

Muscular Mucormycosis in a Child with Leukemia: Findings of MR, US, and Plain Radiography

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= Abstract =

We present the MR, ultrasound, and plain radiographic findings of a muscular mucormycosis, a rare disease but increasing in frequency as a complication of immunosuppressive treatment, in a child with leukemia. Rim-enhancement and hemorrhagic nature of the muscle were demonstrated on MR. The image findings of MR and ultrasound would be helpful in narrowing the differential diagnosis and follow-up examination of the disease.

Key Words: Mucormycosis, Leukemia, MR, Ultrasound

Introduction

Mucormycosis is an uncommon infection in children. This disease usually develops in patients with predisposing conditions such as diabetes mellitus, leukemia, organ transplantation and chemotherapy. The most common manifestations of this infection are rhinocerebral and pulmonary mucormycosis. To our knowledge, image

findings of isolated muscular mucormycosis in children have never been described in the literature. We presented a case of isolated muscular mucormycosis in a child with leukemia that was accurately depicted by MR, ultrasound, and plain radiography, and compared the image and surgical findings.

Case Report

A five-year-old boy underwent chemotherapy for biphenotype acute leukemia during which he complained of a dull pain in the left side of the distal thigh. Physical examination revealed a temperature of 39°C and mild swelling in the lateral aspect of the left thigh, but no local heatness. The blood cultures were all negative for bacteria and fungus.

Plain radiography was unremarkable at the time of presentation. Initial ultrasound demonstrated an ill-defined hypoechoic lesion in the deep muscle layer of the thigh. Follow-up ultrasound after 3 days revealed increased echogenicity of the lesion with a central low echoic area (Figure 1). MR showed the lesion to be

hyperintense on both T1- and T2- weighted images. The central area was isointense on T1-weighted image (Figure 2a) and hyperintense on T2-weighted image. There was peripheral rim-enhancement after Gd-DTPA (Figure 2b).

Illustrations



Figure 1. Longitudinal ultrasound of the distal thigh demonstrates a round hyperechoic lesion (white arrows) with a central hypoechoic area (arrowheads) in the deep muscle layer.



Figure 2a-b. MR images of muscular mucormycosis, a Sagittal T1-weighted image reveals high signal intensity of the lesion (arrows) with a central isointense area (arrowheads) in the vastus intermedius muscle.



b Contrast-enhanced coronal T1-weighted image reveals rim-enhancement of the lesion (arrows).

The patient underwent ultrasound guided biopsy. The pathologic specimens confirmed the presence of hyphae that were considered to be characteristic for Mucorales. The patient was treated with high-dose amphotericin B. Follow-up ultrasound (11 days after the initial examination) showed thickening of the underlying bony cortex with no significant change in the size of the lesion. Subsequent plain radiography also demonstrated solid, thick periosteal new bone formation without any cortical or medullary destruction.

Surgical debridement was performed. The vastus intermedius muscle was found to be swollen and a brownish, ovoid ball, measuring 10×8×5 mm was noted in the central portion of the lesion (Figure 3). The underlying periosteum was thick with findings of periostitis.

Microscopic examination of the excised vastus muscle revealed the central ovoid area of muscle necrosis containing numerous branching hyphae and entirely necrotic skeletal muscle fibers. The surrounding muscle was severely infiltrated by acute and chronic inflammatory cells and revealed areas of hemorrhagic necrosis.

Following surgery, muscle swelling and tenderness were resolved. There was no evidence of a new, additional soft tissue lesion on follow-up ultrasound with subsequent resolution of the periosteal bone formation. The patient was successfully treated with chemotherapy following completion of induction and consolidation therapy.

Discussion

Mucormycosis is one of the most aggressive and lethal of the Mucorales order infections, and death occurred in over half of the reported cases in children (Kline, 1985). Prognosis is directly related to early recognition, aggressive surgical debridement, treatment with systemic amphotericin B, and medical management of the underlying disease (Kline, 1985). An increase in the incidence of mucormycosis has been seen concomitant with an increase in cancer chemotherapy, organ transplant and immunosuppressive treatment, renal failure, and prolonged postoperative courses (Marchevsky & Bottone, 1980). Therefore, early diagnosis of mucormycosis is important considering the management of the disease and other possible complications of underlying disease.

Mucormycosis may be subdivided into several distinct clinical forms, depending on the site of involvement: (1) rhinocerebral; (2) pulmonary; (3) gastrointestinal; (4) cutaneous; or (5) disseminated (Kline, 1985). Regardless of the organ or tissue involved, the pathologic hallmark of mucormycosis is invasion of blood vessel walls, thrombus formation, infarction of surrounding tissue and production of black, necrotic debris (Marchevsky & Bottone, 1980).

The MR appearances in our case reflect the underlying pathology of the disease. The thick peripheral high signal intensity lesion on both T1 and T2-weighted images was attributed to hemorrhagic myositis by the angioinvasion of this patho-

gen. The central, non-enhancing lesion was corresponded to the necrotic debris. These findings may represent a combination of inflammation and infarction due to vascular invasion by fungal elements. These findings were thought to correspond to the rim of hyperintense lesion on both T1 and proton density-weighted images of rhinocerebral mucormycosis described by Press (Press *et al*, 1988).

Although ultrasound revealed nonspecific findings, soft tissue changes and the periosteal bone formation were well correlated with both MR and plain radiography. The follow-up ultrasound examinations were helpful to excluding muscle hematoma which was supposed to be changing in the size and echogenicity with time.

Frank bony destruction with a moth-eaten pattern, but no soft tissue swelling or periosteal new bone formation, has been described as a plain radiographic finding of a mucormycosis osteomyelitis by Moore (Moore *et al*, 1978). In our case, plain radiography demonstrated a benign nature of a solid, thick periosteal reaction due to overlying soft tissue infection.

The main differential diagnoses in this clinical setting include infection with other bacteria or fungi. The presence of hemorrhagic nature is an unusual finding for bacterial infection. Other entities that must be considered in the differential diagnosis are chloroma and hematoma in children with acute leukemia. Chloroma has been previously well documented by

MR images, with the findings of an iso-intense mass on both T1 and T2-weighted images and diffuse, homogeneous contrast enhancement (Pui *et al*, 1994). Subacute hematoma into the muscle cannot be excluded on MR, but follow-up ultrasound examination would be helpful in excluding muscle hematoma.

In summary, although the MR findings of muscular mucormycosis were nonspecific, MR would be helpful in narrowing the differential diagnosis with other complications of the disease. Ultrasound would be useful for follow-up examination during the course and management of the disease.

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