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Cutaneous Mucormycosis Following Trauma at the Surgical Site: A Rare Case

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Authors' contributions

This work was carried out in collaboration between all authors. Author BCV designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. Authors RPM and MKK managed the analyses of the study. Author MKK managed the literature searches. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Mucormycosis is a highly aggressive fungal infection caused by Zygomycetes, belonging to the subphylum Mucormycotina in the order Mucorales. This infection commonly presents an aggressive and rapid course and typically affects immunocompromised patients. Mucormycosis can manifest in different clinical patterns and locations. Although the correct diagnosis is often difficult, early identification is essential for patient survival. Several clinical forms of mucormycosis are recognised. Cutaneous mucormycosis is less common than other clinical forms, but potentially lethal if treatment is not rapid. Tissue examination by histopathology and culture confirms the fungal infection. Standard treatment includes antifungal therapies associated with surgical debridement. We report a case of postoperative cutaneous mucormycosis followed by trauma with lethal outcome.

Keywords: Mucormycosis; aggressive fungal infection; cutaneous mucormycosis; lethal outcome.

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1. INTRODUCTION

MUCORMYCOSIS is an angioinvasive opportunistic infection caused by moulds belonging to the subphylum Mucoromycotina in the order Mucorales [1]. The infection is more common among people with the suppressed immune system as the causative fungus is nonpathogenic for healthy individuals, but it can also rarely occur in immunocompetentpeople [2]. Known risk factors for developing mucormycosis comprise uncontrolled diabetes mellitus, metabolic acidosis, high dose of corticosteroids, prolonged neutropenia, organ transplantation, skin trauma (cuts, scrapes, punctures, or burns), and catheter infection, thus it represents an opportunistic rather than a true pathogen. However, some patients with mucormycosis may have no identifiable risk factors [3]. Rhinocerebral infection is the commonest form of mucormycosis in humans, followed by pulmonary infection. Most common mode of transmission of infection is inhalation of infectious spores which initiates an infection in the respiratory tract, other less common routes of infection include ingestion of spores resulting in intestinal mucormycosis, or by penetrating injuries to the skin [4]. Cutaneous mucormycosis results due to disruption of cutaneous barriers as in trauma, but can rarely be iatrogenic. Cutaneous involvement is less common than the others and mainly occurs in two forms, a “benign” well-localized subcutaneous form and a more fulminant cutaneous infection with necrotizing fasciitis, systemic sepsis and a fatal outcome if the diagnosis and consequently, the appropriate treatment is delayed [5]. Although dermal involvement rarely occurs an early diagnosis and prompt treatment can lead to a reduction of mortality and morbidity [2]. Cutaneous mucormycosis can also be classified according to the extent of the infection as localized when it affects only the skin or subcutaneous tissue; deep extension when it invades muscle, tendons, or bone; and disseminated when it involves other non-contiguous organs. The typical presentation of cutaneous mucormycosis is a necrotic eschar accompanied by surrounding erythema and induration. However, a nonspecific erythematous macule may be the cutaneous manifestation of disseminated disease in an immunosuppressed patient [6]. When cutaneous mucormycosis presents with necrotic eschar, the lesions may mimic pyodermagangrenosum, bacterial synergistic gangrene, or other infections produced by bacteria or fungi [7]. Successful treatment of mucormycosis includes certain strategies, which includes evaluation of patients to recognize risk factors and early signs of infection, removal or reduction of risk factors to deal with any reversible predisposing factors such as management of diabetic ketoacidosis, reducing the level of immunosuppression, surgical debridement, initiation of specific antifungal therapy such as liposomal amphotericin B, in addition to adjunctive therapies like the use of hyperbaric oxygen. Patients with mucormycosis should be treated in a tertiary referral centre with subspecialty units experienced in the care of the condition and the underlying causes.

We report a rare case of Postoperative cutaneous mucormycosis caused by Mucor species following exposure to road traffic accident (RTA).

2. PRESENTATION OF THE CASE

A 45-year-old, previously healthy male presented to our emergency department with a history of RTA with an open wound which resulted in right femur fracture, head injury and right eye globe rupture. The patient was treated conservatively for a head injury, evisceration was done for eye globe rupture and Open Reduction and Internal Fixation (ORIF) was performed on the fractured bone.

Three days after the surgery the patient noticed an extremely painful swelling on the lateral aspect of his right leg, which became extensively ulcerated and rapidly progressed in size with time and was associated with fever. The patient was not a diabetic or hypertensive. There was no history of insect bite, immunosuppressive therapy or any chronic dermatological diseases. On general physical examination, the patient was alert, febrile (38.8°C) with no palpable lymph nodes. Other systems revealed no abnormality. Physical examination revealed a 10 × 15 cm an area of necrosis with oozing pus at the lateral aspect, anterior side of the right femur.

A swab was sent for culture and sensitivity. A gram-stained smear of the sample showed plenty of inflammatory cells and fungal hyphae. 10% Potassium hydroxide (KOH) wet mount examination of the sample showed broad aseptate fungal hyphae against a clear background which is suggestive of fungal infection. Bacterial culture yielded no bacterial growth and the sample was subjected to fungal culture on Sabaurauds Dextrose Agar (SDA)

without cycloheximide which yielded the growth of cottony white to greyish brown fungal growth on the obverse and no pigment on the reverse. A lactophenol cotton blue preparation of the fungal growth showed broad aseptate hyphae with right angle branching and spherical sporangia with no rhizoids suggesting the growth of Mucor and the same was confirmed by slide culture technique (Fig. 1). Repeat sample also yielded the growth of same fungi and the patient was diagnosed as a case of cutaneous mucormycosis. Blood picture, fasting and post-prandial blood sugar levels, renal function tests and liver functions tests were normal at the time of admission but later on patient developed renal impairment. Fungal culture report was immediately informed to the consultant for further management of the patient. The patient was immediately started on a course of intravenous Amphotericin B (0.5 mg/kg/ day) but the infection progressed rapidly and eventually, he developed disseminated mucormycosis and expired after 2 weeks of surgery.

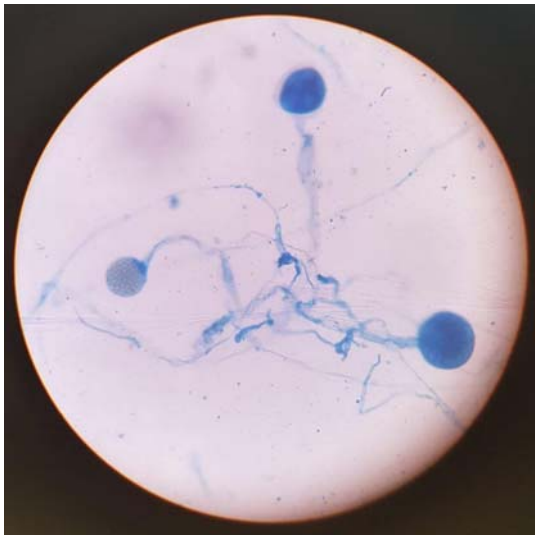

Fig. 1. Lacto-phenol Cotton Blue Preparation of Fungal growth showing Mucor spp.

3. DISCUSSION

Fungi belonging to the order Mucorales are ubiquitous in the environment and have been isolated in up to 22% of hospital air samples [8]. Healthy individuals have strong natural immunity to the Mucorales, mucormycosis usually occurs in immunocompromised patients. Depending upon the primary site of involvement, mucormycosis can be classified as rhinocerebral, pulmonary, gastrointestinal, disseminated and miscellaneous or cutaneous [9]. Cutaneous mucormycosis constitutes about 10% of all cases [10]. Mucorales are incapable of penetrating intact skin, and infection requires direct inoculation through a compromised cutaneous barrier. Patients who are at risk usually have multiple injuries or burns associated with extensive tissue damage or soil contamination [9]. Infection of a clean surgical wound is extremely rare. Our patient is a good example of the natural history of the disease. The patient has following major risk factors: trauma with an open wound, Surgery and Short course of corticosteroids. He also did not react favourably to the initial treatment and showed the rapid progression of the infection, which was complicated by deep tissue invasion, multiple organ failures and succumbed to death.Possibility of infection in our patient could be that the spores were on the skin of the patient as part of the transient flora, and it is known that solutions such as povidone iodine used to prepare skin preoperatively are unable to eradicate the spores and this may have been source of the initial inoculum or may have gained entry through contaminated surgical instruments, sutures, dressing materials. A short course of steroids was given to our patient, which might have predisposed the patient to mucormycosis. Our patient did not have any other risk factors for the development of mucormycosis.The most characteristic feature of mucormycosis is the hyphal invasion of blood vessels. Early tissue biopsy for KOH examination and fungal culture is the gold standard for diagnosis. Treatment of cutaneous mucormycosis involves eliminating all predisposing factors such as discontinuing all immunosuppressive drugs and initiating parenteral antifungal therapy, however early and aggressive surgical debridement reduces overall mortality from 60% to 11% [9]. Surgical treatment should always be combined with a systemic antifungal agent because this increases the overall survival rate of patients to 70%. The first choice is high-dose amphotericin B (1.0–1.5 mg/kg/day) or 3 mg/kg when liposomal formulations are used and are preferred due to their lower nephrotoxicity [9]. Posaconazole in vitro has been shown to have activity against zygomycetes; therefore, it is usually the second-line of treatment in patients with toxicity associated with amphotericin or treatment-refractory infections [9]. Treatment duration should be individualized, and it is almost always continued until disappearance of clinical data. Mortality from cutaneous mucormycosis usually results from the spread of the infection to vital organs not amenable to surgical excision and is

directly dependent upon the speed of diagnosis and institution of treatment. In conclusion, cutaneous Mucormycosis is a true medical challenge. Due to its low frequency, for diagnosis, it requires a high index of clinical suspicion. It should always be included in the differential diagnosis of necrotic wounds unresponsive to antibiotic treatment. The aggressiveness and rapid evolution of this condition require the combination of timely surgical treatment along with systemic antifungal therapy.

4. CONCLUSION

A high index of suspicion is crucial for identifying and preventing progression of the disease and mucormycosis must always be considered as a differential diagnosis particularly when wound infections responding poorly to antibiotic therapy. Extra vigilance should be given to those who are immunocompromised, and also who are receiving short courses of steroids. Early recognition, prompt surgical intervention and initiation of an appropriate antifungal treatment are crucial in the management of this rare case but potentially limb and life-threatening infection.

CONSENT

As per international standard or university standard, patient's written consent has been collected and preserved by the authors.

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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