CORRESPONDENCE

Primary Cutaneous Mucormycosis in a Beta-Thalassemia Patient

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To the Editor,

Cutaneous Mucormycosis (CM) is among the known causes of morbidity and mortality in immunocompromised patients. The infection is caused by Zygomycetes, a member of the order of Mucorales. Cutaneous mucormycosis is the third most common form of this disease behind rhinocerebral and pulmonary forms [1].

Although mostly occurring in immunocompromised and diabetic patients, there are several reports of CM infection in the immunocompetent patients. The most common risk factor is breach in the skin barrier. Desferoxamine therapy is another risk factor for CM [1].

A 24-year-old woman, a known case of beta-thalassemia intermedia presented with a necrotic crusted ulcer on her chin a few days after blunt trauma. She received repeated monthly blood transfusion and Desferoxamine injection. She had history of splenectomy due to hematologic indication one month prior to skin lesion. She was febrile (temperature = 39 °C orally) on admission. There was necrotic ulcerative plaque on chin measuring about 6 cm in

diameter with a few peripheral hemorrhagic vesicles accompanying soft tissue edema (Fig. 1a). Laboratory and Peripheral blood smear examination revealed elevated level of erythrocyte sedimentation rate, anemia, and leukocytosis with neutrophil dominancy and toxic granulation. Gram stain and bacterial culture were negative. Serum iron and ferritin levels were 188 µg/L and 506 ng/mL, respectively. Histopathological examinations of skin biopsy specimen showed epidermal and dermal necrosis, severe neutrophil infiltration and many broad non-septate hyphae which branched irregularly at 90° throughout the dermis and subcutaneous fat (Fig. 1b, c). Unfortunately, species could not be identified by culture. The lesion healed after intravenous injection of conventional amphotericin B deoxycholate (cumulative dose of 575 mg during 14 days) without need to surgical debridement.

Although mostly expected in immunocompromised patients, CM has been frequently reported in immunocompetent ones. There are several cases of posttraumatic CM in immunocompetent patients reported in the literature. Vitrat-Hincky et al. [2] described 75 patients infected by filamentous fungi including mucorales after major trauma. Traffic and farm accidents, natural disaster, fall, and blunt trauma were among the injuries specified in this series and the other studies [1, 2]. There are reports of CM in the immunocompetent patients following bite, surgery, intramuscular injection at site, hit by bull, and site of intravenous cannula. However, CM rarely occurs in immunocompetent hosts without preceding trauma [1].

Primary cutaneous mucormycosis presents after penetrating trauma as erythematous to violaceous lesions proceeding to necrotic ulcers with indurated erythematous borders and malodorous discharge. Cases of superficial mucormycosis may present with mildly raised squamous

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Fig. 1 **a** Clinical presentation. Necrotic ulcerative plaque with overlying hemorrhagic vesicles and soft tissue edema. **b** Histopathological analysis (mid-power). There are some broad non septate hyphae among neutrophilic infiltrate (H&E 200 $\times$).

c Histopathological analysis (high-power). There are some broad focally twisted non-septate hyphae between collagen fibers with degenerative changes (H&E400 $\times$)

borders or targetoid plaques with erythematous margins and ecchymotic or black necrotic centers [3].

Increased iron availability is one of the risk factors for mucormycosis. Diabetic patients with ketoacidosis are also prone to mucormycosis, likely due to elevated available serum iron during ketoacidosis. Iron overload is expected in repeated transfusions or dyserythropoiesis. Patients treated with desferoxamine are also at risk of mucormycosis. Desferoxamine used as an iron-chelator in dialysis recipients or beta-thalassemia patients is a siderophore which mediates iron transport for mucorales as a xenosiderophore. The more recent iron chelators such as deferiprone and deferasirox lack xenosiderophore activity and are even proposed as an alternative antimucorale treatment due to ironstarvation effect to the fungus [3, 4]. However, desferoxamine is still used in some centers in our country.

Primary CM in immunocompetent patients is treated with surgical debridement and antifungal therapy including amphotericin B (drug of choice) or posaconazole [1].

We found another reported case (an 11 years old boy) with beta-thalassemia who developed CM after splenectomy and leg fracture and cured after debridement, amphotericin B infusion and skin graft [5].

The case presented here highlights the importance of considering CM in any immunocompetent patient especially beta-thalassemic patients, particularly if there is

history of even minor trauma and starting rapid and effective treatment.

Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

Informed Consent Informed consent was obtained from all individual participants included in the study.

References

1. Kaushik R (2012) Primary cutaneous zygomycosis in India. *Indian J Surg* 74(6):468–475
2. Vitrat-Hincky V, Lebeau B, Bozonnet E, Falcon D, Pradel P, Faure O et al (2009) Severe filamentous fungal infections after widespread tissue damage due to traumatic injury: six cases and review of the literature. *Scand J Infect Dis* 41(6–7):491–500
3. Petrikos G, Skiada A, Lortholary O, Roilides E, Walsh TJ, Kontoyiannis DP (2012) Epidemiology and clinical manifestations of mucormycosis. *Clin Infect Dis* 54(Suppl 1):S23–S34
4. Kontoghiorghes GJ, Kolnagou A, Skiada A, Petrikos G (2010) The role of iron and chelators on infections in iron overload and non iron loaded conditions: prospects for the design of new antimicrobial therapies. *Hemoglobin* 34(3):227–239
5. Tanphaichitr VS, Chaiprasert A, Suvatte V, Thasnakorn P (1990) Subcutaneous mucormycosis caused by *Saksenaia vasiformis* in a thalassaemic child: first case report in Thailand. *Mycoses* 33(6):303–309