

Case Report

Fungal Infection of Pelvis : an Uncommon Cause for Low Backache

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ABSTRACT

This study reports a case of 43 years male with low back ache which on investigation proved to be due to cryptococcus fungal infection of ilium which is an uncommon cause of low backache. The focus of infection was evacuated, and the bone debrided along with administration of anti-fungal drugs. On follow up of 9 months after surgery and antifungal treatment, the patient had no symptoms and there was regression in size of the lesion in the left iliac bone.

Key Words: Cryptococcus, Fungal infection, Low back ache,

INTRODUCTION

Cryptococcus is a disseminated infection caused by *Cryptococcus neoformans*. *Cryptococcus* is an encapsulated yeast like fungus with common reservoirs being soil and pigeon feces.¹ Although nearly any organ can be affected, the most affected organs by this infection are the lungs and central nervous system. Cryptococcal osteomyelitis is rare, having an incidence of 5-10 % in patients with systemic Cryptococcosis. Isolated Cryptococcal skeletal infection is rare but can lead to significant morbidity and mortality if left untreated.² We present here a rare case of isolated cryptococcal infection of the iliac bone in an immunocompetent host.

CASE REPORT

A 43-year-old male, businessman by profession, presented to us with low back ache for 4 years which was radiating to the left hip and thigh. There were no constitutional symptoms and no history of trauma. Patient did not have any co-morbidities as well as significant travel history. The patient had visited multiple centers in the last 4 years for management of low back ache. All blood investigations including multiple X-rays and MRI L/S spine were normal. He got temporary relief with analgesics and physiotherapy but for 3 months prior to presentation, there was an increase in the severity of pain. An USG abdomen, CT scan abdomen and MRI pelvis were done. USG abdomen showed an oval, lobulated, heterogenous lesion with solid component and septations in left iliac fossa that signified either a neoplastic or infective pathology.

CT scan abdomen showed focal expansile lytic lesion with sclerotic margin in the left iliac blade. MRI pelvis showed

destructive lesion in the left iliac bone (32 x 24 mm) with large exophytic tissue component causing infiltration into the overlying iliac muscle and the finding was suggestive of a neoplastic lesion.

The patient referred to us for the management of the neoplastic lesion. On examination, there was mild tenderness around the left iliac fossa and PSIS, and the terminal flexion of left hip was painful. All other systemic and local examinations were normal. We repeated the MRI pelvis, which showed complex lytic lesion in left iliac bone along with large cystic area under the left iliac muscle in continuity with the intraosseous component displaying restricted diffusion and findings suggestive of abscess with osteomyelitis (Figure 1 and 2).



Figure 1. Representative coronal section of MRI demonstrating a cystic lesion underneath the iliac muscle.

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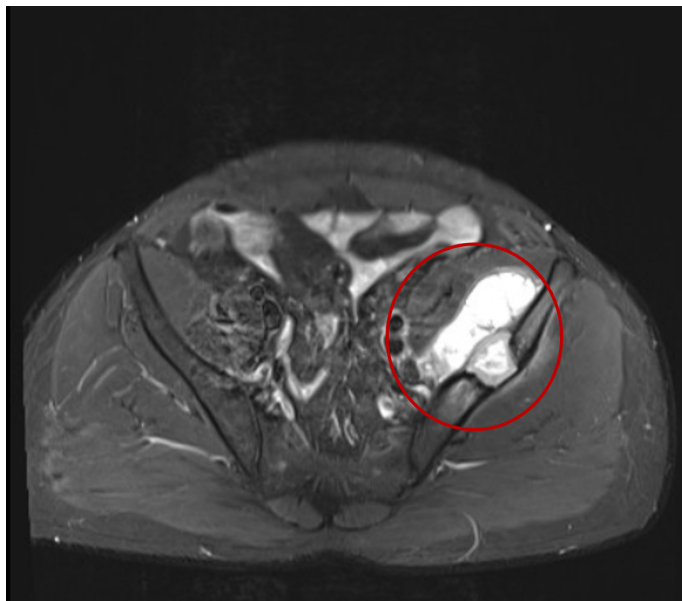


Figure 2. Axial section demonstrating lytic lesion in left iliac bone along with large cystic area under the left iliac muscle in continuity with the intraosseous component.

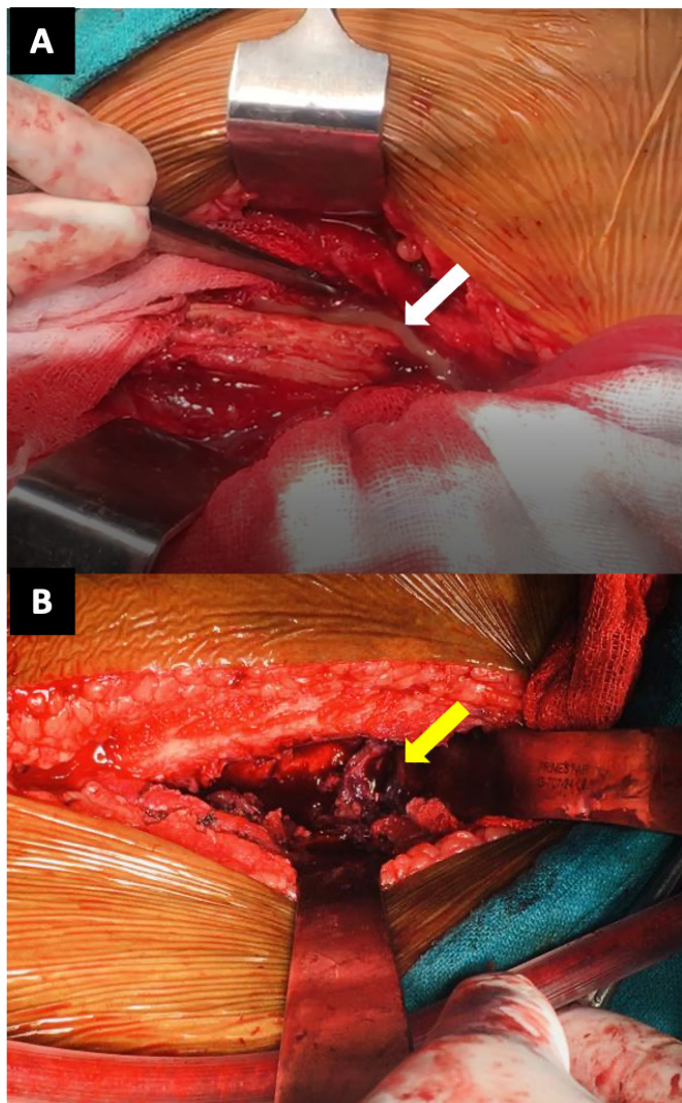


Figure 3. A. Intraoperative pictures showing pus (white arrow). B. Necrotic tissue and osteomyelitis of iliac bone.

An USG guided biopsy was done. No pyogenic bacteria were isolated in pus culture whereas granulation tissue with deep fungal infection was seen in histopathology which was suggestive of cryptococcus infection.

We consulted our physician to rule out any systemic involvement and started antifungal (IV Amphotericin 50mg x OD) for 5 days before going for surgical exploration. Intraoperatively we found pus and necrotic tissue.

Evacuation of pus and debridement of the lesion was done to decrease the fungal load. Pus was sent for culture and tissue for biopsy (Figure 3).

There was no growth in pus culture but findings suggestive of Cryptococcus was seen in histopathology. Fungal culture from the pus showed no growth.

Patient's symptoms improved postoperatively and he was discharged with Tab Voriconazole x 200mg x BD x 9 months. He had no symptoms on 9 months follow up and the MRI shows regression of size of the lesion of left iliac bone and significant decrease in collection in left iliac muscle (Figure 4).

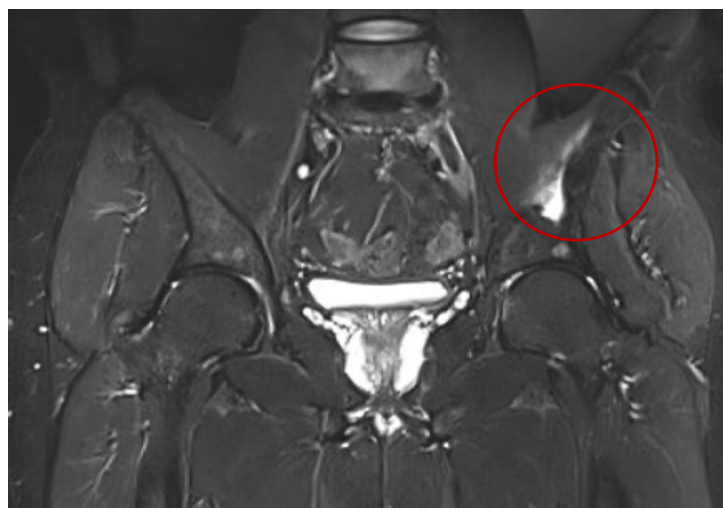


Figure 4. Representative section of MRI at 9 months follow up demonstrating significant regression of lesion.

DISCUSSION

Cryptococcus is usually a disseminated infection caused by *Cryptococcus neoformans*. *Cryptococcus* is an encapsulated yeast like fungus with common reservoirs being soil and pigeon feces.¹ The earliest reported case of human bone disease due to *C. neoformans* was in 1894 by Busse and Buchke, who isolated the organism from a tumor like lesion in tibia.³

Cryptococcal skeletal infection was first reviewed in the literature by Collins in 1950. He reported skeletal involvement in 10% of patients with systemic cryptococcal disease but isolated skeletal entity was rare.⁴

Cryptococcal skeletal infections although rare, can lead to significant morbidity and mortality if left untreated. In a review of

40 patients with skeletal infections caused by *Cryptococcus*, the vertebrae were the most common site involved, followed by femur, tibia and ribs. In this review, only 5 patients had the involvement of ilium.¹ Isolated skeletal cryptococcosis is rare but may occur in healthy individuals after daily exposure to the organism. The development of cryptococcosis was associated with the interaction of fungal agent and the host immunity.⁵ There are essential two types of cryptococcal infections, but the manifestation depends on the host response rather than the strain of the organism.⁶

Cryptococcal skeletal infection is rarely diagnosed at the time of presentation. Infection in immunocompetent individuals is usually mild or subclinical; however overt infection has been described in 10-40% of cases.⁷ In this case, the patient was initially treated for back ache for 4 years without any constitutional symptoms.

Involvement of bone typically manifests as lytic bone lesion with irregular but discrete margins and an absence of periosteal reaction.⁸ The lack of periosteal reaction can be mistaken for noninfectious etiologies such as neoplastic lesion. Histopathology can also establish the diagnosis, but definitive diagnosis is established by culture of the fungus from infected material.⁹ Since in our case the fungal culture was negative, but histopathology finding was suggestive of *cryptococcus*, we treated this patient for fungal infection.

There is no standardized treatment protocol for Cryptococcal infection of specific body sites, except for lung and CNS infection.

The Infectious Disease Society of America (IDSA) 2010 guidelines recommend that treatment of non-meningeal, non-pulmonary cryptococcal infection limited to a single site can be treated with oral fluconazole 400 mg daily for 6-12 months in a immunocompetent host. Treatment similar to that for central nervous system infection is recommended in patients with cryptococemia with involvement of 2 contiguous sites.¹⁰

It is generally felt that most patients with skeletal cryptococcosis warrant some form of systemic therapy and the surgery is important to decrease the fungal burden.¹¹

We treated this case with IV antifungal therapy preoperatively, surgical debridement of the lesion and oral antifungal drugs for 9 months.

CONCLUSION

Intrapelvic pathology should be ruled out as one of the causes of low back ache. Cryptococcal infection should be considered as a differential diagnosis in osteolytic lesions without periosteal reaction in both immunocompromised and immunocompetent individuals. Surgical debridement along with systemic antifungal therapy can be useful in treatment of isolated cryptococcal iliac infection.

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