Case Report

Subcutaneous Mucormycosis in an Immunocompetent Patient

Umesh Kulkarni[1], Sonali Katole[1], Seema Kulkarni[2], Anand Pathak[3], Anand Dank[1]

[1]Department of General Surgery, R.K.Damani Medical College, Chhatrapati Sambhaji Nagar, Maharashtra, India
[2]Department of Microbiology, R.K.Damani Medical College, Chhatrapati Sambhaji Nagar, Maharashtra, India
[3]Department of Medicine, Infectious Diseases, Dr Hedgewar Hospital, Chhatrapati Sambhaji Nagar Maharashtra, India

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Corresponding Author: ***Sonali Katole*** Email: skatole24@gmail.com

ABSTRACT

Mucormycosis is a rare but rapidly progressive fungal infection predominantly affecting immunocompromised individuals. Subcutaneous mucormycosis in an immunocompetent host is extremely uncommon and diagnostically challenging. This case presents previously healthy patient with a rapidly progressing cellulitis over the anterior region of right thigh, debridement revealed necrotic tissue suspicious of fungal infection, consecutive KOH microscopy confirmed Mucormycosis. Patient was managed successfully with serial surgical debridement and systemic antifungal therapy with Isavuconazole, and ultimately split-thickness skin grafting. This case emphasizes the importance of considering Mucormycosis even in immunocompetent individuals with aggressive soft tissue infections, and highlights the role of early surgical intervention with repeated debridement and antifungal therapy.

Keywords: Subcutaneous Mucormycosis, immunocompetent, debridement

1. INTRODUCTION

Mucormycosis is a fatal, opportunistic fungal infection caused by fungi of the order, Mucorales, typically affecting immunocompromised individuals. Also seen in individuals with pre-disposing factors like uncontrolled diabetes mellitus, blood dyscrasias or malignancies, post-transplant patients, trauma or use of corticosteroids [1-2]. Amongst its varied clinical presentations rhinocerebral, pulmonary, gastrointestinal, disseminated, and subcutaneous, the cutaneous variant is seen approximately in 10-20% of cases [3]. The portal of entry for the fungal spores is usually through direct inoculation into the skin after some trauma, which disseminates rapidly if left unattended. Reports of subcutaneous mucormycosis in immunocompetent individuals are very rare, especially without any evident risk factors or trauma [4]. This is an interesting case of primary subcutaneous mucormycosis in an otherwise healthy 26-year-old male patient with no evident history of trauma or any other co-morbidity. This patient was treated successfully with prompt surgical debridement and appropriate antifungals.

2. Case Report

A 26-year-old male visited the general surgery outpatient department with high grade fever and a painful swelling over the anterior aspect of his right thigh for 7 days. The swelling was associated with pain, redness and local warmth. There was no history of evident trauma, diabetes, HIV infection, malignancy, or immunosuppressive therapy. But the patient did report a suspected insect bite at the site.

On general examination, the patient was febrile with stable vital signs. Local examination revealed a tender, tense, erythematous swelling which measured approximately 8 x 6 cm (Figure 1). Laboratory investigations revealed leukocytosis (TLC count: 21,000/mm³). The patient was admitted and started on intravenous Piperacillin-Tazobactam and analgesics. An emergency debridement was performed on the day of admission. A cruciate incision was made over the anterior thigh (Figure 2) and

seropurulent material along with necrotic tissue was removed. Sample was collected under sterile conditions and sent for bacterial culture and sensitivity, which did not yield growth of any microorganisms.

On postoperative day two, wound inspection revealed a blackish necrotic patch approximately of size (3 × 4 cm) with surrounding white debris, and fungal-mold like growth raising suspicion of fungal infection (Figure 3).

Figure 1: Swelling on anterior aspect of right thigh

Figure 2: Cruciate incision over anterior aspect of right thigh

Figure 3: Blackish-necrotic patch with mold-like growth

A second debridement was performed, and tissue was sent for histopathological and KOH examination. The debrided tissue was cleaned with sterile saline solution and processed for microscopy with 10% and 20% potassium hydroxide (KOH) in the microbiology laboratory as per the standard operating procedure. Microscopy revealed broad, aseptate, hyaline, ribbon-like hyphae with irregular wide-angle branching, consistent with Mucorales (Figure 4).

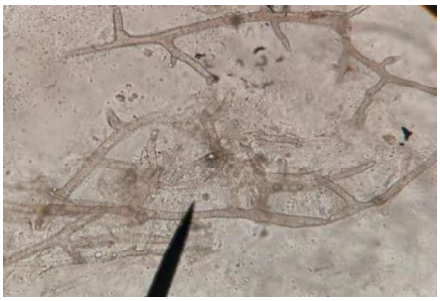

Figure 4: KOH microscopy revealing broad, aseptate hyphae

However, fungal culture on Sabouraud's dextrose agar at both 25°C and 37°C remained sterile after three weeks of incubation. Histopathology examination findings suggested clinical diagnosis of abscess and did not show any evidence of fungal tissue invasion. Further investigations including blood culture, Chest radiography, abdominal ultrasound, and serologic markers for HIV and diabetes mellitus were unremarkable, ruling out systemic fungal infection and immunosuppression. In consultation with an Infectious Disease specialist, systemic antifungal therapy was initiated. The patient received intravenous Isavuconazole 200 mg thrice daily for two days, followed by a maintenance dose of 200 mg once daily for three weeks. Subsequently, he was switched to oral Isavuconazole 200 mg once daily for one month. Simultaneously, the wound was managed with regular sterile dressings using povidone–iodine, hydrogen peroxide, and topical antifungals. Repeated debridements were done as needed until the wound was negative for any evidence of fungal elements and growth on microscopy and culture respectively. Over the next six weeks, healthy granulation tissue developed (Figure 5 a, b).

Figure 5(a, b) Healthy granulation tissue

Repeated debridement resulted in a large raw area associated with significant skin loss.

Primary wound healing seemed difficult, hence, split-thickness skin grafting was performed using an autograft from the contralateral thigh under spinal anesthesia. Skin grafting was opted to facilitate faster wound healing and avoid scarring and fibrosis. There was good acceptance of the graft with a satisfactory outcome (Figure 6). We keep follow up of patient for one year. He did not develop any new infection at local or distant sight and remain asymptomatic for one year.

Figure 6: Satisfactory wound healing after graft acceptance

3. Discussion

Cutaneous Mucormycosis is primarily a disease of the immunocompromised, but rare cases have been documented in immunocompetent healthy individuals too, often linked to trauma [5]. Our case here is unusual in that, a twenty six years-old immunocompetent male patient had no history of diabetes mellitus, any type of immunosuppression or evident trauma. The pathogenesis in these cases remains inconclusive. There are certain hypothesis which suggest some local but temporary immunological deficit, sub-clinical, unnoticed skin injuries or simple environmental exposure too, which may precede Mucorales inoculation leading to infection in healthy individuals [6]. Though the history of insect bite was vague there are certain case reports giving evidence of direct inoculation of mucorales through insect bite causing cutaneous Mucormycosis [7]. The well-known fact that, Mucorales are ubiquitous in the environment supports another possibility of idiopathic origin of Mucormycosis due to accidental lodging of spores in the wound [8]. Diagnosis in this case was supported by repeated evidence of broad, aseptate hyphae on KOH microscopy, strongly suggestive of Mucorales infection. Interestingly though, fungal culture on Sabouraud's Dextrose agar did not yield any growth. This can be attributed to the fact that Mucorales are fragile and lose their viability of the hyphal elements due to necrosis, during processing or due to technical limitations. A study by Mohanty et al highlights the importance of KOH microscopy in the early diagnosis of fungal infection [9]. Also, the literature review confirms absence of growth after fungal culture in more than 50% of cases as documented by studies of Mohanty et al., and Walsh et al., [8, 10]. A study by Rani Singh et al., [11] also showed a higher sensitivity of KOH microscopy than fungal culture. Histopathology demonstrated the presence of necrosis correlating with initial clinical diagnosis of abscess. Findings were not suggestive of tissue-invasion pathognomonic of Mucorales. This could be likely due to low fungal load in the sample, especially where tissue invasion is patchy as seen in our case or if sample sent is not representative of the said pathology. As it was subcutaneous involvement, the sparse fungal elements may have restricted to the superficial layers detected by KOH microscopy and histopathology sample shows only inflammation. Similar correlation was seen in a case report by Belgaumkar et al., [12]. Thus, clinical judgement and repeated microscopic examinations remain critical, especially when culture and histopathology results are not contributory. The treatment of Mucormycosis consisted of aggressive surgical debridement to remove the necrotic tissue combined with systemic antifungal therapy with Isavuconazole. Liposomal Amphotericin B is the conventional first-choice antifungal. Here, Isavuconazole a newer broad-spectrum triazole, was used, which shows good efficacy with fewer adverse effects

[13]. Recent reports promote its use with good outcomes in patients with various underlying

conditions. Also, the choice of Isavuconazole in this patient was justified by its cost- effectiveness and good oral bio-availability which resulted in good patient-compliance [14]. No definite clinical guidelines exist to assist the clinician in duration of antifungal therapy in immunocompetent patients, and treatment must be customized as per clinical resolution [15]. So, in this case, surgical debridement till wound margins became negative for fungal elements along with Isavuconazole were the mainstay of treatment. Early surgical intervention and repeated debridement in our patient, being immunocompetent prevented the characteristic dissemination of Mucormycosis. Patient follow-up was conducted up to one year, and no recurrence was seen. Thus, this case highlights the need for clinicians to consider Mucormycosis in immunocompetent healthy individuals with rapidly progressing necrotic lesions. In spite of negative fungal cultures, repeated microscopic examination remains critical for diagnosis and management. KOH mount microscopy is a sensitive and affordable method to make a rapid presumptive diagnosis. Repeated surgical debridement combined with appropriate antifungal can lead to better outcomes.

4. Conclusion

Subcutaneous mucormycosis, although rare in immunocompetent patients, should be considered in cases of rapidly progressive soft tissue infections with necrosis. KOH microscopy is a reliable presumptive test to diagnose and initiate therapy. Repeated surgical debridement combined with targeted antifungal therapy can result in favorable outcomes.

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Conflicts of Interest

The authors declare no conflicts of interest.

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Consent

Informed written consent was obtained from the patient for publication of this case and associated images and approval of Ethical committee was taken.

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