Case Report

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Myocutaneous Mucormycosis in a Diabetic Burnt Patient Led to Upper Extremity Amputation; A Case Report

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ABSTRACT

Mucormycosis is a rare opportunistic fungal infection that can implicate cranial sinuses, brain, lungs, gastrointestinal tract and skin. Although it can occur in patients with competent and incompetent immunity such as patients with diabetes mellitus, lymphoma, leukemia and burns, but it has an aggressive, malignant and lethal course in patients with incompetent immunity. To enforce the importance of burn in patients with underlaying diseases such as diabetes, we are going to report a rare case of diabetic burnt patient complicated by right upper extremity myocutaneous mucormycosis. We selected this case to emphasis the importance of underlying disease (diabetes mellitus) with cutaneous burn, aggressive treatment of fungal infection in these patients and referring such case to burn center to prevent catastrophic results. A 50-year-old woman was introduced to us after several days of medical and surgical care of right upper extremity and trunk split-thickness burn. Due to gross muscle necrosis of right upper extremity and poor general condition of the patient, she was taken to the operating room that led to right upper extremity amputation and several times of aggressive debridement to save her life. Pathologic report was indicative of mucormycosis. We can conclude from this case that: 1) Burn, even partially thickness and with little body surface area, should be referred to burn center for better care 2) No response to usual medical treatment should make us more sensitive to consider the unusual causes of infection such as fungi 3) Suspected dead tissues should be excised aggressively especially if suspiciousness to wound sepsis and fungal infection is present especially in an immunocompromised patient.

Keywords: Mucormycosis; Extremity amputation; Partial thickness burn; Diabetes; American Burn Association (ABA).

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Introduction

Mucormycosis is a rare opportunistic fungal infection that can implicate cranial sinuses, brain, lungs, gastrointestinal tract and skin [1-6]. Although it can occur in patients with competent and incompetent immunity such as patients with diabetes mellitus, lymphoma, leukemia and burns; however it has an aggressive, malignant and lethal course in patients with incompetent immunity [1, 3-7]. Mucormycosis is caused by a fungus commonly found in soil and decaying plants [1]. It can enter the body

through air, gastrointestinal tract or skin [6]. This infection is more common in developing countries than in Europe [3]. In a system review by Vaezi *et al*., 98 cases of mucormycosis evaluated in Iran from 1990 to 2015. Diabetes mellitus was the most common underlying disease (48.9%) and rhinocerebral (48.9%), pulmonary (9.2%) and cutaneous (9.2%) were the most common clinical types. Overall mortality was 40.8% and disseminated infection (75%) was the most common cause of mortality.

Mucor involvement of skin is still rarer compared to other organs such as cranial sinuses and usually presents as a firm and painful skin lesions with a dark center [1]. Biopsy is required for diagnosis [1], which is confirmed by smear and culture [3]. Fixation with KOH and staining with PAS will show fungal hyphae in skin [2]. Early diagnosis and timely treatment is very effective in reducing mortality [1, 2] and surgeon should remove all the dead and infected tissues [1]. Concomitant with surgery, systemic anti-fungal therapy is necessary [1-3]. As mentioned above, diabetes is a disease in which the susceptibility to rare infections such as mucormycosis increases. In diabetes associated with burns, breakdown of skin barrier and immune-lowering synergy between these two conditions increase risk of infection. So, burn in diabetic patients should be transferred to a burn center for special care. To enforce the importance of burn with diabetes, we are going to report a rare case of diabetic burnt patient complicated by right upper extremity myocutaneous mucormycosis.

Case Presentation

A 50-year-old diabetic woman with 4-5% (TBSA) 2nd degree burn in the right shoulder, arm and right upper hemithorax was referred to a specialist for management. After examination by physician (specialist), patient was sent home due to partial thickness and small size burn with medication and outpatient follow-up. Due to worsening symptoms and wound appearance, she went to a general surgeon and debridement of necrotic tissues of her right arm and shoulder (2-3% TBSA) was done on the tenth day after burn. After debridement by surgeon, 6 days after debridement, she was introduced to our hospital (Ghotbodin Burn Center, Shiraz, Iran) due to poor response to treatment. The patient was admitted to Burn Center. Patient was mildly confused, ill looking and with tachycardia and high grade fever (39-39.5° C) at arrival. There was a deep wound on right arm with exposed dead gray colored muscle without contraction. Her serum WBC and blood sugar were 16,000/dl and 250-350 mg/dl respectively. Other laboratory data such as serum electrolytes, kidney and liver function tests all were in normal value range. Patient's hyperglycemia was poorly controllable by high dose insulin. Total parenteral nutrition was started for her to support nutritional status. Due to gross muscle necrosis and systemic signs, multi-drug antibiotic regimen (imipenem, vancomycin, and penicillin) to cover gram positive, gram negative and anaerobes was administered and after initial stabilization, the patient was taken to the operating room. Intra-operative findings included necrosis around all the muscles of right arm (Figure 1) extending to the forearm muscles, and chest wall soft tissue with involvement of lateral of right breast. So, all of the necrotic tissues and muscles excised and due to the extent of necrosis of the right upper extremity, upper extremity inevitably amputated 5 cm distal to shoulder joint and the wound kept open. Despite extensive debridement to reach red and living tissue after the first operation and intensive wound care after it, unfortunately the wound color changed to black, gray and dark brown after 2 days (Figure 2A). Patient's general condition became worse, blood sugar increased, and hyperthermia recurred. So, we have to do debridement in the operating room again and extensive debridement of infected soft tissues (including muscles) of right lateral chest wall, and right breast was done; right arm amputated at shoulder joint and all necrotic areolar tissues and lymph nodes of the right axilla evacuated. Pathology report was "Ulceration, granulation with necrosis and abscess formation with fat necrosis". Due to the clinical suspicion of fungal infection, fluconazole was started 100 mg 2 times a day. Six days after 2nd debridement, due to necrosis around and below right scapula and clavicle and chest wall, right scapula and clavicle removed and chest wall re-excision to grossly red and viable tissue.

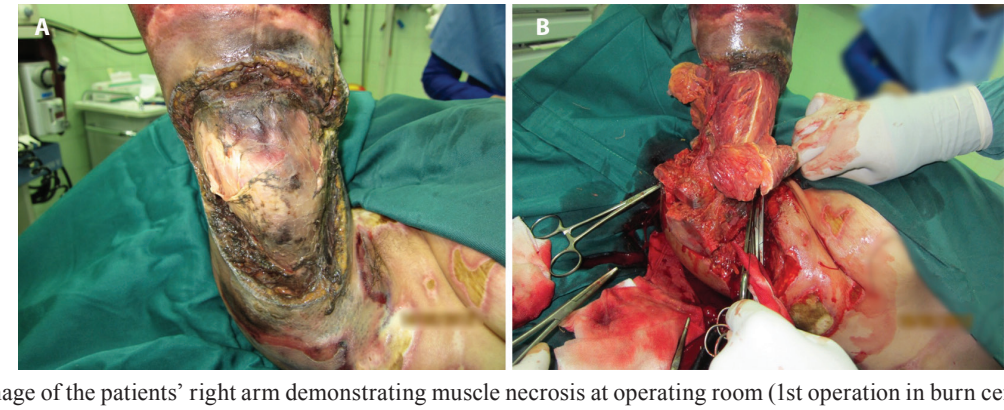

Fig. 1. An image of the patients' right arm demonstrating muscle necrosis at operating room (1st operation in burn center) (A); gross necrosis of all muscles of the right arm after incision during first operation in the operating room (B).

Although the progression of tissue necrosis was less thereafter, but then we have to do debridement of right chest wall and shoulder every 4-5 days for a few times. Chest x-ray didn't show any internal thoracic involvement due to proximity of the right external chest wall pathology. Forty-eight to seventy-two hours after each debridement, we saw scattered black and brown foci in the wound that coalesced together rapidly (Figures 2A, B, C). Biopsy of the lesions with bacteriological (aerobic and anaerobic) and fungal cultures were done with no positive results. Three times blood cultures were done and no organism grew. Due to recurrence of necrosis we did repeated excision of necrotic tissues (Figure 2 B and C]. We used silver sufadiazine, mafenide acetate and nystatin cream, dilute H2O2, sodium hypocholorite 0.025% for local wound care. A few days after the last debridement, the disease process stopped and after formation of the proper stable granulation tissue (Figure 2 D), skin graft to the wound with a mesh of 1 to 1/5 was done (Figure 3). On the 17th day of admission, one of the last pathology reports showed mucormycosis as definite tissue diagnosis (sections from pathologic specimen showed complete dermal and epidermal fat necrosis along with many broad hyphae with no septation and acute angle branching) and systemic anti-fungal drug changed from fluconazole to voriconazole 200 mg/d. Finally, patient discharged with good condition 33 days after admission. On subsequent visits the patient's general condition was good. One month after discharge the patient complained sternal bulging, but chest CT only showed soft tissue bulging over sternum that disappeared on subsequent follow-up visits with no need for tissue diagnosis or culture.

Discussion

Mucormycosis is a rare fungal infection in patients with underlying diseases such as diabetes [1-8]. It may invade vessels and cause deeper tissue necrosis [4]. However, patients with moderate to severe burns (over 20% of TBSA) due to reduced systemic immunity (cellular and humoral) are more susceptible to the risk of infection. As we know, burn with underlying disease such as diabetes mellitus should be referred to the burn center [American Burn Association (ABA) burn center referral criteria] because such patients usually have more complicated course and needs more specialized care in burn center. As we saw in this case, the patient was visited by a specialist and managed as a small size, superficial burn and cared with outpatient visits. Ignoring the standards seemingly simple but important, can lead to grave results. So we have to stress that performance of burn referral criteria is very important and we should notice that diabetes and major underlying diseases coexisted with burn can compromise benign course of small sized and partially thickness burn. It is important to consider that these guidelines are the results of experiences of researchers around all of the world and for many

Fig. 2. The follow-up images of the right proximal arm amputation stump after right upper extremity amputation and extensive debridement: 3 days after operation (A); 11 days after operation demonstrating glenoid fossa at lower and left aspect of the wound (disarticulation) (B); 16 days after the operation demonstrating excised right scapula, clavicle and most part of lateral side of breast (C). Right chest wall wound 25 days after the operation. Wound is red and nearly no gross necrotic tissues are present (D).

Fig. 3. Right chest wall wound 38 days after right upper extremity amputation and extensive debridement (follow-up visit). Wound is covered with skin graft.

years. On the other hand, if patient with burn and concomitant major systemic disease has symptoms and signs of infection, locally or systemically, the cause should be identified and treated appropriately and promptly. If it presents unusually or responds poorly to ordinary treatment, it is better to consider less common and more aggressive causes of infection. Mucormycosis is a rare, but aggressive fungal infection in immunocompromised patients that we should consider it when response to usual treatment is poor or presentation is unusual. Control of underlying disease (hyperglycemia in diabetes mellitus) and preliminary stabilization of vital signs, aggressive & repeated surgical intervention (such as debridement), and usage of appropriate drugs are important to limit the disease or stop it as soon as possible. Even extremity amputation, although an ominous decision, should be performed if indicated to preserve patient's life. Closed observation of the patient after operation to detect and treat any recurrence of disease is also very important. Another point is that all necrotic soft tissues, especially progressive forms and with systemic symptoms should be carefully excised and if needed repeated at short intervals before reappearance of systemic symptoms and signs.

So, in brief, burn injury concomitant with underlying disease such as diabetes mellitus is one of 10 indications of referring the patient to burn center based on American Burn Association burn center referral criteria to burn center. This is very import because due to increased morbidity and even mortality in these group of patients they should be managed in a burn center that is a special hospital for burnt patients. Outpatient management or management in hospitals other than burn hospital can increase complications. Another important point that should be noticed is considering fungal disease (in our special case, mucormycosis) as a cause of infection. Many physicians may miss fungal infections and it is very important to consider fungal infections in immunocompromised patients such as patients with extensive burn or burn and diabetes. And the last very important point in extensive serial debridement of all of necrotic tissues infected by fungals (mucormycosis in our case). Any hesitation in such approach can compromise patients's life.

We can conclude from this case: 1) Burn, even partially thickness and with little body surface area in patients with underlying diseases such as diabetes mellitus, should be referred to the burn center 2) Non-responsiveness to usual treatment should make us more sensitive to consider the unusual causes of infection such as fungi 3) Suspected dead tissue should be excised aggressively to save patient's life.

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