppressive drugs. Pyomyositis is mostly caused by Staphylococcus aureus up to 90% of the cases. S. aureus is in most cases a community-acquired methicillin-resistant strain. Pyomyositis usually affects a single muscle, although the involvement of multiple muscles is not unusual (12%-40% of cases). Bulky muscles, particularly of the thigh, are the most commonly infected site. Other muscles, including the forearm, sternocleidomastoid and intercostal muscles are occasionally involved.

Three clinical stages of pyomyositis are known. The first stage is an invasive stage, involving muscle pain with oedema, low-grade fever and general malaise. The second stage is a suppurrative phase, where patients experience severe muscle tenderness and swelling, a high spike in fever and other severe systemic symptoms. During this stage, abscess formation is detectable, and it is when most cases are clinically diagnosed. However, classical signs of an abscess may be absent due to the tense overlying muscles. If treatment is delayed, the infection disseminates and progresses to the final stage, which is also the most severe stage. During this last stage, patients often experience bacteraemia, septic shock, acute renal failure, multiple abscesses and even death. The diagnosis of pyomyositis depends on findings obtained from imaging, including ultrasonography, CT and MRI or pathological findings. However, it is difficult to diagnose during the early stage due to its non-specific features. Therefore, it is necessary for clinicians to be alert for patients showing risks. Systemic lupus erythematosus (SLE) is a chronic autoimmune disease, which renders patients susceptible to infections due to impaired immunological responses, which are related to the disease itself. As the disease progresses, when superfluous immune reactivation damages multiple organs in patients with SLE, defence mechanisms fighting against pathogens including bacteria, viruses and fungi become impaired. Initially, white cell function defects play a very important role. Additionally, cytokine abnormalities also influence an SLE patient’s susceptibility to infection, including decreased phagocytosis, reduced production of IL-8 and IL-12 by polymorphonuclear leucocytes and complement deficiency as well as defective chemotaxis, membrane recognition and attachment to microorganisms. Third, the presence of antimannose-binding lectin (MBL) autoantibodies in patients with SLE can influence MBL plasma levels and their functional activity. MBL activates the complement system, so the polymorphisms of the MBL-2 gene have been associated with infections. In addition to the disease itself, immunosuppressive agents used to treat SLE, such as steroids and cyclophosphamide are known to predispose patients with SLE to infection. The risk factors for infection are lupus activity state, hypocomplementaemia, use of high-dose steroids and immunosuppressive agents, fever, high-level erythrocyte sedimentation rate and of C reactive protein (CRP), an abnormality in white blood cell (>4×10^9/L or >10×10^9/L) and a low level of albumin.

Timely diagnosis of pyomyositis in patients with SLE is challenging not only because of its non-specific features but also due to its mimicky of SLE flare-up and inflammatory myositis. Chest wall involvement is an uncommon condition and has been identified in only one of 25 reported SLE cases with pyomyositis in the available literature. Here, we have reported on a case of SLE with a large extent of chest wall pyomyositis, where the patient fully recovered after surgical intervention and antibiotic treatment.

CASE PRESENTATION
A 64-year-old woman with SLE was admitted to our hospital due to progressive pain in the right lower neck, painful swelling at the right shoulder for 3 days as well as fever without chills for 1 day. She had received acupuncture over the right shoulder 2 days prior to the onset of fever. She denied any...
Case report

Figure 1  Axial views (A and B) and sagittal view (C) obtained from multidetector CT on hospital day 8. They demonstrate (A) a large area of an abscess formation over inner part of the right upper chest wall, particularly the pectoralis muscles, which also compressed to the right lateral part of superior vena cava (star); (B) abscess compressed to distal end of the right subclavian vein (star); (C) abscess extended to right lower neck and mediastinum (star).

Figure 2  (A) Intraoperative photography showed a partially necrotic pectoralis major muscle and presence of extensive necrotic tissues between the pectoralis minor and major muscles. (B) The surgical wound was sutured on hospital day 22, with a tube put in place for continuous drainage.

Table 1  Differential diagnosis of external compression to vein, deep vein thrombosis and lymphangitis in SLE patients with unilateral limb oedema

| Clinical manifestation | Swelling or oedema | Upper extremity/lower extremity* | Deep vein thrombosis†‡ | Lymphangitis§¶ |
|------------------------|--------------------|---------------------------------|------------------------|----------------|
|                        |                    | Lower extremity†                |                        | Lower extremity† |
| Pain                   | +                  | +/− $                           |                        | −              |
| Fever                  | +/- §              | −                               | −                      | −              |
| Laboratory             |                    |                                |                        |                |
| D-dimer                | –                  |                                |                        |                |
| CRP/ESR                | +                  |                                |                        |                |
| APS profile            | –                  |                                |                        |                |
| Therapy                | Removal of lesion  |                                |                        | Anticoagulation |
|                        |                    |                                |                        | Steroids       |

*Pyomyositis most often occur in the lower extremity, including the thigh, calf and gluteal muscles, although any muscle group can be involved, including the upper extremity muscles.
†Only 4%–10% of all deep vein thromboses were located in the upper extremities. Lower extremity deep vein thrombosis was diagnosed in the range of 20%–30% for patients with APS seen in large cohort studies.
‡Pain was felt in 86% and 19% of patients diagnosed with deep vein thrombosis.
§Twelve to 21 days after the initial onset of symptoms.
¶Thirty two per cent (21/66) of patients with positive lupus anticoagulant (LA) and/or positive anticardiolipin antibodies (aCLs) were diagnosed with deep vein thrombosis. Patients with LA+/aCL− showed a higher prevalence of DVT (53%) as compared with LA+/aCL+ (27%) and LA−/aCL+ (22%).

APS, antiphospholipid syndrome; CRP, C reactive protein; ESR, erythrocyte sedimentation rate; SLE, systemic lupus erythematosus.
On hospital day 5, her vital signs became stable, but she had an intermittent low-grade fever and progressive right upper limb edematous changes without erythema. However, there was no turning pale, pulseless, nor did she experience any numbness. Four sets of blood cultures were taken, all of which yielded no bacterial growth. Because her progressive right upper limb oedema was suspected to be due to deep vein thrombosis, a multidetector CT (MDCT) was performed on hospital day 8. Those results revealed an extensive abscess at the pectoralis muscle with adjacent fat stranding. The abscess extended upward to the right lower neck (levator scapulae muscle) and the mediastinum as

| Number | Reference/year | Age (years) | Symptoms at presentation | Lesion location | Therapy | Outcome |
|--------|----------------|-------------|--------------------------|----------------|---------|---------|
| 1      | Ushijima et al/198518 | 14          | Fever, Swelling          | Quadriceps, Gluteal | Antibiotics ID | Recovery |
| 2      | Shames and Fast/19898  | 59          | Fever, Pain              | Gluteal         | Antibiotics ID | Recovery |
| 3      | Shamiss et al/199011  | 19          | Fever, Swelling, Pain, Oedema | Posterior thighs, calves | Antibiotics ID | Recovery |
| 4      | Bonafede et al/199213  | 31          | Swelling, Pain           | Upper arm, thigh | Antibiotics ID | Recovery |
| 5      | Dede et al/199314   | 23          | Swelling                 | Calf            | Antibiotics   | Recovery |
| 6      | Yoshino et al/199415  | 44          | Pain                     | Gluteal         | Antibiotics ID | Recovery |
| 7      | Belzunegui et al/199515 | 27         | Fever, Pain, Palpable mass | Erector spinae | Antibiotics | Recovery |
| 8      | Gordon et al/199512 | 48          | Fever                    | Pectoralis major and minor, subclavus, intercostal muscles | Antibiotics | Death |
| 9      | Claudepierre et al/199616 | 32       | Fever, Pain, Swelling    | Quadriceps      | Antibiotics ID | Recovery |
| 10     | Ushida et al/200117 | 21          | Pain                     | Psoas           | Antibiotics ID | Death |
| 11     | Teh et al/200218    | 25          | NA                       | Thigh, calf     | NA            | Recovery |
| 12     | Garcia Hernández et al/200319 | 33       | Fever, Pain              | Iliopsoas       | Antibiotics ID | Recovery |
| 13     | Jidpugdeebodin and Punyagupta/200429 | 31         | NA                       | Shoulder, axilla, arm, forearm | NA | Recovery |
| 14     | Ravindran and Duke/200920 | 34        | Fever, Pain, Swelling    | Pronator teres | Antibiotics | Recovery |
| 15     | Collier et al/201021  | NA          | Fever, Pain, Swelling    | Sternoceildomastoid | Antibiotic | Recovery |
| 16     | El Baaj et al/201022  | 47          | Fever, Pain              | Quadriceps      | Antibiotics ID | Recovery |
| 17     | Manzoor/201023      | 23          | NA                       | Rectus femoris  | NA            | Recovery |
| 18     | Sokolove et al/201024  | 39         | Fever, Pain              | Quadriceps      | Antibiotics | Recovery |
| 19     | Souza et al/201125  | 25          | Fever, Pain              | Iliacus         | Antibiotics ID | Recovery |
| 20     | Blay et al/201426  | 16          | Fever, Pain              | Vastus intermedius | Antibiotics | Recovery |
| 21     | Chebbi et al/201427  | 52          | Fever, Pain              | Iliacus, Gluteal | Antibiotics | Recovery |
| 22     | Simopoulou et al/201428 | 46         | Fever, Pain, Swelling Ulcer with purulent discharge. | Vastus lateralis | Antibiotics ID | Recovery |
| 23     | Meesiri S/20167     | 14          | Fever, Pain              | Gastrocnemius   | Antibiotics ID | Recovery |
| 24     | Modi MA et al/201329  | 45          | Swelling                 | Quadriceps      | Antibiotics ID | Recovery |
| 25     | Chen YH et al/201830  | 48          | Pain, Swelling           | Flexor hallucis, flexor digitorum | Antibiotics ID | Recovery |

ID, incision and drainage; NA, not available.

Table 2 Summarised clinical features and therapies of 25 cases of systemic lupus erythematosus with pyomyositis7-31
well as down to the right deep lateral thoracic wall (intercostal muscles).

Notably, the abscess had compressed the superior vena cava and right subclavian vein, but no evidence of deep vein thrombosis was found (figure 1). A fasciotomy involving drainage and debridement was performed on the same hospital day as the MDCT, day 8. Extensive necrotic tissues were observed at the pectoralis minor and major muscles (figure 2). Negative pressure wound therapy (NPWT) was also applied. A pus culture yielded methicillin-resistant *S. aureus*. Vancomycin was prescribed for 28 days in order to control infection. Serial debridements were performed in combination with continuous NPWT for a span of 2 weeks. The patient’s symptoms improved, including right lower neck and right upper limb swelling, and were completely resolved 6 days after the first debridement. The surgical wound was sutured on hospital day 22, with tubing put in place for drainage. Beginning on hospital day 29, antibiotics were switched to oral linezolid at 600 mg every 12 hours. She was discharged on hospital day 33.

Her condition remained stable during subsequent outpatient department (OPD) visits, with the drainage tubing being later removed. A follow-up CT scan performed 3 weeks after discharge showed a complete resolution of both the chest wall abscess and mediastinal abscess. The antibiotic linezolid was finally discontinued. No recurrence of pyomyositis was noted during OPD follow-up visits over the next 3 months.

**DISCUSSION**

Pyomyositis is an infection primarily involving skeletal muscles. The evolution of myositis could be clinically classified into three stages. In the first stage, non-specific symptoms may be complained of, including focal muscle pain with a wooden consistency, erythema, oedema, low-grade fever and general malaise, while many differentials should be concerning, including thrombotic events, haematoma, muscle strain and osteomyelitis. Our patient experienced progressive warmth, swelling and oedema without erythema over her right lower neck and right upper limb and a mimicking of classic symptoms for deep vein thrombosis. Positive findings for phospholipid antibodies occur in up to 40% of patients with SLE. A significant proportion of patients with SLE is also diagnosed with primary Antiphospholipid syndrome. Pyomyositis should also be differentially diagnosed for patients with SLE. Another differential diagnosis given patients with SLE with unilateral limb oedema is lymphangitis. The comparison between external compression to vein (eg, abscess), deep vein thrombosis and lymphangitis is shown in table 1.

Regarding pyomyositis, up to 90% of patients are diagnosed at the second stage, also known as the purulent stage. However, during this stage, a lack of specific signs and symptoms as well as atypical manifestations may also be observed in patients with SLE. In SLE child patients, pyomyositis is extremely rare with a prevalence of only 0.35% (1/289). However, the prevalence of pyomyositis in adult patients with SLE remains unclear. The diagnosis of pyomyositis is sometimes delayed or even missed due to its unfamiliarity to physicians. Any or all adverse conditions could result in high rates of complications and morbidity. Therefore, obtaining a timely diagnosis of pyomyositis in patients with SLE can be fraught with challenges. For a clinician, pyomyositis should be taken as a differential diagnosis for patients with an immunosuppression and immunocompromised state, like SLE. Imaging assessments, including ultrasonography, CT scans and MRIs, are important in diagnosing pyomyositis. Ultrasonography is a simple, easily available, non-radiative imaging tool used for diagnosis, although it may miss spotting early myositis, deep muscle abscess or lesions, where its ultrasound beam fails to detect. Alternatively, a contrast-enhanced CT scan and MRI can provide a comprehensive assessment of muscle lesions. If clinical symptoms, including fever, local oedema, swelling and erythematous change, do not improve after empirical antibiotics treatment, repeating an image study, such as a CT scan, is necessary for both detection of disease progression and determining the time when surgical intervention becomes necessary. Regarding our present case, the first CT scan showed no abscess formation in the chest wall. However, a subsequent CT scan performed 7 days later revealed the extensive involvement of pyomyositis with abscess formations. An accurate assessment of the location and extent of muscle abscess is essential for both proper treatment and preoperative planning.

For pectoralis pyomyositis, particularly in stages 2 and 3, further management should include not only immediate and appropriate antibiotics therapy according to the culture of the purulent materials but also early drainage of any purulent discharge as well as surgical debridement. Our patient’s clinical condition improved rapidly after surgical intervention when compared with antibiotics treatment alone. A similar case report on a 48-year-old SLE diagnosed woman with chest wall pyomyositis revealed that she had received antibiotics treatment, CT-guided aspiration and drainage of her abscess. She died due to pneumonia and sepsis while experiencing with multiple organ failure. Other clinical features and therapies surrounding 25 cases of patients with SLE with pyomyositis are summarised in table 2. In our present case, the patient successfully recovered from severe chest wall pyomyositis after serial debridement and proper antibiotic treatments. This result indicates the importance of surgical intervention in patients with SLE with chest wall pyomyositis. NPWT is a recent wound treatment modality that provides negative suction pressure for the continuous drainage of an abscess and any wound discharge. The therapy could improve wound closure after a fasciotomy has been performed for treating acute compartment syndrome in patients experiencing necrotising fasciitis. The time interval from fasciotomy to wound closure averages 11.8 days for patients with compartment syndrome using NPWT. There is no need for using a skin graft or flap, with no additional morbidity being reported. Our patient experienced normal, successful wound healing using NPWT after fasciotomy for pyomyositis. The time interval from fasciotomy to wound closure was 14 days.

In conclusion, pyomyositis may mimic deep vein thrombosis and be misdiagnosed in patients with SLE with unilateral limb oedema.

**Learning points**

- **When a patient with systemic lupus erythematosus (SLE) shows unilateral limb oedema in the presence of fever and warm skin, infections such as an abscess or pyomyositis should be taken into consideration.**
- **Pyomyositis may mimic deep vein thrombosis and be misdiagnosed in patients with SLE experiencing unilateral limb oedema.**
- **An imaging assessment such as a CT scan, along with the identification of causal pathogens, is crucial for the diagnosis of pyomyositis.**
- **Wile regards to pyomyositis, early effective antibiotic treatment and surgical intervention can provide a satisfactory outcome.**
Imaging assessment such as a CT scan and identification of causal pathogens are crucial for diagnosis. A successful outcome depends on early and effective antibiotic treatment as well as surgical intervention.

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Case reports provide a valuable learning resource for the scientific community and can indicate areas of interest for future research. They should not be used in isolation to guide treatment choices or public health policy.

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