

Mucormycosis in a liver allograft after liver transplantation for secondary biliary cirrhosis due to bile duct injury

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Abstract

Introduction: Mucormycosis refers to a group of opportunistic mycoses that occur generally in immunocompromised patients and are caused by Mucorales, ubiquitous filamentous fungi with broad, thin-walled, sparsely septate, ribbon-like hyphae.

Case report: A 57-year-old man with a history of secondary biliary cirrhosis due to inadvertent bile duct injury during cholecystectomy. He was referred to our center and underwent LT on June 2018. Due to severe coagulopathy he underwent exploratory laparotomy and abdominal packing for 48 hours. He recovered with good liver function and LFT's with a tendency towards normalization. On post op day 8 the patient presented an episode of fever and a CT scan was performed showing a large zone of hypoperfusion with bubbles of gas in the liver dome. A percutaneous biopsy was taken for cultures. Preliminary results reported a filamentous fungus and liposomal amphotericin b was initiated with the suspicion of mucormycosis. The patient remained afebrile and asymptomatic. After 5 days of treatment a new image was performed, and progression of the lesion was noticed, due to these findings the patient was taken to the OR for surgical debridement. Involvement of the liver dome and diaphragm was noticed and a non-anatomic hepatectomy was performed. After surgery the patient required increasing amounts of vasopressors. Despite all the support he progressed to multiple organ failure and finally expired. The product of hepatectomy confirmed the diagnosis of mucormycosis (*Rhizopus* sp).

Discussion: Despite all the efforts the patients' clinical condition deteriorated after surgery showing the high mortality rate in liver transplant recipients that has been reported of 50 to 100%.

Keywords: liver transplantation, fungal infections, mucormycosis, secondary biliary cirrhosis

Introduction

Fungal infections after liver transplantation are becoming more frequent with an incidence between 6 and 38%. Among these fungal infections, mucormycosis represents 2 to 4 % of cases [1-3].

Mucormycosis refers to a group of opportunistic mycoses that occur generally in immunocompromised patients and are caused by Mucorales, ubiquitous filamentous fungi with broad, thin-walled, sparsely septate, ribbon-like hyphae [1]..

Case Report

A 57-year-old man with a history of secondary biliary cirrhosis due to inadvertent bile duct injury during cholecystectomy and underwent a Y-en-Roux hepaticojejunostomy a week after. Posteriorly, he presented multiple episodes of cholangitis that

required long stay hospitalizations, percutaneous procedures and transhepatic biliary drainages. He was referred to our center and underwent LT on June 2018. The donor was a 17-year-old male with brain dead secondary to trauma. Cold ischemia time was 10 hours; warm ischemia time 39 minutes, transfusion of 6 PBRC during surgery and induction was done with basiliximab. Twelve hours after the transplantation, he underwent exploratory laparotomy and abdominal packing, due to severe coagulopathy. He recovered with good liver function and LFT's with a tendency towards normalization. On post op day 8 the patient presented an episode of fever and a CT scan was performed showing a large zone of hypoperfusion with bubbles of gas in the liver dome (Figure 1A). A percutaneous biopsy was taken for cultures. Preliminary results reported a filamentous fungus and liposomal

amphotericin b was initiated with the suspicion of mucormycosis. The patient remained afebrile and asymptomatic. After 5 days of treatment a new image was performed, and progression of the lesion was noticed, due to these findings the patient was taken to the OR for surgical debridement (Figure 1B). Involvement of the liver dome and diaphragm was noticed and a non-anatomic

hepatectomy was performed (Figure 1C and 1D). After surgery the patient required increasing amounts of vasopressors. Despite all the support he progressed to multiple organic failures and finally expired. The product of hepatectomy confirmed the diagnosis of mucormycosis (*Rhizopus sp*) (Figure 2).

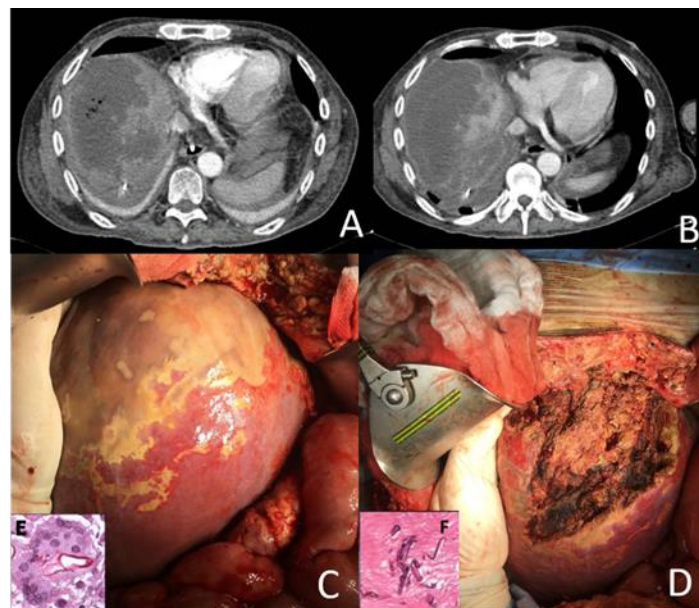


Figure 1. A) CT scan with large zone of hypoperfusion with bubbles of gas in the liver dome. B) CT scan after 5 days of treatment, no gas but progression of lesion. C) Involvement of the liver dome and diaphragm was noticed. D) A non-anatomic hepatectomy was performed.

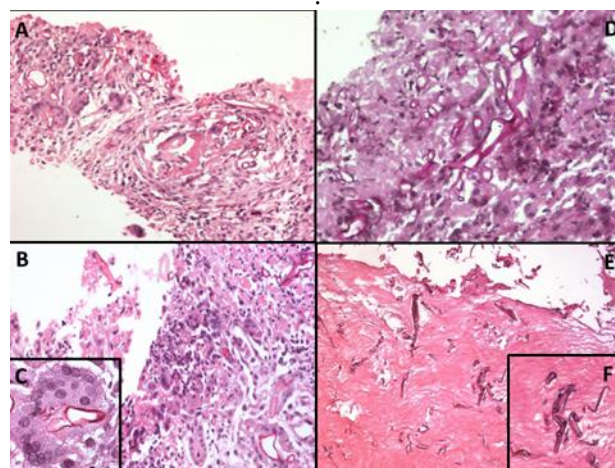


Figure 2. Hematoxylin and eosin staining of hepatic biopsy. **A and B)** 10x extensive ischemic necrosis with hyphae. **C)** 100x multinucleated giant cell with hyphae inside. **D)** Periodic Acid-Schiff Positive Stain, 40x. **E)** 10x. **F)** 20x. Hematoxylin and eosin staining of diaphragm biopsy with the presence of filamentous hyphae.

Discussion and conclusion

Rarely, mucormycosis affects the liver allograft, however, a few cases have been reported [3]. In a review including 116 of mucormycosis in solid organ transplant recipients, infection was localized in 87% of patients and disseminated in 13% [4]. The most frequent sites are rhino-sino-orbital (31%), pulmonary (24.1%), cutaneous (15.5%) gastrointestinal (11.2%) and renal allograft (5.2%) [4]. Several risk factors have been described including diabetes mellitus, cholestasis, hypertransfusion, acute rejection, high-dose steroids or use of OKT3, renal failure,

bacterial infection, retransplantation, malnutrition, Candida and CMV infection [5]. Three modes of transmission have been reported: inhalation, ingestion and percutaneous introduction of spores [6]. Treatment requires liposomal amphotericin B and complete excision of the affected tissue and exceptionally a case with just medical treatment and good outcome has been reported [2,5]. Despite all the efforts the patients' clinical condition deteriorated after surgery showing the high mortality rate in liver transplant recipients that has been reported of 50 to 100% [2].

This case showed a rare case of the disease localized in the liver allograft that progressed despite medical and surgical treatment. Even though surgical debridement is considered as a must for this kind of infections, we keep wondering what would have happened if we only continued with medical treatment (just one successful case). The patient was “doing well” clinically before the surgery but the lesion was progressing in the CT scan, at this point we can only wonder but maybe with surgery, the only thing that we achieved was “unleashing the beast”.

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