

Disseminated *Lomentospora prolificans* infection presenting with arterial exsanguination

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ABSTRACT

Lomentospora prolificans is an opportunistic fungal pathogen found especially in Australia, Spain, Portugal, California, and the southern United States. Although it causes a wide spectrum of infections, disseminated *L. prolificans* infection remains very rare. The diagnosis and medical management are challenging. No clear guidelines are available for management. The treatment options are limited and mortality is high, especially for immunocompromised patients. In the present case report, we have highlighted the rare vascular effect of disseminated *L. prolificans* infection. We have reported the case of a 48-year-old immunocompromised man who had presented with a right groin mass. After extensive workup, the patient was found to have disseminated *L. prolificans* infection causing a superficial femoral artery pseudoaneurysm. A multidisciplinary team was involved to provide medical and surgical care. However, the patient died after treatment failure and withdrawal of support. (J Vasc Surg Cases Innov Tech 2021;7:785-9.)

Keywords: Mycosis; Infections; Aneurysm; Infected; Immunocompromised host; Scedosporium; Septicemia

Lomentospora prolificans (formerly *Scedosporium prolificans*) is an opportunistic fungal pathogen found especially in Australia, Spain, Portugal, California, and the southern United States.¹ *L. prolificans* can cause severe infections in immunocompromised and immunologically normal persons.² It has been found in soil, sewage, and air and is currently an emerging human pathogen.² It can manifest as a sinopulmonary infection, an extrapulmonary localized infection, or disseminated infection.¹ The clinical outcome depends on the patient's condition and immunologic status. The timing of the diagnosis and initiation of antifungal treatment are important factors determining the outcome. Multiple cases of disseminated fungal infection have been reported. However, no clear guidelines are available to help clinicians determine the appropriate treatment. Mortality has been high owing to the aggressiveness of the pathogen and its resistance to antifungal treatment. Vascular complications have rarely been reported. In the present report, we have described the effects of disseminated *L. prolificans* infection and its vascular complication.

The patient's family provided written informed consent for the report of the patient's case details and imaging studies.

CASE REPORT

A 48-year-old man had presented with swelling and pain over his right groin, with a secondary complaint of a left medial distal shin mass. During the previous several months, he had developed a small bulge of the right inner thigh that had remained stable until it had increased in size during the previous 3 weeks. His medical history was significant for end-stage renal disease after three renal transplants, hypertension, hyperlipidemia, paroxysmal atrial fibrillation, and pneumocystis pneumonia. He had been receiving immunosuppression therapy with tacrolimus and prednisone. Other than the pulsatile right groin mass, the findings for the remainder of the abdominal examination were unremarkable. Computed tomography of abdomen and pelvis without contrast enhancement revealed a right thigh lobular proximal soft tissue lesion measuring 4.9 × 4.4 cm with fluid attenuation (Figs 1 and 2). Ultrasound revealed a right groin hematoma with a proximal right superficial femoral artery (SFA) pseudoaneurysm measuring 4.2 × 2.9 cm (Fig 3). The patient was admitted to the intensive care unit and broad-spectrum intravenous antibiotics were started. The vascular surgery service performed incision and drainage on the right proximal thigh abscess, the pseudoaneurysm was excised, and a right proximal SFA to distal SFA bypass was created using a reversed ipsilateral great saphenous vein. The vein was tested, and all bleeding points were sutured. It was then reversed and sutured to the proximal SFA in an end-to-side fashion. It was tunneled through the groin and down to the distal SFA and then sutured to the distal SFA in an end-to-side fashion. The infection was in the mid-SFA. No obturator bypass was required because the common femoral artery was not involved by the infection. The common femoral artery was far enough from the infected field that the bypass was anastomosed proximally there and tunneled

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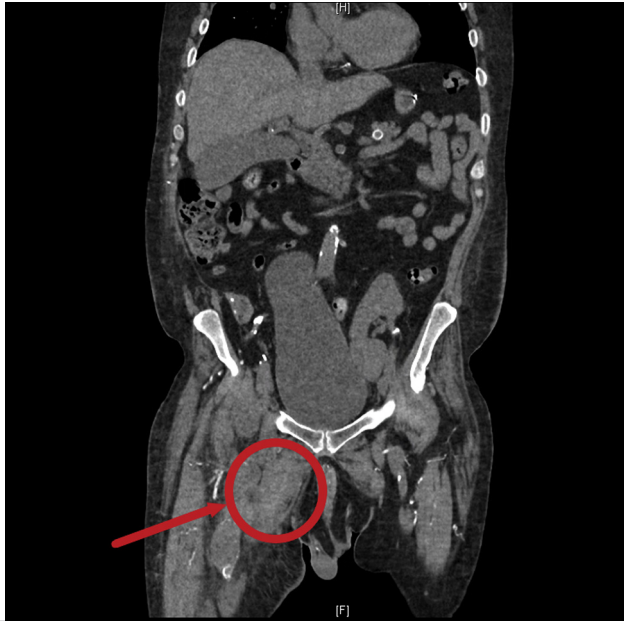


Fig 1. Computed tomography scan of the abdomen and pelvis without contrast enhancement. Right thigh soft tissue lesion measuring 4.9×4.4 cm. (red arrow).

away from the infected area to be anastomosed distally in another clean field in the very distal SFA.

On day 3 of admission, the patient had developed an expanding right proximal thigh hematoma. No surgical bleeding vessel was identified; however, a generalized oozing from the abscess cavity was present, and an abscess lateral to the femoral vein was found, which was drained. The bypass anastomoses were intact. The abscess cultures were initially identified as mold. The infectious disease service initiated posaconazole and amphotericin B and continued the broad-spectrum intravenous antibiotics. The tacrolimus dose was decreased. An echocardiogram revealed no valvular vegetations or evidence of endocarditis. The antifungal therapy was switched to voriconazole and terbinafine after the cultures had revealed *L. prolificans*.

On day 5, the patient experienced pulsatile bleeding at right distal thigh incision. A small bleeding arterial branch was ligated at the bedside. However, the patient became hypotensive. The right thigh and groin were explored intraoperatively. The proximal anterior thigh surgical incision revealed an old hematoma and generalized oozing from pseudoaneurysm cavity. Hemostasis was achieved with electrocautery and thrombin Gelfoam (Pfizer, New York, NY). The distal incision revealed another hematoma with fresh blood and a hole in the distal end of the vein bypass, which was repaired. On day 7, incision and drainage was performed on the left medial distal shin mass, and the cultures grew *L. prolificans*. Magnetic resonance imaging of the brain revealed multiple areas with brain abscesses. The neurosurgery team performed right frontal craniotomy with ventricular drain placement for the right frontal brain abscess, which was positive for *L. prolificans*. On day 12, the patient was found to have a pulsatile bleeding and pulseless electrical activity. Treatment was initiated for hemorrhagic shock, and

cardiopulmonary resuscitation was performed. The common femoral artery was clamped at the bedside, and the right thigh hematoma was evacuated in the operating room. The proximal bypass anastomosis had partially dehisced and was ligated, and a sartorius muscle flap was used to close the proximal anastomosis site. An audible Doppler signal to the leg at the level of the ankle was present.

On day 18, he developed acute leg ischemia. Above-the-knee amputation was offered to the family with the knowledge that it was unlikely to alter the clinical course, and they elected to proceed with the amputation. During the surgery, purulent drainage occurred in the thigh muscles, and the cultures grew *L. prolificans*. The patient remained in critical condition for several days. On day 23, his family elected to change his code status, and the patient died. The clinical timeline is shown in Fig 4.

DISCUSSION

L. prolificans causes local infection in immunocompetent persons and disseminated infection in immunocompromised patients.³ The most common infections are soft tissue and bone infections.² The local infections tend to be secondary to direct trauma or pulmonary infections.¹ Although the prevalence of infection is rare, the mortality rate has been high (87.5%).⁴ The risk factors include lung disease, human immunodeficiency virus, and malignancy.⁵ Most (70%) infections occur in patients immunocompromised because of hematologic malignancy or solid organ transplantation.⁶ The entry point is through the respiratory tract, central venous catheters, or sites of trauma and ulcers.²

The main symptoms of disseminated infections are fever, central nervous system (CNS) involvement, skin lesions, and muscular pain.⁷ *L. prolificans* has particular neurotropism, and CNS disease is considered a poor prognosticator.⁷ Lung infections were seen mainly in those with cystic fibrosis and those who had undergone solid organ transplantation. Other infections include osteomyelitis, arthritis, cutaneous wound infections, ocular infections, external otitis, and endocarditis.⁷

The main treatment of infection is antifungal therapy.⁷ One study reported that amphotericin B, posaconazole, isavuconazole, and echinocandins are not effective and that voriconazole has an intermediate effect (Fig 5).⁸ Bhat et al⁹ reported success with voriconazole and terbinafine for a patient with CNS disease. Jenks et al¹⁰ reported an association between survival and the use of combination therapy (amphotericin B and posaconazole). They also reported that terbinafine-based regimens were significantly associated with treatment success and survival.¹⁰ The treatment choice should be determined by the type of infection, whether the infection is disseminated, and the underlying cause of the infection source.

Vascular complications of disseminated fungal infections are rare. Kelly et al⁵ described a pedunculated mass in the ascending aorta in an immunocompromised

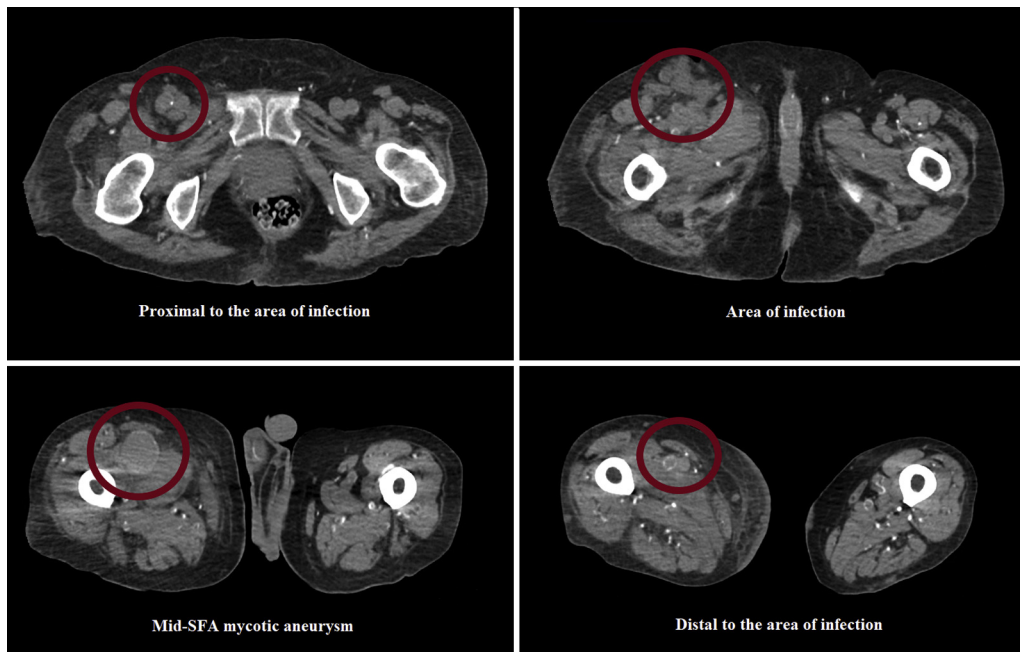


Fig 2. Computed tomography scans of the abdomen and pelvis without contrast enhancement showing area of infection (red circle) and mid-superficial femoral artery mycotic aneurysm (red circle).

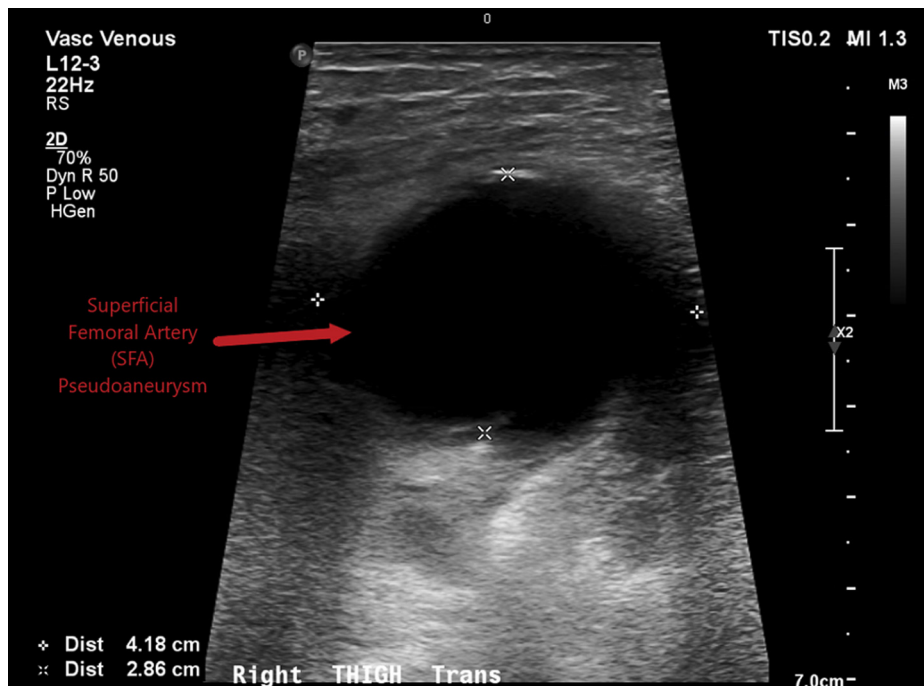


Fig 3. Ultrasound scan of the lower extremity.

patient with positive blood cultures for *L. prolificans*. The patient developed infective endocarditis and embolic phenomenon with cerebral lesions.⁵ Jenks et al¹⁰ reported that hematologic malignancies were the most frequent underlying disease, with the lung the most

frequently involved organ. Similarly, Penteado et al¹¹ indicated that the lungs, urinary tract, central nervous system, and heart were the most commonly involved organs. Ochi et al¹² performed an autopsy of a patient who had died of multiorgan failure despite the

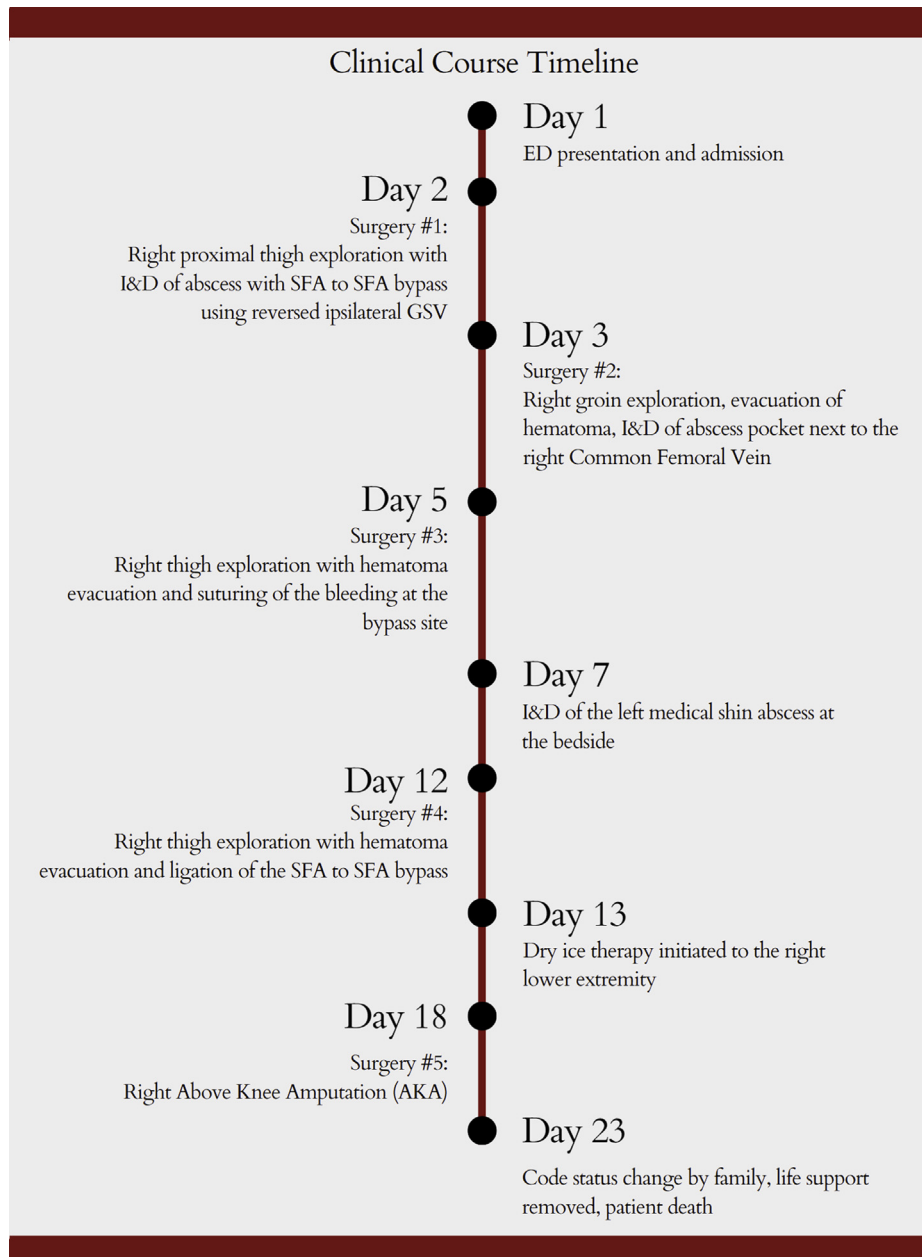


Fig 4. Clinical course timeline. *ED*, Emergency department; *GSV*, great saphenous vein; *I&D*, incision and drainage; *SFA*, superficial femoral artery.

Fungal Pathogen	Amphotericin B	Voriconazole	Posaconazole/Isavuconazole	Echinocandins
<i>L. prolificans</i>				

Inactive (High MIC)

Intermediate

Active (Low MIC)

- *In vitro* antifungal activity of antifungal drugs against non-Aspergillus molds.

- MIC: Minimum Inhibitory Concentration.

- Adapted from:

Lamoth F, Kontoyannis DP. 2019. Therapeutic challenges of non-Aspergillus invasive mold infections in immunosuppressed patients. *Antimicrob Agents Chemother* 63:e01244-19.

Fig 5. Effect of antifungal drugs against *Lomentospora prolificans*. (Reprinted, with permission, from Lamoth and Kontoyannis.⁸)

combined use of voriconazole and terbinafine. The autopsy revealed numerous mycotic emboli within multiple organs and endocarditis caused by *L. prolificans*.¹² Hurst et al¹³ reported a case of invasive aspergillus of the sphenoid sinus with a mycotic aneurysm that extended intradurally and caused fatal subarachnoid hemorrhage. Azar et al¹⁴ reported the case of an immunocompromised patient who had developed an internal carotid artery mycotic aneurysm, associated with a proven invasive fungal infection (presumptively *Mucorales*) of the sphenoid sinuses. The patient survived after undergoing coil embolization with parent vessel sacrifice of the aneurysm combined with liposomal amphotericin B.¹⁴ To the best of our knowledge, after an extensive literature search, our case is the first reported case with such extensive vascular involvement and complications related to arterial and venous bleeding caused by *L. prolificans* infection. Increasing awareness of this disease might help in its early diagnosis and treatment. Further studies are needed to determine the optimal treatment required to control such infections.

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