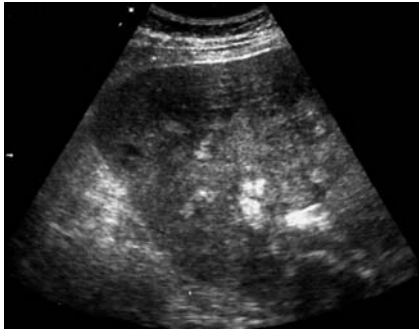


## Fatal case of biliary cast syndrome in nontransplant patient



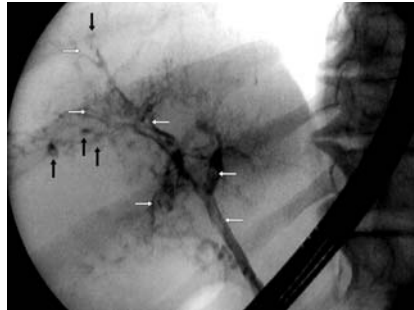
**Fig. 1** Ultrasound of the liver.



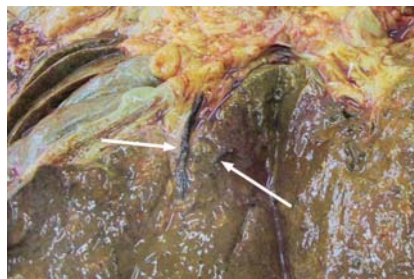
**Fig. 2** Computed tomography.

A 69-year-old male, with no history of liver disease, was admitted after several days of low-grade fever, weakness, and jaundice, preceded by dyspepsia and diarrhea that occurred during recent holidays in North Africa. Laboratory tests revealed anemia (hemoglobin 7.7 g/dL), inflammation (C-reactive protein 148 mg/L, white blood cells 16.6 g/L), and liver injury (alkaline phosphatase [ALP] 426 U/L, gamma glutamyl transpeptidase [GGT] 160 U/L, alanine aminotransferase [ALT] 90 U/L, aspartate aminotransferase [AST] 114 U/L, bilirubin rapidly rising to 22 mg/dL). All possible etiologies of liver injury were excluded. Abdominal ultrasound and computed tomography revealed an enlarged liver with multiple focal lesions requiring differentiation between abscesses, metastases, or hamartomas (▶ **Fig. 1** and **2**).

Endoscopic retrograde cholangiography showed irregular filling defects in extra- and intrahepatic bile ducts, with cystic dilations of intrahepatic ducts, suggestive of microabscesses (▶ **Fig. 3**). Outflow of pus with sludge was observed after



**Fig. 3** Endoscopic retrograde cholangiography. Irregular filling defects (biliary casts) visible in both extra- and intrahepatic bile ducts (white arrows). Black arrows show cystic dilation of intrahepatic bile ducts presumed to be microabscesses.



**Fig. 4** Autopsy. Arrows show biliary casts protruding from bile ducts on the cross-section of the liver.

sphincterotomy. *Escherichia coli* was cultured from bile. Repeated attempts to remove the casts with a Dormia basket were unsuccessful.

The patient's condition deteriorated despite intensive care, and he died of progressive liver failure. On autopsy, the biliary tree, including small branches, was filled with cast replicating the ductal system (▶ **Fig. 4**). Liver histopathology showed diffuse bile stasis in hepatocytes and portal fibrosis with mild inflammation.

Biliary cast syndrome is defined as the presence of casts (caused by retained lithogenic material) within the intra- and/or extrahepatic bile ducts, causing obstruction and cholangitis, eventually complicated by multiple strictures, ductal dilation, and/or liver microabscesses. Biliary cast syndrome used to be a common fatal complication in the early years of liver transplantation, and was related to

poor graft preparation [1,2]. When possible, biliary cast syndrome is treated endoscopically or surgically, including by liver transplantation [3–5].

There are only a few reports of biliary cast syndrome in nontransplant patients [3–5]. In the presented case the infection responsible for travellers' diarrhea and presumably cholangitis was the most probable triggering event in the development of casts. The involvement of small intrahepatic branches made endoscopic treatment ineffective and was responsible for liver failure.

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### Bibliography

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