Saksenaea vasiformis infections: Case report and literature review

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Abstract
Since the first human infection by Saksenaea vasiformis in 1976 another 26 cases have been reported. Here is a report of a new case which involved an Ecuadorian adolescent who suffered serious burns after a car accident. It developed as a localized cutaneous infection which was successfully treated with surgical debridement and amphotericin B. This is the second report of this infection from South America and the third involving a burn patient. The previously reported 27 cases are reviewed.

Key words: Saksenaea vasiformis, Mucorales, zygomycosis

Introduction
Several genera of the order Mucorales, mainly Rhizopus, Mucor, Absidia, Rhizomucor, Apophysomyces and Saksenaea, are able to cause acute opportunistic infection of rapid evolution which are frequently fatal [1, 2]. The infection may be localized or disseminated. Rhinocerebral form of the diseases is most commonly seen. Less frequent are pulmonary, gastrointestinal, cutaneous and other. Predisposing factors are poor nourishment, poorly controlled diabetes mellitus, kidney insufficiency, use of corticoids, antibiotics, deferoxamin, trauma and burns [2]. The primary cutaneous zygomycosis usually follows traumatic implantation of the fungus by destruction of the skin barrier produced by surgery, use of catheter, severe burns, etc. The most commonly involved body sites are legs, arms, head and abdomen [2, 3]. Several Saksenaea vasiformis infections have been reported in recent years. The first case attributed to this species was reported in 1976 [4] and 26 more cases have been published. It is a rare fungus which has been associated with several cases of localized but also of disseminated infections. It is a disease of worldwide distribution associated to soil contact and its clinical pattern varies from a mild chronic infection to a severe and fatal acute infection. We report the second case of S. vasiformis infection in South America and the third involving a burned patient and review the previously reported 27 cases.

Case report
A 13 year-old male from the rural area of Víeques, in the Ríos Province, Ecuador, was admitted with severe burns due to a domestic accident affecting 65% of the cutaneous surface. Efforts to extinguish the flames included rolling the patient on the ground then applying oil to prevent water or plasma loss. The patient was initially treated at the
Hospital Regional de Vinces with analgesics and cleaning of the wounds and then transferred to the "Roberto Gilbert Elizalde" children's Hospital in Guayaquil. On admission, the patient was conscious, pale, and with a respiratory rate of 28 breaths/min and a pulse rate of 80 beats/min. Twenty-four hours after admission, he was anxious and showed distal frailty, regular perfusion, edema, fever, tachycardia, systolic murmur, microscopic haematuria, leukopaenia and lymphopaenia. He received antibiotic treatment with oxacillin. After Klebsiella pneumoniae was isolated from blood, treatment was changed to cefepime and some improvement was observed. Surgical cleaning of the lesions and dermal grafting was performed. The patient showed fever and malaise. The total white blood cell count was 30,000/mm$^3$ with a predominance of neutrophils (93%) and lymphocytes (6%). After a week, an area of approximately 8 cm diam. localized in the sacrum region showed lysis of the graft and fetid purulent exudates. Serial cultures of the exudates were performed and colonies of a single fungus compatible with a zygomycete were grown. A deep debridement of the involved tissue and treatment with amphotericin B intravenously at dose of 1 mg/kg/day for 14 days was performed, with an evident improvement of the lesions and the patient was discharged.

Exudates were initially cultured in routine media for bacteria, but after 48 h of incubation only a hyaline fungus was developed. Direct preparations of the exudates mounted in KOH and Wright-stained revealed the presence of broad, non-septate, hyaline hyphae, typical of zygomycetes (Figure 1a). The exudates were cultured on Sabouraud dextrose agar (SDA; Difco Laboratories, Detroit, Mich.), with and without chloramphenicol and cycloheximide, and incubated at 28 °C. After 48 h a fungus was grown only on SDA, which hyphae showed the same morphological features mentioned before. However, sporulation was not observed. Sporulation was induced by putting small blocks of SDA agar with the fungal mycelium into distilled water. After 21 days at 28 °C numerous typical vase-shape sporangia of S. vasiformis were developed (Figure 1b), from which numerous cylindrical sporangiospores released through the sporangial opening (Figure 1c).

Discussion

Up to now 28 cases of S. vasiformis infection affecting both immunocompetent and immunosuppressed patients have been reported (Table 1). This is indeed a small number of published cases, considering that this fungus is commonly found in soil samples over the world [2], and probably underestimates the true incidence of this infection in human beings. This could be explained by the fact that this fungus fails to sporulate in routine mycological media, and usually requires the use of special culture techniques such as that of 'floating' of agar blocks of mycelial growth in water for its identification. Once the typical sporangium is developed, the definitive identification of the species is very easy. If not, and only the mycelium is produced, it may be confused with any other zygomycete or discarded as a contaminant [5].

The 28 reported cases corresponded to 19 males (67.8%) and 9 females (32.1%). The median age (range) of the patients was 42.7 years (3 month to 71 years). In most of the cases (19 cases) the portal of entry of the fungus was through a break in the cutaneous barrier. The majority of underlying risk factors were any type of trauma including needle stick [6] and insect or spider bites [7, 8]. Most of the patients were immunocompetent (20 cases), although in some cases some degree of immunosuppression was present. One patient had granulocytic leukaemia [9], one suffered acute lymphoblastic leukaemia [10], three diabetes mellitus [7, 11, 12], one bladder carcinoma [13] and one thalassemia with splenectomy [14]. In one case the immune status of the patient was not indicated [13]. However, the fact that the infection disseminated in only one of these immunocompromised patients [9] seems to demonstrate that although the fungus exhibited a strong predilection for angioinvasion [15], its ability for spreading into the human body is not very high. Two other cases of disseminated infection affecting immunocompetent patients have been reported [16, 17]. In the three cases of disseminated infection where the causes were not identified, it is likely that patients resulted in infection by inhalation of a considerable mass of spores probably when underground or gardening. All patient died as a result of the infection.