

Case Report

A rare etiology of cauda equina syndrome

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Abstract

Fungal infections of the spine are very rare and usually seen in immunocompromised patients. Acute cauda equina syndrome presenting in an immunocompetent patient is usually due to a prolapse of the intervertebral disc. Infective pathology caused by *Mycobacterium tuberculosis* with epidural collection can also have a similar presentation. We present a case of spinal epidural abscess caused by *Aspergillus fumigatus*, presenting as acute cauda equina syndrome. To the best of our knowledge, spinal aspergillosis presenting as cauda equina syndrome in an immunocompetent patient has not been reported before in the English-language based medical literature. Surgical decompression with antifungal treatment with oral itraconazole yielded a good recovery.

Key words: Cauda equina syndrome; fungal epidural abscess; vertebral osteomyelitis; aspergillosis; laminectomy

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Introduction

Vertebral osteomyelitis and epidural abscesses are most commonly caused by pyogenic organisms, particularly *Staphylococcus aureus* [1]. Non-pyogenic origins of such cases may be due to *Mycobacterium tuberculosis* or to a fungus, especially *Candida* or *Aspergillus* [2,3]. Cases of fungal spinal infections are on the rise owing to an increasing number of patients receiving immunosuppressants after organ transplantation, HIV infection, and intravenous drug abuse [2,4]. Spinal aspergillosis presenting as acute cauda equina syndrome in an immunocompetent patient has not been reported before in the English-language based medical literature, to the best of our knowledge. Such cases should be treated with urgent surgical decompression. The drug of choice for invasive aspergillosis has historically been intravenous amphotericin B, which is highly nephrotoxic, but cases have also shown improvement from treatment

with newer anti-fungal drugs such as itraconazole [5-7].

Case report

A 45-year-old male patient presented with weakness of both lower limbs with loss of bladder and bowel control for one day. He had a history of low backache for one month. There was no history of cough, fever, weight loss or close contact with a patient of tuberculosis. There was no history of repeated infections or receiving corticosteroids for a prolonged duration in the recent past. The general physical examination did not reveal any abnormality. There was no localized spinal tenderness, swelling, or discharging sinus in the back. Straight leg raising tests on both sides were positive at 60 degrees. The extensors and flexors of the toes on both sides had Medical Research Council (MRC) grade 3/5 power (*i.e.*, the patient was able to perform the action against gravity but not against resistance). Sensations were decreased below the L5 dermatome on both

Figures 1 and 2. T1 and T2 weighted sagittal and axial MRI scans, respectively, showing a large multiloculated extradural abscess extending from L3 to S2 level (indicated by arrows). Altered signal intensity is seen in the L3, L4 and L5 vertebral bodies.

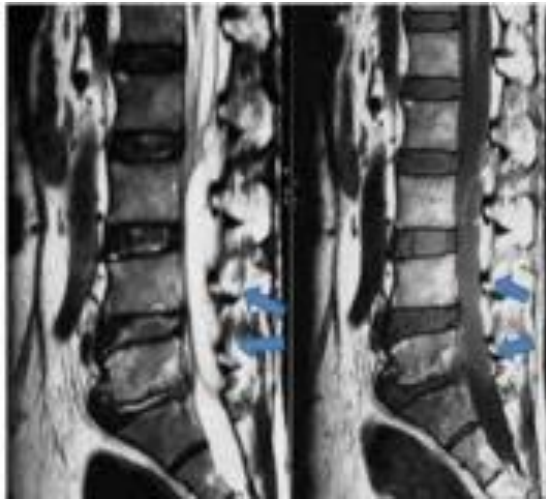


Figure 1

sides with perianal anesthesia and the ankle jerk was absent on both sides. A clinical diagnosis of cauda equina syndrome was made. The routine lab investigations were within normal limits except the erythrocyte sedimentation rate, which was raised (65 mm after one hour by Wintrobe's method). Radiographs of the lumbo-sacral spine showed degenerative changes with reduction of disc space at the L5-S1 level. An MRI of the lumbo-sacral spine showed a multilocular extradural collection extending from the L3 to the S1 vertebra, with altered signal in the body of the L3-L5 vertebrae (Figures 1 and 2). An ELISA test for HIV I and II antibodies was negative.

Urgent surgical decompression was planned and a laminectomy was performed at the L3-L5 levels. The cauda equina and dura were found to be covered with thick grayish granulation tissue without any pus collection. The granulation tissue was removed by decompressing the cauda equina and sent for culture and histopathological examination. The histology showed the presence of hyphae, indicating a fungal infection with absence of granulomas. The culture showed growth of *Aspergillus fumigatus*. CT scans of the chest, skull, and paranasal sinuses were also conducted but did not show any abnormality. Upon further work-up, serum complement levels, immunoglobulin levels and Hb electrophoresis yielded normal values.

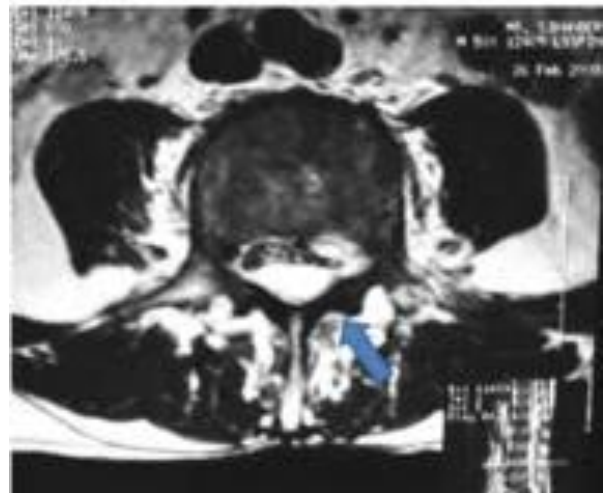


Figure 2

The patient was started on oral itraconazole at 200 mg twice a day. Complete motor and sensory recovery was seen within a week of surgical decompression. The patient regained bladder sensations and voluntary control after three weeks of surgical decompression; however, a detailed urodynamic study was not performed. The drug was continued for a period of three months. At the 36-month follow up, the patient was asymptomatic.

The patient was informed that the data concerning the case would be submitted for publication, and written, informed consent authorizing radiologic examination and photographic documentation was taken.

Discussion

Cauda equina syndrome is a serious neurological condition in which there is an acute loss of function of the neurological elements of the spinal canal below the termination of the spinal cord. Cauda equina syndrome of sudden onset is regarded as a medical emergency. Surgical decompression by means of laminectomy or other approaches may be undertaken on an urgent basis if a compressive lesion, e.g. prolapsed disc, epidural abscess, tumor or hematoma is demonstrated [8,9].

A prolapsed intervertebral disc is a common cause of cauda equina syndrome wherein the patient presents with history of backache and sudden onset of weakness with bladder and bowel involvement [8]. Infective pathology with epidural collection as a cause of sudden onset cauda equina syndrome is

uncommon, especially when the patient does not have any history of fever, weight loss or features of an immunocompromised status [9]. Spinal tuberculosis is a common cause of epidural abscess in a country such as India where tuberculosis is highly prevalent [10]. Fungal infection presenting with epidural collection can also be considered as a rare possibility. It is difficult to distinguish between fungal infection and tuberculosis both clinically and radiologically [4,10]; hence a laboratory diagnosis is important to start appropriate therapy [11].

Treatment of fungal spondylitis is often delayed because of difficulty with diagnosis, as fungal organisms are slow growing and are difficult to identify by culture. A delay in the diagnosis leads to poorer results in terms of neurological recovery [12].

A few cases of aspergillosis of the spine have been reported in the literature. *Aspergillus* commonly invades an immunocompromised host, which may be due to long-term steroid or immunosuppressive therapy, malignancy, organ transplantation, widespread use of antibiotics, intravenous drug abuse, acquired immunodeficiency syndrome or chronic granulomatous disease [11-14]. Occasionally, *Aspergillus* invades an immunologically competent individual, in whom the prognosis is better [11]. Stratov *et al.* reviewed the English-language based literature between 1966 and 2001 and identified 41 cases of osteomyelitis in immunocompetent hosts [15], whereas only five cases of *Aspergillus* vertebral osteomyelitis with an extradural abscess in immunocompetent patients were reported up to 2004 and none of them presented with acute cauda equina syndrome [16]. Extradural (epidural) aspergillosis is an uncommon manifestation of CNS aspergillosis. It arises usually from extension into the epidural space from a vertebral abscess [17].

Management of such cases is a challenge. Although most treatments are based on individual case reports and brief case series, surgical drainage with systemic antifungal therapy is considered to be standard practice. Historically, amphotericin B (AMB) has been used because of its long-standing availability, but its effectiveness has been questioned when not combined with surgery. This could be explained by the low concentrations AMB attains in the bone [18]. On the other hand, numerous studies including a large, randomized controlled trial [19] have demonstrated the superiority of voriconazole over AMB in the treatment of invasive aspergillosis with improved survival and lower toxicity. Therefore, according to the Infectious Diseases Society of

America guidelines on the treatment of aspergillosis released in January 2008, voriconazole is recommended as the primary treatment of invasive aspergillosis, including CNS aspergillosis and osteomyelitis, with a recommendation for a strength of B-II for the latter [20]. Itraconazole, although successfully used in some case-reports, is recommended as an alternative therapy of invasive aspergillosis for refractory cases or cases intolerant to routine antifungal therapy.

Different therapeutic modalities have been proposed for the treatment of spinal aspergillus osteomyelitis. Although it has been primarily treated medically, certain cases may require surgical intervention. It has been suggested that, when the only symptom is back pain without significant instability or neural compression, medical treatment alone is sufficient. When spinal instability or symptoms of spinal cord or radicular compression are present, surgical decompression is indicated [21].

Our case presented with acute cauda equina syndrome and urgent decompression was required to prevent irreversible damage to the neural elements. Amphotericin B has been used for the treatment of spinal aspergillosis in most reported cases. It has to be given by the intravenous route and is reported to have very high nephrotoxicity. Itraconazole is a newer drug which is given orally and is well tolerated. A few authors have reported good results with the use of itraconazole in spinal aspergillosis [5-7,21]. The patient presented here was also treated by oral itraconazole and responded well without any signs of toxicity.

Conclusion

Fungal spinal infection should be considered as a differential diagnosis in infective cauda equina syndrome. Granulation tissue/pus should be sent for histopathological diagnosis in every such case as spinal aspergillosis can rarely present as acute cauda equina syndrome. Urgent surgical decompression with antifungal medication should be the treatment in such cases.

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