

Hyperbilirubinemia 73 mg/dl after Acute Suppurative Cholangitis is not always Fatal, a Case Report

Aya Mohammed Mahros¹, Amr Abdel Shafy², Mohammed H. Emara¹

¹Department of Hepatology, Gastroenterology and Infectious Diseases, Faculty of Medicine, Kafrelsheikh University, Kafrelsheikh, Egypt.

²Department of Nephrology, Faculty of Medicine, Kafrelsheikh University, Kafrelsheikh, Egypt

Corresponding Author
Aya Mohammed
Mahros

Mobile:
+201004039954
E mail:
yoye_85@hotmail.com

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Obstructive jaundice is a common health problem with various complications including liver dysfunction as well as systemic complications. Proper treatment of the underlying etiology should be the primary goal in management. However, different therapeutic approaches have been tried in lowering the bilirubin serum levels when treatment of the underlying cause is not feasible or unsatisfactory. These approaches include extracorporeal liver support (e.g. molecular adsorbent recirculating system (MARS), plasma exchange, and charcoal. To the best of

our knowledge, this is the first case of marked hyperbilirubinemia that was successfully managed with combined ERCP and plasma ultrafiltration. This is a case of acute suppurative cholangitis following choledocholithiasis treated by combined biliary drainage with ERCP and ultrafiltration and although this exceptional high serum bilirubin and impaired renal function, the patient recovered..

BACKGROUND

Obstructive jaundice is a common problem with different etiologies. It does occur due to interference with bile flow from the liver down to the duodenal papilla [1]. The hazards of obstructive jaundice include liver dysfunction as well as systemic complications. Disruption of the intestinal mucosal barrier as a consequence of the decrease of bile in the gut results in an increased absorption of endotoxins and inflammatory cytokines (TNF- α , IL-6) production. As a result, systemic inflammatory response syndrome occurs which may lead to multiple organ dysfunction syndrome. Fatal complication could occur such as hemodynamic instability and acute renal failure, cardiovascular suppression, immune compromise, coagulation disorders [2], so urgent and effective management is mandatory to avoid further organ damage, especially permanent liver damage [3].

Proper treatment of the underlying condition should be the primary goal in management [4]. However, different therapeutic approaches have been tried in lowering the bilirubin serum levels when treatment of the underlying cause is not feasible or unsatisfactory. These approaches include extracorporeal liver support (e.g. molecular adsorbent recirculating system (MARS) [5], plasma exchange, and charcoal [1, 6]. Plasma exchange is a safe and effective method for the clearance of bilirubin and have been tried in many cases with acceptable success rates [7, 8].

Acute suppurative cholangitis, conveys sepsis in the biliary system and is associated with a wide range of morbidity and mortality mainly in the elderly and high risk populations. The

disease clinically is characterized by fever (and chills), jaundice, pain, shock [9]. In most of the cases it follows obstructive jaundice of any etiology, although it is one of the documented adverse events of ERCP and seldom occur without obstruction or interventions [10].

We describe a case of marked hyperbilirubinemia (73 mg /dl) following cholelithiasis acute suppurative cholangitis successfully treated by combined plasma ultrafiltration and internal drainage through ERCP and biliary stenting.

Case presentation

Male patient 42 years old heavy smoker known to have liver cirrhosis for four years presented to our outpatient clinic at Kafrelsheikh University Hospital with one month right upper quadrant colicky pain radiate to the right shoulder, fever and jaundice of one month duration. The condition was associated with clay stool and dark urine. On examination, the patient was very toxic, with deep jaundice and splenomegaly but without ascites. Laboratory evaluation revealed: total bilirubin 55mg/dL, direct bilirubin: 39mg/dL, S. creatinine: 3 mg\dl. Ultrasound examination revealed intrahepatic biliary channel dilatation (IHBC), dilated common bile duct (CBD): 16 mm with multiple stones inside. ERCP was done with pus drainage and multiple

stone extraction and plastic stent insertion; cholangitis was diagnosed. The patient was discharged on demand against medical advice, refused to stay at hospital and did not receive the prescribed antibiotics. Ten days later, the patient came back with sever right hypochondrial pain, deep jaundice and fever. Total bilirubin was 73mg/dL and direct bilirubin: 60 mg/dL. Serum creatinine was 5 mg/dl. Abdominal US revealed no IHBC dilatation. MRCP revealed distal CBD stricture and residual stones. Two sessions of plasmapheresis were done as a trial to reduce the markedly elevated bilirubin. As regards plasmapheresis was done by membrane plasmapheresis using a plasma flux filter P1 with about around 2400 ml plasma volume exchanged during the session. The hemodialysis machine used for plasmapheresis was Fresenius 4008s hemodialysis machine

Two days after the second session, total bilirubin was: 37 mg\dl, Direct bilirubin: 29 mg\dl. Follow up abdominal ultrasound showed non functioning stent, ERCP was done with removal of the old upward migrated occluded plastic stent and insertion of a new plastic stent 10 F/10 cm. The patient was kept on antibiotics and follow up; and 3 days after the second ERCP showed, total bilirubin dropped to 15 mg/dL and 10 mg/dL direct bilirubin. Follow up the patient after 1 months revealed a functioning stent with total bilirubin 2 mg/dL. Three months later his labs were 1.2 mg/dl

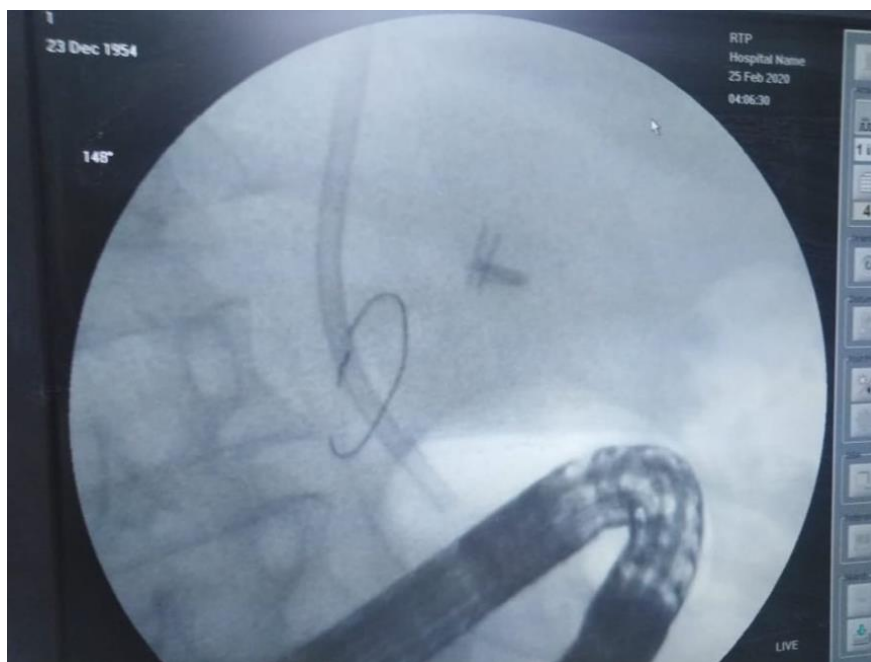


Figure 1: ERCP image showing upwards migration of stent.

DISCUSSION

Our case developed progressive post ERCP cholestasis mostly due to inadequate antibiotic treatment of cholangitis due to premature discharge of the patient from the hospital without receiving the antibiotics. Renal impairment subsequently developed and this markedly worsened the general condition of the patient and threatened his life. The patient was managed with plasma ultrafiltration followed by a drop of the total bilirubin. Unfortunately obstruction of the stent occurred and ERCP was done with stone extraction, extraction of occluded upward migrated stent (**Fig. 1**). Follow up serum bilirubin was done for 3 months till it came to normal.

The presented case here had some interesting clinical aspects. **First**, the patient presented by one month history of improperly treated cholangitis. We do not have enough data about this one month history of illness, but we expect that the underlying chronic liver disease, that is quiet high in our community, directed physicians treating him to thought in hepatic decompensation. Consequently we alarm all physicians treating patients with chronic liver disease to exclude other diagnoses to avoid further delays in proper management. **Second**, the patient was admitted to our hospital with very high serum bilirubin (53 mg / dl) due to suppurative cholangitis and was obvious with the pus flowing from the CBD at time of sphincterotomy. In fact, we achieved internal drainage but the patient insists on discharge and ignored antibiotic therapy. This point of extreme importance because suppurative cholangitis in patients with chronic liver disease would result in high morbidity and mortality [11] and this had been reported in many publications and consequently clinicians treating patients with acute suppurative cholangitis should begin aggressive antibiotic therapy [12].

Third, the exceptionally high level of bilirubin (73 mg/dl) reported in this patient in second admission. In fact, we searched the literature and we did not find similar high level. However, a case report by Daniyeh Khurram and his colleges reported marked elevation of direct bilirubin to the level of (17 mg/dl) due to ceftriaxone use [13]. Whereas Guimarães and his colleges described a case of marked hyperbilirubinemia not responding to plasmapheresis due to sickle

cell anemia with reported serum bilirubin level of 53 mg/dl [14].

Fourth, it seems also that this high bilirubin level affected the renal function with high creatinine level. Although broad spectrum antibiotic therapy was initiated, the general condition did not improve. At that time, we sought advice of our nephrology team. They performed two sessions of plasma ultrafiltration and consequently serum bilirubin dropped to 37 mg /dl. It was reported in the literature that different methods of dialysis may benefit patients with hyperbilirubinemia. The majority of cases were presented with acute liver cell failure treated with MARS [15].

In fact, plasmapheresis or ultrafiltration was tried in treatment of severe hyperbilirubinemia in the centers lacking the facility of artificial ad bioartificial liver support. Novelli tried plasma ultrafiltration to treat patients with high serum bilirubin awaiting liver transplantation [15] and also it was tried in patients with cholestasis with variable rates of success [7]. To the best of our knowledge, this is the first case of ultrafiltration in managing acute suppurative cholangitis with this exceptionally high level of serum bilirubin.

Fifth, the collaborative teamwork. We assume that this patient would have been lost if not managed by the team work between gastroenterologist, endoscopist and nephrologist. Unfortunately, in communities lacking this collaborative health care system, patients like our patient would be lost. Our hospital support this team activity and this reflected on the good outcome we obtained. This case reflect that marked hyperbilirubinemia is not always fatal with proper management.

Sixth, the importance of follow up. Over a 3 months period this critically ill patient was recovered. However, we encourage not only our juniors but also our patients not to be depressed and insist on good outcome provided that proper team management is applied.

Conclusion, this is a case of acute suppurative cholangitis following choledocholithiasis treated by combined biliary drainage with ERCP and ultrafiltration and although this exceptional high serum bilirubin and impaired renal function, the patient recovered.

Abbreviations:

ERCP: endoscopic retrograde cholangiopancreatography, **MARS:** molecular adsorbent recirculating system, **CBD:** common bile duct, **IHBC:** intrahepatic biliary channel, **MRCP:** magnetic resonance cholangiopancreatography.

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Ethical consideration: An informed consent was freely given by the patient.

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