

Obstructive putty-like cast of the biliary tree

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Abstract: A 62-year-old woman was presented at the emergency department with cholangiosepsis. She had a history of liver metastases for which she had a left hemihepatectomy and loco regional recurrence at the liver hilum for which she received chemoradiation therapy MR-imaging of the liver showed multiple intrahepatic fluid collections/abscesses, together with significantly dilated intrahepatic bile ducts. For her significant hepatic function impairment she received antibiotic treatment and percutaneous drainage. The differential diagnosis was local tumor recurrence or post radiation fibrosis causing outflow obstruction centrally in the liver hilum. During the admission the serum bilirubin values kept rising. During surgery surgeons decided to perform a hepatotomy and connect a loop of the jejunum directly on the liver parenchyma, in order to allow better drainage. Intra operatively, after the hepatotomy, we encountered green-brownish putty-like material. By traction a 'cast' of a large part of the biliary tree was removed.

Keywords: Biliary tree; obstruction; cast; jaundice; cholangiosepsis



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A 62-year-old woman was presented at the emergency department with sepsis, jaundice, general discomfort, abdominal pain and dyspnoea. She had a history of coloncarcinoma for which she underwent a left hemicolectomy [1993], liver metastases for which she had a left hemihepatectomy [2005] and loco regional recurrence at the liver hilum for which she received chemoradiation therapy [2010]. MR-imaging of the liver showed multiple intrahepatic fluid collections/abscesses varying in size, together with significantly dilated intrahepatic bile ducts. The patient was admitted with the diagnosis cholangiosepsis with significant hepatic function impairment for which she received antibiotic treatment and percutaneous drainage. Contrast enhanced 3D rotation cholangiography showed communication between the anteromedial segments and the posterolateral segments of the biletree in the right hemi-liver. The differential diagnosis was abscesses, local tumor recurrence or post radiation fibrosis causing outflow obstruction centrally in the liver hilum.

During the admission the two percutaneously placed transhepatic cholangio drains barely produced any bile fluids and serum bilirubin values kept rising. During

surgery surgeons decided a resection was impossible, as a consequence of too many portal vein collaterals, so another solution to drain the bile was needed. Surgeons decided to perform a hepatotomy towards a superficial biliary duct in the liver and connect a loop of the jejunum directly on the liver parenchyma, in order to allow better drainage. Intra operatively, after the hepatotomy, we encountered green-brownish putty-like material (*Figure 1*). By traction a 'cast' of a large part of the biliary tree was removed (*Video 1*).

The presence of biliary sludge, stones or casts in the biliary tree causing obstruction are referred to as the biliary cast syndrome (BCS). This syndrome was first described in 1975, refers to the development of hardened, dark material within the biliary ductal system that takes the physical shape of bile ducts (1,2). Biliary casts are uncommon; when they do develop they are more commonly reported in orthotopic liver transplant patients. An accurate estimation of the occurrence of bile casts in non-liver transplant patients is not possible due to the scarcity.

The exact pathogenesis of biliary cast remains uncertain and a point of debate among physicians. Biliary infection, bile duct damage and ischaemia, fasting and haemolysis may



Figure 1 Picture taken intraoperative. Putty-like obstructive cast extracted by traction from the bile tree. TOP, top of image; *, towards the right hepatic duct; #, cast of intrahepatic cavity.

all be playing a role in biliary cast formations and stasis.

Biliary cast formation in non-transplant patients is highly unusual; until recent there were only two reported cases of BCS in the literature (3,4). In the non-liver transplant population, very sick patients that are being treated in intensive care units have been shown to develop biliary sludge more frequently and in a much faster pace. In particular patients who undergone abdominal surgery are at risk (5,6). The higher incidence may in part be explained by fasting or total parental nutrition (TPN) that is no uncommon treatment in these patient groups. Fasting and TPN promotes gallbladder hypocontractility due to less secretion of cholecystokinin (7). In theory everything that hinders or interferes with the outflow of bile may enhance the formation of a cast. Foreign bodies such as biliary stents or percutaneously placed transhepatic cholangio drains like the ones placed in this case play a primary role in, stone or cast development. Bacterial attachment on these foreign bodies facilitates initiation of biliary sludge formation in patients with long lasting bile stasis (8).

After diagnosis with for instance ultrasound initial management should preferably be done via endoscopic removal of the obstructive cast, recent literature reported that 60% of the cast patients were successfully treated with endoscopic methods (9). However, there is no recommended standard procedure described in the literature this in combination with the low incidence of the syndrome makes that each case should be viewed individually and thus going for laparotomy isn't necessarily wrong (10). For instance



Video 1 Intra-operative video of the removal of the cast from the biliary tree.

in patients without bile duct access solving the problem endoscopically or percutaneously is very unlikely, those patients will need a laparotomy.

Conclusions

Biliary cast formation is uncommon; especially in non-liver transplant patients. The multidisciplinary HPB team didn't even considered the possibility of an obstructing biliary cast even though most of the risk factors for biliary cast formation are present in the case. Not recognizing the biliary cast in advance by the team did result in performing a hepatotomy with the construction of a biliodigestive anastomoses. Although eventually this surgical procedure turned out to be futile; the patient died unfortunately of ongoing liver failure most likely due to dissolving bile duct walls. Concluding in this case ongoing bile duct obstruction even with percutaneously drains may result in cast formation with life threatening potential.

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