Case report

A 61 year old man presented with progressive lower left abdominal pain and constipation since a few months. Colonoscopy showed a large obstructing tumour in the descending colon, and a hemicolecctomy was performed. Histology showed an inflammatory reaction, possibly caused by a fungal or parasitic infection, but no definite identification of an organism was made. Postoperatively his complaints disappeared, but after a few weeks he developed abdominal discomfort in his right upper abdomen, and six weeks postoperatively a CT scan revealed a large mass with a diameter of 6 cm central in the right liver lobe. Treatment with metronidazole, directed against an amoebic liver abscess, was unsuccessful. A subsequent four-week course with fluconazole resulted in a small decrease of the liver abscess, without clinical improvement. He developed a marked eosinophilia (27.7%). A liver biopsy was performed and the patient was referred to a university hospital.

A repeated CT scan showed a large mass with a diameter of 9 cm central in the liver, with extension to the right liver lobe (figure 1A). Review of the slides from the colonic mass and the liver biopsy showed similar features, with extensive necrosis and a mixed inflammatory cell infiltrate containing histiocytes, multinucleated giant cells and numerous eosinophils. In Grocott stained sections, many unusually large hyphae could be recognized which were surrounded by strongly eosinophilic material in haematoxylin and eosin stained sections (Splendore-Hoeplli phenomenon) (figure 2).

On the basis of this morphology, a presumptive diagnosis of infection with Basidiobolus spp. was made. A percutaneous cholangiodrain was placed to treat the cholestasis caused by the hepatic mass. A few days later, the patient developed a septic shock, probably of hepatic origin. Blood cultures yielded Escherichia coli and Clostridium perfringens. The patient was treated with broad-spectrum antibiotics. The presumed basidiobolomycosis was treated with
amphotericin B intravenously, because the preferred therapy, intravenous itraconazole, was contra-indicated because of severe renal insufficiency. A few days after initiation of the antifungal therapy the patient died of multiple organ failure. Postmortem autopsy showed signs of extensive fungal infection of the liver (figure 1B), gallbladder and sigmoid colon. Culture of liver, gallbladder and sigmoid colon yielded *Basidiobolus ranarum*.

Basidiobolomycosis is a rare disease caused by the fungus *Basidiobolus ranarum*, an environmental saprophyte, member of the class *Zygomycetes*, order *Entomophthorales*, found worldwide (1). Usually basidiobolomycosis is a subcutaneous infection that is transmitted through traumatic inoculation (1). Gastrointestinal basidiobolomycosis is rare with only 13 cases reported worldwide in adults (2,3) and 10 in children. (4,5). Only a few cases of retroperitoneal (6,7,8) or pulmonary (9) basidiobolomycosis have been reported. In gastrointestinal basidiobolomycosis the colon is the most frequently involved part of the gastrointestinal tract, and patients usually present with mild abdominal pain with a subacute onset, eosinophilia, and on histopathologic examination inflammatory changes with many eosinophils (2). In contrast to pediatric patients only very few of the reported adult patients also had a hepatic mass (4,5,10). Definitive diagnosis requires culture of the organism, serological testing via an immunodiffusion method can be helpful (1). Because a fungal infection is not always suspected, in a number of patients the diagnosis must be made on histology alone. The fungal morphology and the Splendore-Hoepli phenomenon, although not entirely specific, are characteristic histological features.

There are no prominent risk factors for this disease (2). For treatment surgery is usually required, followed by prolonged antifungal therapy (1,2). The preferred drug is itraconazole (2).
There was difficulty in reaching the diagnosis. The case provides a teaching point, because although gastrointestinal basidiobolomycosis is a rare disease, the clinical presentation of our patient was characteristic for this disease. The prognosis of gastrointestinal basidiobolomycosis is usually favourable, our patient is the second in whom the outcome was fatal (9). Better familiarity with this condition may prevent a fatal outcome like in our patient.
References


Authors contributions and conflicts of interest:

1) I declare that I participated in the clinical care of the patient. I participated in the writing of the case report and I have seen and approved the final version. I have no conflicts of interest. Guido E.L. van den Berk, Infectious disease specialist in training.

2) I declare that I participated in reviewing the samples of colon and liver of the patient described. I participated in the writing of the case report and I have seen and approved the final version. I have no conflicts of interest. L. Arnold Noorduyn, Pathologist.

3) I declare that I participated in the clinical care of the patient described and that I participated in reviewing the samples from colon and liver. I participated in the writing of the case report and I have seen and approved the final version. I have no conflicts of interest. Ruud J. van Ketel, Microbiologist.

4) I declare that I participated in the clinical care of the patient. I participated in the writing of the case report and I have seen and approved the final version. I have no conflicts of interest. Willem A. Bemelman, Surgeon.

5) I declare that I participated in the clinical care of the patient. I participated in the writing of the case report and I have seen and approved the final version. I have no conflicts of interest. Jan M. Prins, Infectious disease specialist.

Authors contributions and conflicts of interest
6) I declare that I participated in the clinical care of the patient. I participated in the writing of the case report and I have seen and approved the final version. I have no conflicts of interest. Jeannouel van Leeuwen, Surgeon
Consent from next of kin

To whom it may concern.

Hereby I, Mrs Rojer, wife of Mr Rojer, the patient described, declare that I agree with the publication of this case report.

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