

# 『Spinal Aspergillosis of the Thoracic Spine:』 Case Report

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## Abstract:

**OBJECTIVE AND IMPORTANCE:** Fungal infection of the spine is a rare entity, and is usually associated with opportunistic infection in immuno-compromised patients. The authors present a case of spinal aspergillosis in a diabetic patient.

**CLINICAL PRESENTATION:** The subject was a 74-year-old man with a history of left lung tuberculosis who was diagnosed as having diabetes mellitus 20 years ago. His diabetes was controlled by diet and insulin therapy. On admission, he suffered from a moderate fever, paraparesis and sensory impairment of the bilateral lower extremities. A thoracic MRI showed a mass lesion was constricting the dural sac in the T6-9 thoracic spine region.

**INTERVENTION:** A laminectomy was performed and antifungal agents (fluconazole and amphotericin B) were administered, but the treatment was ineffective, with the patient dying of multiple organ failure about three months after the operation.

**CONCLUSION:** The outcome in cases of *Aspergillus* spinal infection is generally poor, in spite of the recent progress of medical treatment or surgical technique, so the importance of early diagnosis and initiation of treatment for this intractable disease are emphasized.

**Key words:** Aspergillosis, Fungal infection, Spondylitis, Thoracic spine

The occurrence of any form of fungal infection in a spinal lesion is rare, usually resulting from an opportunistic infection in immuno-compromised hosts. The most common pathogenic causes are *Candida* or *Aspergillus*. While some good outcomes using an antifungal treatment have been reported, the prognosis of the fungal spinal infection is generally poor. The invasion of the infection into the spinal canal, once established, appears to be prone to spread easily and surgical debridement typically yields a dis-

appointing outcome. We here report a case of spinal aspergillosis in a patient with diabetes mellitus.

## CASE REPORT

### History and Examination

A 74-year-old man with a history of left lung tuberculosis dating from 20 years ago, was diagnosed at the same time as having diabetes mellitus. He was

treated by anti-TB chemotherapy for the former, with the diabetes being controlled by diet and insulin therapy. In March 2001, he experienced a general malaise, loss of appetite and decrease in body weight, consequently presenting at a local hospital, where he was admitted in April. After admission, he exhibited a slight fever and his laboratory data showed a raised erythrocyte sedimentation rate (ESR; 144 mm/hr) and C-reactive protein (CRP; 12.3 mg/dl). His white blood cell count was 9,800 cells/mm<sup>3</sup> and cultures of his blood, urine, sputum and gastric juice showed negative for any infection, suggesting there was no malignancy. Anti-bacterial treatment by ciprofloxacin hydrochloride proved ineffective in reducing his fever. In May, almost one month after his initial admission, he complained of paraparesis and urinary retention. Four days after this, suffering from paresthesia and a numbness in his thigh region, along with an elevation in his paraparesis, he was transferred to our hospital.

At the time of transfer, a neurological examination showed severe paraparesis (1-2 on left, 3-4 on right), deprivation of pain and deep sensation, deep tendon reflex, and an associated deterioration of tactile awareness in the bilateral lower extremities. A thoracic MRI showed an abnormal mass lesion in the T6-9 vertebrae. Hypointensity could be evidenced on T1-weighted images and isointensity was visible on T2-

weighted images (Fig. 1). Gd-DTPA enhanced MRI showed that the enhanced lesion had invaded the spinal canal and was constricting the dural sac.

### Procedure

The patient was treated by a T5-9 laminectomy, based on the diagnosis of vertebritis and an epidural abscess on the thoracic lesion. Degenerative adipose tissue was seen in the epidural cavity and the dural sac was strangulated. Almost all abnormal adipose tissue was totally resected and the dural sac was decompressed by surgical debridement.

### Postprocedural Course

His neurological status improved slightly and muscle power in his left lower extremity recovered to 60% postoperatively. However, some twelve days after the operation, he experienced a sudden onset of paraplegia. In a resultant pathological examination nineteen days after the operation, spinal aspergillosis was diagnosed (Fig. 2) and antifungal treatment (200 mg per day of fluconazole and 25 mg per day of amphotericin B) was initiated. Despite this being, no symptomatic improvement was evidenced. A follow-up MRI showed intradural invasion of the lesion two

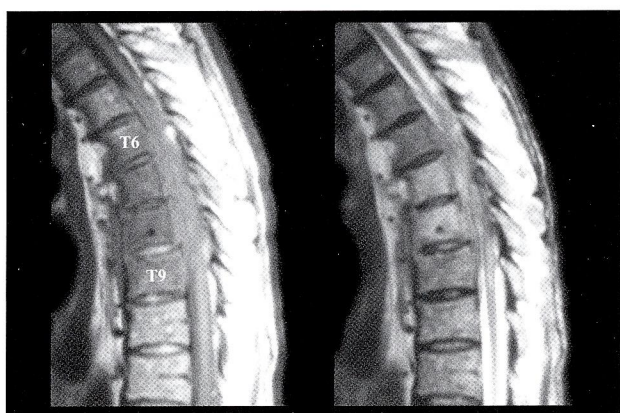


Fig. 1 T1 weighted (*left*) and T2 weighted (*right*) MRI of thoracic spine, revealing the abnormal mass in the vertebrae and epidural space.

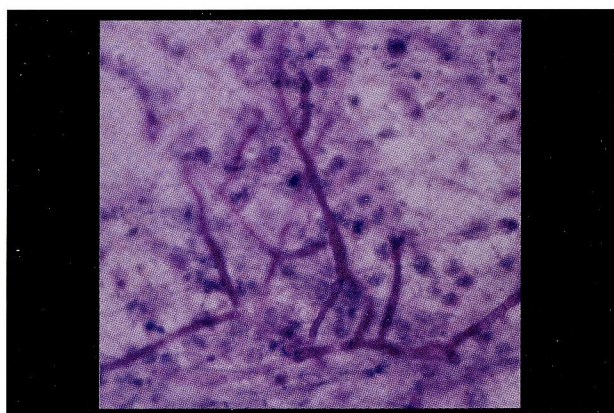


Fig. 2 Photomicrograph revealing the hyphae of aspergillus, PAS stain, original magnification  $\times 400$ .

months after the operation (Fig. 3). This was further complicated by an onset of pneumonia at the same time, and the general deterioration of his condition. Finally, 112 days after the operation, he died of multiple organ failure.



Fig. 3 T1 weighted (*left*) and T2 weighted (*right*) MRI of thoracic spine two months after the operation, revealing intradural invasion of the lesion.

## DISCUSSION

Vertebral osteomyelitis and discitis are commonly due to pyogenic infection, particularly *Staphylococcus aureus*, which accounts for 60% of spinal infections<sup>26</sup>. While the incidence of fungal infection is rare, the rate of occurrence has risen over the last decade as the population of immuno-compromised patients has increased<sup>6</sup>. Spinal infection due to *Aspergillus* is exceedingly rare<sup>16</sup>, and the incidence of aspergilloma in the general population is estimated to be less than 0.01%<sup>11</sup>. Aspergillus disc space infections tend to occur most commonly (80%) in immuno-compromised hosts. This includes a wide range of patients, including those treated by steroids or immuno-suppressive drugs on a long-term basis, those with a malignant tumor, and kidney or heart transplantees. Additionally, other patients at risk typically include those that use antibiotics heavily, intravenous drug users, AIDS patients, alcoholics, people with hepatic cirrhosis, and

those suffering from chronic granulomatous disease<sup>1-3,7,9,10,13,18-22,24</sup>. Because of its rarity and the lack of medical awareness, aspergillus spondylitis is often misdiagnosed as tuberculous spondylitis, especially since the clinical and radiographic findings for the two are similar<sup>4,9,14,17</sup>. Spinal tuberculosis, however, classically begins in the anterior inferior portion of the vertebral body, then spreads beneath the anterior longitudinal ligament to involve the adjacent vertebral body with secondary narrowing of the disc space<sup>17</sup>. In invasive aspergillois, the lesion expands circumferentially and destroys all the surrounding spinal structures, i.e., vertebral bodies, discs, and neural arches, as well as all the contiguous structures, e.g., ribs, thoracic wall, lungs, etc., as seen in our case and other reported cases<sup>12,17</sup>. These two entities must be distinguished, because their chemotherapeutic management differs, especially as there are now reports of successful treatment of aspergillus spondylitis using antifungal agents alone<sup>3,7,14,17</sup>.

Morbidity and mortality in patients with aspergillus spondylitis, however, continue to be very high<sup>4,7,12,17,25</sup>. This is especially so when extensive vertebral destruction and chest wall invasion have occurred, as seen in our case<sup>12,17,25</sup>. Spinal epidural abscesses resulting from *Aspergillus* have a poor prognosis for recovery from paraparesis or paraplegia, even if decompression is performed<sup>9,22</sup>. As a delay in diagnosis leads to less than favorable results in terms of final neural recovery<sup>8</sup>, the importance of early diagnosis cannot be too strongly emphasized<sup>17</sup>. Patients with immuno-suppressive defects or those infected with *Aspergillus fumigatus* generally have a poor prognosis, whereas healthy hosts tend to survive<sup>3,24</sup>.

The most useful laboratory test to confirm the diagnosis involves quantifying the erythrocyte sedimentation rate (ESR)<sup>16</sup>. If the ESR is typically elevated, the WBC is normal<sup>3,9</sup>. This tendency was seen in our case. A decrease in ESR parallels clinical and radiological improvement<sup>16</sup>.

MRI findings also normally tend to show hypointensity of vertebral bodies and intervertebral discs on T1-weighted images. Similarly, hyperintensity of the intervertebral disc on T2-weighted images with an abnormal configuration (ie, absent intranuclear cleft), and hyperintensity of the vertebral endplates at the abnormal disc level on T2-weighted images can be typically seen<sup>15,26</sup>. The absence of hyperintensity within the intervertebral discs on T2-weighted images and the preservation of the intranuclear cleft are findings associated with nonpyogenic or TB spondylitis<sup>23,26</sup>.

Conservative therapy does not seem adequate or appropriate in obtaining good long-term outcome in *Aspergillus* spinal infection cases and additionally carries the risk of more serious systemic complications<sup>13,16</sup>. In contrast to tuberculosis, invasive aspergillosis responds especially poorly to drug therapy<sup>14,17</sup>. Amphotericin B given in doses of 0.5 mg/kg/day and 5-flucytosine given in doses of 60 to 100 mg/kg/day for 6 to 12 weeks are considered to be the standard regimen<sup>3</sup>. Cortet et al. claims successful treatment using non-invasive drug therapy, administering itraconazole either alone or in combination with amphotericin B or 5-flucytosine. The reported recommended dose of itraconazole in the treatment of disc space infection was 3 to 5 mg/kg/day<sup>3</sup>.

Others have reported that even after application of antifungal agents, persistent back pain and a persistently elevated sedimentation rate typically necessitates surgical debridement<sup>16</sup>. Surgical treatment with a posterior approach, laminectomy, and debridement is more common than an anterior approach or a direct lateral extraperitoneal approach<sup>3,9</sup>. Some authors conversely note that anterior decompression and stabilization yields predictably better results than laminectomy in patients with spinal cord compression<sup>5,8</sup>. They argue that with an infection having destroyed the anterior and middle columns of the vertebrae, a laminectomy removing posterior elements will only exacerbate further instability of the spine<sup>5,8</sup>.

## CONCLUSION

The outcome in cases of *Aspergillus* spinal infection is generally poor, in spite of the recent progress of medical treatment or surgical technique, so authors emphasize the importance of early diagnosis and initiation of treatment for this intractable disease.

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