Lymphology 58 (2025) 29-33

PYOMYOSITIS OF THE PIRIFORMIS MUSCLE SECONDARY TO CELLULITIS COMPLICATING LYMPHEDEMA

M. Tsujita, Y. Suzuki, T. Kato, K. Kishi

Department of Plastic and Reconstructive Surgery, Keio University School of Medicine, Tokyo, Japan (MT, YS, TK, KK).

ABSTRACT

A woman in her 70s with a 20-year history of postoperative edema and repeated cellulitis after surgery and chemotherapy for ovarian cancer presented with pain, redness, and swelling in her left lower leg. She was admitted with dehydration, disorientation, and elevated inflammation. After antibiotic treatment, redness of the lower extremities gradually improved. However, the patient complained of severe back pain after 10 days of treatment. Computed tomography (CT) revealed an abscess of the right pisiform muscle and patient was diagnosed with pyomyositis of the piriformis muscle. On hospital day 18, CT-guided percutaneous drainage of the abscess was performed. At the 6-month follow-up, there was no recurrence of the abscess. It is difficult to diagnose pyomyositis of the piriformis muscle via physical examination. This extremely rare case involves pyomyositis of the piriformis muscle as a result of cellulitis due to lymphedema. If antibiotic treatment for cellulitis in a patient with lymphedema is unsuccessful and severe pain persists, early imaging diagnosis should be considered.

Keywords: pyomyositis; lymphedema; piriformis muscle

INTRODUCTION

Pyomyositis of the piriformis is a rare disease secondary to bacteremia. Patients with lymphedema often develop cellulitis which in mild cases is relieved with oral antibiotics. However, septic shock can occur in some cases, requiring intensive care (1).

We present a case in which antibiotic therapy improved cellulitis of the lower extremities, but pain persisted leading to diagnosis of pyomyositis of the piriformis muscle secondary to cellulitis. This case is an extremely rare case of lymphedema-associated cellulitis followed by an abscess.

CASE PRESENTATION

The patient was a woman in her 70s with International Society of Lymphology (ISL) stage III secondary lymphedema of the lower left extremity. Twenty years prior, she had undergone abdominal total hysterectomy, bilateral salpingo-oophorectomy, pelvic and paraaortic lymph node dissection, and postoperative chemotherapy for ovarian cancer. She had experienced edema since operation, which worsened over the past two years, and she had repeated cellulitis in her left lower extremity. She had a medical history of systemic lupus erythematosus (SLE) and Sjögren syndrome and has undergone long-term administration of prednisolone (5 mg). As the next step in treatment, lymph node transfer was considered owing to recurrent cellulitis.

At the time of the initial consultation, the patient had stiff edema of her entire left lower

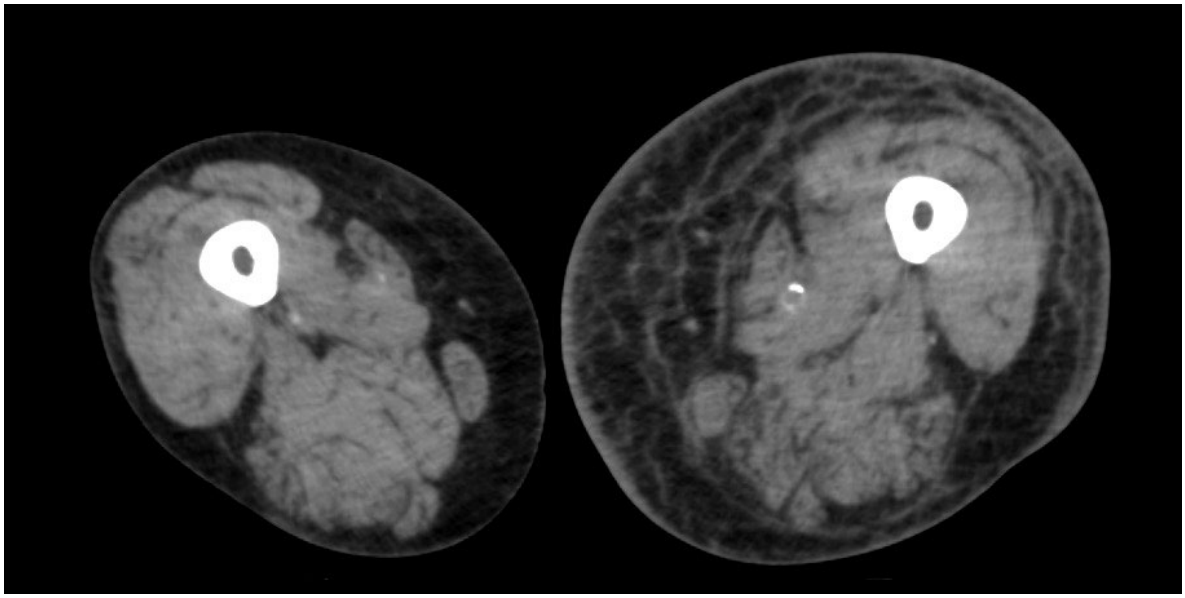

Fig. 1. *Computed tomography (CT) of lower extremity. Extensive subcutaneous tissue swelling and high dense adipose tissue were noted which is indicative of cellulitis.*

extremity and was diagnosed with ISL stage III lymphedema. Lymphoscintigraphy showed dermal backflow localized to the lower leg, which was classified as Maegawa classification type 4 (2).

As a first step in lymphedema treatment, lymphaticovenular anastomosis (LVA) was performed. LVA can improve the patient's condition with a small incision, even in severe lymphedema cases, if lymphatic vessels with good function can be identified (3). Therefore, although the likelihood of improvement with LVA in this patient was low, we decided to proceed with LVA first as a less invasive approach. In this case, the functional lymph vessels were degenerated, and the procedure was not effective. During outpatient follow-up, the patient presented to our clinic with fever, swelling of the lower extremities, and cellulitis. She was diagnosed with recurrent cellulitis and was admitted to our outpatient clinic. Physical examination revealed disorientation with temperature of 36.7°C and blood pressure of 88/55 mmHg. She presented with erythema, swelling, and pain in the entire left lower extremity with lymphatic leakage in the lower leg.

Investigations

Blood samples showed a markedly elevated inflammatory response with white blood cell counts 11.5x10^3 /μL, C-reactive protein 46.58 mg/dL, and decreased renal function with blood urea nitrogen 72.9 mg/dL and creatinine 2.20 mg/dL. Computed tomography (CT) demonstrated extensive subcutaneous tissue swelling and a high density of adipose tissue in the left lower extremity, indicative of cellulitis (*Fig. 1*). As there were no obvious abscesses or gas, the patient was urgently admitted to the hospital with a diagnosis of prerenal renal failure due to cellulitis, sepsis, and dehydration with swelling in the left lower extremity.

TREATMENT

The patient was treated with 3.0 g × 3/day of sulbactam ampicillin. Gradually, erythema of the lower extremities improved, and cephazolin-sensitive *Staphylococcus schleiferi* was detected in blood and wound cultures. On the 5th day of hospitalization, the patient was switched to 2 g × 2/day of cephazolin. How-

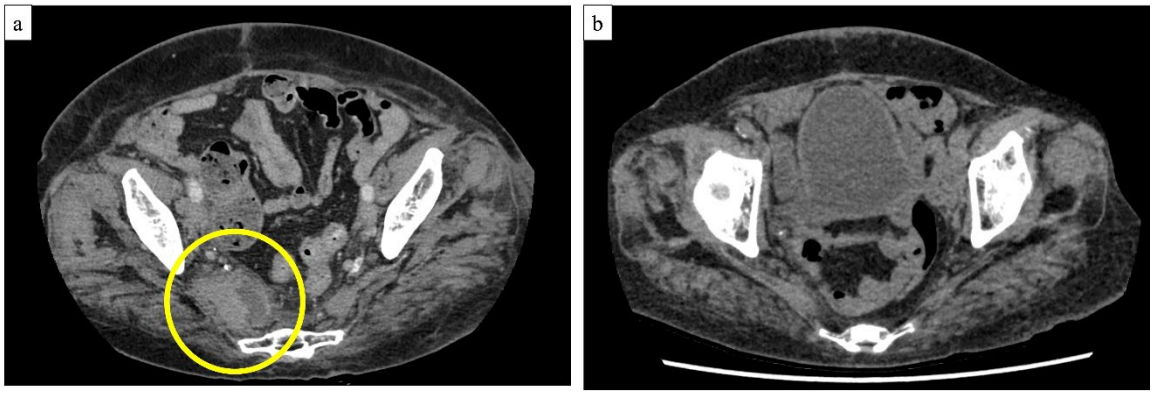

Fig. 2. *Computed tomography (CT) of pelvic area. (a) The abscess is confirmed within the right pisiform muscle (circle) diagnosing pyomyositis of the piriformis muscle. (b) After discharge, the abscess observed during hospitalization disappeared.*

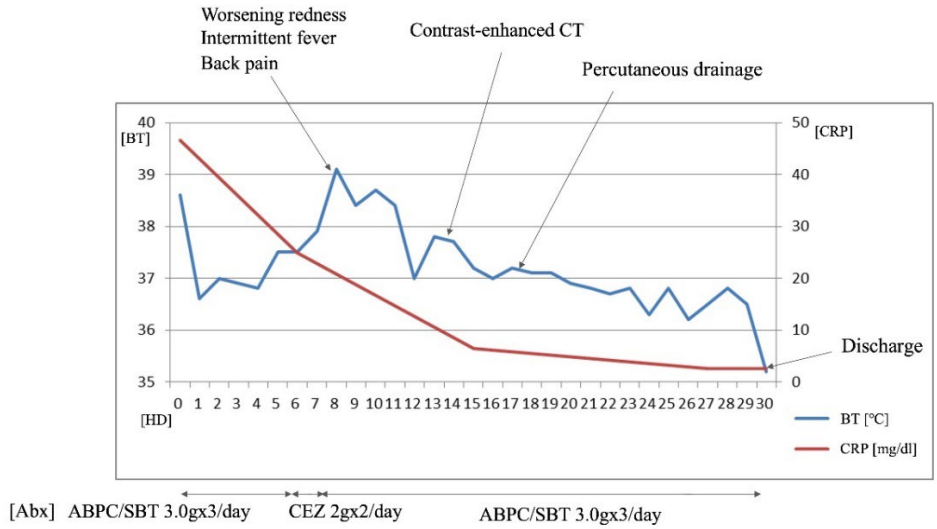

Fig. 3. *Timeline depicting post-hospitalization events. Abx: Antibiotics; ABPC/SBT: Sulbactam ampicillin; CEZ: Cephazolin; BT: Body temperature; CRP: C-reactive protein; HD: Hospital day*

ever, on the 6th day of hospitalization, the patient developed worsening redness of the left lower extremity, intermittent fever of approximately 39°C, and complaints of back pain; therefore, antibiotics were switched back to sulbactam ampicillin (3.0 g × 3/day).

On the 10th day of admission, the patient's left lower extremity erythema had improved; however, intermittent fever and back pain persisted, severely limiting her mobility. Contrast-enhanced CT was performed to evaluate the abscess formation, including pyogenic spondylitis. A low-absorption area was found within the right pisiform muscle, and pyomyositis of the piriformis muscle was diagnosed (*Fig. 2a*). On day 18 of hospitalization, CT-guided percutaneous drainage of the abscess was performed, which resulted in marked improvement in pain, and patient was discharged from the hospital. (*Fig.3*)

Outcome and follow-up

After discharge, the inflammation improved and abscess observed during hospitalization disappeared (*Fig. 2b*). Vascularized lymph node transfer was performed after discharge. One year later, cellulitis and abscess did not recur.

DISCUSSION

Pyomyositis of the piriformis muscle is one of the rarest forms of pyomyositis and is a subacute infection of the muscle. Most patients (72%) present with fever, 40% with difficulty walking, and 32% with buttock pain as subjective symptoms; however, all symptoms are nonspecific and often difficult to diagnose (4). Sepsis can occur secondary to pyomyositis, especially in the case of a piriformis abscess, which is caused by the excessive use of pelvic muscles (5-7), gynecological surgery (8), poor posture (9), and Crohn's disease (10). However, to the best of our knowledge, there have been no reports of pyomyositis of the piriformis muscle secondary to cellulitis in patients with lymphedema. The risk of cellulitis is 70 times higher in patients with lower extremity lymphedema than in those with healthy lower extremities (11). In addition to lymphedema, cellulitis occurs owing to skin injuries and tinea pedis (11-13). The most common bacteria associated with cellulitis are β-hemolytic streptococci and *Staphylococcus aureus* (14). Cellulitis often resolves with antibiotics, cooling, and rest. However, in rare cases, it can become severe owing to septic shock. The CREST classification is a known severity scale for cellulitis, and this case was judged to be Stage IV because it was accompanied by sepsis (15).

Based on these findings, the present case is extremely rare. However, muscle abscesses occur in patients on long-term steroid therapy in immunocompromised conditions such as human immunodeficiency virus infection, malignancy, or rheumatic diseases (16, 17). The organisms responsible for abscesses were staphylococci (61%), gram-negative rods (16%), streptococci (12%), and 2% fungi (18). In this case, *S. schleiferi,* which is rare in humans except in the immunocompromised was detected. This patient was on steroids for SLE and Sjögrens syndrome. Not only did the lymphedema contribute, but prolonged steroid therapy may have also weakened the patient's immune system. This weakness may have been a factor in the development of this uncommon muscle abscess.

These conditions may have contributed to the rarity of this disease. An abscess that was not observed on CT upon admission appeared during the course of the patient's illness, suggesting that the patient developed cellulitis owing to lymphedema, which led to sepsis and pyomyositis of the piriformis muscle. In this case, percutaneous drainage allowed for early symptomatic improvement.

Therefore, this possibility should be considered if back pain persists after lymphedema cellulitis. Imaging studies such as CT and magnetic resonance imaging (MRI) should be performed as soon as possible, and drainage should be performed if an abscess is present. The piriformis muscle stretches between the thigh bone and the spinal column and can trap the sciatic nerve. As a result, sciatic pain caused by piriformis syndrome can sometimes be mistaken for other conditions. Additionally, there is a possibility of pyomyositis in cases of piriformis syndrome. Therefore, imaging studies for back pain are valuable. MRI is useful for early detection and should be considered (19). However, at our hospital, CT scans were more readily ordered; thus, we opted for a CT scan instead of an MRI.

CONCLUSION

Here, we report a rare case of pyomyositis of the piriformis muscle secondary to cellulitis in a patient with lymphedema. It is difficult to distinguish pyomyositis of the piriformis muscle from other types of abscesses through physical examination alone. Early imaging should be considered when antibiotic therapy for cellulitis in patients with lymphedema is unsuccessful and new symptoms develop.

DISCLOSURE OF CONFLICT OF INTEREST

All authors declare no competing financial interests exist.

REFERENCES

1. Cutfield, T, H Walter, M Hobbs, et al: Association of the Dundee severity classification with mortality, length of stay and readmission in adult inpatients with cellulitis. J. Antimicrob. Chemoth. 74 (2019), 200-206. https://doi.org/10.1093/jac/dky400
2. Maegawa, J, T Mikami, Y Yamamoto, et al: Types of lymphoscintigraphy and indications for lymphaticovenous anastomosis. Microsurg. 30 (2010), 437-442. https://doi.org/10.1002/micr.20772
3. Yang, JC, SC Wu, WC Lin, et al: Supermicrosurgical lymphaticovenous anastomosis as alternative treatment option for moderate-to-severe lower limb lymphedema. J. Am. Coll. Surgeons 230 (2020), 216-227.
4. Salehi, M, F Ghiasvand, MM Feizabadi, et al: The piriformis abscess: a case-based review. Iran J. Microbiol. 13 (2021), 252-256. ttps://doi.org/10.18502/ijm.v13i2.5988
5. Chusid, MJ, WC Hill, JA Bevan, et al: Proteus pyomyositis of the piriformis muscle in a swimmer. Clin. Infect. Dis. 26 (1998), 194-195. https://doi.org/10.1086/517062
6. Giebaly, DE, S Horriat, A Sinha, et al: Pyomyositis of the piriformis muscle presenting with sciatica in a teenage rugby player. BMJ Case Rep. 2012 (2012), bcr1220115392. https://doi.org/10.1136/bcr.12.2011.5392
7. Okubo, T, S Yabe, T Otsuka, et al: Multifocal pelvic abscesses and osteomyelitis from community-acquired methicillin-resistant *Staphylococcus aureus* in a 17-year-old basketball player. Diagn. Micr. Infec. Dis. 60 (2008), 313-318. https://doi.org/10.1016/j.diagmicrobio.2007.10.008
8. Chong, KW, BK Tay: Piriformis pyomyositis: A rare cause of sciatica. Singap. Med. J. 45 (2004), 229-231.
9. Elhagar, A, I Kamar, MFH Elsheikh, et al: Unusual case of lower back pain-piriformis myositis: A case report and literature review. Pan. Afr. Med. J. 32 (2019), 4. https://doi.org/10.11604/pamj.2019.32.4.17808
10. Jwa, HJ, HJ Song, H Jun, et al: Gluteal and presacral abscess due to Crohn's disease with multiple fistulas. Korean J. Gastroenterol 80 (2022), 267-272. https://doi.org/10.4166/kjg.2022.088
11. Dupuy, A, H Benchikhi, JC Roujeau, et al: Risk factors for erysipelas of the leg (cellulitis): Case-control study. BMJ 318 (1999), 1591-1594. https://doi.org/10.1136/bmj.318.7198.1591
12. Semel, JD, H Goldin: Association of athlete's foot with cellulitis of the lower extremities: Diagnostic value of bacterial cultures of ipsilateral interdigital space samples. Clin. Infect. Dis. 23 (1996), 1162-1164. https://doi.org/10.1093/clinids/23.5.1162
13. Björnsdóttir, S, M Gottfredsson, AS Thórisdóttir, et al: Risk factors for acute cellulitis of the lower limb: A prospective case-control study. Clin. Infect. Dis. 41 (2005), 1416-1422. https://doi.org/10.1086/497127
14. Jeng, A, M Beheshti, J Li, et al: The role of beta-hemolytic streptococci in causing diffuse, nonculturable cellulitis: A prospective investigation. Medicine 89 (2010), 217-226. https://doi.org/10.1097/MD.0b013e3181e8d635
15. Clinical Resources Efficiency Support Team (CREST). Guidelines on the management of cellulitis in adults. Belfast: CREST 2005.
16. Ngor, C, L Hall, JA Dean et al: Factors associated with pyomyositis: A systematic review and meta-analysis. Trop. Med. Int. Health 26 (2021), 1210-1219. https://doi.org/10.1111/tmi.13669
17. Crum, NF: Bacterial pyomyositis in the United States. Am. J. Med. 117 (2004), 420-428. https://doi.org/10.1016/j.amjmed.2004.03.031
18. Gomez-Reino, JJ, JJ Aznar, JL Pablos, et al: Nontropical pyomyositis in adults. Semin. Arthritis Rheu. 23 (1994), 396-405. https://doi.org/10.1016/0049-0172(94)90090-6
19. Shanmuga, JS, SS Rajkumar, VS Kumar, et al: Pyomyositis of the piriformis muscle – A case of piriformis syndrome. Indian J. Radiol. Imaging. 31 (2021), 1023-1026.

Yushi Suzuki
Department of Plastic and Reconstructive Surgery
Keio University School of Medicine
35 Shinanomachi, Shinjuku-ku, Tokyo, Japan
Phone number: +81-3-5363-3814
Email: fi080150@keio.jp