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Phlegmasia Cerulea Dolens: A Rare Complication of Cutaneous Mucormycosis

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Abstract

Mucormycosis is a rare, opportunistic, aggressive infection caused by fungi which frequently infects the brain, sinuses, or lungs, rarely skin, GIT. Primary cutaneous infection in humans is limited to patients with severe immunocompromised status. This is a case of middle aged immunocompetent patient with primary cutaneous mucormycosis infection of the anterior abdominal wall with involvement of bilateral inguinal lymph nodes causing deep vein thrombosis and eventual gangrene of lower limb. The potential life threatening condition was averted with timely intervention, although his limb could not be salvaged. Diagnosis and timely management of mucormycosis still remains a challenge. The key to the diagnosis lies is the identification of the important risk factors and hallmark symptoms and a quick histopathological diagnosis. Awareness, early recognition, extensive and aggressive diagnostic procedure, prompt surgical intervention and initiation of an appropriate antifungal treatment are crucial in the management of this uncommon but potentially limb and life threatening infection.

Keywords: Mucormycosis • Immunocompetent • Cutaneous • Histopathological

Introduction

Mucormycosis is a rare, opportunistic, aggressive infection caused by fungi of the order Mucorales. Mode of transmission is most often through inhalation, contaminated food, or contamination of open wounds which frequently infects the brain, sinuses, or lungs, rarely skin, GIT [1]. Primary cutaneous infection in humans is limited to patients with severe immunocompromised status, diabetes mellitus, trauma, burn, prolonged ICU stay and prolonged steroid therapy but the disease in immunocompetent hosts is still rare [2]. We report a rare case of primary cutaneous mucormycosis infection in an immunocompetent patient of the anterior abdominal wall with involvement of bilateral inguinal lymph nodes and causing deep vein thrombosis of unilateral limb.

Case Report

A 40 year old patient presented to emergency with complaints of an enlarged non-healing ulcer in anterior abdominal wall (Figure 1A) for 3 month followed by progressive enlargement in bilateral inguinal region for 60 days (right more than left) (Figure 1B). Patient had an episode of fever; there was discoloration of the right lower limb progressing from leg to involve the upper thigh and inguinal region for 20 days (Figure 1C). There was no history of recent trauma, chronic illness, substance abuse and prolonged drug intake. On examination, abdomen was soft, diseased limb peripheral pulses present. Patient was admitted in emergency, was haemodynamically stable, intravascular fluids, empirical antibiotic therapy started after taking samples for culture and sensitivity from the wound. The patient underwent debridement of the abdominal wound and an incisional biopsy of the right inguinal lymph node was performed simultaneously. The debrided tissue and the inguinal lymph node were sent for histopathological examination. On blood investigation patient had mild neutrophilic leukocytosis, raised D dimer, rest of the routine investigations KFT, LFT were within normal limits. Patient also underwent COVID-19 testing despite being asymptomatic for it, which was also negative; screening for HIV/Hepatitis B, C infection was negative. On Duplex scan of the right lower limb, Iliofemoral DVT was detected, with intact arterial circulation, following which the patient was put on anticoagulation. There was no improvement in symptoms after 5 days of appropriate treatment and despite antibiotics escalation based on the culture report which suggested polymicrobial infection. On Day 6 of admission, histopathological report revealed the demonstration of ribbon-like hyaline, predominantly aseptate hyphae with wide-angle branching (Figure 2) and hyphae invasion of the blood vessel with impending thrombus (Figure 3), ocnfirming the diagnosis of mucormycosis. As no other organ involvement was detected, it was diagnosed as a case of primary cutaneous mucormycosis. Treatment with injection Liposomal Amphotericin B was instituted immediately (300 mg/day for 5 days) and there was dramatic improvement in clinical parameters. The abdominal wound recovered well after total three debridement followed by split skin grafting, patient eventually required the amputation of the lower limb. Post amputation clinical parameters improved and a potential life threatening condition was averted with timely intervention and the patient was discharged after another 10 days of inpatient treatment.

Discussion

Mucormycetes are scattered throughout the environment, as in soil, in

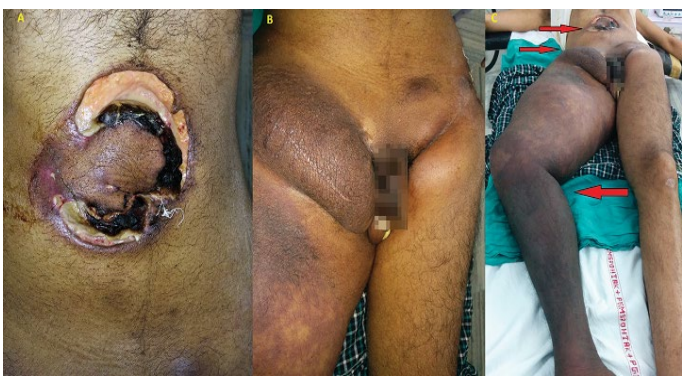

Figure 1. A) Abdominal wall ulcer, B) massive inguinal lymphadenopathy and C) bluish discoloration of the right lower limb.

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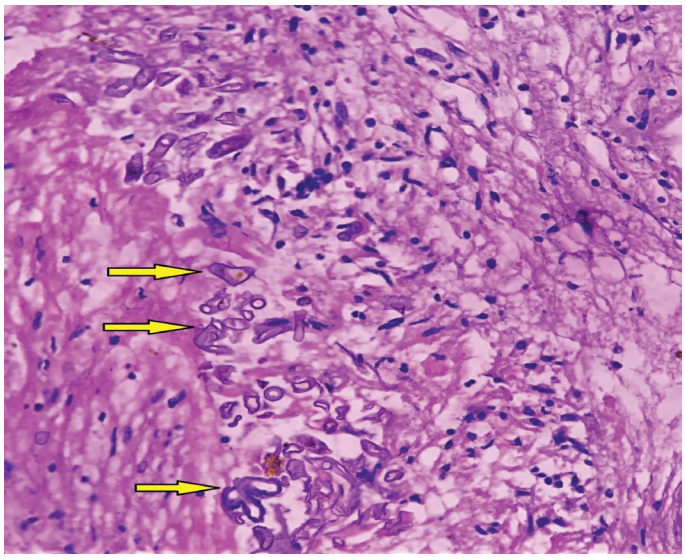

Figure 2. Histopathological image ribbon-like hyaline, predominantly aseptate hyphae with wide-angle branching.

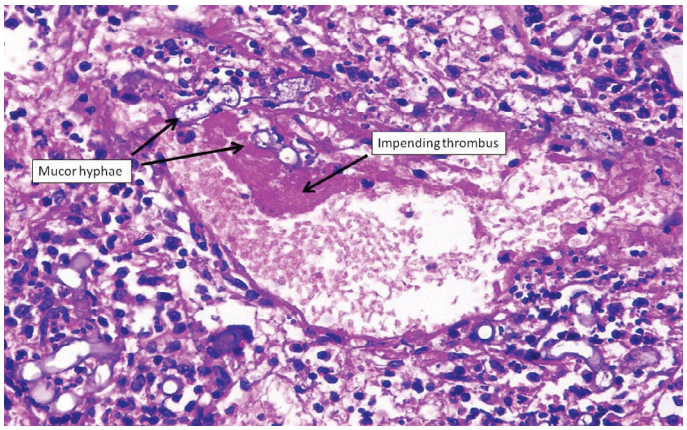

Figure 3. Histopathological image showing hyphae invasion of the blood vessel with impending thrombus.

decaying organic matter, such as leaves, animal dung, and compost piles. Cutaneous mucormycosis, although less commonly, constitutes about 10% of all cases [3,4]. Mucorales are not capable of penetrating intact skin, and infection requires direct inoculation through a compromised cutaneous barrier. Cutaneous mucormycosis occurs in two forms: primary with no other organ involvement and secondary, due to distant sites of rhinocerebral, pulmonary or gastrointestinal. Majority of the cutaneous presentations are acute and as surgical emergencies, but can also rarely have a chronic indolent course, with signs and symptoms developing over weeks. In proportion of cases developing in immunocompetent patients with no underlying disease, trauma or burns were the usual predisposing factor resulting in cutaneous disease. The trauma can be trivial (injection sites, animal bites, gardening) or major, including road traffic accidents, natural disasters and surgery [5]. In immunocompetent patients, infections usually remain localized and can be managed if recognized and treated timely but on the contrary ignorance of the warning signs and virulence of the organism can lead to dissemination and fulminant infection. The pathognomic feature is characterized by hyphae invading the blood vessels, with rapid production of thrombosis, inflammatory occlusion and tissue necrosis [6]. Cutaneous mucormycosis has three clinical stages, which are based on the degree of vascular invasion achieved by the fungus with involvement of the major arteries in stage III [7]. In the above described case, the etiology of the phlegmasia cerulea dolens is most likely due to the massive inguinal lymphadenopathy causing obstruction to the venous drainage, secondary to abdominal wall wound infection. However, owing to the aggressiveness of the infection a direct vascular invasion of the major vessel causing secondary venous stasis can also be attributed as the probable cause. Patient has no associated illness which could lead to an immunocompromied status. Although the option of intravascular catheter directed thrombolysis was contemplated [8], but the patient limb was beyond salvage, it had been more than 3 weeks since the onset of symptoms, and moreover patient was unwilling to be referred to higher centers with such facilities due to financial issues. Diagnosis and timely management of mucormycosis still remains a challenge. Histology forms the core to the diagnosis and although culture is an essential tool, it is still unreliable and also various other molecular methods of diagnosis are still evolving. The key to the diagnosis lies is the identification of the important risk factors and hallmark symptoms. In management of mucormycosis, the prognosis depends upon the following factors which are critical to eradicate the infection:

- Prompt and accurate diagnosis
- The degree of the associated comorbidity or chronic illness and correction of predisposing factors such as hyperglycemia, neutropenia, reduction of dose of temporary suspension of immunosuppressive drugs.
- Appropriate timing of therapeutic intervention such as urgent surgical debridement
- Antifungal treatment

Conclusion

In conclusion, awareness, early recognition, extensive and aggressive diagnostic procedure, prompt surgical intervention and initiation of an appropriate antifungal treatment are crucial in the management of this uncommon but potentially limb and life threatening infection.

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