

INTRODUCTION

Coronavirus infection disease 2019 (COVID-19), caused by severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2), was first known as a predominantly respiratory tract infection but now we know that it has the capacity to affect multiple organ systems. According to the literature, gastrointestinal symptoms are associated with COVID-19 in 12% of patients.¹ Anorexia, nausea, vomiting, diarrhea are gastrointestinal manifestations commonly associated with this condition and there is known more and more about biliary complications of the disease. Liver chemistry abnormalities were observed in 15-20% of patients and include elevation of liver enzymes and total bilirubin¹. In the critically ill COVID-19 patient the development of biliary tract disease similar to secondary sclerosing cholangitis is a rarely seen complication (secondary sclerosing cholangitis-critically ill patient, SSC-CIP).⁵ An even more rare complication is formation of biliary casts.⁶

In this report, we present a patient with recurrent abdominal pain, fever and elevated liver enzymes of unknown cause. Abdominal ultrasound showed normal bile ducts. Magnetic resonance cholangiopancreatography revealed long, tubular filling defect in central extrahepatic bile duct (CBD). During endoscopic retrograde cholangiopancreatography (ERCP) a biliary casts were found and removed from CBD. We found out that the patient had experienced a prolonged course of severe critical illness related to COVID-19 with long hospitalisation prior to this episode.

CASE DESCRIPTION

A 71-year old male with a past medical history of hypertension, cardiac failure, diabetes and chronic kidney disease had experienced a prolonged hospitalisation with severe critical illness related to COVID-19 at the beginning of 2021.

In October 2021 he presented to the hospital with signs of acute pancreatitis. Some small gallstones were found in gallbladder on abdominal ultrasound and no biliary dilatation. Endoscopic ultrasound showed no stones in biliary tract. After recovery cholecystectomy was performed.

In January 2022 patient presented with signs of cholangitis – right upper quadrant abdominal pain, fever and elevated liver chemistry tests were noticed. Laboratory studies for active or chronic non -SARS-Cov-2 viral infections were negative. Because of prolonged elevation of liver enzymes MRCP (magnetic resonance cholangiopancreatography) and liver magnetic resonance with intravenous contrast material were performed. Intrahepatic and common bile duct were not dilated. Some short strictures of intrahepatic biliary tree were suspected. About 8 cm long, thin, “worm-like” filling defect was found in CBD and some tiny hyperintense focuses in liver parenchyma, suggestive of small abscesses.

Endoscopic retrograde cholangiopancreatography (ERCP) revealed mild dilatation of CBD and left intrahepatic ducts and irregular filling defects in extrahepatic and left intrahepatic bile ducts. During ERCP sphincterotomy was done and biliary casts were found and removed. Because of cholangitis he was treated with antibiotics and was discharged after significant improvement.

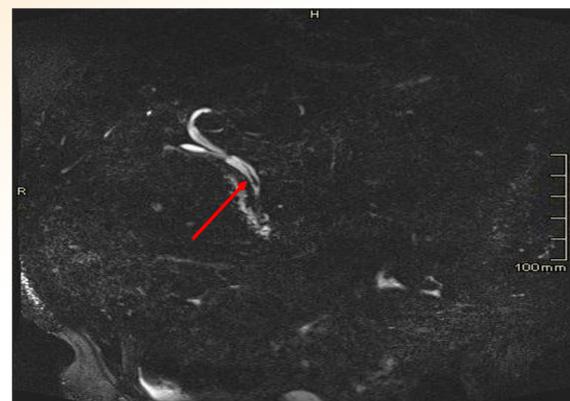


FIG 1: MRCP; Filling defect in CBD, „worm-like“.



FIG 2: ERCP
Biliary cast during removal.
Photo: L. Strniša, MD, Dep. of Gastroenterology



FIG 3: Part of biliary cast after removal.
Photo: L. Strniša, MD

DISCUSSION

Biliary cast syndrome (BCS), first described in 1975, was previously known almost exclusively as a rare complication of orthotopic liver transplantation, especially during the first few years of application of the technique and was associated with increased morbidity, mortality and rejection of the graft.^{7,8} A hardened, dark material, taking the physical shape of bile ducts, develops within biliary ductal system. The causes of the formation are not clearly understood. It is believed that biliary obstruction, infection, a cholesterol-saturated bile and haemolysis play a part, but ischemia is probably one of the main triggers of the syndrome.⁹ Systemic bacterial or fungal infection, which causes intrahepatic cholestasis, may also be a predisposing factor to the formation of casts in the bile ducts after liver transplantation.¹⁰ This can even progress to liver fibrosis and cirrhosis and requires retransplantation.

Liver enzymes elevation occur frequently in patients with COVID-19. Cholangiocyte epithelial expression of angiotensin converting enzyme-2 (ACE-2),¹¹ the host receptor for SARS-CoV-2, may lead to direct viral damage to cholangiocytes (necrosis and destruction of the biliary epithelium, ductular reaction and progressive fibrosis of portal tracts) and eventual chronic cholestasis and liver injury. The microvascular features of hepatic artery endothelial swelling, portal vein phlebitis and sinusoidal obstruction also play a role.

CONCLUSION

Post-COVID-19 cholangiopathy is a new entity, which should be considered as a potential complication of the COVID-19 infection, especially after a severe disease. There were reports of prolonged and severe cholestasis during recovery from critical cardiopulmonary COVID-19 recently. Some of these patients had clinical and histologic features similar to secondary sclerosing cholangitis of the critically ill patient (SSC - CIP) and a few of them had biliary cast syndrome.^{5,6,12} Biliary cast syndrome is treated endoscopically or surgically, including by liver transplantation.

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