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CASE REPORT

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Infectious myositis involving the piriformis in a patient with rheumatoid arthritis

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Abstract A 49-year-old Japanese woman treated with oral corticosteroids, methotrexate, and infliximab for malignant rheumatoid arthritis was admitted because of fever and low back pain. The white blood cell count and C-reactive protein concentration were elevated. Lumbar and pelvic computed tomography showed enlargement of the piriformis muscle including a hypodense area consistent with gas formation. The patient was treated successfully for infectious myositis with intravenous antibiotics.

Key words Immunosuppression · Infectious myositis · Infliximab · Piriformis muscle · Rheumatoid arthritis

Introduction

Infectious myositis is a subacute deep bacterial infection of skeletal muscle. Also called tropical myositis, primary pyomyositis, pyogenic myositis, and suppurative myositis, this infection previously was considered a tropical disease that was rare in temperate regions such as Japan, but nontropical cases appear to be increasing.[1] The quadriceps, gluteus, and iliopsoas muscles are most commonly affected; other muscles, especially the piriformis, rarely are involved. Here we describe an occurrence of infectious myositis of the piriformis during treatment for rheumatoid arthritis.

Case report

A 49-year-old Japanese woman diagnosed with rheumatoid arthritis in 1997 was treated with oral corticosteroids, methotrexate, and infliximab. Her illness fulfilled criteria for malignant rheumatoid arthritis (MRA: RA with vasculitis)[2] and represented stage III and class 3 in the Steinbrocker classification. Her medical history included many additional conditions such as diabetes mellitus, hypertension, lumbar spinal canal stenosis from L3–4 spondylolisthesis, digital flexor tendon rupture, bacterial cellulitis, ureteral lithiasis, pyelonephritis related to a temporary nephrostomy, and refractory skin ulcer. The patient also had undergone a right total knee arthroplasty. Treatment with infliximab injections was started in August 2003. After the 16th injection on October 18, 2005, she developed low back pain, which an orthopedic evaluation on October 25 attributed to spondylolisthesis. On the next day the pain was worse, and increasingly was exacerbated by walking. When she consulted the emergency department on October 28, the patient had intense pain, was unable to walk, and was febrile (37.4°C). She denied trauma. The white blood cell (WBC) count was 16900/mm^3, and the serum C-reactive protein (CRP) concentration was elevated at 12.1 mg/dl. She was admitted to the orthopedic department with a diagnosis of purulent discitis, and was treated with intravenous antibiotics (imipenem/cilastatin sodium). Despite this treatment, she had a high fever (39.2°C) on the next day, as well as a sharp increase in CRP elevation (42.2 mg/dl). Methicillin-susceptible *Staphylococcus aureus* (MSSA) was detected in a blood culture. She was transferred to the medical department for further treatment.

On examination, height was 156 cm and weight was 68 kg (body mass index, 27.9). Heart rate was 94 beats/min and blood pressure 110/60 mmHg. The abdomen was soft and distended. Bilateral pretibial pitting edema was assessed as 2+. A neurologic examination could not be performed because of severe pain. Initial laboratory evaluation upon transfer showed elevated values for liver function parameters (γ-glutamyl transpeptidase, 286 IU/l; alkaline phosphatase, 414 IU/l); WBC, 13300/mm^3 including 90.7% neutrophils; lactate dehydrogenase, 245 IU/l; erythrocyte sedimentation rate, 137 mm/h; and CRP, 42.2 mg/dl. Endocarditis was ruled out by transthoracic echocardiography. Lumbar and pelvic computed tomography (CT) after intravenous administration of a radiographic contrast agent demonstrated enlargement and relative enhancement of the

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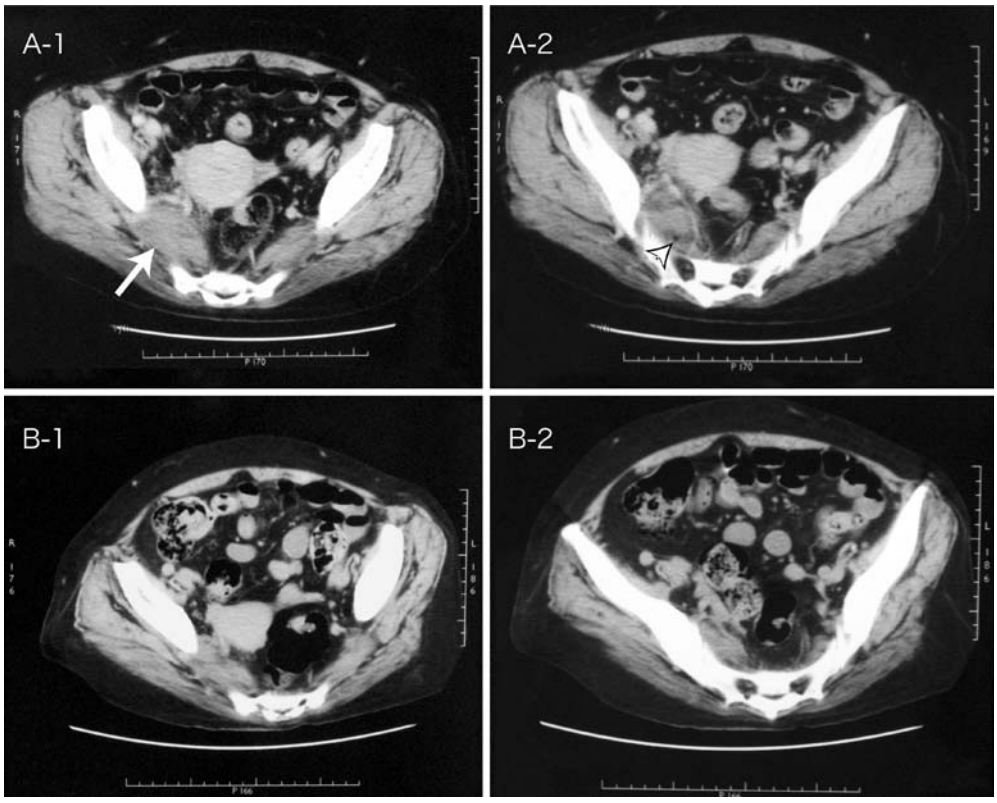

Fig. 1A,B. Abdominal computed tomography obtained with intravenous contrast administration. **A** Hospital day 5. The piriformis muscle is enlarged and shows relative enhancement (*arrow*), also containing a hypodense area (*arrowhead*) considered to represent gas formation. **B** Hospital day 33. Abnormalities indicating myositis have resolved

piriformis (Fig. 1A-1, arrow); a hypodense area in this muscle (Fig. 1A-2, arrowhead) was considered to represent gas formation by bacteria.

Needle drainage guided by CT or ultrasonography was considered but deferred, since the abscess was small and located near the sciatic nerve, with risk of nerve injury. The location also was unfavorable for operative drainage. Intravenous antibiotic therapy was changed (piperacillin, clindamycin, sulbactam/ampicillin, and panipenem/betamipron).

The clinical course is shown as a time line in Fig. 2. On hospital day 20 *Candida albicans* was detected in blood cultures, and fosfluconazole was added to the regime. Gradual clinical improvement followed. According to CT on day 33, myositis appeared to have resolved (Fig. 1B). By day 39, CRP had decreased to 0.4 mg/dl. As myositis had been treated successfully, the patient was transferred to the orthopedic department for management of spondylolisthesis on day 53.

Discussion

Infectious myositis, a subacute, deep bacterial infection of skeletal muscle, has also been termed tropical myositis, primary pyomyositis, pyogenic myositis, and suppurative myositis; the first of these alternative names reflects its previous status as a tropical disease. To an increasing extent, this form of myositis no longer is rare in temperate regions including Japan. The most common site of infection is the quadriceps (26.3%), followed by the gluteus and iliopsoas muscles.[1] In our patient the site of infection was the piriformis muscle, a very rare location; we could find only a few reports in which the piriformis was affected. In these cases, one patient had a perineal laceration, another had undergone dilation and curettage for a missed abortion, and the rest had no known source of infection. No reported case involved rheumatoid arthritis or immunodeficiency (Table 1).[3–7] While the sciatic nerve usually passes inferior to the piriformis, the sciatic nerve divides with part of the nerve passing through this muscle in about 12% of persons.[8] Accordingly, the piriformis usually is avoided in surgical procedures;[9] for that reason our patient's abscess could not be drained.

The most common causative organism is *Staphylococcus aureus* (70%–90%),[1,10] which appeared to be at least partly responsible in this case. *Streptococcus* species represent the second most common cause. Other causative organisms include *Neisseria gonorrhoeae*, *Salmonella enteritidis*, *Prevotella melaninogenica*, *Yersinia enterocolitica*, *Klebsiella oxytoca*, *Klebsiella pneumoniae*, *Aeromonas hydrophila*, *C. albicans* (also cultured from this patient), *Clostridium septicum*, *Escherichia coli*, *Haemophilus influenzae*, *Citrobacter freundii*, *Serratia marcescens*, *Fusarium*, *Proteus*, *Mycobacterium tuberculosis*, and aerobic diphtheroids.[1,10] Cases involving multiple pathogens, possibly including the present one, have been reported.

Although infectious myositis is considered to be caused by transient bacteremia rather than local extension of a contiguous infection, bacteremia only rarely causes muscle infection since striated muscle is relatively resistant to bacterial infection. However, an underlying muscle abnormality may predispose to intramuscular abscess formation.

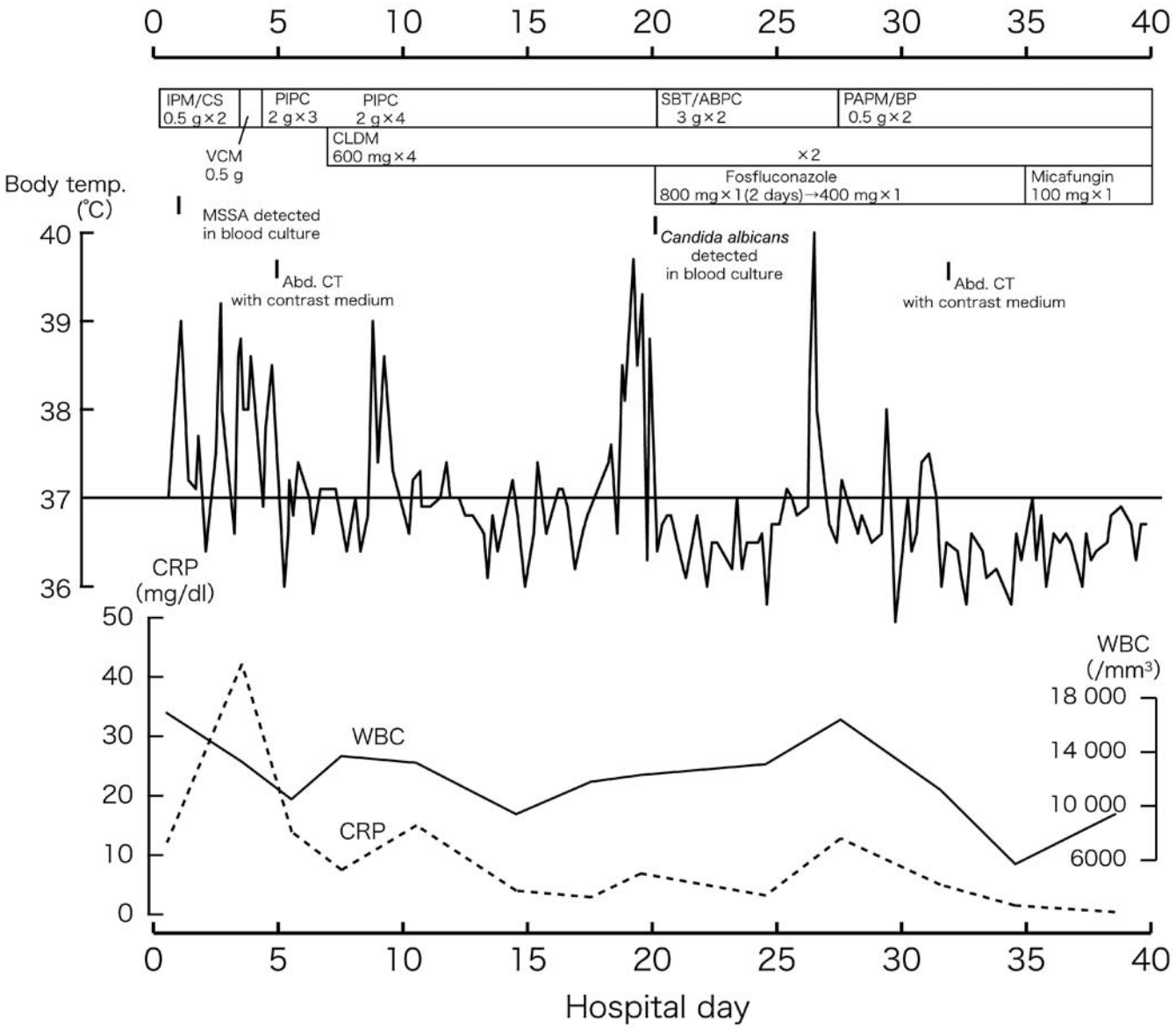

Fig. 2. Clinical course. *IPM/CS*, imipenem/cilastatin sodium; *VCM*, vancomycin; *PIPC*, piperacillin; *SBT/ABPC*, sulbactam/ampicillin; *PAPM/BP*, panipenem/betamipron; *CLDM*, clindamycin; *MSSA*, methicillin-susceptible *Staphylococcus aureus*; *CRP*, C-reactive protein; *WBC*, white blood cells; *Abd. CT*, abdominal computed tomography

These factors may include a preceding viral or parasitic infection, diabetes mellitus, malnutrition, thiamine deficiency, scurvy, beriberi, intense exercise, or local trauma.[10] Other risk factors include hematopoietic disorders (lymphoma, leukemia, neutropenia, aplastic anemia), compromised immune function (primary immunodeficiency or secondary immunosuppression from chemotherapy given in treating cancer or in organ transplantation), intravenous drug use, and connective tissue diseases. Various infrequent causes also have been suspected, such as intramuscular hemorrhage, alcoholism, Felty's syndrome, rheumatoid arthritis, atopic dermatitis, endocarditis, septic arthritis, renal failure, decubitus ulcers, and osteomyelitis.[1,10]

Our patient had multiple risk factors including malignant rheumatoid arthritis treated with corticosteroid (prednisolone 14 mg/day), methotrexate (6 mg/week), and infliximab (3 mg/kg), as well as diabetes mellitus. We know of no reported cases of infectious myositis involving infliximab administration for RA, but administration of corticosteroid or methotrexate has been described as an important predisposing factor inducing immune compromise. Infliximab had been administered in accord with guidelines for RA therapy,[11] beginning at a time when WBC was 10500/mm^3; lymphocytes, 1155/mm^3; beta-D-glucan, negative; and Disease Activity Score 28 (DAS 28), 7.67. Seven months later DAS 28 had decreased to 6.19 despite reduction in prednisolone from 20 mg/day to 15 mg/day. The patient also had a history of bacterial cellulitis, ureteral lithiasis, pyelonephritis, and refractory skin ulcer. Although glycemic control was fairly good (hemoglobin A1c, 6.1%; urine sugar, negative), malnutrition and alcohol excess were suspected. She consumed an estimated 1.5 l of "shochu" daily (alcohol con-

Table 1. Reported cases of infectious myositis involving piriformis

First author, year, ref.	Age/sex	Pathogen	Treatment	Outcome	Underlying disease	Other factors
Chen 1992[3]	42/M	*Staphylococcus aureus*	Surgery, cephalosporin	Cured	(–)	None known
Kinahan 1995[4]	22/F	negative blood culture	Imipenem	Cured	Perineal laceration, gestational thrombocytopenia	Epidural catheter insertion
Chusid 1998[5]	17/M	*Proteus mirabilis*	Cefotaxime, tobramycin	Cured	(–)	Athlete (swimmer)
Burkhart 2003[6]	69/M	MSSA	CT-guided aspiration (failed), cefazolin	Cured	(–)	Tennis player
Chong 2004[7]	30/F	MSSA	Vancomycin (patient allergic to penicillin)	Cured	(–)	History of dilation and curettage for a missed abortion
Present case	49/F	MSSA, *Candida albicans*	Piperacillin, ampicillin, panipenem, clindamycin, fosfluconazole, micafungin	Cured	MRA treated with corticosteroid, methotrexate, and infliximab; DM	History of spondylolisthesis, tendon rupture, bacterial cellulitis, ureteral lithiasis, pyelonephritis, and skin ulcer

MSSA, methicillin-susceptible *Staphylococcus aureus*; MRA, malignant rheumatoid arthritis; DM, diabetes mellitus.

tent, 300 g). Nutrition also was poor; total serum protein and cholinesterase were decreased (5.4 g/dl and 105 IU/l, respectively). Finally, despite lack of previous reports of infectious myositis linked to infliximab therapy for RA, serious and even fatal infections such as abscess and sepsis have been linked to infliximab. The patient had received 16 infliximab injections over 26 months. Overall, considering her recurrent infections (bacterial cellulitis, pyelonephritis, and skin infection), the patient was immunocompromised and susceptible to deep bacterial infections such as infectious myositis.

Most patients with infectious myositis can be treated successfully with intravenous antibiotics for 7 to 10 days. On the other hand, our highly susceptible patient required a longer period of intravenous antibiotic therapy for resolution. When intravenous therapy is concluded, an oral antibiotic, usually a first-generation cephalosporin, usually is given for at least 6 weeks. When we treat patients with rheumatoid arthritis under conditions of immune suppression, vigilance for unusual infections is warranted, as in this case.

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