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CASE REPORT

Case of disseminated pyomyositis in poorly controlled type 2 diabetes mellitus with diabetic ketoacidosis

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ABSTRACT

Primary pyomyositis is a pyogenic and uncommon infection of skeletal muscle, which is mainly observed in tropical areas and/or human immunodeficiency virus patients. In non-human immunodeficiency virus infected patients, the most common cause is diabetes mellitus. Because of its rarity, the accurate diagnosis is often challenging. *Staphylococcus aureus* is the most common causative bacteria. According to the severity, pyomyositis is divided into three stages, and the late stage is occasionally lethal. The present case was compatible with the most advanced stage. Therefore, it was very difficult to save her life without precise and timely diagnosis. Furthermore, in the invasive stage, surgical drainage and broad-spectrum antibiotics should be given for a long enough period. Here, we report a case of a Japanese woman who developed disseminated abscesses under poorly controlled diabetic conditions accompanied by ketoacidosis, but was successfully treated without any sequelae.

INTRODUCTION

Pyomyositis is a primary pyogenic infection of skeletal muscle, an uncommon cause of musculoskeletal infection. Although relatively common in tropical areas, it is very rare in temperate areas[1]. In contrast, pyomyositis is observed in immunodeficient hosts, particularly in the human immunodeficiency virus (HIV) infected condition[2]. In non-HIV infected cases, the most common cause is diabetes mellitus[2]. In general, skeletal muscle tissue is resistant to bacterial infection, even in the severe bacterial septic condition[3]. In approximately 80% of cases, the location of the infection is unifocal, and the lower extremities, especially the thigh, are the most common site[2]. In the early stages of pyomyositis, there are no typical features of this disease. Thus, the accurate and timely diagnosis of pyomyositis is challenging. Because of its rarity, it is often misdiagnosed as muscle hematoma, cellulitis or neoplasm[4]. Therefore, prompt imaging procedures, aggressive surgical interventions and appropriate antibiotic therapy are very important to cure this disease without any complications. Here, we report a case of a Japanese woman who developed multiple abscesses, which were, surprisingly, disseminated over 30 parts throughout the body, under poorly controlled diabetic conditions accompanied by ketoacidosis, but was successfully treated with prompt and appropriate therapy.

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CASE REPORT

A 26-year-old woman was referred to Kawasaki Medical School Hospital, Kurashiki, Japan, because of a high fever and right thigh pain. She had mild pain of the right thigh, a sense of thirst and general fatigue. Furthermore, her bodyweight reduced by 6 kg during 2 weeks, and severe thigh pain, and high-grade fever developed and progressively worsened. Her medical history was unremarkable, except for adjustment disorder with no medication. She was a never smoker and did not consume alcohol. She had untreated type 2 diabetes, but did not have any history of trauma or illicit drug use. Her grandmother had adult-onset type 2 diabetes, but except for this there was no family history of diabetes including maturity-onset diabetes of the young and mitochondrial diabetes. On admission, her body mass index was 23.0 kg/m^2 (height 156 cm and body weight 56 kg). Her temperature was 38.7°C. Blood pressure was 135/90 mmHg and pulse rate was 116/min. She was drowsy, and

complained of polydipsia and severe thirst. There were several dehydration findings, such as diminished skin turgor and dry tongue. Emergent computed tomography (CT) showed a giant mass in the right adductor muscle (Figure 1a,b). The drastic elevation of plasma glucose (470 mg/dL) and glycated hemoglobin levels (15.2%) were shown with severe ketoacidosis. Despite prehospital administration of insulin (total 50 U) and bicarbonate, blood gas analysis still showed metabolic acidosis (pH 7.339, HCO_3^- 10.9 mEq/L, $PaCO_2$ 20.8 mmHg, PaO_2 107.5 mmHg, base excess –12.8 mEq/L, lactate 1.22 mEq/L under oxygen inhalation 2 L/min by nasal cannula). Anti-GAD antibody was negative. Leukocytosis and an increased C-reactive protein (28.21 mg/dL) were also observed. Renal and liver function, serum creatinine kinase, and myoglobin levels were not elevated. In addition, HIV serology test was negative.

Based on these findings, we diagnosed this patient as primary pyomyositis, and immediately started broad-spectrum antibiotics therapy (doripenem 3.0 g/day, clindamycin 1200 mg/day). Aggressive hydration with saline (4.5 L/day) and intravenous continuous insulin infusion was immediately started. Furthermore, we carried out surgical drainage and removed 700 mL of yellowish pus. The blood cultures and pus grew methicillin-sensitive *Staphylococcus aureus* and *Candida albicans*. Therefore, we added intravenous fluconazole. After a 1-week good clinical course, we stopped drainage and discontinued clindamycin treatment. Soon after, the patient had a high fever and systemic muscle pain. Surprisingly, the follow-up CT showed multiple pyomyositis disseminated over 30 parts throughout her body, as well as a kidney abscess (Figure 1c). We immediately restarted clindamycin, and changed fulconazole to micafungin with additional percutaneous renal drainage. We used broad-spectrum antibiotics and an antifungal drug intravenously for a total of 66 days. There was no clear evidence of osteomyelitis, which is closely associated with pyomyositis, but in consideration of the possibility of osteomyelitis, we decided to carry out long-term antibiotics and antifungal therapy. Indeed, despite a good clinical course, low-grade fever and persistent mild elevation of CRP (approximately 1.5 mg/dL) were observed. Therefore, we continued antibacterial and antifungal agents until the normalization of these infectious markers. In the findings of follow-up CT with the enhancement, the origin bulky abscess in the adductor

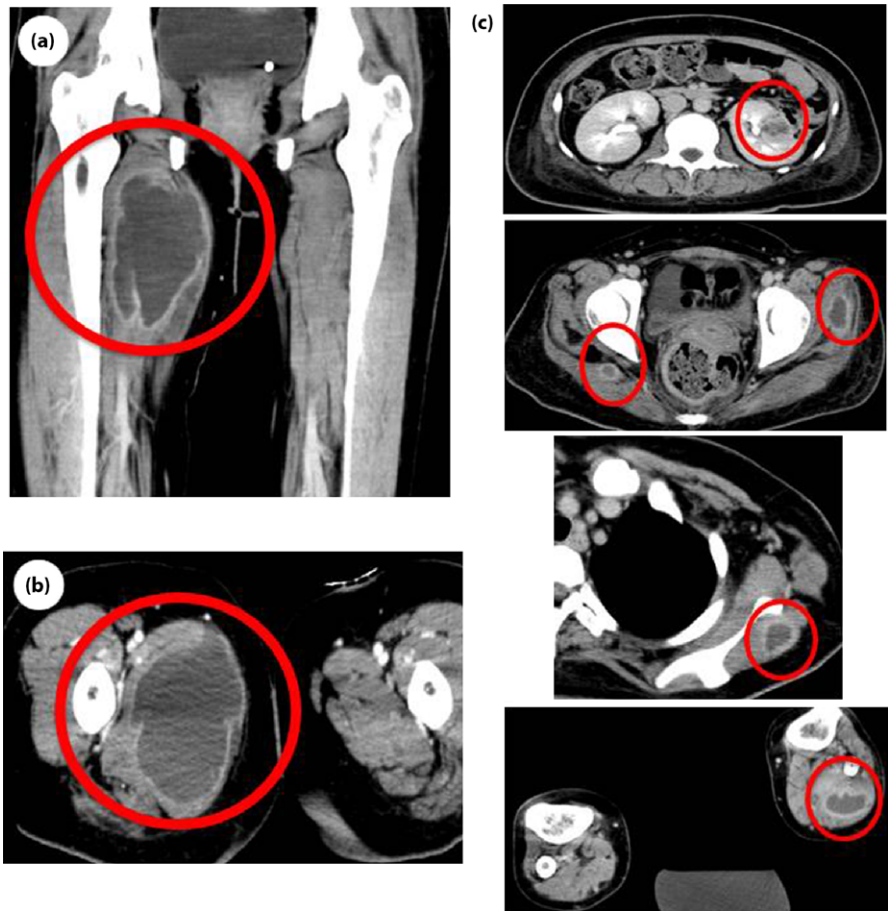

Figure 1 | (a, b) Computed tomography with contrast enhancement in the right thigh on admission. (a) Frontal slice. (b) Mid-femoral cross sectional slice). (c) Disseminated abscesses throughout the patient's body including the kidney and muscle (on day 7).

muscle and systemic intramuscular abscess completely disappeared (Figure 2). On day 66 of the hospitalization, when the value of C-reactive protein remained within the normal range for 7 days, we stopped all intravenous agents. The patients was discharged on day 96 without any sequelae.

DISCUSSION

It is known that skeletal muscle is resistant to bacterial infection, but under various immunodeficient conditions, such as severe lymphopenia with HIV infection or diabetic ketoacidosis, the host's defense system against microbiota is destroyed, which easily leads to opportunistic infection. Indeed, there is a close relationship between diabetes itself and the incidence of serious infections[5,6], especially sepsis[7]. In addition, because of its rarity, the accurate and timely diagnosis of pyomyositis is challenging. In the present patient, severe hyperglycemic state with diabetic ketoacidosis and severe infectious signs led to the diagnosis of pyomyositis. Furthermore, typical CT imaging of the bulky abscess in the right adductor muscle was helpful for the diagnosis. To the best of our knowledge, there have been no reports showing as many as 30 disseminated abscesses. Because there seemed to be no other additional immunodeficiency cause, we assume that the delay of the visit to the medical institution was the main cause of pyomyositis observed in this patient.

According to the severity, pyomyositis can be divided into three stages[1]. Stage 1 is the invasive stage where local symptoms and low-grade fever are observed, but other physical findings are typically absent. This stage usually lasts from 1 to 10 days. However, only a minority of patients (<2%) visits a medical institution at this stage[1]. Stage 2 is the suppurative stage with abscess formation. Around 2–3 weeks after the initial onset, patients complain about progressive fever and severe pain of the affected sites. Stage 3 is the late stage, where sepsis and dissemination of the infection are observed if the abscess remains untreated in the previous stages[1]. This progressive stage can be occasionally associated with lethal conditions, especially septic shock, multiple organ failure and rhabdomyolysis[6,8]. Our case was compatible with stage 3. Our patient was very afraid of undergoing all treatment, and hesitated to visit a medical institution. We believe it is likely that such a delay progressed the pyomyositis in this patient to the most severe and life-threatening condition. Therefore, it would be very difficult to save her life, if the only one piece of the following situation is missing: precise and timely diagnosis, intensive surgical approach, long-standing antibiotics therapy, and general supportive care. As observed in the present patient, *Staphylococcus aureus* is the most common (approximately 70%) causative bacteria in both tropical and temperate conditions[4].

Treatment of pyomyositis depends on the stage of the disease. In the invasive stage, antistaphylococcal antibiotics should be given for 2–4 weeks[1]. Surgical drainage is often necessarily in the suppurative phase. A key feature of prognosis of pyomyositis is recurrence, but it is uncommon (<30% of all cases). Indeed, there are only very few case reports of recurrent pyomyositis[9]. In general, a threshold of a 6-month interval is required to distinguish relapse from reinfection. The mortality rate is <1.5% in the early stage, but it becomes as high as 15% in the late stage[10]. Therefore, we have to continue antibiotics and antifungal agent for a long enough period. In fact, the present patient was successfully treated with surgical interventions, in combination with long-standing broad-spectrum antibiotics

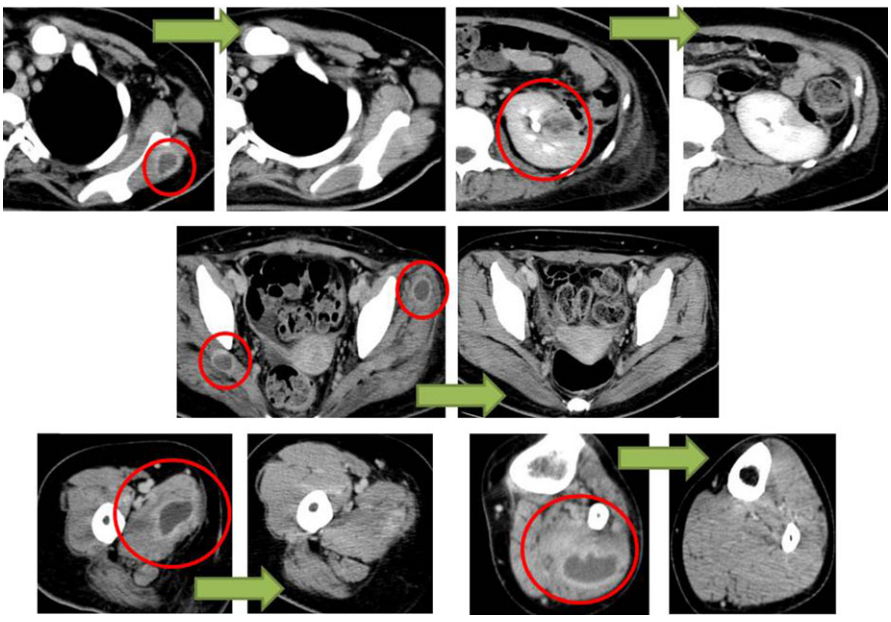

Figure 2 | Complete disappearance of origin and disseminated abscesses. Left panel, computed tomography on admission or day 7; right panel, computed tomography on day 96.

and antifungal therapy. Fortunately, during the long-standing broad-spectrum antibiotics and antifungal therapy, there were no adverse events including the appearance of resistant bacteria, pseudomembranous colitis, liver dysfunction and electrolytes abnormalities.

Pyomyositis is a rare infectious disease, but physicians should be aware of its possibility in poorly controlled diabetic patients. In addition, the present case strongly suggests that we should continue appropriate therapies until the complete disappearance of abscesses.

DISCLOSURE

The authors declare no conflict of interest.

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