Mucormycosis infection following intravenous access in the forearm

Ronit Wollstein MD¹, Alka Palekar MD²

R Wollstein, A Palekar. Mucormycosis infection following intravenous access in the forearm. *Can J Plast Surg* 2010;18(2):e30-e32.

Mucormycosis is an opportunistic infection that is often fatal, requiring aggressive local control as well as systemic therapy. A rare case of a forearm infection originating in a traumatic intravenous access portal is described in the present study. The *Mucor* species infection prevented liver transplant, and the patient passed away. In the present case, it was decided to limit the resection to the skin and subcutaneous tissue based on a frozen section and the viability of the biopsied tissue. With consistently rising numbers of immunocompromised patients, awareness and familiarity with mucormycosis in the extremities is important. Knowing that a minimal traumatic event may precede the infection could assist in prevention and early diagnosis. Guidelines for pathological and clinical diagnosis and treatment need to be further clarified.

Key Words: *Mucormycosis; Trauma; Upper extremity*

Mucormycosis is a rare opportunistic infection classically described in the nasopharynx (1,2). It has been described in the cerebral, pulmonary and gastrointestinal areas, and rarely in the extremities. Infection is often fatal, and treatment requires aggressive local control and systemic therapy.

We describe a patient with a forearm infection that originated from an area of traumatic intravenous (IV) access, preventing liver transplant. With constantly rising numbers of immunocompromised patients, awareness and familiarity with this condition in the extremities is increasingly important.

CASE PRESENTATION

A 48-year-old woman with alcoholic cirrhosis was admitted for acute encephalopathy and decompensation. Background diseases included end-stage liver failure. During her hospitalization, she developed septic shock – the source of which was pneumonia. She required endotracheal intubation and dialysis for renal failure.

Following the appearance of ecchymosis on her right forearm in the area of IV access, she developed an area that seemed necrotic. This patch spread quickly and increased in size over a period of hours. She was taken to the operating room for biopsy and culture; the lesion included the skin and subcutaneous tissue. The subcutaneous fat appeared viable, but was flecked with tiny black spots. The underlying fascia and muscle were biopsied and sent for frozen section. There was no fungus or necrosis detected and, therefore, the excision was limited to the skin and subcutaneous tissue. The *Mucor* species infection prevented liver transplant and the patient's family decided to

Une infection à mucormycose après l'obtention d'un accès intraveineux dans l'avant-bras

La mucormycose est une infection opportuniste souvent fatale qui exige un contrôle local agressif et un traitement systémique. Dans la présente étude, les auteurs décrivent un cas rare d'infection de l'avant-bras découlant de l'obtention d'un accès intraveineux portal traumatique. En raison de l'infection par l'espèce *Mucor*, on n'a pu procéder à une greffe du foie, et le patient est décédé. Dans le cas présent, il avait été entendu de limiter la résection à la peau et aux tissus sous-cutanés d'après une coupe congelée et la viabilité de la biopsie tissulaire. Puisque le nombre de patients immunocompromis est en hausse constante, il est important de connaître la mucormycose des membres et d'y être sensibilisé. Le fait de savoir qu'un événement traumatique minime peut précéder l'infection peut contribuer à prévenir l'infection et à poser un diagnostic rapidement. Il faut apporter davantage d'éclaircissements sur les lignes directrices et sur le diagnostic pathologique et clinique ainsi que sur le traitement de cette infection.

Figure 1) High-power magnification of the skin with the fungus (hematoxylin and eosin stain). Only a small intact portion of the epidermis is seen at the top left, and there is fungal invasion of the blood vessel with some seen in the surrounding tissue. No inflammation or cellular changes are noted; however, there is fat necrosis

withdraw care. No further surgery was performed and the patient passed away. Her family declined a postmortem examination.

Pathology

Several specimens were evaluated. These showed skin and subcutaneous tissue with varying degrees of denudation of the overlying epidermis, with extensive colonization by the hyphae and chlamydospores of *Rhizomucor* species (Figure 1). The hyphal

¹Department of Surgery, Division of Plastic and Reconstructive Surgery; ²Department of Pathology, University of Pittsburgh Medical Center, Pittsburgh, Pennsylvania, USA

Correspondence: Dr Ronit Wollstein, 3550 Terrace Street, Pittsburgh, Pennsylvania 15261, USA. Telephone 412-648-2381, fax 412-648-1987, e-mail wollst1@verizon.net

Figure 2) Low-power magnification of fungus with periodic acid-Schiff special stain showing both components: the hyphae and chlamydozoospores (shown in pink)

Figure 3) High-power magnification of fungus with periodic acid-Schiff special stain showing both components: the hyphae and chlamydozoospores

elements were broad, twisted and ribbon-like, with variable width from 5 μ m to 25 μ m in diameter and nonparallel walls. True septa were not present, although wrinkles and folds in the cell wall appeared as septa. These hyphae also showed nondichotomous, irregular and haphazard branching, which was irregularly spaced with lateral mycelium growing nearly at 90°. The presence of spherical to ovoid, 15 μ m to 50 μ m diameter, thick-walled hyaline chlamydozoospores was noted. Vascular invasion (Figures 2, 3 and 4) and infarction of the adjacent tissue with extensive fat necrosis was evident in all the sections, with no evidence of inflammatory reaction.

DISCUSSION

Although widely described in the transplant literature, cases of limb involvement are rare (3-9). Our patient was immunocompromised due to sepsis in a patient with chronic liver failure. There was an initial insult to the forearm with an IV access. In a case described by Raizman et al (10), the infection seemed to have started similarly in an arterial line and caused necrosis of the hand distally. As in the present case, our patient had

Figure 4) Low-power magnification of fungal invasion of blood vessel with Grocott special stain, which is a silver stain. The fungus appears black with this stain

multiple organ failure and, unfortunately, the final outcome was the same (10). A case of localized mucormycosis was described following an intramuscular injection of corticosteroids. Again, a traumatic incident preceded the infection. The authors concluded that the infection remained localized due to the mildness of the underlying disease (11).

Mucor species infections most often cause suppurative necrosis in the tissues and, occasionally, granulomatous reactions. However, none of the sections in our case demonstrated these changes. No inflammatory cell infiltrate or inflammatory response was noted in our specimens probably due to the immunocompromised status of our patient. The immune status of the patient should, therefore, be taken into account during pathological evaluation.

Mucormycosis must be specifically differentiated from *Aspergillus*, which also invades blood vessels, but is characterized by straight septate hyphal forms with acute angle branching. All diagnoses of fungal infections should confirm the species by culture because histological identification is imprecise, particularly in immunocompromised patients who develop increasingly frequent infections associated with organisms that were previously believed to be nonpathogenic.

In our case, we decided to limit the resection to the skin and subcutaneous tissue based on a frozen section and the viability of the biopsied tissue. It was not clear from this particular patient whether this was the ideal approach because the mere occurrence of the fungus precludes liver transplant.

CONCLUSIONS

A minimal traumatic event may precede the infection. Treating physicians should be aware of this possibility and be proactive in its prevention and early diagnosis. Those cases that survived the infection necessitated a radical resection (in the hand, this might indicate amputation) as well as systemic amphotericin treatment (5,10,12-14). The determination of the necessary extent of resection needs to be further elucidated. From this case and in reviewing the literature, it seems that the general immune status of the patient determines the final outcome more than the particular treatment provided by the surgeon.

REFERENCES

1. Raji MR, Agha FP, Gabriele OF. Case report. Nasopharyngeal mucormycosis. *J Comput Assist Tomogr* 1981;5:767-70.
2. Hoover LA, Hanafee WN. Differential diagnosis of nasopharyngeal tumors by computed tomography scanning. *Arch Otolaryngol* 1983;109:43-7.
3. Tobon AM, Arango M, Fernandez D, et al. Mucormycosis (zygomycosis) in a heart-kidney transplant recipient: Recovery after posaconazole therapy. *Clin Infect Dis* 2003;36:1488-91.
4. Clark FL, Batra RS, Gladstone HB. Mohs micrographic surgery as an alternative treatment method for cutaneous mucormycosis. *Dermatol Surg* 2003;29:882-5.
5. Harada AS, Lau W. Successful treatment and limb salvage of mucor necrotizing fasciitis after kidney transplantation with posaconazole. *Hawaii Med J* 2007;66:68-71.
6. Moses AE, Leibergal M, Rahav G, et al. *Aeromonas hydrophila* myonecrosis accompanying mucormycosis five years after bone marrow transplantation. *Eur J Clin Microbiol Infect Dis* 1995;14:237-40.
7. Peterson PK, Dahl MV, Howard RJ, et al. Mucormycosis and cutaneous histoplasmosis in a renal transplant recipient. *Arch Dermatol* 1982;118:275-7.
8. Reddy IS, Rao NR, Shankar Reddy VM, et al. Primary cutaneous mucormycosis (zygomycosis) caused by *Apophysomyces elegans*. *Indian J Dermatol Venereol Leprol* 2008;74:367-70.
9. Wilcken DE, Mackie JD, Pussell BA, et al. An unusual infection after renal transplantation. *Med J Aust* 1986;145:513-7.
10. Raizman NM, Parisien M, Grafe MW, et al. Mucormycosis of the upper extremity in a patient with alcoholic encephalopathy. *J Hand Surg Am* 2007;32:384-8.
11. Jain JK, Markowitz A, Khilani PV, et al. Localized mucormycosis following intramuscular corticosteroid. Case report and review of the literature. *Am J Med Sci* 1978;275:209-16.
12. de Oliveira-Neto MP, Da Silva M, Fialho Monteiro PC, et al. Cutaneous mucormycosis in a young, immunocompetent girl. *Med Mycol* 2006;44:567-70.
13. Moran SL, Strickland J, Shin AY. Upper-extremity mucormycosis infections in immunocompetent patients. *J Hand Surg Am* 2006;31:1201-5.
14. Foley FD, Shuck JM. Burn-wound infection with phycomycetes requiring amputation of hand. *JAMA* 1968;203:596.