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Case Report

# Concurrence of polyostotic fibrous dysplasia and spinal aspergillus in non-immunocompromised adult patient: Case report

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## Abstract

Aspergillus, a rare agent in spinal infections, is often transmitted via inhalation. It can be traced as an infectious agent in immunocompromised patients. While in non-immunocompromised patients, it is highly unlikely to cause spondylodiscitis. Radiological findings remind tuberculosis. The recommended medical treatment is applied with Itraconazole and Amphotericin B. Surgical indication involves the presence of progressive neurological deficit, instability and biopsy requirement.

Fibrous dysplasia was first reported in 1938 by Lichtenstein and is a benign developmental disorder of the skeletal system with uncertain etiology. Polyostotic type involved more than one bone, while the monostotic type occurs by involving only one bone structure. Spinal involvement may lead to collapse fractures and deformity development and the most common complaint is pain.

In our case, these two disorders occur concurrently, causing bone destruction and severe pain, and no similar cases were found in the literature.

**Keywords:** Fibrous Dysplasia; Spinal Aspergillus Ifection; Non-Immunocompromised.

## INTRODUCTION

Aspergillus, a rare agent in spinal infections, is often transmitted via inhalation. It can be traced as an infectious agent in immunocompromised patients. While in non-immunocompromised patients, it is highly unlikely to cause spondylodiscitis (1). Bone involvement is rare. It is transmitted through direct adjacency from lungs in children, whereas haematogenously in adults. Spinal involvement is seldom and mostly thoracic spine is involved. Radiological findings remind tuberculosis (2).

It is characterized by narrowing disc range, adjacent vertebrae involvement and the presence of paraspinal abscesses. Epidural abscess and associated neurological deficit development is highly likely. The recommended medical treatment is applied with Itraconazole and Amphotericin B. Surgical indication involves the presence of progressive neurological deficit, instability and biopsy requirement (3). It may be located in the thoracic vertebrae and epidural space and cause pressure. By hematogenous spreading or local invasion; it causes spinal involvement(4). Vertebral body involvement may imitate spinal tuberculosis. However, during recovery proliferative changes occur that were not observed in tuberculosis. Due to collapsed and destructed vertebral body; pressure onto the cord and spinal instability may occur. In T1-weighted sequences, loss in disc hyperintensity and intranuclear cleft persistence may be observed. The diagnosis is done via direct biopsy and identifying the agent, and the treatment is applied by surgery and medical treatment (5).

Fibrous dysplasia was first reported in 1938 by Lichtenstein and is a benign developmental disorder of the skeletal system with uncertain etiology (6). In the involved region or regions, the normal bone medulla is replaced by fibroosseous tissue. The disorder is divided into subcategories often based on the number of bones involved. Polyostotic type involved more than one bone, while the monostotic type occurs by involving only one bony structure. When the facial bones are added in this picture together with endocrinological disorder and pigmentation of the skin, the disseminated type of the disorder is called the Mc Cune Albright syndrome. This disorder that can develop at any age is typically observed at the 1st and 2nd decades (7). Fibrous dysplasia constitutes approximately 7% of all benign bone lesions and spinal involvement is quite low and is often observed in monocytic form (8). Spinal involvement may lead to burst fractures and deformity development and the most common complaint is pain. Direct graphies and CT are determinants for

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diagnosis and bone scintigraphy is a guide for differential diagnosis (9). Most commonly, bisphosphonates, primarily pamidronate, are used for treating polyostotic disorder. Several authors reported successful medical treatment with bisphosphonate therapy for fibrous dysplasia patients. (10) In order to surgically treat the bone fractures, stabilization can be performed in the spine via vertebroplasty or transpedicular screw application (11). In our case, these two disorders occur concurrently, causing bone destruction and severe pain, and no similar cases were found in the literature.

## CASE REPORT

The 55-year-old male patient had a history of an operation for L3-L4 lumbar disc herniation causing pain in the lower back and the left leg three months ago. He had decreased pain after the operation; however, he has experienced severe pain in the lower back and both legs for approximately forty five days after the procedure and he was examined by control spinal MR. The patient had a story of cranial left frontal fibrous dysplasia and his spinal radiological examinations revealed polyostotic fibrous dysplasia located in the entire vertebral column, costal surfaces and skull base, loss of height in T10, T12 and L1 corpuses, partial laminectomy on the left at the L3-4 level, and abscess formation at the left peritecal/paraspinal area with spondylitis at this level. (Figure 1).

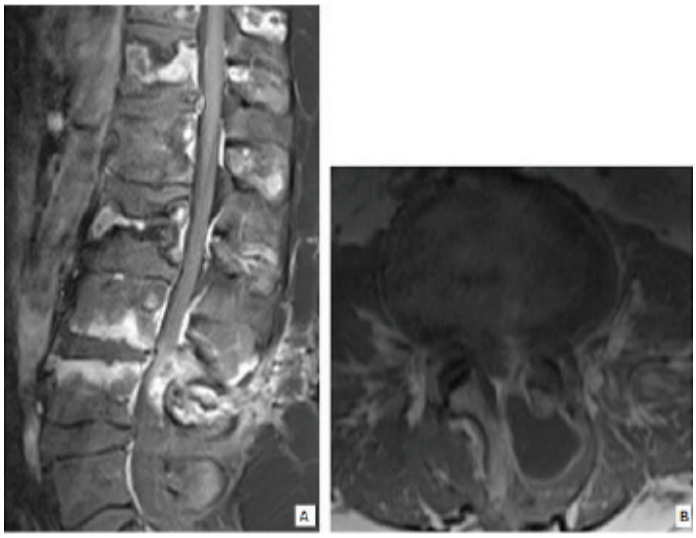

**Figure 1.** T10, T12 and L1 corpuses, partial laminectomy on the left at the L3-4 level, and abscess formation at the left peritecal/paraspinal area with spondylitis at this level.

The patient received an early diagnosis of fibrous dysplasia, spondylodiscitis and paraspinal abscess and his examination revealed swelling in the lumbar region and 3/5 muscular strength in all the bilateral lower extremity muscle groups. The blood tests of the patient revealed WBC (7.8 ; 10^9 / L ), sedimentation (6 mm) , and CRP (0.32 mg/dl) assay results within normal limits. The patient was operated via posterior route and the grayish brown dense pus material in the paraspinal area was sampled and removed. The paraspinal muscles, L3-4 disc space, left lamina and the surrounding bony tissue were cleaned via debridement. In the biopsied material, aspergillus fumigatus was identified. The histopathological diagnosis of the patient was reported to be acute inflammatory cells infiltrating the fibrovascular tissue and necrotic fibrous tissue . (Figure 2). Amphotericin B treatment was started as medical therapy.

The patient received antifungal therapy for 6 months, the motor deficit was abandoned and the complaints of lower back and leg pain decreased. The patient's control spinal MR images showed that the spondylodiscitis was regressed. (Figure 3).

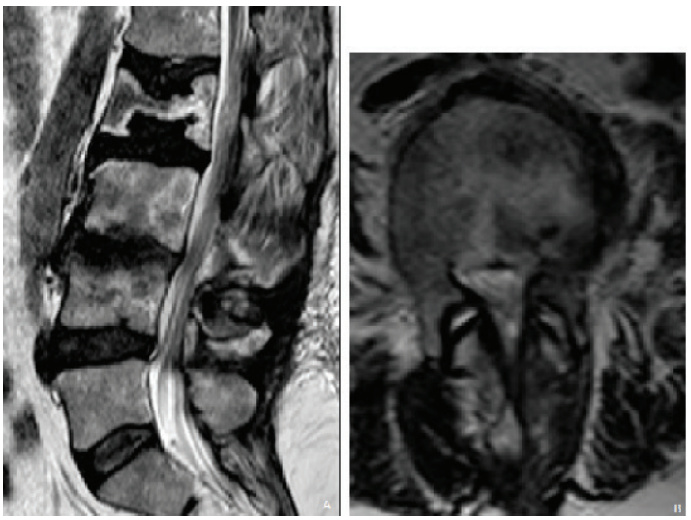

**Figure 2.** Control spinal MR images showed that the spondylodiscitis was regressed

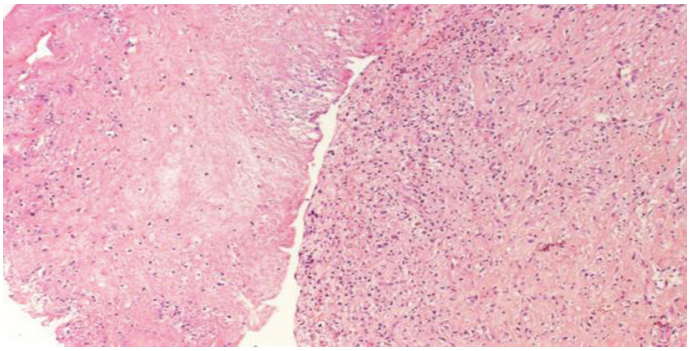

**Figure 3.** Histopathologic examination: Acute inflammatory cells infiltrating the fibrovascular tissue and necrotic fibrous tissue on the left side (H&E,X100)

## DISCUSSIONS

The frequency of fungal infections has risen over the last decade relatively to the increase in the number of immunocompromised patients. While in non-immunocompromised patients, spondylodiscitis caused by fungal infections is often diagnosed late (12). The most common reason for fungal infections is Candida and rarely Aspergillus. These infections are frequently observed in the spine as spondylodiscitis or osteomyelitis (13). Fungal organisms grow slowly and are hard to detect by culture. Thus, histopathological study is important for the verification of the diagnosis (12). Eismont et al. reported fungal organisms as the agent in 61 cases in the

retrospective vertebral infection study they carried out (14). Vinas et al. (15) in their systematic compilation stated that A.fumigatus was the agent in 71% of the 39 vertebral infections caused by Aspergillus types in the literature of 1966-1998. Vinas et al. (15) detected underlying diseases including hematological malignancy, chronic granulamatosis, organ transplantation, corticosteroid use, surgical intervention history, tuberculosis in most of the patients in their study, while no immunosuppressants were reported in three patients as in our case. Fibrous dysplasia and Aspergillus infections may interfere in the clinical diagnosis (16), however in our case there was a co-existence of Aspergillus infection and fibrous dysplasia. In Aspergillus infections, spinal involvement is seldom and radiological findings remind tuberculosis (2).

It is characterized by narrowing disc range, adjacent vertebrae involvement and the presence of paraspinal abscesses. The recommended medical treatment is applied with Itraconazole and Amphotericin B. Surgical indication involves the presence of progressive neurological deficit, spinal instability and biopsy requirement (3). By hematogenous spreading or local invasion, spinal involvement occurs (4).

Vertebral body involvement may imitate spinal tuberculosis. However, during recovery proliferative changes occur that are not observed in tuberculosis. Due to collapsed and destructed vertebral body; pressure onto the cord and spinal instability may occur. Fibrous dysplasia constitutes approximately 7% of all benign bone lesions and spinal involvement is quite low and is often observed in monocytic form (8). Spinal involvement may lead to collapse fractures and deformity development and the most common complaint is pain. Surgical treatment is indicated for clinical pictures including pain that cannot be eliminated by medical treatment, neurological deterioration, vertebral collapse and/or cord compression (11).

As the bone quality is not satisfactory, internal stabilization is recommended to enable long-term stabilization. However, stabilization cannot be applied to every case due to poor bone quality of these patients. Vertebroplasty may be performed as an alternative treatment for patients who develop neurological deficit and who do not get better despite the medical treatment (11). In our case, since clinical recovery was observed with medical treatment following surgical decompression, additional surgical intervention was not made for stabilization.

## CONCLUSION

Development of an infection after spinal surgery is among expected complications. However, infection development with aspergillus has been rarely reported, its concurrence with fibrous dysplasia is interesting. In our case, aspergillus infection, which frequently causes infections in immunocompromised individuals, was observed in a patient with polyostotic fibrosis, which is not caused by immunosuppression and rarely has spinal involvement. The patient developed spondylodiscitis after the lumbar disc herniation operation and that he had two factors that cause destruction in spinal structures concurrently was found significant.

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