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Cryptococcal Ilium Osteomyelitis and Pelvic Abscess in an Immunocompetent Host

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Abstract

Osteomyelitis and abscess due to *Cryptococcus neoformans* is an unusual presentation of cryptococcal illness. Infection with this organism is rare in immunocompetent hosts. We describe the case of a young female with cryptococcal ilium osteomyelitis and associated pelvic abscess.

Keywords: *Cryptococcus neoformans*; Osteomyelitis; Ilium; Pelvic abscess

Introduction

Cryptococcus neoformans is an encapsulated fungus found in soil contaminated with pigeon excreta. Although nearly any organ system can be affected, pulmonary and central nervous system infections are by far the most common. We report the case of an 18-year-old female with no known immunodeficient condition with cryptococcal ilium osteomyelitis and pelvic abscess.

Case Presentation

An 18-year-old African American female with a history of iron deficiency anemia, presented to our institution with a history of left hip pain that had been ongoing for 1 year. The pain was described as dull and not precipitated by any trauma. Her pain had gradually worsened since the initial onset. No other constitutional symptoms such as fever, chills, night sweats or weight loss were reported. She denied any associated hip erythema, induration or drainage. She had several emergency room visits previously for the complaint with no clear diagnosis for her symptoms.

Prior to her admission to our institution, she was seen at an emergency room at another hospital. Computed tomography of the pelvis done there revealed an ill-defined lucency throughout the central portion of the left iliac wing with multiple areas of cortical irregularity and periosteal elevation. A large abscess measuring 15.9 x 6.8 x 5.6 cm was noted in the left iliopsoas musculature (Figure 1). Reactive bulky left pelvic sidewall and inguinal lymphadenopathy was noted. She was discharged to the follow up at the orthopedic clinic where aspiration of the area was performed and yielded 400 cc of pus like fluid. She was admitted to our facility for surgical drainage, debridement and initiation of intravenous antimicrobials.

Upon admission, patient was a febrile and white blood cell count was 7.3×10^6 cells/L with 80% neutrophils. C reactive protein was elevated at 135mg/L, ESR was 122mm/hr and chest x-ray was normal.

The patient underwent debridement and irrigation of the left ilium and drainage of the abscess. Approximately 600 cc of purulence was suctioned from the abscess and erosive changes were noted in the ilium. She underwent repeat debridement and irrigation of the ilium 3 days later.

Histopathology of the surgical material revealed granulomatous inflammation, and yeast-like organisms on direct examination consistent with *Cryptococcus*. This was confirmed by gomori methamine silver (GMS) and mucicarmine stains (Figure 2). Material from the initial aspiration and that obtained from the surgery grew *Cryptococcus neoformans* from both fungal and routine aerobic culture. Serum cryptococcal antigen using latex agglutination test was positive with a value of 1:64.

Further investigations for disseminated disease included a lumbar puncture, which was negative for cryptococcal antigen and fungal culture. CT abdomen and pelvis revealed prominent mesenteric and small periaortic retroperitoneal lymphadenopathy and splenomegaly. CT brain and CT chest were unremarkable. Blood cultures were negative.

A search for any underlying predisposing condition was done. HIV serotesting was negative,

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Figure 1: CT pelvis with osteolytic lesion in left iliac wing and large pelvic abscess.

quantitative immunoglobulins did not demonstrate any deficiencies, CD4 count was normal, quantiferon gold testing for tuberculosis was negative and angiotensin converting enzyme was within normal range.

The patient was started on intravenous liposomal amphotericin B at 3mg/kg and flucytosine 25mg/kg orally every 6hrs. Due to severe gastrointestinal distress, the flucytosine was discontinued after 1 day and this was substituted with fluconazole 800mg orally daily. After 14 days treatment was changed to monotherapy with fluconazole 400mg daily which was continued for 2 months. The dose of fluconazole was changed to 200mg with a planned duration of one year. On subsequent clinic visits, patient was clinically doing well with resolution of pain and normalization of inflammatory markers. Patient was lost to follow up after 4 months.

Discussion

Cryptococcal neoformans is an encapsulated fungus that has a wide geographic distribution and is typically found in soil-frequented pigeons. The portal of entry is the lung from where it can disseminate systemically, most commonly to the central nervous system; however direct inoculation is also possible [1]. It is prudent to rule out any possible immunodeficiency in patients with cryptococcal infection. Conditions that increase risk for cryptococcosis include HIV infection, isolated CD4 lymphocytopenia, malignancies, stem cell and solid organ transplantation, cirrhosis, renal failure, chronic lung disease, diabetes, sarcoidosis, and treatment with glucocorticoids or tumor necrosis factor-alpha inhibitors [2]. Infection in immunocompetent individuals is usually mild or subclinical; however overt infection has been described in 10-40% of cases involving non-HIV associated disease [2].

Cryptococcal osteomyelitis typically follows hematogenous seeding from other infected sites. Involvement of bone has been reported in 5-10% of cases as part of a systemic infection and typically manifests as lytic bone lesions with irregular but discrete margins and an absence of periosteal reaction [3]. The lack of periosteal reaction can be mistaken for noninfectious etiologies such as neoplastic lesions. In our case, there was some periosteal elevation noted on imaging. Patients typically present with swelling and pain of the surrounding soft tissue. Usually there is no erythema but some local warmth may be present. Sinus tract formation is rare [4]. 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 rib [5]. In this review, only 5 of the patients had involvement of the ilium. Approximately 25% of patients had multifocal skeletal lesions and the median duration of symptoms before diagnosis was 3

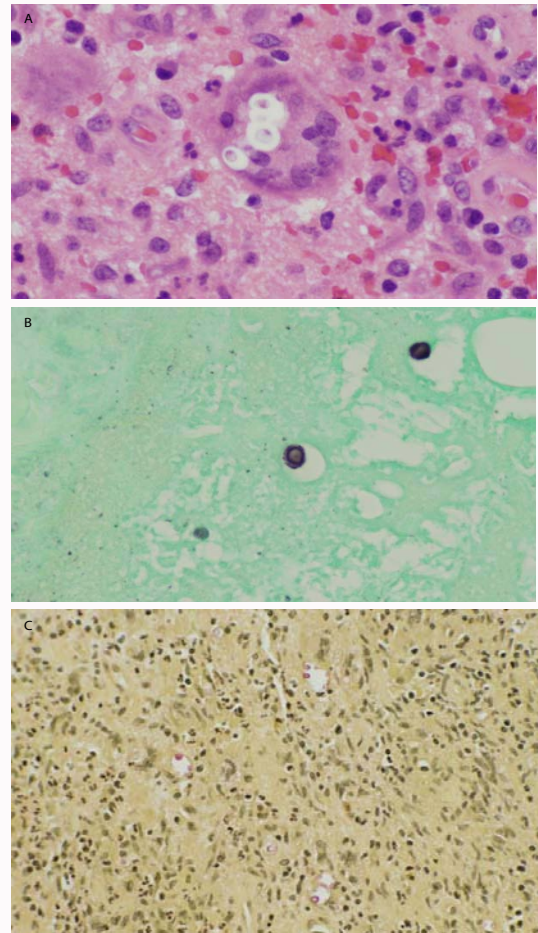


Figure 2: (A): H&E stain; (B): GMS stain; (C): Mucicarmine Stain.

months. Underlying immunosuppressive conditions were present in about 50% of the cases.

Definitive diagnosis is established by culture of the fungus from infected material. Histopathology can also establish the diagnosis. The organism appears as spherical, irregular in size measuring 5-20 micrometers in diameter and appears as narrow-based budding yeast. The cells typically exhibit a thick polysaccharide capsule which is represented by an empty space surrounding the organism [6]. Several stains can be used to aid in the identification of the fungus including Wright's stain, GMS, Calcoflour White, Alician blue, mucicarmine, Fontana Mason and India ink. Cryptococcal antigen positivity can also be useful in diagnosis.

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 400mg daily for 6-12 months in an immunocompetent host [7]. Treatment similar to that for central nervous system infection is recommended in patients with cryptococemia and involvement of 2 contiguous sites or a high fungal burden based on an elevated cryptococcal antigen of $\geq 1:512$. In this situation, the recommended treatment is Amphotericin B 0.7-1 mg/kg IV daily and oral flucytosine 100mg/kg per day in 4 divided doses for 2-4 weeks as induction therapy followed by consolidation therapy with fluconazole at a dose of 400mg daily for 8 weeks followed by fluconazole 200mg daily for 6-12 months for maintenance treatment.

The distinction between disseminated and local disease is often quite difficult since the majority of skeletal cryptococcal infections likely result from hematogenous spread from the lungs. 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 and to prevent leptomeningeal involvement [8,9]. Our patient was treated with the treatment recommended for CNS infection. Since our patient was intolerant of flucytosine, this was substituted with high dose of fluconazole at 800mg daily for the induction period. In our case, aggressive surgical debridement in conjunction with concurrent antifungal chemotherapy was utilized. In a case series of 40 patients with cryptococcal osteomyelitis, those who underwent both surgical and medical treatment had better outcomes compared to those patients who received medical treatment alone [5].

Outcome of cryptococcal osteomyelitis in HIV negative patients is favorable in treated patients [10]. In the review by Liu, the majority of patients were cured of infection with appropriate treatment [5].

In summary, we present a case of a young immunocompetent female with cryptococcal osteomyelitis of the ileum and a large pelvic abscess. Although osteomyelitis due to this organism is rare, involvement of the ilium with an associated pelvic abscess is even more unusual. Cryptococcal osteomyelitis may lead to significant morbidity and mortality, it should be considered as a differential diagnosis in osteolytic osseous lesions in both immunocompetent and immunocompromised individuals.

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