Thoracic Interdural Aspergillus Abscess Causing Rapid Fatal Spondylitis in the Presence of Posterior Mediastinitis

Joon-Seok Kim, M.D.,¹ Sung-Bum Kim, M.D.,¹ Hyeong-Joong Yi, M.D.,¹ Won-Sang Chung, M.D.²

Department of Neurosurgery, Thoracic and Cardiovascular Surgery, Hanyang University Medical Center, Seoul, Korea

Most primary spinal abscesses, irrespective of pathogens and anatomical locations, have better prognosis than that of secondary abscesses with spondylitis. We report a 68-year-old man, previously undertaken pulmonary resection due to tuberculosis, presented with paraparesis. Imaging studies showed primary intraspinal abscesses at T-1 and T-3 vertebral levels, semi-invasive pulmonary Aspergillosis and inflammation of the posterior mediastinum. Operative procedure and histopathological examination revealed interdural Aspergillus abscess. Despite chemotherapy, he deteriorated progressively, and spondylitis developed at corresponding vertebrae. He eventually died 6 weeks postoperatively due to pulmonary complication. The authors intended to inform that such an extradural inflammatory lesion of Aspergillus abscess should be treated carefully.

KEY WORDS: Aspergillosis · Interdural abscess · Posterior mediastinitis · Spondylitis.

Introduction

spergillus frequently involves the spine with the typical radiographic features of nonsuppurative vertebral osteomyelitis¹⁾. Primary abscess, including spinal epidural abscess, without involvement of surrounding musculoskeletal structures, occurs less commonly, invariably needs surgical intervention, and shows better prognosis than the secondary counterpart⁷⁾. Rapid fatal development of the secondary abscess with full-blown spondylitis from the pure primary abscess, particularly in immunocompetent Aspergillus patient, is not likely to occur. Among numerous factors responsible for this condition, we suggest the role of concomitant pulmonary (posterior mediastinum) inflammation. We briefly summarized case history and discussed the possible explanation to this phenomenon.

Case Report

A 68-year-old man, without chronic debilitating illness, presented with progressive bilateral leg paralysis of 1-

- Received: August 19, 2004
 Accepted: September 13, 2004
- Address for reprints: Sung-Bum Kim, M.D., Department of Neurosurgery, Hanyang University Medical Center, Haengdangdong 17, Seongdong-gu 133-792, Seoul, Korea

Tel: 02) 2290-9790, Fax: 02) 2281-0954 E-mail: sungbumi7@hmc.hanyang.ac.kr week duration. Prior to this admission, he had been well and free of locomotive limitation. He had a significant medical history of right upper lobectomy due to the pulmonary tuberculosis 35 years ago, and received anti-tuberculous medication for another 3 years to attain complete "cure". At admission, vital signs showed non specific finding except high fever up to 38.6°C Blood tests revealed a peripheral leukocytosis(18,000), erythrocyte sedimentation rate(ESR) 148mm/h, and C reactive protein(CRP) 35mg/dl. On examination, he showed paraparesis (Gr. 2), hypoesthesia below T-4 cord level, bilaterally positive Babinski signs, urinary retention, and adynamic ileus. Chest radiograph showed a right upper lobe fibrosis and posterior mediastinal widening. A "crescent" sign indicated aspergillosis of semiinvasive type. MRI of the thoracic spine showed two isolated round intraspinal masses with peripheral enhancement, located at the right side of T1 and T3 levels. The lesions abutted on the posterior vertebral bodies, but without concurrent signal changes within them. Radionuclide bone scan also failed to reveal any vertebral involvement (Fig. 1).

Aspergillus epidural abscess was suspected, and operative procedure was scheduled. Because there were posterior mediastinitis, previous operative scarring at the rightside of lung and lack of vertebral involvement, posterior laminectomy and abscess drainage was planned. Following laminectomy T1 to T3, very small amount of epidural granulation was found on thinned outer dura, but responsible

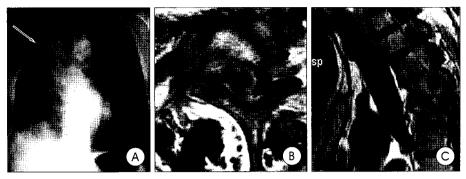


Fig. 1. A: Initial plain chest radiograph shows contracted right lung field and mediastinial widening with crescent sign in the right upper lung (arrow). T2—weighted axial (B) and sagittal magnetic resonance images (C) at admission show two isolated intraspinal lesions with peripheral enhancement at right side of T1 and T3 vertebral level.

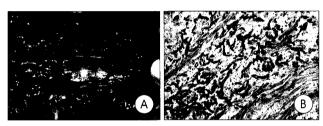


Fig. 2. A : Intraoperative photograph reveals suppurative material and granulation (arrow) and cut margin of the inner side of the dura (arrowhead). B : Photomicrogram of surgical specimen demonstrates numerous intermingled fungal hyphae of Aspergillus species (H & E, original magnification, x100).

mass was not identified. Dura was incised to reveal swollen cord with stony hard consistency, and dural thickening was identified at the right side. Incision to the thickened inner side of the dura revealed suppurative abscess material and granulation within the interdural space. There was no fistulous connection between abscess cavity and vertebral bodies. Histopathological examination confirmed numerous intermingled fungal hyphae of Aspergillus species (Fig. 2).

The patient's postoperative course was remained unchanged, and medical treatment with Itraconazole (400mg/day) was commenced and ensued for 27days. Intravenous Amphotericin B was discarded due to elevation of blood urea nitrogen and creatinine. Magnetic resonance imaging, postoperative 15th day, showed relief of the mass effect exerted by the abscesses, however, the extent of the mediastinal inflammation was more progressed and suspicious high signal intensities were shown within the vertebrae.

Five weeks after the first operation, he was readmitted for dyspnea and painful ascending paralysis of the arms. All blood tests revealed evidence of fulminating infection (peripheral leukocytosis with left shift, elevated ESR and CRP). Examination showed complete paraplegia, anesthesia below T-4 cord level, dysesthetic pain and weakness of both

elbows and shoulders. Chest radiograph showed total haziness on the right lung field, suggesting complicated invasive pulmonary aspergillosis. MRI revealed frank vertebral osteomyelitis (T1-3), propagation of the intradural abscess with ascending spread of cord swelling to the lower cervical cord. Bone scan also exhibited hot uptake spots at the T1 to T3 vertebral bodies, mainly right side (Fig. 3). Despite intensive therapy and systemic support, the patient's

condition continued to deteriorate, and he died of the adult respiratory distress syndrome on day 8 readmission.

Discussion

A spergillosis, an infrequent opportunistic fungal infection primarily occurred in immunocompromised patients, involves the spine with the typical radiological features of the nonsuppurative osteomyelitis¹⁾. Spinal column is one of the most commonly involved sites of Aspergillus, and it is usually secondary to the preexisting infection focus³⁾. This patient showed primary abscess, without involvement of nearby structures, on the initial presentation⁷⁾, and underwent surgical decompression followed by postoperative chemotherapy. His clinical course and radiological findings, however, ended in more wide spread form; secondary abscess and demise.

Concerning pathogenesis of spondylitis, there are two major spreading patterns; direct contagious contact from the adjacent pulmonary infection and hematogenous spread^{1,9)}. Very rapid evolution of destructive process generally has been attributable to the hematogenous origin, and it is frequent in chronically ill patient with poor nutrition and immunosuppression, in contrast to the present case^{2,13)}. Development of aspergilloma in the upper thoracic vertebrae possibly reflects direct spread from pulmonary infection, especially in patients with preexisting lung cavity such as tuberculous^{6,13)}. A "crescent" sign is representative of semiinvasive or chronic necrotizing aspergillosis, although distinction between two is not always possible^{6,8)}. Invasive pulmonary aspergillosis, characterized by multiple pulmonary nodules due to proliferation of fungal hyphae, exhibits destruction of all paraspinal structures, can also manifest "crescent sign" in relatively late course, and therefore,

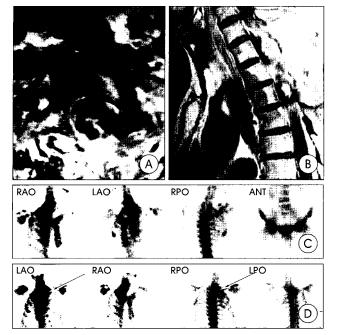


Fig. 3. T2-weighted axial (A) and sagittal magnetic resonance images (B) at secondary admission show aggravation of abscesses with loss of cord integrity by progressive abscess growth at the right side. Also note conspicuous high signal intensity change within the vertebrae. Radionuclide bone scan at the first admission (C) shows no foci of hot uptake lesion, except previous scarring due to right lobectomy, whereas scan obtained at the second admission (D) shows hot uptake foci at upper thoracic vertebrae (T1 T3) mainly at right side (arrows).

scrupulous differential diagnosis is crucial^{8,11}. Disseminated aspergillosis, infection of two or more noncontiguous organs, is also known to involve spinal cord, but localized involvement is rare^{12,14}.

We found a case dealing intradural Aspergillus abscess following epidural steroid injection¹⁰⁾, however, did not find any English literatures concerning the 'interdural' abscess. We thought this entity as an extended form of epidural abscess. Intraoperative findings (thinner outer dural wall and small amount of epidural granulation) account for transdural spread of Aspergillus, although we could not completely exclude the fistulous connection that located anterior to the spinal cord. And, when we consider both the low virulence of Aspergillus and initial disease spectrum of the pulmonary aspergillosis (semi-invasive), contagious spread alone is less likely to cause such a rapid occurrence of spondylitis. There should have been additional factors attributable to this unusual situation; localized abscess and semi-invasive lung aspergillosis in immunocompetent patient on initial presentation. Aspergillus mediastinitis, albeit extremely rare, usually extends to the spinal cord from spondylitis, and frequently produces anterior spinal artery syndrome by mediastinal vascular invasion¹⁴. Secondary spondylitis seemed to extend to the spinal cord with consequent formation of necrosis and ascending infection in this patient. It might be suggested that the presence of inflammation of the posterior mediastinum result in such sequels, in a certain but not fully understood manner.

Although intravenous Amphotericin injection has been mainstay for the management of Aspergillus infection, its nephrotoxicity precluded prolonged use. Instead, Itraconazole emerged as a promising alternative, although its pharmaceutical potency is still questionable^{4,8)}. Surgery is recommended in the presence of neurologic compromise, unstable spinal column, and medically refractory abscess^{2,3)}. However, postoperative chemotherapy is still mandatory in most cases. Prognosis is chiefly related to the patients' general immunological condition¹³⁾, and because of irreversible demyelination, vacuolization and localized necrosis, early diagnosis is important for preventing permanent neurologic deficits and guaranteeing patient's survival^{5,12)}.

Conclusion

In this case, we suggest the role of concomitant inflammation of the posterior mediastinum in rapid development of spondylitis, although there was still inconclusive evidence of active hematogenous spread in this immunocompetent patient. Clinicians should be very careful of such inflammatory lesion outside of the spinal cord when predicting prognosis of Aspergillus abscess.

References

- Baldwin NG, Donald FC: Fungal and parasitic infections of the spine.
 In: Batjer HH, Loftus CM, eds. Textbook of neurological surgery.
 Baltimore: Lippincott Williams & Wilkins, 2002, Vol 4, pp3263-3267
- Bridwell KH, Campbell JW, Barenkamp SJ: Surgical treatment of hematogenous vertebral aspergillus osteomyelitis. Spine 15: 281-528, 1990
- Castelli C, Benazzo F, Minoli L, Marone P, Seghezzi R, Carlizzi CN: Aspergillus infection of the L3-L4 disc space in an immunosuppressed heart transplant patient. Spine 15: 1369-1373, 1990
- Cattelan AM, Loy M, Tognon S, Rea F, Sasset L, Cadrobbi P: An unusual presentation of invasive aspergillosis after lung transplantation. Transpl Int 13: 183-186, 2000
- Guermazi A, Benchaib N, Zagdanski AM, Hocqueloux L, Rili M, Molina JM, et al: Cerebral and spinal cord involvement resulting from invasive aspergillosis. Eur Radiol 12: 147-150, 2002
- 6. Hendrix WC, Arruda LK, Platts-Mills TA, Haworth CS, Jabour R, Ward Jr GW: Aspergillus epidural abscess and cord compression in a patient with Aspergilloma and empyema. Am Rev Respir Dis 145: 1483-1486, 1992
- Khan SH, Hussain MS, Griebel RW, Hattingh S: Title comparison of primary and secondary spinal epidural abscesses: a retrospective analysis of 29 cases. Surg Neurol 59: 28-33, 2003
- 8. Miller WT: Aspergillosis: a disease with many faces. Semin Roentgen

- 31:52-66,1996
- Park YS, Kim SB, Cho YE, Jung WH: Thoracic Intradural Aspergillus Abscess: Case Report. J Korean Neurosurg Soc 34: 72-74, 2003
- 10. Saigal G, Donovan Post JD, Kozic D: Thoracic intradural aspergillus abscess formation following epidural steroid injection. AJNR 25A: 642-644, 2004
- 11. Sharma OP, Chwogule R: Many faces of pulmonary aspergillosis. Eur Respir J 12: 705-715, 1998
- Sheth NK, Varkey B, Wagner DK: Spinal cord aspergillus invasioncomplication of an aspergilloma. Am J Clin Pathol 84: 763-769, 1985
- 13. Vinas FC, King PK, Diaz FG: Spinal aspergillus osteomyelitis. Clin Infect Dis 28: 1223-1229, 1999
- 14. Wells WJ, Fox AH, Theodore PR, Ross LA, Stanley P, Starnes VA: Aspergillosis of the posterior mediastinum. Ann Thorac Surg 57: 1240-1243, 1994