



Aspergillus Septic Arthritis of the Hip in an Immunocompetent Middle-aged Female with Undiagnosed Recurrent Pulmonary Aspergillosis

Pil Whan Yoon, MD, Joo Ho Song, MD, Kang Sup Yoon, MD*,
Jae Suk Chang, MD, Hee Joong Kim, MD[†], Kee Hyung Rhyu, MD[†]

Department of Orthopaedic Surgery, Asan Medical Center, Seoul, Korea
Department of Orthopaedic Surgery, SMG-SNU Boramae Medical Center, Seoul, Korea*
Department of Orthopaedic Surgery, Seoul National University Hospital, Seoul, Korea[†]
Department of Orthopaedic Surgery, Kyung Hee University Hospital at Gangdong, Seoul, Korea[‡]

We present a case of *Aspergillus* septic hip arthritis in an immunocompetent patient with undiagnosed recurrent pulmonary aspergillosis who underwent arthroscopic surgery. Biopsy specimens of synovium revealed fungal hyphae, confirming *Aspergillus* infection. *Aspergillus* septic hip arthritis can occur in immunocompetent patients, and arthroscopy can be a noninvasive surgical option in these cases.

Key Words: Hip, *Aspergillus*, Arthritis, Infectious, Arthroscopy

Although *Aspergillus* species are ubiquitous in the environment and exposure to their spores is a daily occurrence, *Aspergillus* infections are rare and occur primarily in immunocompromised hosts¹⁾. Septic arthritis by invasive aspergillosis is relatively rare; most cases are associated with compromised host defenses¹⁾. We present the case of *Aspergillus* septic arthritis of the hip in an immunocompetent adult patient with undiagnosed

recurrent pulmonary aspergillosis, and describe the outcome of hip arthroscopy and its intraoperative findings.

CASE REPORT

A 49-year-old female presented with a 3-month history of progressive pain and limited range of motion of the right hip, showing no fever and no indication of localized heat, redness, and soft tissue swelling around the hip. An anteroposterior hip radiograph showed a slight narrowing of the joint space in the right hip (Fig. 1A). A magnetic resonance image revealed marked joint effusion, and edematous changes of the bone marrow and muscles around the right hip joint (Fig. 1B). Her laboratory results were as follows: white blood cells (WBC), 12.2/mm³; erythrocyte sedimentation rate, 69 mm/hr; C-reactive protein, 2.56 mg/dL.

We decided to perform a hip arthroscopy on both diagnostic and therapeutic purpose, suspecting septic arthritis based on the images and laboratory findings. Arthroscopic findings included intra-articular hematoma

Submitted: June 26, 2015 **1st revision:** August 4, 2015

Final acceptance: August 12, 2015

Address reprint request to

Kee Hyung Rhyu, MD

Department of Orthopedic Surgery, Kyung Hee University Hospital at Gangdong, 892 Dongnam-ro, Gangdong-gu, Seoul 05278, Korea

TEL: +82-2-440-6156 **FAX:** +82-2-440-6296

E-mail: khrhyu@gmail.com

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/4.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

and hyperemic synovium overlaying the entire acetabular labrum and joint capsule (Fig. 2A). There was some hypertrophic synovial tissue in the acetabular fossa invading the subchondral bone and erosion of acetabular cartilage (Fig. 2B). A synovectomy was performed using an arthroscopic shaver and a

radiofrequency probe. Synovial biopsy samples were taken for culture and histological examination. High-volume irrigation using 30 L of normal saline was performed. The cultures contained no bacterial, fungal, or mycobacterial growth. However, biopsy specimens revealed fungal hyphae in the fibrous tissue and chronic

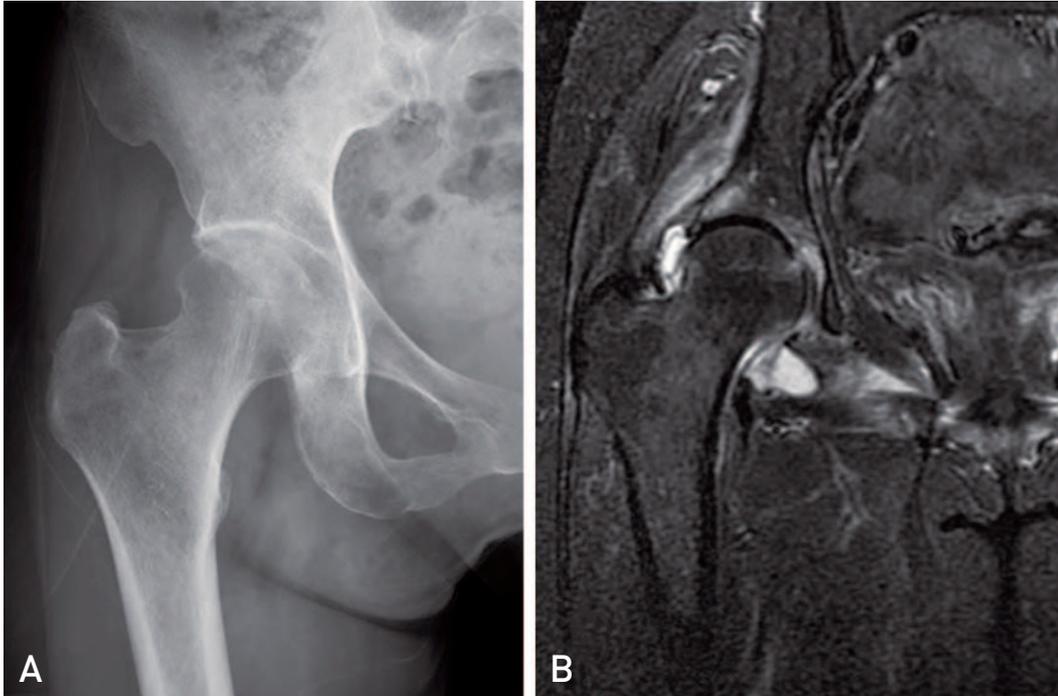


Fig. 1. (A) Anteroposterior hip radiograph showing a slight narrowing of the joint space. (B) Magnetic resonance image showing marked joint effusion, and edematous changes of the bone marrow and muscles around the hip joint.

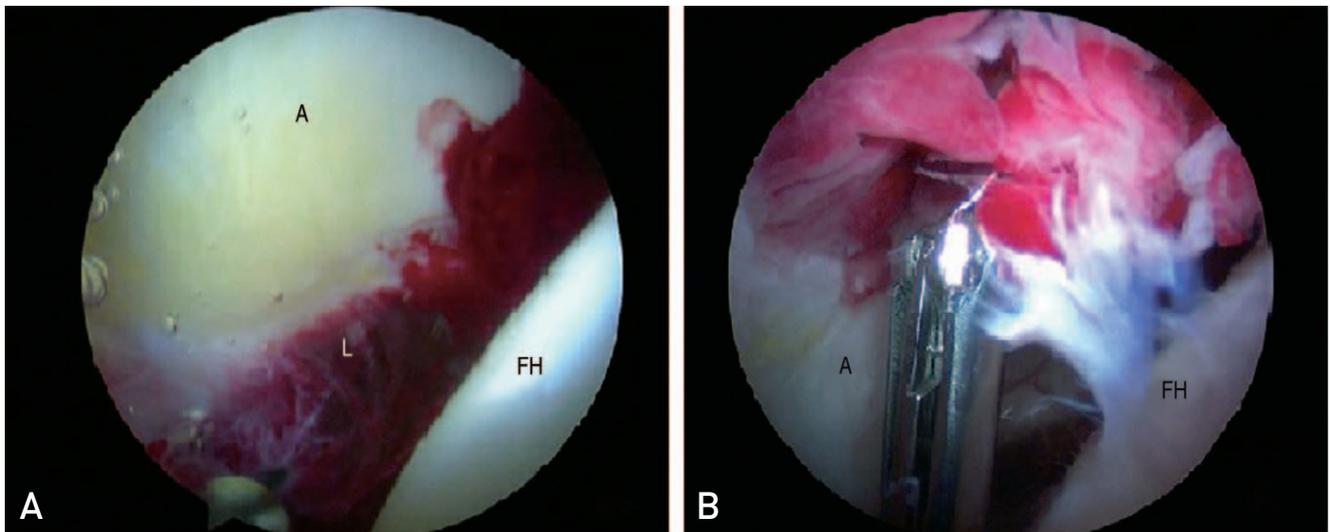


Fig. 2. (A) Arthroscopic view of a right hip through the anterolateral portal showing severe villous synovitis in the acetabular fossa. Synovial biopsy samples were taken through the posterolateral portal for culture and histological examination. (B) Arthroscopic view showing erosion of the acetabular cartilage and revealed subchondral bone. A: acetabular cartilage, FH: femoral head, L: labrum.

active inflammation with abscess formation, confirming a diagnosis of *Aspergillus* septic arthritis (Fig. 3). She had no apparent immune deficiency; an human immunodeficiency virus (HIV) test was negative, immunoglobulin G and subtypes, and CD4 and CD8 counts were normal. Because her past medical history revealed three previous episodes of bronchogenic cysts, of an unknown cause, which had been treated with surgical or percutaneous drainage at other hospital in 2003, 2007, and 2010, we retrospectively performed a thorough review of her other hospital records and found

previously undiagnosed reports of *Aspergillus fumigatus* in intraoperative cultures from 2003 and 2010. The last episode of bronchogenic cysts occurred 5 months before the onset of the pain in the right hip and was treated by sclerotherapy after percutaneous drainage; however, the patient had never been prescribed any antifungal agents.

After the hip arthroscopy, intravenous amphotericin B was administered for 2 weeks, followed by voriconazole, orally administered for 6 months. Hematologic and biochemical test results had normalized at 4 months after surgery; however, at 9 months after surgery she

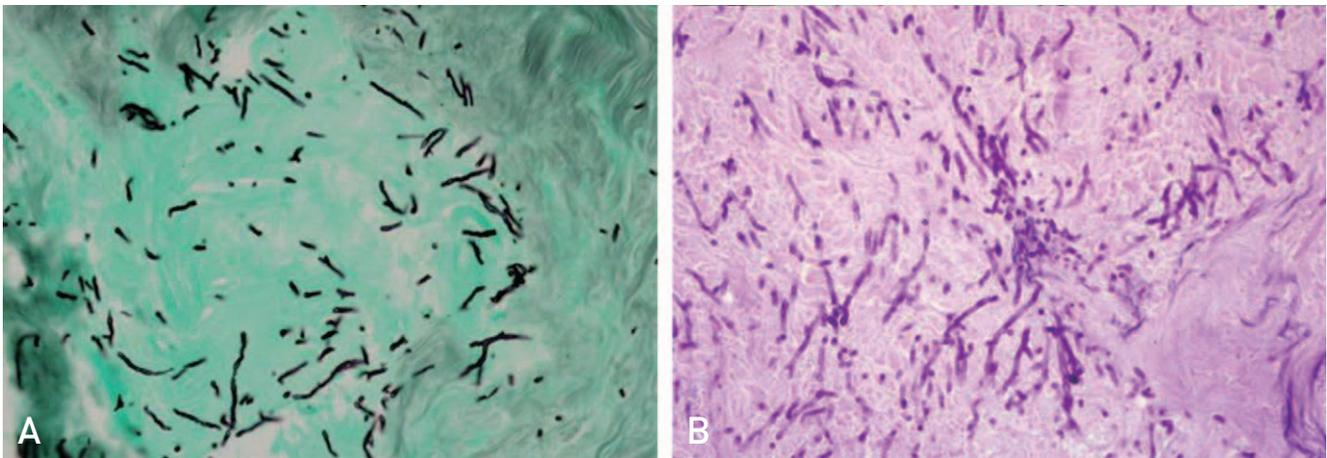


Fig. 3. Biopsy specimens reveal fungal hyphae in the fibrous tissue and chronic active inflammation with abscess formation. (A) Gomori methenamine silver stain, (B) periodic acid-Schiff stain; $\times 400$.

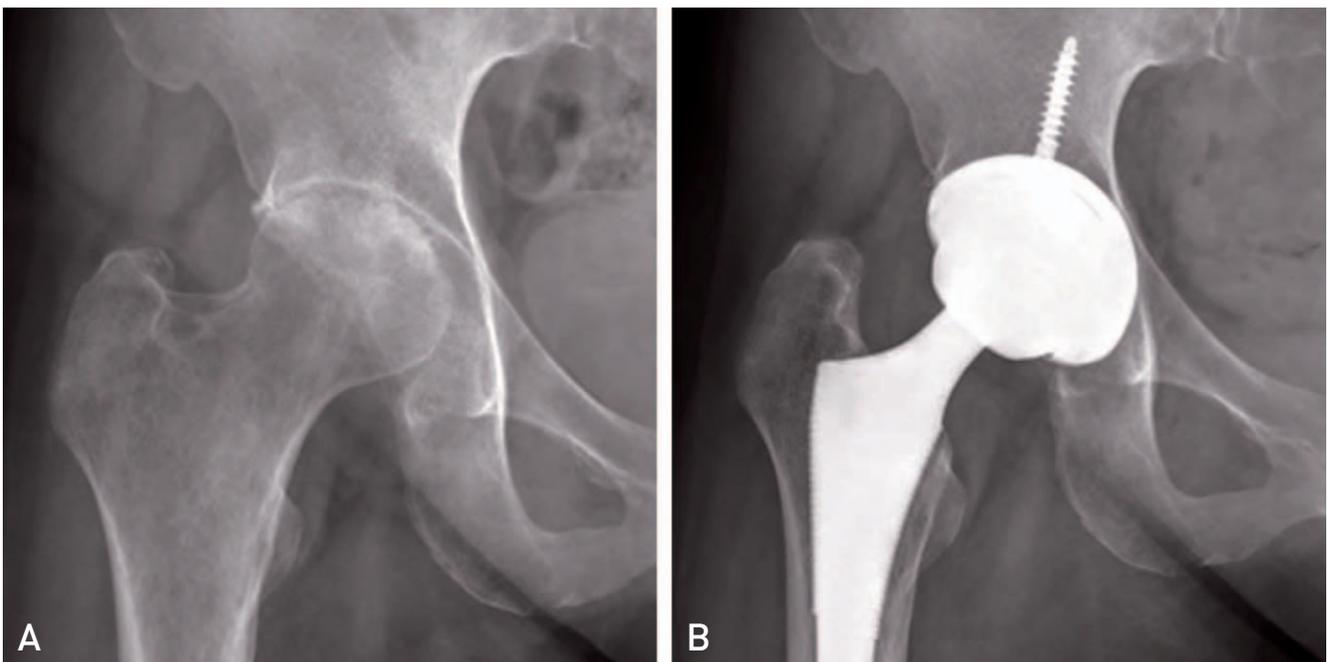


Fig. 4. Anteroposterior hip radiographs before (A) and after (B) total hip arthroplasty.

underwent a total hip arthroplasty (THA) due to persistent pain with severe joint space narrowing visible on plain radiographs (Fig. 4). Postoperative cultures did not have any fungal growth and fungal organisms were not found in histological examinations. Nineteen months after the THA, the patient presented no signs of infection recurrence.

DISCUSSION

We describe a case of *Aspergillus* septic arthritis of the hip in a middle-aged immunocompetent female with a history of undiagnosed recurrent pulmonary aspergillosis. Articular aspergillosis is rare and usually occurs in association with compromised host defense and intra-articular steroid injections¹. To our knowledge, only 24 cases of *Aspergillus* septic arthritis have been reported¹⁻⁴. The knee joint was most commonly affected, followed by the shoulder; however, septic hip arthritis caused by *Aspergillus* was reported in only one patient who had received a liver transplantation². Our patient had no apparent underlying immunodeficiency and no history of intra-articular steroid injections. She did have three previous episodes of bronchogenic cysts caused by *Aspergillus fumigatus* which was identified after a thorough review of other hospital records. Although the bronchogenic cysts had been treated with surgical or percutaneous drainage, she had not taken any antifungal agents. Pulmonary aspergillosis is rare in non-immunocompromised and non-neutropenic patients, with few cases being reported in the literature¹. In most previously reported cases, *Aspergillus* septic arthritis was caused by disseminated diseases linked to hematogenous spread. We assumed that without antifungal treatment for the recurrent pulmonary aspergillosis, the microorganism was able to spread hematogenously to create *Aspergillus* septic arthritis of the hip in this patient. *Aspergillus fumigatus* is the most common pathogen responsible for *Aspergillus* septic arthritis; however, *Aspergillus terreus* and *Aspergillus flavus* were also identified in the knee and shoulder joints³. Although there was no fungal growth on cultures from the synovial fluid and tissues, identification was made after a synovial biopsy and histopathology showing typical acute *Aspergillus* branching hyphae. In case review about joint infection by *Aspergillus fumigatus*, Golmia et al.¹ found that 9 cases out of 14 were confirmed on basis of culture

results and other 4 cases were diagnosed by tissue biopsy. They also reported that among 4 cases which had results of joint fluid analysis, 2 cases showed WBC count of joint fluid less than 50,000/mm³, and only 1 case showed differential count of neutrophil more than 90%.

There is no consensus on the type of antifungal therapy for treatment of *Aspergillus* septic arthritis. Recently, voriconazole has become the drug of choice for invasive aspergillosis¹; however, Stratov et al.⁵ performed a literature review and highlighted the importance of surgery combined with the use of antifungal agents for *Aspergillus osteomyelitis*. Of the previous 24 patients with *Aspergillus* septic arthritis, 13 were treated by open synovectomy and 4 had arthroscopic surgery (3 knees and 1 shoulder joint)¹⁻⁴. Our patient was treated with arthroscopic debridement and synovectomy, and this is the first case of *Aspergillus* septic arthritis of the hip joint to be treated by hip arthroscopy.

Although the *Aspergillus* infection was successfully controlled with the hip arthroscopy and the 2-week intravenous amphotericin B, and 6-month oral voriconazole, our patient underwent a THA because of persistent pain with severe narrowing of the joint space, as seen on plain radiographs. The poor prognosis of this patient may be due to the delayed diagnosis and treatment of the *Aspergillus* infection, because her initial plain radiographs showed narrowing of the joint space of the affected hip, and the arthroscopic findings revealed invasive synovitis with cartilage destruction.

As in this case, septic arthritis caused by fungi tends to be more indolent than bacterial septic arthritis, and diagnostic tools routinely used for septic arthritis such as joint fluid analysis or culture may not be helpful¹. This forces surgeons to be alert to the possibility of fungal joint infection, as late diagnosis is often followed by poor prognosis.

In summary, another case of *Aspergillus* septic arthritis is reported, representing the second case involving the hip joint. Orthopedic surgeons should be aware that *Aspergillus* septic arthritis of the hip can occur even in an immunocompetent patient, and hip arthroscopy can be a noninvasive surgical option in such cases.

REFERENCES

1. Golmia R, Bello I, Marra A, Hamerschlak N, Osawa A,

- Scheinberg M. *Aspergillus fumigatus* joint infection: a review. *Semin Arthritis Rheum.* 2011;40:580-4.
2. Kim T, Lee SR, Cho OH, et al. A case of septic hip arthritis caused by *Aspergillus fumigatus* in a liver transplant recipient. *Infect Chemother.* 2008;40:170-4.
 3. Yu OH, Keet AW, Sheppard DC, Brewer T. Articular aspergillosis: case report and review of the literature. *Int J Infect Dis.* 2010;14:e433-5.
 4. Dal T, Tekin A, Deveci Ö, Bulut M, Firat U, Mete M. *Septic arthritis caused by Aspergillus fumigatus in an immunosuppressive patient: A case report and review of the literature.* *J Microbiol Infect Dis.* 2012;2:29-32.
 5. Stratov I, Korman TM, Johnson PD. Management of *Aspergillus* osteomyelitis: report of failure of liposomal amphotericin B and response to voriconazole in an immunocompetent host and literature review. *Eur J Clin Microbiol Infect Dis.* 2003;22:277-83.