

Case Report: Aspergillus Spondylodiscitis After Spinal Stenosis Surgery: A Case Report



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ABSTRACT

Background and Importance: Spondylodiscitis is an inflammatory disease of the body of one or more vertebrae and intervertebral disc. The fungal etiology of this disease is rare, particularly in patients without immunodeficiency. Delay in diagnosis and treatment of this disease can lead to complications and even death.

Case Presentation: A 63-year-old diabetic female patient, who had a history of spinal surgery and complaining radicular lumbar pain in both lower limbs with a probable diagnosis of spondylodiscitis, underwent partial L2 and complete L3 and L4 corpectomy and fusion. As a result of pathology from tissue biopsy specimen, Aspergillus fungi were observed. There was no evidence of immunodeficiency in the patient. The patient was treated with Itraconazole 100 mg twice a day for two months. Pain, neurological symptom, and laboratory tests improved.

Conclusion: The debridement surgery coupled with antifungal drugs can lead to the best therapeutic results.

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Highlights

- Aspergillus spondylodiscitis is a rare reason of low back pain.
- Immunosuppression, diabetes and pervious spine surgery place patients at high risk for Aspergillus spondylodiscitis.
- Laboratory finding (elevated ESR and CRP) and magnetic imaging resonance finding can help early diagnosis.
- The treatment is antifungal drug such as Amphotericin B and Itraconazole and debridement and fusion, if necessary.

Plain Language Summary

Fungal infections of spine are a rare cause of back pain and in a patient with this complain and history of diabetes and pervious spine surgery, it should be considered. Imaging and some blood tests help us to diagnose but the fungi should be seen in spine tissue sample for a definitive diagnosis. Delay in diagnosis can lead to irreversible damage to the patient. Treatment includes taking antifungal drug and removing infected tissues and stabilizing the spine, if necessary.

1. Background and Importance

Spondylodiscitis is a rare disease, and is responsible for 2-7% of pyogenic osteomyelitis cases. The incidence ranges from 1/10000 per year to 1/250000 per year, and it occurs three times higher in males [1, 2]. The incidence of invasive fungal infection has been increasing since 1980, and it is believed that it is due to an increase in the population of patients with immune system dysfunction [3]. Therefore, the prevalence of Aspergillus – associated vertebral osteomyelitis is increasing in the population of patients with immune system disorders. However, due to nonspecific symptoms, there are still many challenges in diagnosis and its treatment [4].

Aggressive aspergillus rarely infects bony structures, but when occurs, the spine is the most common site of involvement [5]. Spinal fungal infections are relatively uncommon, and it has a prevalence of 0.05%-1.6% [1]. Until 2009, fewer than 50 cases of Aspergillus osteomyelitis have been reported in the literature, in which the majority of patients suffered from immune dysfunction, and only in 10 of them, patients had this type of fungal infection without immune dysfunction [6]. It has been reported that 34% of patients with Aspergillus osteomyelitis were without any predisposing or immunosuppressive factors [7].

Causes of vertebral Aspergillus can be classified into three major categories; trauma-induced infection, spinal surgery or epidural injection, and spread of pleural effusion that affects the thoracic spine. A quarter of Aspergillus osteomyelitis occurs due to spinal procedures [7].

Back pain is the most common symptom and most patients have disc inflammation and osteomyelitis at one or more vertebral levels. According to imaging results, it is difficult to diagnose Aspergillus and bacterial infection [4]. Aspergillus spondylodiscitis may be mistaken for tuberculosis spondylitis [1]. Most physicians recommend surgical treatment if there is progression of symptoms or neurological deficits [1, 8]. Patients who received both surgical and medical treatment showed more acceptable results [1]. The duration of antifungal therapy has not been established. Infectious Diseases Society of America (IDSA) guide line recommends at least 6-8 week antifungal therapy with amphotericin B or voriconazole in immunocompromised patients with Aspergillus spondylodiscitis [1].

2. Case Presentation

A 63-year-old female patient referred to Medical Center of Poursina because of severe low back pain and radiculopathy. She had diabetes for 8 years and was treated with metformin 500 mg twice a day. She had not taken Immunosuppressive drugs and corticosteroids. She had surgery of lumbar canal stenosis in 2017 and 2018. Between the surgeries, she had radicular low back pain. Three months after the second surgery, the patient was admitted again because of severe low back pain and a probable diagnosis of spondylodiscitis.

Physical examination

In the initial study, the patient reported a low back pain score of 7 out of 10 on the VAS score. Vital signs

were stable and without any fever. Clinical examination showed that the left lower extremity muscles force in distal and proximal was 3/5, and in other limbs in distal and proximal 5/5. She had radicular pain and paresthesia in the S1 nerve root pathway. There was no cervical pain and no sphincter dysfunction. Deep tendon reflexes were normal. Huffman, Clonus, and Babinski nerve tests were normal both in right and left sides. The patient had no spasticity and rigidity.

Paraclinical examination

MRI revealed spondylodiscitis at L4 vertebra (Figure 1). The initial CRP and ESR tests were 37 mg/L and 114 mm/h, respectively. Anti-Brucella IgG, Anti-Brucella IgM, Anti-HIV, Anti-HCV and Blood Culture tests were negative.

The patient underwent partial L2 and complete L3 and L4 corpectomy and L2 / L3, L3 / L4 discectomy with anterior retroperitoneal approach and fusion (Figure 2). Sampling from disc and vertebral body was performed for pathology and PCR for detection of fungi and tuberculosis. The debridement and irrigation of vertebrae were performed. On the fourth day after surgery, she was transferred to a cardiac center due to chest pain and ECG changes according to the advice of a cardiologist. Pathologic studies

revealed *Aspergillus* fungi in the specimen (Figure 3), the patient was treated with Itraconazole antifungal drug 100 mg twice a day for two months.

Outcome and Follow-up

In the postoperative follow-up, the patient's pain was reduced from a score of 7 to 2, and the latest test showed that CRP was decreased to 12 mg/l and ESR was also 120 mm/h. Paresthesia and lower extremity forces were also improved.

3. Discussion

The term spondylodiscitis means initial infection of intervertebral disc with a pathogen and secondary infection of vertebral body. The disease is very rare [1, 9]. Studies showed that any age group may be affected, but it is more frequent in the fifth to seventh decades of life [1, 10].

Today, there is an increasing prevalence of spondylodiscitis due to increased life expectancy associated with chronic debilitating diseases, immunosuppressive therapies, increased use of body implants, and a variety of spinal surgery [2, 4].



Figure 1. Magnetic resonance imaging scans of the lumbosacral spine of a 63-year-old woman who presented with low back pain, numbness and weakness of both lower limbs

A. The images show osteomyelitis involving the L4 vertebral bodies and surrounding tissues; B. Sagittal T1-weighted MRI B; C. Sagittal T1-weighted MRI with GAD that shows L4 body enhancement C; sagittal T2-weighted MRI.



Figure 2. Post-operative lumbosacral x-ray (anterior-posterior view)



Aspergillus exists as a safe microorganism in the upper respiratory tract and is rarely invasive. It is common in people with immunodeficiency disorder. Bone fungal infection can be due to (iatrogenic) treatment or direct trauma infection [6]. Contamination during surgery and other procedures in immunocompromised patients is usually caused by air pollution [5].

The *Aspergillus* infection of the vertebral body and intervertebral disc is one of the rare causes of spinal osteomyelitis. These types of infections are mostly limited to patients with predisposing factors such as leukocyte dysfunction, neutropenia-related malignancy, antibiotic and steroid therapy, respiratory *Aspergillus*, and surgical manipulation. Diabetes is known to be a detrimental factor in phagocytic function. Therefore, it may be effective in causing infection and growth of microorganisms [6].

Sethi et al. (2012) reported the largest number of patients without immunodeficiency with *Aspergillus*

spondylomyelitis [11]. D'Agostin et al. reported, in their 7-year prospective study of 81 patients with spondylodiscitis, only 6 of them had fungal infections [2]. In a study by Gabrielli, 310 cases of *Aspergillus* osteomyelitis from 1939-2013 were studied. The results of this study showed that 49% of people had *Aspergillus* osteomyelitis in the spine area. Also, 13% of people in this study had no risk factors for *Aspergillus* osteomyelitis [12].

Early detection of *Aspergillus* spondylodiscitis remains a challenge and needs high suspicion [1]. Definitive diagnosis of *Aspergillus* spondylodiscitis is performed by biopsy. However, biomarkers such as galactomanan and serum beta D-glucan have recently been used to confirm the diagnosis [1]. Nandeesh et al., conducted early detection of *Aspergillus* osteomyelitis by Fine-needle Aspiration (FNA) Cytology. This procedure led to early detection and timely treatment without any life-threatening complication [6].

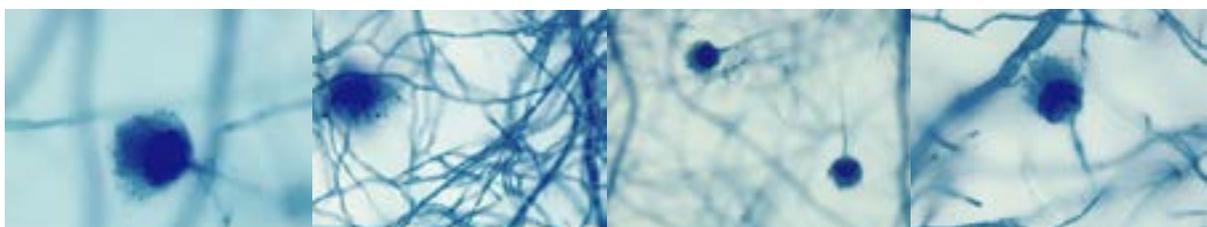


Figure 3. Dissectomy specimen showing *Aspergillus*



When fungal contamination occurs in the body, polymorphonuclear leukocytes play the most important role in removing hyphae [13]. The oxidative process done by phagocytes is required for the release of invading *Aspergillus* [13, 14]. Diabetes is known to be a detrimental factor in phagocytic function. Therefore, it may play a pivotal role in causing infection and making a suitable environment for the growth of microorganisms [6]. The prevalence of neutrophil oxidative dysfunction and phagocyte function in diabetic patients can make them more susceptible to *Aspergillus* [13]. In Gabrielli's study on 310 cases of *Aspergillus* osteomyelitis, 6% were diabetic [12].

In this patient, the only significant predisposing factor is a history of diabetes as well as spinal surgery. Previous reports have indicated that the duration of symptoms until a definitive diagnosis ranged 5-7 months. In this patient, there were nonspecific signs and symptoms such as radicular low back pain and parenthesis. Due to the heart problems that occur in this time, it took 9 months before a definitive diagnosis. Laboratory tests are often less helpful in the diagnosis of *Aspergillus* infection, and the final diagnosis is confirmed by microscopic examination of the fresh tissue specimens and culture in Sabouraud agar medium in which fungi are observed in 74% of the cases [13]. In this patient, a definitive diagnosis was made by observing the fungi in an intraoperative biopsy specimen.

Aspergillosis is highly fatal in patients with immune system dysfunction and amphotericin B should be prescribed intravenously at the maximum tolerable dose [13]. Treatment of *Aspergillus* osteomyelitis is multimodal, which includes improvement of chronic hyperglycemia, surgical intervention, and rapid starting of antifungal medication [14]. Whereas the British Infectious Disease Society recommends the use of amphotericin in combination with Flucytosine in severe cases of *Aspergillus* osteomyelitis, in stable patients Itraconazole has been recommended [7]. The patient was treated with Itraconazole 100 mg twice daily for two months.

4. Conclusion

Fungal Spodylodiscitis is particularly rare in patients with an appropriate immune system. Only a small number of patients without immune system disorders have been reported to date. Delay in diagnosis can lead to life-threatening risks. Diagnosis and treatments for this disease are challenging. It seems that debridement surgery combined with antifungal drugs has the best curable results. This diagnosis should also be considered in patients with chronic low back pain who have risk factors for fungal infections.

Ethical Considerations

Compliance with ethical guidelines

The participant was informed about the purpose of the research and its implementation stages; she was also assured about the confidentiality of her information. Moreover, she was allowed to leave the study whenever she wished, and if desired, the results of the research would be available to her.

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Authors' contributions

Conception and design: Mohsen Aghapoor, Babak Alijani; Data collection: Mahsa Pakseresht-Mogharab; Drafting the article: Mohsen Aghapoor, Mahsa Pakseresht-Mogharab; Critically revising the article, Reviewing the submitted version, and Approving the final version of the manuscript: All authors.

Conflict of interest

The authors declared no conflict of interest.

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