

A Rare Case of Osteomyelitis Caused by *Scedosporium Apiospermum* in the Right Foot of an Immunocompetent Patient

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Background: Fungal osteomyelitis is rare in clinical practice, and osteomyelitis caused by *Scedosporium apiospermum* is even rarer, which is easy to misdiagnose, and it is resistant to many antifungal drugs, which makes it tricky to treat. Early diagnosis and accurate treatment are essential.

Case Presentation: A 38-year-old healthy male has been experiencing recurrent pain in his right foot, accompanied by skin ulcers and exudate, for the past five years. He has been diagnosed with bacterial osteomyelitis at other hospitals as well as at our hospital. In the first stage, osteomyelitis lesion removal + vancomycin bone cement tamponade was used, and the infected bone tissue was taken for microbial culture and morphological observation, and identified as *Scedosporium apiospermum*. The patient was cured after postoperative treatment with voriconazole. No further signs of infection or *Scedosporium apiospermum* were detected during the second stage of bone reconstruction surgery, and the incision healed with grade A healing and no further signs of osteomyelitis, such as bone destruction, were detected after bone reconstruction surgery.

Conclusion: This is a rare case of *Scedosporium apiospermum* osteomyelitis of the right foot, which was successively misdiagnosed and finally cured by surgery and antifungal treatment with voriconazole. Given that *Scedosporium apiospermum* is extremely rare and resistant to antifungal drugs, this case highlights the importance of microbiologic culture and pathologic examination, surgical debridement, and precise antifungal treatment.

Keywords: foot, *scedosporium apiospermum*, osteomyelitis, surgical debridement, antifungal treatment

Introduction

Fungal osteomyelitis is an inflammatory disease characterized by bone infection and destruction caused by fungi. Due to its rarity and atypical clinical and radiological manifestations, early diagnosis and treatment of fungal osteomyelitis are extremely challenging. In the absence of definitive etiological evidence, many cases are often delayed in diagnosis or misdiagnosed, leading to severe illness and difficult treatment. *Scedosporium apiospermum* is the asexual stage of *Pseudallescheria apiosperma*, a conditionally pathogenic fungus widespread in environments such as sewage, wetlands, and decaying plants.^{1,2} This fungus is increasingly recognized as an emerging, clinically rare, highly invasive, and virulent saprophytic filamentous fungus. The disease is more common in immunocompromised patients, such as those undergoing bone marrow or organ transplantation, AIDS patients, and those with leukemia, but it can also occur in immunocompetent individuals.³ Clinically, fungal osteomyelitis is relatively rare, and osteomyelitis caused by *Scedosporium apiospermum* is even more uncommon. Furthermore, it is resistant to multiple antifungal drugs, making treatment challenging. Herein, we report the clinical manifestations, laboratory results, radiological findings, and diagnostic and treatment process of a case of *Scedosporium apiospermum* osteomyelitis in the right foot of an immunocompetent patient admitted to our department, to draw the attention of healthcare professionals.

Case Presentation

Medical History

A 38-year-old male patient from Chongqing, China, who was previously healthy, unfortunately, suffered a puncture wound on his right foot caused by an iron object from a garbage pile in 2005. After the injury, he underwent debridement and suturing at a hospital in Guangdong, China. Postoperatively, the wound had persistent exudation and healed slowly, finally healing after two months of dressing changes. In October 2018, the patient developed redness, swelling, heat, and pain in his right foot without any apparent cause. He self-administered painkillers and antibiotics for symptomatic treatment, but the efficacy was poor. In January 2019, he went to the First Affiliated Hospital of a Medical University for treatment, where he underwent bone necrosis debridement and iliac bone grafting. Postoperatively, the wound ruptured and had recurrent exudation, which healed after three months of dressing changes and other symptomatic treatments. In January 2023, the patient experienced pain in his right foot again, which was not alleviated with rest and affected his walking. He subsequently sought treatment at the outpatient clinics of the First and Second Affiliated Hospitals of the Medical University, where he received symptomatic treatment with medication for pain relief, resulting in a slight improvement of symptoms. In September 2023, a 3×1.5cm mass appeared at the incision site on his right foot, which was cystic and surrounded by redness, swelling, and heat, occasionally exuding reddish-yellow secretions. Consequently, the patient was hospitalized in our hospital at 09:12 on October 10, 2023. Past medical history: The patient was healthy, with no chronic diseases or immune system disorders. Physical examination: A 3×1.5cm mass was visible at the mid-section of the surgical scar on the dorsum of the right foot, which was cystic and contained a small amount of fluid. The surrounding area was red, and swollen, and had elevated skin temperature with obvious tenderness. Peripheral blood circulation, sensation, and motor function were normal. Preoperative examinations: On February 2, 2023, a foot X-ray at the Second Affiliated Hospital of the Medical University showed diagnostic findings of bone membrane hyperplasia, irregular thickening of the cortical bone, and partial bone destruction in the lateral cuneiform and metatarsal bones, with internal fixation shadows in the lateral cuneiform bone (Figure 1A–C). On October 11, 2023, a CT scan of the right foot revealed diagnostic findings of an abnormal shape of the lateral cuneiform bone with screws inside, irregular cystic bone destruction in the lateral cuneiform, cuboid, and the bases of the third and fourth metatarsal bones, unclear trabecular bone, thickened and sclerosed cortical bone of the third metatarsal, irregular articular surface of the third tarsometatarsal joint, narrowed joint space, increased bone density in the navicular, intermediate cuneiform, and medial cuneiform bones, some trabecular bones were unclear, and there was soft tissue swelling on the dorsum of the foot, suggesting a chronic infectious lesion (Figure 2A–C).

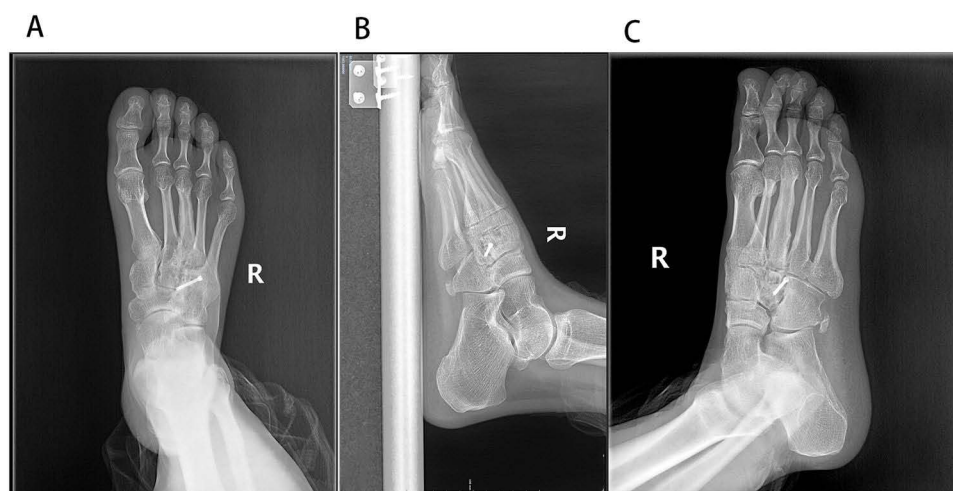


Figure 1 Pre-operative DR. (A) anteroposterior projection, (B) lateral projection, and (C) oblique projection.

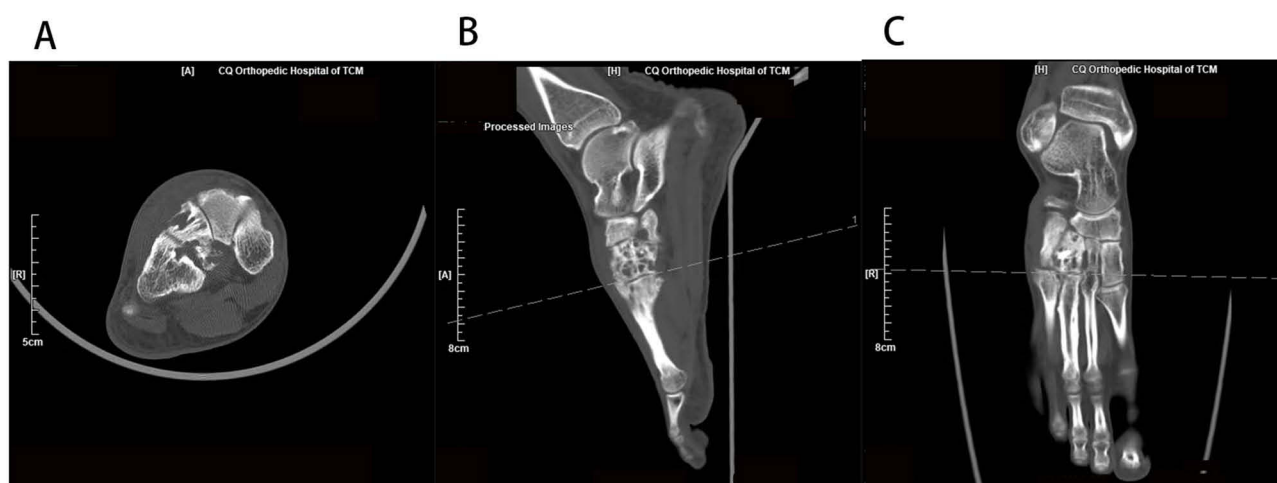


Figure 2 Pre-operative CT. (A) Coronal view, (B) sagittal view, and (C) axial view.

Treatment and Prognosis

After admission, complete examination and exclusion of contraindications to surgery, one-stage surgery was performed on October 13, 2023: right lateral cuneiform internal fixation removal + right lateral cuneiform, middle cuneiform, navicular, dice bone, 3rd metatarsal, and 4th metatarsal osteomyelitis lesion removal + vancomycin cement tamponade + Kirschner's pin external fixation (Figure 3A and B), and intra-operative conditions are shown in (Figure 4A). Postoperatively, cefuroxime sodium + levofloxacin for injection was given to fight infection, microbial culture and identification of *Scedosporium apiospermum* was considered (Figure 5A and B), and pathologic diagnosis of fungal spores and hyphae-like material was seen (Figure 6), and according to the results of the fungal culture and pathology, it was changed to voriconazole for injection as antifungal treatment, and the patient's incision was continued to be taken voriconazole by oral intake for 6 weeks after discharge from the

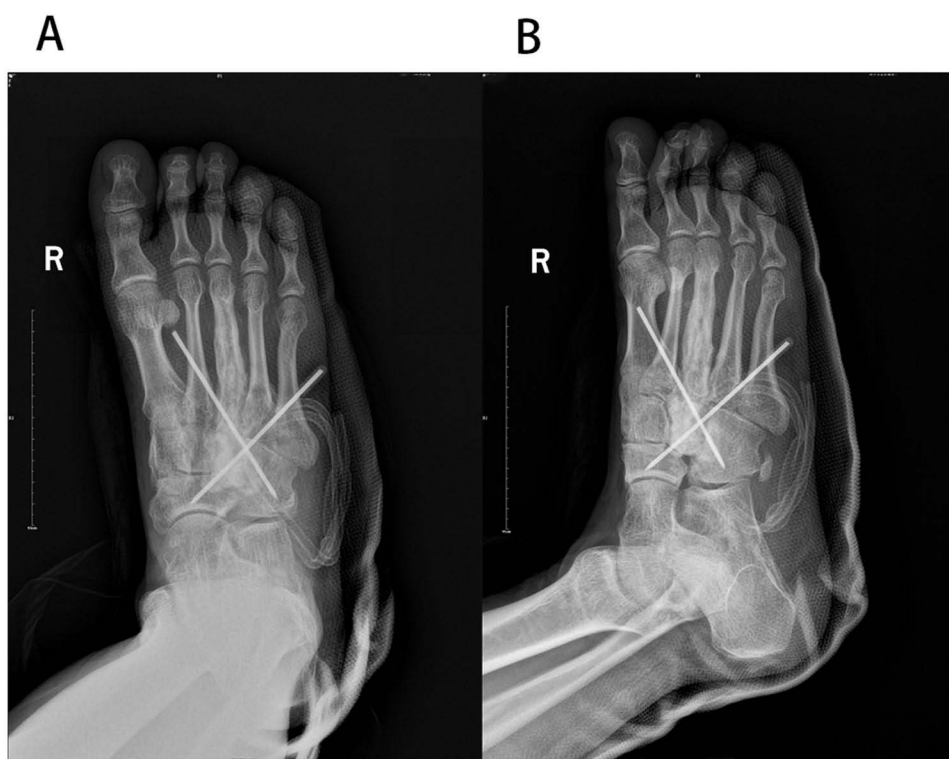


Figure 3 Postoperative DR for one-stage surgery. (A) anteroposterior projection, (B) oblique projection.

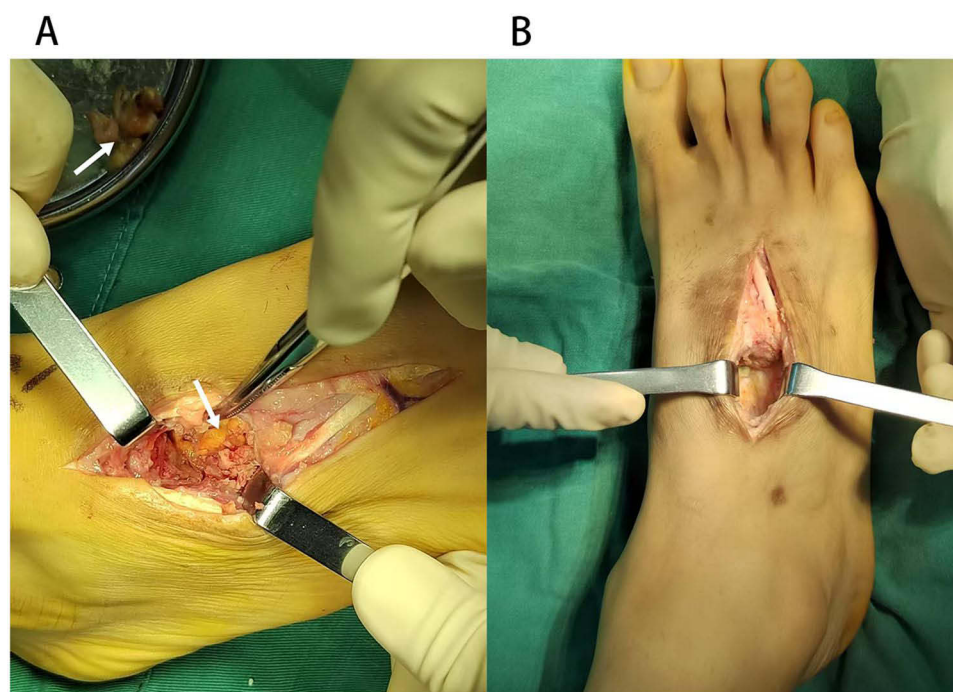


Figure 4 (A) Intraoperative findings during the first-stage surgery: Bone destruction and complete necrosis of the lateral cuneiform bone of the right foot, with partial sclerosis and destruction of the base bone, navicular bone, third and fourth metatarsals, and middle cuneiform bone. A small amount of yellow pus and granular, crab roe-like material (indicated by the arrow) were observed. **(B)** Intraoperative findings during the second-stage surgery: The wound healed well with no redness or swelling. Upon cutting through the skin, no exudate was observed around the bone cement, and the induced membrane was clean without secretions. There was no significant bone destruction or sclerotic bone.

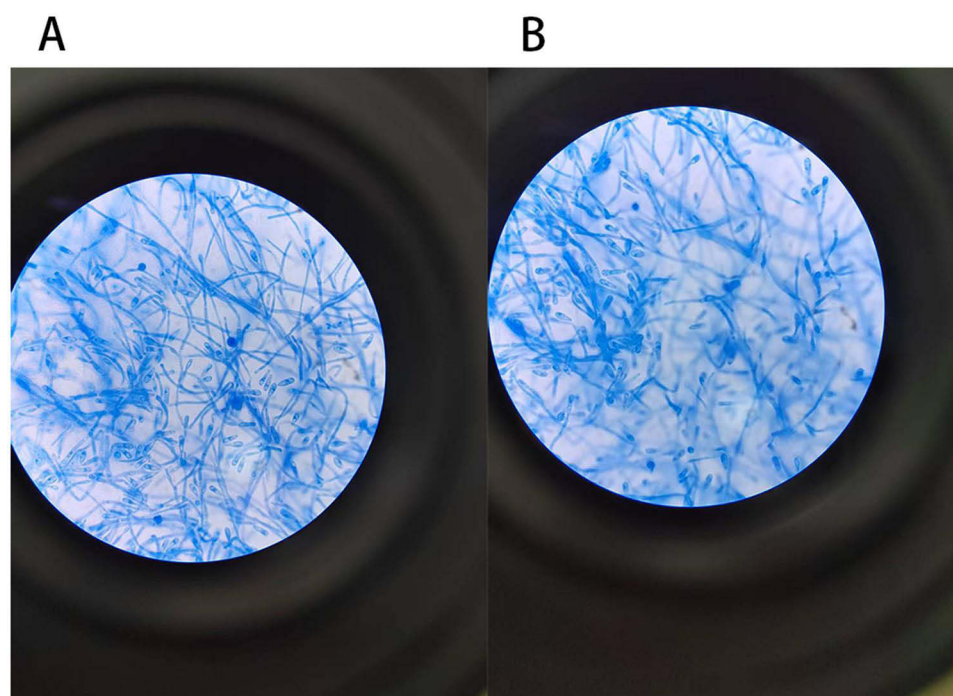


Figure 5 (A–B) October 18, 2023 The microbial culture and identification showed that after 3 days of culture, no bacterial growth was observed in the routine culture. Upon extending the culture period, a small amount of *Scedosporium* growth was observed, morphologically considered *Scedosporium apiospermum*.

hospital, and the patient's incision was grade A healing. After the infection was controlled, a second-stage surgery was performed on October 10, 2024: antibiotic bone cement removal of the right foot + bone grafting of the tarsal and metatarsal bone defects from the iliac bone + internal fixation of the tarsal and metatarsal plate screws

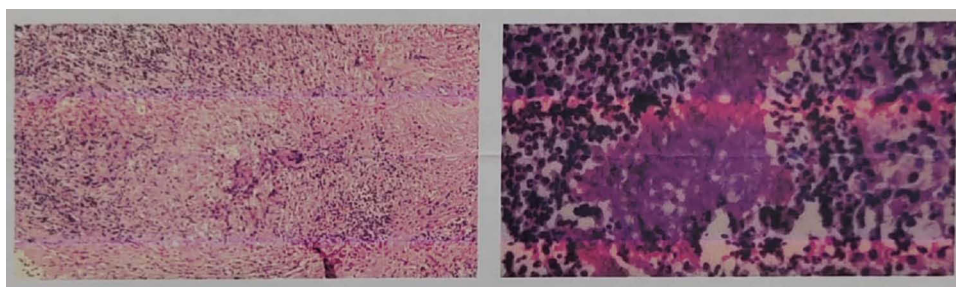


Figure 6 October 19, 2023 The pathological diagnosis report indicates: “The submitted tissue from the right foot shows acute suppurative inflammation with abscess formation, necrosis, and granuloma formation. Fungal spores and hyphal-like structures are visible within. Please correlate with laboratory tests. Fluorescent immunohistochemical staining: positive (+)”.



Figure 7 Postoperative DR for second-stage surgery. (A) anteroposterior projection, (B) lateral projection, and (C) oblique projection.

(Figure 7), and the intra-operative situation is shown in (Figure 4B). After the second stage of bone grafting, the wound healed at grade A, the fungal culture was negative. On review 5 months after the second stage surgery, no further signs of osteomyelitis such as bone destruction were found. (Figure 8).



Figure 8 5-month postoperative review of DR after second-stage surgery. (A) anteroposterior projection, (B) lateral projection, and (C) oblique projection.

Discussion

Clinically, bacterial osteomyelitis is relatively common, whereas fungal osteomyelitis is rare. Fungal osteomyelitis typically presents with local symptoms such as pain, tenderness, swelling, and pus discharge. Radiologically, it often manifests as lytic lesions, bone destruction, and bone erosion.⁴ The clinical symptoms are atypical, and the radiological findings are generally similar, with sporadic case reports in the clinical literature. In the absence of microbiological evidence, early diagnosis and treatment of fungal osteomyelitis are challenging. Osteomyelitis caused by *Scedosporium apiospermum* is even rarer and is easily misdiagnosed in clinical practice. Therefore, in this case, the patient underwent debridement of necrotic bone and iliac bone grafting at another hospital, but the treatment failed. Upon admission to our hospital, bacterial osteomyelitis was initially considered, with the possibility of fungal infection not ruled out. Before the bacterial culture results were available, conventional treatment for osteomyelitis was administered: vancomycin-impregnated bone cement was used to fill the defect after debridement, and intravenous cefuroxime sodium and levofloxacin was administered postoperatively for infection control. Antifungal treatment with voriconazole was not initiated until *Scedosporium apiospermum* was confirmed microbiologically. Currently, the diagnosis of *Scedosporium apiospermum* infection relies on non-specific CT scans, lengthy and insensitive invasive biopsy for fungal culture, and time-consuming histopathological examination.⁵ Therefore, rapid laboratory isolation of the pathogen is crucial for patient diagnosis and treatment.

Infections caused by *Scedosporium apiospermum* commonly occur in the eyes, lungs, intracranial region, sinuses, skin, joints, and spine. In this case, following surgical debridement and antifungal treatment with voriconazole, the wound healed well (Class A healing), and during the second-stage surgery, the induced membrane was clean, with no signs of infection or pathogens detected. Follow-up after the second-stage bone grafting surgery did not reveal any signs of bone destruction or osteomyelitis. Therefore, early diagnosis and treatment are crucial for the prognosis of patients.⁶ Given the rarity of the disease in this patient, the department organized multiple full-department discussions and actively adopted a multidisciplinary team (MDT) approach, particularly seeking the assistance of the clinical laboratory department and the pharmacy department. Fungal osteomyelitis in immunocompetent individuals is extremely rare⁷. The transmission mechanism primarily involves inhalation or traumatic inoculation with contaminated material into the skin or deeper structures^{8,9}.

Scedosporium apiospermum can cause localized infections following trauma¹⁰ or surgery, as seen in this immunocompetent patient who developed an infection after an injury or surgery. In immunocompromised individuals, however, it can cause severe disseminated disease.

Scedosporium apiospermum is naturally resistant to fluconazole, itraconazole, and amphotericin B, while voriconazole has excellent bacteriostatic activity against *Scedosporium apiospermum*. Voriconazole is approved by the US Food and Drug Administration (FDA) for the treatment of patients with severe *S. acne* infections who are intolerant to other antifungal agents.¹¹ The European Society for Clinical Microbiology and Infectious Diseases Fungal Infections Study Group (EFISG) and the European Confederation of Medical Mycology (ECMM) 2013 joint guideline level A recommendations for the use of voriconazole are both voriconazole.¹² Our patient was treated postoperatively with voriconazole antifungal therapy based on laboratory cultures, and the incision healed with grade A. No further pathogens were found in the second stage of surgery. The standard clinical management of osteomyelitis involves long-term antibiotic therapy as well as surgical resection of the infected tissue.¹³ Therefore, in limited *Scedosporium apiospermum* osteomyelitis, surgical excision of the infected tissue should be an important part of treatment in addition to antifungal therapy.^{14,15} Because fungal osteomyelitis is rare, clinicians should consider the possibility of fungal infection if it does not respond to antibacterial therapy in the clinic.¹⁶ Early diagnosis, surgical debridement and appropriate antifungal therapy based on fungal species can lead to better clinical outcomes.¹⁷

Conclusions

This case underscores the importance of rapid laboratory isolation of the pathogen, early diagnosis, surgical debridement, and precise antifungal therapy for fungal osteomyelitis. Clinically, bacterial osteomyelitis is common, while fungal osteomyelitis is rare. If there is no response to antibacterial treatment, clinicians should consider the possibility of fungal infection. Although human infections caused by *Scedosporium apiospermum* have mostly been reported as individual

cases in recent years, *Scedosporium* species remain relatively unfamiliar to clinical healthcare providers. Considering the severity of *Scedosporium apiospermum* infections and the uniqueness of treatment options, clinical staff should be fully vigilant and take *Scedosporium* infections seriously, ensuring timely and effective treatment to safeguard patient safety.

Abbreviations

AIDS, Acquired Immune Deficiency Syndrome; MDT, Multidisciplinary Team.

Ethics Approval and Consent to Participate

Informed consent for the publication of medical histories and photographs was obtained from the patient. Additionally, approval for case publication was granted by the Ethics Committee of Chongqing Orthopedic Hospital of Traditional Chinese Medicine.

Consent for Publication

Written informed consent has been provided by the patient to have the case details and any accompanying images published. Institutional approval was not required to publish the case details.

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Disclosure

The authors report no conflicts of interest in this work.

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