Cutaneous Zygomycosis Due To Saksenaea Vasiformis In A Patient With Paraparesis, Burns And Pressure Ulcer

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Abstract

Cutaneous zygomycosis is a devastating clinical condition. Most of the reported cases involve some kind of cutaneous trauma predisposing to the infection. In debilitated bedridden patients, development of pressure ulcers is a common complication. The present case adds the emerging pathogenic fungus, Saksenaea vasiformis as a cause of infection in pressure ulcer, with burns as an added local predisposing factor.

INTRODUCTION

Cutaneous zygomycosis is a devastating clinical condition. A review in the 2006 listed 30 human cases of cutaneous infection caused by Saksenaea vasiformis [1]. Three of these have been have been described in patients with burns but none in pressure ulcers. The first case of Saksenaea vasiformis infection in India was reported in 1988 [2]. The reports of cutaneous zygomycosis due to Saksenalia vasiformis indicate its distinct predilection to the skin and subcutaneous tissue. Most of the reported cases involve some kind of cutaneous trauma predisposing to the infection. In debilitated bedridden patients, development of pressure ulcers is a common complication. The present case adds the emerging pathogenic fungus, Saksenaea vasiformis as a cause of infection in pressure ulcer, with burns as an added local predisposing factor.

CASE REPORT

A forty-five year old male was admitted in our tertiary care hospital in India with complaints of weakness of both lower limbs for three months, pain in lower back and inability to control urine and stool for three months. He also had a large ulcer on the lower back for two months. The patient was transferred from a peripheral hospital to our centre. The patient had a history of vehicular accident fifteen years back following which he experienced increasing pain in the lower back. Due to the pain in the lower back, the household remedy for pain by applying hot brick was tried on the bedridden patient. Probably due to the associated sensory loss, the extent of heat due to the brick was not sensed by the patient resulting in deep burns. The burns were followed by ulcer formation on the lower back.

On examination, the patient was conscious but disoriented and had grade II/V power in lower limbs with peripheral neuropathy. He had poor tone in anal sphincter and had an ulcer 15 cm by 20 cm on the lumbosacral region extending to the perianal area. The ulcer was deep, with exposed ligaments, muscle and bone (Stage IV ulcer) [3] and it was contaminated with incontinent stools (Fig. 1).

The blood investigations showed normal sugar level and haemoglobin, with a raised white blood cell count to 54,200/mm$^3$ and a neutrophil predominance of 82%. The
MRI of lumbosacral region was suggestive of degenerative changes at L4-L5 and L5-S1 level compressing the thecal sac at L5 nerve root with encroachment of the left neural foramen. Focal canal stenosis was seen at L4-L5 level. Since the wound appeared unhealthy and was non-healing, a swab from the ulcer was sent for fungal culture, which was negative. Surgical debridement of the ulcer was done and the debrided material was received for mycological examination and culture. On examination in potassium hydroxide, broad, sparsely septate fungal hyphae [A] were seen amidst necrotic tissue and muscle fibers (Fig.2). On Gram staining, pus cells, gram positive cocci and gram negative bacilli were seen. On fungal culture, by the fourth day on Sabouraud’s dextrose agar, a white cottony fast growing fungus with sparsely septate hyphae was isolated. The fungus failed to sporulate, making identification difficult. Subculture and slide culture was made on tap water agar which is a nutritionally deficient medium. When growth appeared, this was exposed to sunlight for two hours and reincubated. When the slide culture and the growth on the agar was observed, it showed the typical morphological features of Saksenaea vasiformis. After nearly a month after isolation, the sporulation could be induced on tap water agar with exposure to sunlight for 2 hours once in the course of incubation and the fungus was identified.

Since the direct examination of the tissue had shown zygomycetous fungal elements, the patient was treated with amphotericin B and antibacterial antibiotics as per the susceptibility pattern of the bacteria in blood and wound, however in the course of treatment, amphotericin B had to be stopped due to the deteriorating medical condition. The blood culture of the patient had grown Escherichia coli. Blood for fungal culture was negative. The patient succumbed to infection 13 days after admission, with bacterial septicemia and zygomycosis.

**DISCUSSION**

Sixteen percent of all zygomycetous infections involve cutaneous tissue, with an associated mortality of 16%. In the pathogenesis of the cutaneous infections, local predisposing factors appear to play a major role rather than a metabolic or systemic illness, or an immunocompromised condition [4]. Saksenia vasiformis appears to have a predilection for traumatised tissue. Eleven out of 20 cases reviewed by Al-Hedaithy had history of injury to the skin in the form of an injection, arterial site puncture, tattoo, surgery etc. prior to developing the infection [5]. Burns have been reported as a predisposing factor in three patients [1]. In our presently described case, the burnt necrotic tissue as well as the neurological and bedridden condition of the patient appear to have resulted in an ulcer on the lumbosacral area. Contamination of the wound due to incontinence of stools and urine predisposed to secondary bacterial infection adding to the complication. Fungal infection also set in, in the traumatized tissue resulting in a devastating and ultimately fatal infection. The patient also had bacterial septicemia which contributed to his death. Patients with reduced sensation do not feel the developing bedsore and continue to lie on it with resultant maceration and worsening of the condition. Fungal infection is known to occur in pressure sores, like in diabetic feet. In a study, fungal infection was found in 65% with a majority of yeast infection and moulds in 38%. Basidiobolus ranarum was the only zygomycetous mould recovered in this study [6].

To the best of our knowledge, our case is the fourth case of Saksenaea vasiformis associated with burns and the first with pressure ulcer. Paradoxically, in one case of chronic subcutaneous nodular infection by Saksenia vasiformis probably following a thorn prick reported by Al-Hedaithy [5], infliction of burns as traditional therapy appeared to have healed the lesions. In our case however, the burnt tissue with pressure necrosis appeared to be the nidus of infection. Hence burns could be a double edged sword and patients should be cautioned about the possible consequences of such alternate therapies.

S. vasiformis grows easily on routine culture media but does not easily sporulate. Induction of sporulation includes sterile distilled water method, Czapek’s agar, saline agar etc. Our
attempt to induce sporulation in the fungus was successful when tap water agar and exposure to sunlight was used. Two new species of Saksenaea have very recently been proposed, S. oblongispora and S. erythrospora based on molecular phylogeny [7].

A greater awareness of such an unusual agent of infection in pressure ulcer and burns and the appropriate sampling of the tissue, (preferably tissue, not swabs) will result in a timely diagnosis which could be life saving. Since this infection occurs generally in traumatized tissue and the fungus is present in soil, proper sterilization techniques for skin piercing instruments, prevention of contact of traumatized tissue with soil, proper care in pressure ulcers etc. should reduce the incidence of such a devastating and potentially life threatening infection.

References
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