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Case Report

Liver cirrhosis requiring transplantation in the context of hepaticojejunostomy stricture after a traumatic bile duct injury [☆]

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ABSTRACT

Biliary injury secondary to trauma is frequently associated with long-term complications. Liver transplantation is rarely indicated but might be the best therapeutic option in severe or intractable cases. We report the case of a 19-year-old male referred for liver transplantation due to biliary injury after abdominal trauma. A Roux-en-Y hepaticojejunostomy was initially performed without immediate complications. Anastomotic stricture developed requiring several trials of biliary dilatation and stenting through a percutaneous approach. The presence of liver cirrhosis and the intractability of this complication culminated in the decision of liver transplantation. The authors present clinical course, complications and interventional procedures that were used in a judicious step-up approach.

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Case report

A healthy 14-year-old male suffered a laceration of his extrahepatic biliary tree after a blunt abdominal trauma and was evacuated from Cape Verde to Portugal after failure of treatment with cholecystectomy with clinical deterioration and jaundice. A Roux-en-Y hepaticojejunostomy was performed for biliary diversion because the injury extended to the common hepatic duct. A hepatobiliary scintigraphy confirmed bil-

iary drainage after surgery and follow-up with blood tests and abdominal ultrasound was performed every 12 months. The patient did not show any clinical, biochemical, or radiological signs of liver cirrhosis at this stage. Two years later he remained asymptomatic but presented a mild conjugated hyperbilirubinemia associated with a discrete elevation of the liver enzymes. An abdominal ultrasound revealed hepatomegaly, but the patient returned to his country and was lost to follow-up. Four years after the hepaticojejunostomy he presented with aggravated obstructive jaundice (total bilirubin-

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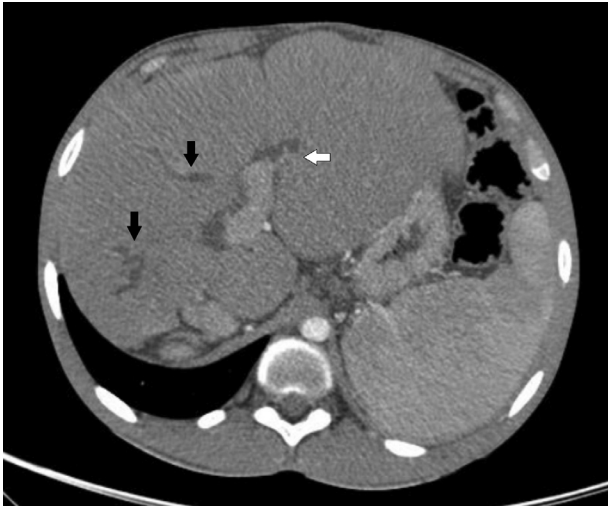


Fig. 1 – Abdominal computed tomography showing dilation of the intrahepatic biliary ducts (black arrows) and portal vein thrombosis (white arrow).

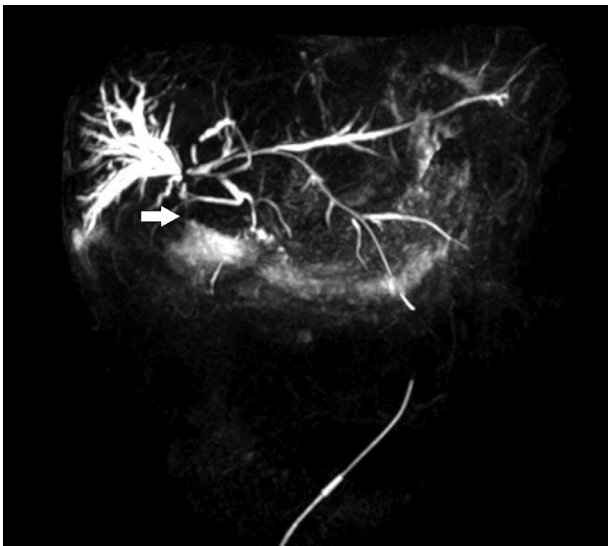


Fig. 2 – Magnetic resonance cholangiopancreatography showing the biliary stricture at the site of the anastomosis (white arrow) along with marked dilation of the intrahepatic biliary tree.

bin of 22 mg/dL), pruritus, and physical signs of chronic liver disease, without abdominal pain, fever, or chills. An abdominal computed tomography (CT) revealed enlargement of the left lobe with atrophy of the right lobe, an irregular surface, thrombosis of the right branch of the portal vein, and dilation of the intrahepatic biliary ducts (Fig. 1). A magnetic resonance cholangiopancreatography (MRCP) showed an anastomotic stricture with marked dilation of the intrahepatic biliary tree (Fig. 2).

An underlying procoagulant condition was excluded (negative anticardiolipin and anti-beta 2-glycoprotein antibodies, normal proteins C and S levels, and absence of JAK2 mu-

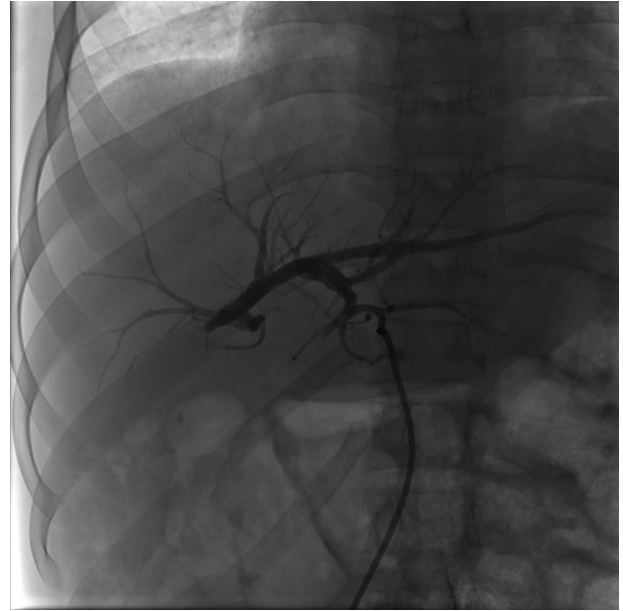


Fig. 3 – Image at the end of the percutaneous transhepatic biliary drainage procedure.

tation). Pylephlebitis due to recurrent ascending cholangitis could not be excluded and the patient was treated empirically with broad-spectrum antibiotics.

Endoscopic resolution of the stricture was considered, but the altered surgical anatomy weighed heavily against this treatment option. A percutaneous transhepatic biliary drainage (PTBD) was attempted instead, but the procedure failed to resolve cholestasis, with subsequent referral to a liver transplantation center due to exhaustion of biliary drainage possibilities and the need for an expert opinion. In our center, another percutaneous biliary drainage was performed with immediate efficacy in terms of improvement of obstructive jaundice and resolution of pruritus (Fig. 3).

However, cholestasis persisted with a gradual increase in total bilirubin and liver enzymes. A subsequent MRCP showed persistent dilation of the intrahepatic biliary ducts. Another percutaneous drainage was tried but the stricture was unsurpassable. An abdominal ultrasound and subsequent CT scan revealed a cirrhotic liver and dilation of the biliary tract. A surgical revision was considered and the patient was discussed by a multidisciplinary team. Five years after the traumatic biliary injury, liver transplantation was regarded as the best therapeutic option due to the condition's intractability and the secondary biliary cirrhosis. Other etiologies were excluded, namely viral hepatitis, Wilson's disease, hemochromatosis, autoimmune etiologies, and sickle cell disease.

At the age of 19 years old, an orthotopic liver transplant was performed. The piggy-back technique was used along with a continuous hepaticojejunostomy without immediate complications. A post-transplant abdominal ultrasound with Doppler showed no dilation of the biliary tract and vascular patency. The patient was discharged from the hospital 18 days later with a total bilirubin of 5 mg/dL and normal prothrombin time and platelet count. One-year post-transplant the patient

is asymptomatic and with a markedly improved quality of life. The liver graft is functioning and the total bilirubin is consistently within normal values.

Discussion

Bile duct injury can occur after abdominal trauma and represents one of the most frequent life-threatening injuries in abdominal trauma patients [1,2]. Biliary injuries can be intrahepatic, extrahepatic, or a combination of both. Extrahepatic biliary injuries are uncommon, occurring in 0.1% of adult and 0.009% of pediatric trauma, and encompass injuries of right and left hepatic ducts, common hepatic and common bile ducts, and cystic duct and gallbladder [3].

Even though extrahepatic biliary tree injury secondary to abdominal trauma is rare, it carries a high morbidity rate since long-term complications are common and resource-consuming [3,5,7]. Prompt recognition of bile duct injuries is crucial to avoid the potentially impaired quality of life and associated financial burden. The most recognized long-term complications are intra-abdominal abscess, cholangitis, biliary cirrhosis, portal hypertension, sepsis, and hepatic or multiple organ failure. Patients with post-traumatic biliary leaks have longer hospital stays, more imaging and interventional procedures, and higher hospital charges compared to similar patients who have sustained blunt or penetrating trauma and do not have bile duct injury [5,8].

Treatment options vary depending on the type of bile duct injury, but include endoscopic, percutaneous, and surgical interventions [5]. Percutaneous or endoscopic interventions should always be considered first, sometimes functioning as a bridge therapy to reconstructive surgery. In major injuries, characterized by a transection or laceration of more than 25% of the bile duct, or when nonoperative management is ineffective, surgery is usually necessary, most commonly Roux-en-Y hepaticojejunostomy [5,9]. Minor injuries that only involve the gallbladder and/or the cystic duct-common hepatic duct junction can be treated with cholecystectomy. Delaying surgical repair or referral for liver transplantation in severe cases can be deleterious [1,3–5,9,10].

Hepaticojejunostomy strictures will occur in up to 20% of patients, most after 1 year or more [1–3]. An anastomotic stricture can result in chronic exposure to hepatotoxic bile acids at the canalicular membrane, possibly leading to fibrosis [6]. A timely diagnosis is vital in order to avoid the development of secondary biliary cirrhosis. The management of anastomotic strictures is initially focused on reestablishing biliary drainage and a step-up approach is recommended, moving on to surgical revision if PTBD with balloon dilatation and stent placement fails [3].

Liver transplantation is the most extreme form of surgical management of extrahepatic biliary tree injury, with limited data supporting its use in an early phase. Commonly, biliary drainage is optimized and the lack of other surgical options is revised before consideration for transplantation. Generally accepted indications for liver transplantation in this context are secondary biliary cirrhosis, acute liver failure, and massive or complex injuries [2,6].

Patients transplanted for secondary biliary cirrhosis have a 3-year survival greater than 70%. Criteria for liver transplantation in the setting of secondary biliary cirrhosis are not well established but the presence of fibrosis and portal hypertension in the setting of an intractable biliary stricture or a poor quality of life are unanimously considered in current literature [6]. Many patients with complicated biliary tree injury are referred for liver transplantation with a prolonged history of chronic intrahepatic cholestasis and advanced liver disease, after years of poor quality of life and multiple procedural interventions.

This case report illustrates how demanding a case of extrahepatic biliary tree injury can be, especially after the failure of the initial management. These patients undergo innumerable imaging and interventional procedures and manifest worse health-related quality of life scores when compared to similar trauma patients who do not have biliary injury [1]. Even after clinically successful treatment, the quality of life may be impaired and a strict follow-up is necessary. Some complicated cases culminate in refractory and intractable stenosis of the hepaticojejunostomy with secondary biliary cirrhosis.

In order to achieve the best possible outcomes in severe extrahepatic biliary tree injury, timely referral to a tertiary center with expertise in hepatobiliary surgery and liver transplantation is essential, not only with the purpose of timely and appropriate evaluation for liver transplant but also to facilitate the access to the multidisciplinary treatment that these patients need [1,3,4,10]. Nowadays liver transplantation is the last line of treatment, but an earlier referral for this treatment may be needed in order to improve outcomes in severe, complicated, or intractable cases, especially after surgical repair is performed.

Patient's perspective

I am happier than ever after having received the liver transplant. Since the accident back in my country that I feel like I lost my childhood... I hated feeling that my clinical situation was always in need of reevaluation. I also understood that the many doctors I visited throughout the years always looked uneasy and worried while looking at my blood analysis and imaging scans. I do not want anyone to go through what I had to, but if they do I hope that a definite solution is offered sooner to them. I admit that I was not the most "dedicated" patient... Even though I was asymptomatic I knew that I shouldn't miss the follow-up visits. However, I was so much happier and free back in my country that I begged my parents to live there, away from the computed tomography machines and the hospital's appointments.

Today I still feel like a teenager but I am completely independent. My parents were always very supportive of me and I am currently studying to enter university. Life is finally getting back on track!

Ethical considerations

Written informed patient consent was obtained for publication of the case details and all accompanying data and images. All information possibly revealing the subject's identity was avoided. Our manuscript complies with manuscript complies with the Declaration of Helsinki and all efforts were made to conceal the identity of the patient.

Patient consent

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