Salmonella Crepitant Pyomyositis in a Patient with Systemic Lupus Erythematosus

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ABSTRACT
A case of Salmonella serogroup B crepitant pyomyositis in systemic lupus erythematosus patient with prolonged treatment of corticosteroid and immunosuppressive drugs was reported. She presented with severe crepitant soft tissue swelling and pain of the left arm extending up to the shoulder and axilla within 15 days. She was successfully treated with a combination of antibiotics and surgical debridement. Focal soft tissue infection is rare in salmonellosis. Over 80 percent of Salmonella soft tissue infection occurs in adult patients with preexisting underlying illness mainly diabetes mellitus and human immunodeficiency virus infection. The mortality rate is high, and the relapses are common. Only 3 cases of gas-producing Salmonella soft tissue infection were previously reported in the literatures. (J Infect Dis Antimicrob Agents 2004;21:11-5.)

CASE REPORT
A 31-year-old Thai woman was admitted to Vichaiyut Hospital, Bangkok, because of painful swelling of the left arm for 15 days.

She had been diagnosed systemic lupus erythematosus (SLE) with renal involvement for 4 years, and had been treated with high dose of prednisolone and intravenous cyclophosphamide. Pulse therapy of 1 g of intravenous methylprednisolone for 3 days was started for active nephritis with mild improvement 1 month prior to this admission. She was then treated with oral prednisolone 45 mg daily, warfarin 1.5 mg daily and intravenous cyclophosphamide 400 mg monthly.

Fifteen days prior to admission, she noted swelling and pain at the left arm which extended up to her shoulder. She denied a history of trauma. She also had chronic watery diarrhea for several months. Three days prior to admission, intravenous dexamethasone 4 mg and cyclophosphamide 400 mg were given to the patient due to a diagnosis of active SLE and reflex sympathetic dystrophy of the left arm.

One day prior to admission, she was admitted to the hospital because the swelling extended further to the axilla.

Physical examination revealed a cushingoid appearance with body temperature of 36.9°C, pulse rate of 80/min and blood pressure of 100/60 mmHg. The left arm was tenderness and marked swelling which extended from the hand to the shoulder and axilla. Crepitus was also noted over the swelling arm and forearm. Other examination was normal.

Laboratory tests showed hematocrit of 31.3 percent, white blood cell counts (WBC) of 6,100 mm$^3$.

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(80% neutrophils, 4% band form, 9% lymphocytes, 7% monocytes), platelet counts of $150 \times 10^3/\text{mm}^3$, partial thromboplastin time of 56.5 sec (control: 31.9 sec), prothrombin time of 99.2 sec (control: 12.8 sec), blood sugar of 1,179 mg/dl, creatinine of 2.3 mg/dl, sodium of 124 mEq/L, bicarbonate of 27 mEq/L and albumin of 1.9 g/dl. Urinalysis showed red blood cells of 100 cells/high-power field (HPF) and WBC of 1-3 cells/HPF.

Chest and left arm radiography showed gas in the fascial planes and muscles of the left arm and axilla (Figure 1-3).

A diagnosis of active SLE with diabetic hyperosmolarity and gas-forming necrotizing fasciitis

Figure 1. Chest radiography showed gas infiltration in the fascial plains and muscles of left axilla.

Figure 2 and 3. Radiography of the left arm showed swelling and large amount of gas in the fascial plains and muscles of the left arm and forearm.
and pyomyositis was made. Blood glucose levels, blood coagulation, fluid and electrolyte imbalance were corrected. Needle aspiration at her left arm was done but failed to obtain any specimen. Meropenem 1 g every 12 hours plus metronidazole 500 mg every 8 hours were given intravenously. Surgical drainage of the left arm was done immediately when the patient’s general condition was stabilized. A long incision of the skin deep down to fascia and muscles of the left arm and shoulder revealed a lot of gas and greenish white color, odorless discharge. There was no gross myonecrosis. Gram’s staining of the discharge revealed gram-negative bacilli. Culture specimens of pus, blood and urine subsequently grew Salmonella serogroup B which was sensitive to trimethoprim/sulfamethoxazole, amikacin, ceftriaxone, levofloxacin, imipenem and piperacillin/tazobactam.

After the surgery, granulocyte colony-stimulating factor and intravenous immunoglobulin were given because she had severe sepsis and leukopenia (WBC of 800 cells/mm$^3$). She gradually improved three days after the surgery, and intravenous antibiotics were continued for 14 days. She was discharged 20 days after admission, and oral levofloxacin of 300 mg daily was prescribed for 10 days. Two weeks after the discharge, she was in a very good condition and the wound was completely healed without any complication.

**DISCUSSION**

Pyomyositis is an infectious process of the muscle, most frequently caused by Staphylococcus aureus.$^{1-3}$ Other less common causative bacteria include group A Streptococcus and mixed aerobic-anaerobic bacteria.$^4$ Aerobic bacteria in particular gram-negative bacilli and anaerobic bacteria other than clostridial species may produce gas in the deep tissue. Gas formation is not usually observed in infections caused by gram-positive cocci. The portal of entry of these bacteria is usually not known, although secondary bacteremia from the break in integrity of skin and mucous membrane barrier of the gastrointestinal or urogenital tract might be the source of this infection.$^{1-4}$

Our patient with active SLE had prolonged treatment with high-dose corticosteroid and cyclophosphamide. Fever is not recognized, probably due to the effects of corticosteroid. This results in a delayed diagnosis until the definite symptoms and signs of infection were noted. Host factors including cell-mediated immunity, macrophage and neutrophil functions play important role in defense against Salmonella infection.$^5$ SLE, diabetes mellitus and acquired defects in cell-mediated immunity from corticosteroid and immunosuppressive drugs predispose our patient to Salmonella infection. The previous history of chronic diarrhea may have induced transient Salmonella bacteremia and dissemination throughout the body.

Focal suppurative extraintestinal complications have been recognized in 7-10 percent of all infections due to non-typhoidal salmonellosis.$^{6,7}$ They occur in patients with local or generalized preexisting conditions including acquired immunodeficiency syndrome (AIDS) and other immunocompromised patients.$^{6-9}$ A study of focal non-typhoidal salmonellosis with a follow-up of more than 9 years showed 10 of 38 (26.3%) patients with AIDS had focal suppurative infection, compared to 19 of 490 (3.9%) patients without human immunodeficiency virus (HIV) infection.$^{10}$ There is a higher rate of bacteremia associated with gastroenteritis in AIDS patients than in immunocompetent individuals. The site of focal infection includes osteoarticular, urinary, pleuro-pulmonary, intraabdominal, central nervous, cardio-vascular and soft tissue infections.$^{6,9}$ Focal soft tissue infection is extremely rare, accounting for 6-12 percent of all focal Salmonella infections and mostly occurs in adults.$^{6,7}$ Collazos et al.$^{11}$ reported only 30 cases of Salmonella pyomyositis during the past four decades. Surprisingly, one-third of these were from Spain. The median duration of symptoms prior to the diagnosis was 14 days, compared to 15 days in our patient. Five of 29 (17%) patients presented without fever, similar to our patient. The most common site of muscle involvement was the psoas muscle, accounting for 17 of 30 (55%) patients.
There were other associated suppurative foci including aneurysm, bone and joint in 15 of 30 (50%) patients. Bacteremia was detected in two-thirds of these patients. A history of previous injuries was observed in 6 patients, and gastroenteritis was reported as a source of muscle infection in 11 patients. The frequency of association could be underreported due to a lack of data in most reports. Our patient had diarrhea, which could have been the primary portal of entry of *Salmonella*. *S. enteritidis* and *S. typhimurium* account for the most common species, in 15 of 30 (50%) and 6 of 30 (20%) of patients, respectively. *S. typhi*, *S. bonn*, *S. newport*, *S. cholerasuis* and *Salmonella* serogroup B were observed in one case each.

*Salmonella* has been rarely associated with gas-producing infection. Almost all *Salmonella* with the notable exception of *S. typhi* are able to ferment carbohydrate and produce gas. Only three cases of gas-forming soft tissue infection due to *Salmonella* were previously reported in the literatures. David et al\(^1\) reported the first case of gas-forming abscess of the thigh and buttock due to *S. typhimurium* in a patient with SLE treated with corticosteroid. Quale and Lonano\(^2\) reported the second case of *Salmonella* serogroup B crepitant myonecrosis in a patient with diabetes mellitus. And Jorring et al\(^3\) reported the third case of *S. enteritidis* gas-forming myonecrosis in the leg of diabetic patient. Our patient is the fourth reported case.

Early diagnosis and treatment especially with a combination of antibiotics and prompt adequate surgical drainage are essential to reduce the mortality in pyomyositis. Collazos et al\(^4\) have reported a high mortality rate (30%) associated with *Salmonella* muscle infection, compared to pyomyositis caused by other bacteria. Relapse is also common, probably due to the older age, underlying medical conditions and frequent bacteremia among these patients.

In conclusion, we reported a case of *Salmonella* serogroup B crepitant pyomyositis in SLE with a successful treatment by a combination of antibiotics and surgery.

References
