Development of bilateral gluteal pyomyositis during treatment of acute pyelonephritis in a patient with diabetes

Ji Hye Kim¹, Ho-Young Yhim², and Ji Hyun Park³

¹Department of Internal Medicine, Presbyterian Medical Center, Jeonju; Divisions of ²Hematology and Oncology and ³Endocrinology and Metabolism, Department of Internal Medicine, Chonbuk National University Medical School, Biomedical Research institute of Chonbuk National University Hospital, Jeonju, Korea

To the Editor,

Pyomyositis (PM) is a bacterial infection of the skeletal muscle and arises due to hematogenous spread rather than contiguous infection. PM can lead to abscess formation and sepsis. Staphylococcus aureus is the causative organism in more than 75% of cases. PM due to Escherichia coli has rarely been reported in immunocompromised patients [1]. We herein report a case involving a patient with longstanding diabetes who developed bilateral gluteal PM associated with transient E. coli bacteremia.

A 66-year-old female was admitted with a 3-day history of fever and pain in both flank areas. She had no history of antecedent trauma or foreign travel. The patient had a 15-year history of type 2 diabetes that had been treated with glimepiride, metformin, and an \(\alpha\)-glucosidase inhibitor. On examination, she was pyrexic (38.3°C) and tachycardic with a heart rate of 106 beats per minute and blood pressure of 120/70 mmHg. The patient had an increased white blood cell count (17,400/mm\(^3\)), an elevated erythrocyte sedimentation rate (ESR; 110 mm/hr), and poorly controlled diabetes mellitus (glycated hemoglobin of 9.4%). Urinalysis showed one to four red blood cells and 26 to 30 white blood cells per high-power field with nitrate positivity. Knocking tenderness in both flank areas was noted.

Renal ultrasonography showed normal renal parenchymal echogenicity and no perinephric fluid collection. Due to empirical suspicion of pyelonephritis, she was immediately started on intravenous ciprofloxacin after two blood and urine samples were obtained for culture. She was supported by intravenous fluids and subcutaneous insulin injections into the abdomen and arm. The blood and clean-catch urine cultures obtained on admission grew E. coli, which was susceptible to all antibiotics tested, except ampicillin. Because of the patient’s persistent flank pain and chills, diclofenac sodium was intramuscularly injected into her bilateral gluteal region on four separate occasions. Her flank pain and fever gradually improved with antibiotic therapy and disappeared 7 days after admission. Her urinalysis findings, complete blood count, ESR, and C-reactive protein (CRP) levels showed improvement.

The patient presented with a 3-day history of increasing nonspecific right buttock pain and low-grade fever 11 days after starting ciprofloxacin. Clinical examination revealed mild tenderness over the right buttock with normal overlying skin. Laboratory in-
vestigation identified leukocytosis of 18,000/mm³ with 84.4% neutrophils, 10.6% lymphocytes, 4.3% monocytes, 1.0% eosinophils, and 0.3% basophils. The serum CRP concentration was 47.3 mg/dL (reference range, < 5). Computed tomography revealed the presence of a focal area of low attenuation and air-fluid formation as well as peripheral rim enhancement after injection of contrast medium within the right gluteus muscle; a smaller but similar lesion was present in the muscle of the opposite side (Fig. 1A). Additionally, multifocal, wedge-shaped, low-attenuation lesions were present in the right kidney. However, no perirenal abscess or fluid collection was found adjacent to the right kidney (Fig. 1B), suggesting a possible diagnosis of PM and abscess formation. The patient was switched from ciprofloxacin to piperacillin/tazobactam because of the possibility of drug resistance due to antibiotic treatment. Surgical debridement revealed 80 mL of pus, and the culture grew E. coli (sensitive to ciprofloxacin, cephalosporins, and piperacillin/tazobactam but resistant to amoxicillin). On pathologic examination of the surgical debris, many inflammatory neutrophils had infiltrated the fibrotic soft tissue and destroyed the normal architecture. The patient was switched from piperacillin/tazobactam to imipenem because of drug-induced skin eruptions and leukopenia. Intravenous antibiotics and insulin therapy resulted in complete recovery after 6 weeks with no residual effects or sequelae.

Although the pathogenesis of PM is obscure in most cases, the infection is believed to be a complication of transient bacteremia [1,2] because it develops without an obvious penetrating injury or any other clear portal of entry in the vast majority of patients. However, bacteremia alone is not sufficient to cause PM. Striated muscle is relatively resistant to secondary infection following hematogenous infection. In experimental animal models, PM secondary to bacteremia occurs only in muscle traumatized by pinching, electric shock, or ischemia [2]. These findings are compatible with those of Smith and Vickers [3], who reported only two muscle abscesses among 327 patients who died of staphylococcal septicemia. They suggested that a concomitant muscle abnormality was required to produce susceptibility. In the present case, the patient presented with localized buttock pain and swelling in association with fever on day 11. Because the underlying muscle involvement was deep, there was a lack of skin erythema or warmth. Therefore, we mistook her buttock pain for postinjection muscular pain during the early stages of PM because this painful buttock area was identical to the injection site. The earlier episode of acute pyelonephritis may have been the source of the bacteremia because blood cultures positive for E. coli were documented. It has been hypothesized that minor muscle trauma by intramuscular injection of nonsteroidal anti-inflammatory drugs may be a predisposing factor for bacterial inoculation in the bilateral gluteal muscles during transient bacteremia.

Figure 1. (A) Contrast-enhanced computed tomography shows areas of low attenuation that represent myonecrosis or abscess formation in the bilateral gluteus maximus muscles (arrows). (B) Computed tomography shows multifocal, wedge-shaped, low-attenuation lesions in the right kidney (arrowhead). However, there are no perirenal abscesses or fluid collections adjacent to the right kidney.
Diabetes mellitus may also predispose patients to skeletal muscle damage. Diabetic patients develop inherent muscle defects that may increase the risk of muscle infection. Local vascular insufficiency and hypoxia secondary to diabetic microvascular disease may increase the risk of muscle infection and abscess formation. Numerous in vitro studies have documented abnormalities of cellular and humoral immunity in patients with diabetes, although the clinical relevance of these abnormalities is controversial and unproven. A review of the PM literature suggests that its incidence in diabetes increased from 8% of cases in 1971 to 1991 to 31% in recent reports [4].

While infectious PM can be caused by a variety of pathogens, including viral and parasitic ones, it is usually caused by gram-positive bacteria, especially S. aureus and, less frequently, Streptococcus pyogenes. PM caused by gram-negative bacteria is uncommon. PM due to E. coli is quite rare, and even fewer reports of its occurrence in patients with hematologic malignancies and diabetes mellitus are noted in the literature [1].

PM is a rare extraintestinal manifestation of deep-tissue E. coli infection reported in immunocompromised hosts. In the present case, metastatic spread to the gluteus muscle by E. coli bacteremia developed in the setting of uncontrolled diabetes and traumatized muscle despite treatment with ciprofloxacin, to which the E. coli isolate was sensitive. According to a review of previous cases, extraintestinal infections in elderly or immunocompromised patients can be caused by E. coli variants, the genotypes and phenotypes of which differ from those of archetypal extraintestinal pathogenic E. coli (ExPEC). We did not perform phylogenetic grouping or virulence factor gene analysis of the isolates. In the future, a better understanding of the biological basis of ExPEC infections may contribute to the development of a method of prevention and improvement of the high morbidity, mortality, and costs associated with extraintestinal E. coli infections [5].

In summary, PM due to E. coli, which is associated with transient gram-negative bacteremia by acute pyelonephritis in patients with type 2 diabetes mellitus, is an uncommon infectious disease that may cause complications if not diagnosed at an early stage and treated appropriately. PM should also be suspected in diabetic patients complaining of muscle pain and fever. For prevention, the serum glucose level should be strictly controlled, and muscle trauma, such as intramuscular injection, also should be avoided during the period of bacteremia.

**Keywords:** Diabetes mellitus; Pyomyositis; Escherichia coli

**Conflict of interest**

No potential conflict of interest relevant to this article was reported.

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