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A BILE DUCT STENT BROKEN DURING REPEAT PREGNANCY IN A POST-LIVER TRANSPLANT PATIENT

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A young female patient who developed anastomotic biliary stricture following an orthotopic liver transplantation was observed. A self-expandable metallic stent was placed to correct the stricture. At the 8th month of her repeat pregnancy, the stent broke asymptomatically into half. Fortunately, the second childbirth, like the first one, had no complications. Eighteen months later, due to obstruction of fragments by sludge and gallstones, re-stenting was performed with a coated biliary stent. Four years and five months later, recurrent jaundice occurred due to occlusion of the second stent. This was addressed by surgical removal of both stents. Two years after surgery, the bile ducts remain completely patent. We found only two cases in literature on a similar extremely rare biliary stenting complication. It has been suggested that stent deformation may be related to pregnancy. The feasibility of using stenting in benign biliary strictures in some clinical situations is discussed.

Keywords: orthotopic liver transplantation, biliary stricture, biliary stenting, broken stent, pregnancy.

Biliary strictures occur in 10–15% of patients following orthotopic liver transplantation (OLTx) [1, 2]. Early anastomotic strictures are associated with technical errors in the operation, and late ones are related to excessive development of scar tissue in the anastomosis area. Modern methods of interventional radiology and endoscopy allow to eliminate this complication minimally and invasively in a number of cases.

We have observed successful correction of anastomotic biliary stricture that developed after OLTx. To correct it, several endobiliary and surgical interventions were required.

CASE

A 24-year-old female patient was diagnosed in 2005 with liver cirrhosis (HCV, HBV), Child–Pugh B, portal hypertension, parenchymal jaundice. In May 2006, she was placed on the waiting list, and in September 2006 OLTx was performed from a cadaveric donor. Biliary reconstruction consisted of anastomosis between the donor's common bile duct and the recipient's hepatic duct end-to-end on a T-shaped drain. She was discharged on day 25 with satisfactory graft function.

In January 2007, 5 months after OLTx, an increase in cholestasis markers was noted. Ultrasound examination of the liver revealed dilatation of the lobular bile ducts up to 8 mm, narrowing in the anastomosis area. Bile duct stricture was diagnosed. An attempt at endoscopic catheterization was unsuccessful. Percutaneous transhepatic cholangiography confirmed the presence of a 10 mm long and 2 mm diameter stricture in the anastomosis area (Fig., a), cholangio drainage was installed.

The patient insisted on removing the drainage because she was planning to become pregnant and then give birth in the near future. We performed dilatation and stenting of the anastomosis area with an uncoated 8 × 60 mm metal stent (S.M.A.R.T® CONTROL, Cordis) with removal of the external-internal drainage (Fig., b, c). Cholestasis phenomena were arrested, bile ducts were markedly reduced in diameter. She was discharged on day 6.

Pregnancy in August 2007, successful Caesarean section delivery in April 2008 (boy). In October 2010, she had another pregnancy, also without complications. A routine ultrasound scan at 8 months revealed an angular deformation in the middle of the stent (60°) with a slight detachment of its fragments. Given the long gestational age, absence of jaundice and complete bile duct patency, it was decided not to perform surgery. In June 2011, she had another cesarean section delivery (girl). At routine examination in October 2011, there were no signs of impaired bile duct patency.

In October 2012, 4 years and 9 months after stenting, obstructive jaundice developed. Ultrasound examination revealed bile duct dilation, stent deformation with impaired patency. X-ray examination confirmed that the stent was broken in half. An attempt at endoscopic removal was unsuccessful; a papillosphincterotomy was performed, and several gallstones were removed. Percutaneous cholangiography revealed that the proximal (closer to the hilum) fragment of the stent deformed and

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blocked the bile ducts with the formation of sludge and multiple gallstones (Fig., d). On the second attempt, all the stones were successfully washed away, an external-internal drainage was installed.

In the next 6 months, we performed 5 balloon plasty procedures on the bile ducts with no success: the ducts were straightened on an 8 mm balloon; but after removing the instrument, angular deformity was formed again; biliary manometric test [3] showed preservation of biliary hypertension above the obstruction. Given the patient's extremely negative attitude to the operation and ineffectiveness of bilioplasty, it was decided to re-stent. A 8 × 60 mm covered biliary stent Wallflex (Boston) was placed so as to match the fragments and eliminate

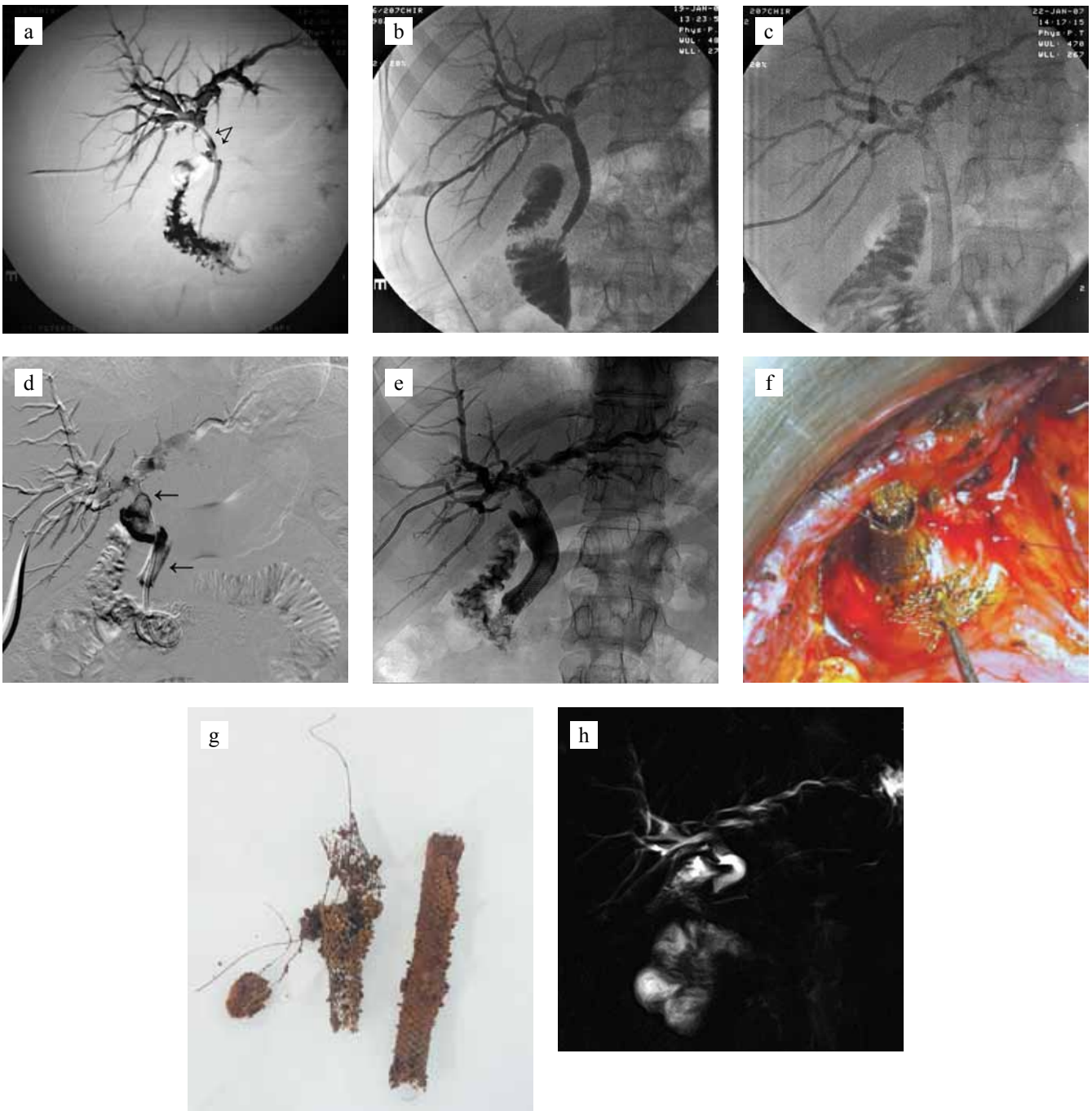

Fig. Radiographic images and intra/postoperative photographs of a patient with post-transplant biliary stricture: a) percutaneous transhepatic cholangiography showing 2-cm anastomotic biliary stricture (arrows); b) after placing a bare nitinol stent S.M.A.R.T. CONTROL, contrast agent flows freely into the duodenum; c) position of stent in the bile ducts; d) fragments of broken stent (arrows), obturated by sludge and stones, cause bile duct obstruction; e) covered biliary stent Wallflex placed coaxially through the broken fragments: normal biliary drainage restored; f) intraoperative photo: a fragment of the nitinol stent captured with forceps, biliary stent occluded by sludge and stones; g) photograph of removed stents; h) magnetic resonance cholangiography 2 years after surgery: bile ducts are completely patent

bile flow obstruction (Fig., e). Hyperbilirubinemia was controlled, the drainage was removed 2 months later, after confirmation of good bile duct patency.

Recurrent jaundice occurred in January 2018, 4 years and 7 months after restenting. Magnetic resonance imaging revealed the cause to be stent occlusion by sludge and gallstones. Open surgery – laparotomy, choledochotomy, stone and stent removal (Fig., f, g) – was performed 2 weeks later. It was decided to leave the distal part of the first stent because it had completely epithelialized and did not interfere with duct patency, and attempts to remove it could significantly complicate the course of the intervention. The operation was completed with leaving the control bile drainage, which was removed after 3 months.

Control examinations (the last one was in May 2020) showed unobstructed duct patency (Fig., h). The patient was feeling well and was receiving 125 mg of Sandimmune on maintenance therapy.

DISCUSSION

Significant anastomotic biliary stricture developing after OLTx is a life-threatening complication that must be corrected. Treatment methods include endoscopic (mostly) or percutaneous transhepatic bilioplasty with or without stenting, surgical reconstruction [1–2, 4–6]. If these methods are unsuccessful, liver resection together with the stricture or even retransplantation may be required.

Treatment of stricture in our patient can be roughly divided into three stages. The first one was percutaneous drainage with insertion of bare metal stents in 2007. Most authors preferred to treat strictures either with balloon plasty, at the same time noting the disadvantages of such approach in the form of treatment duration (6 months and more) and lack of prognostic criteria of success [1–2]. Primary surgical reconstruction comes with a high risk of complications, especially against the background of significant immunosuppressive therapy, and also not always a positive final outcome [1–2, 4]. Stenting rapidly addressed the problem without surgical risk, but the risk of stent occlusion after some time should have been considered [5–7].

Taking into account these factors and the patient's persistent desire to get rid of the problem as soon as possible, we chose stenting, having warned the patient that additional interventions might be required in the future. The jaundice was quickly stopped. The patient successfully gave birth to two children. The stent worked adequately for 4 years and 3 months before it failed.

The second stage of treatment was required due to recurrent jaundice. As a rule, complications from the stent are caused by its "clogging" with sludge/stones or by "neointimal" sprouting. Cases of stent failure have been described in hepatobiliary tumors deforming the stent as the malignant process progresses.

A broken metal biliary stent in a benign stricture is an extremely rare event. We found only two detailed observations in the literature [8, 9]. K. Kawakubo et al. [8] described a patient with benign biliary stricture after liver resection, which was successfully eliminated with an uncovered metal stent. After 4 years, signs of recurrent cholangitis appeared, which was the reason for hospitalization. Endoscopic retrograde cholangiopancreatography (ERCP) demonstrated that the stent was torn in half and the distal of the stent part was floating in the dilated common bile duct. After balloon dilatation of the papilla, the stent fragment was successfully removed with endoscopic forceps.

I. Zuber-Jerger and F. Kullmann [9] observed a 93-year-old patient in whom, after unsuccessful treatment of cholangitis stricture with repetitive plastic stenting, a self-expandable metal stent was also endoscopically installed. One year later, the patient developed abdominal pain, fever, and jaundice. Duodenoscopy showed an incomplete fracture of the stent. It had migrated into the duodenum. This fragment was removed using an Nd:YAG laser, after which a second stent was placed.

In our case, the stent did not migrate from the bile ducts. It can be assumed that the possible cause of its breaking was ductal deformation during two pregnancies. This reason is also supported by the timing of stent fragmentation: the 8th month of the second pregnancy, when the enlarged uterus presses on the liver as much as possible and pushes it to the subcostal area. This mechanism has not yet been considered in literature.

As for treatment, we considered all possible options. An attempt at endoscopic access was unsuccessful. Given the technical possibility of repeated prosthetic replacement, and the appearance of modern covered stents designed specifically for the biliary tract by that time, we chose this option, considering that in case of failure, open surgery will not be ruled out. The patient fully understood the risks of both interventions and preferred a less aggressive approach. The stent was installed coaxially through the fragments such that the fragments of the first stent were almost perfectly aligned, the construction remained passable for 4 years and 7 months.

The third stage consisted in surgical removal of the stents occluded by sludge. Fortunately, there was no need to extend the operation to complex liver reconstruction or resection. There were no signs of bile ducts dysfunction for more than 2 years.

On the whole, the patient's treatment should be considered successful. On one hand, three procedures were required to completely cure the biliary stricture, but two of them were minimally invasive and allowed to provide a good, full-fledged quality of life for 9 years. Unfortunately, attempts at endoscopic stent removal were unsuccessful. The patient underwent a small open surgery without any complications. The total follow-up period

was 13.5 years. Both children, 12 and 9 years old, are growing and developing normally.

The above observation suggests that the use of standard methods (bilioplasty, installation of a temporary stent with endoscopic removal after a few months) is not always possible and effective in a particular clinical situation. Most researchers recommend refraining from stenting benign strictures, but do not deny that there are situations where this method can be justified [1, 2, 6, 7, 10]. We fully share this point of view: there are correct recommendations, and there is a specific clinical situation. The most important thing is the final treatment outcome. An extremely important factor is having a multidisciplinary medical team that would determine the options and sequence of treatment procedures [1, 2].

Introduction of biosoluble stents is expected to open up a certain perspective in the treatment of such patients in the future. The first outcomes of their use look very promising [11, 12].

The authors declare no conflict of interest.

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