

The role of portoenterostomy in the treatment of patients with multiple bile duct trauma: Case series and review of the literature

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ABSTRACT

BACKGROUND: Hepaticojejunostomy is a challenging and complex procedure to be administered with the confidence, in conditions which contain a large number of bile duct damaged by benign pathologies or major bile duct trauma.

METHODS: Here, our clinical series of portoenterostomy (PE), in which we applied in patients who had aggressive hilar dissection for hilar benign biliary pathologies and major bile duct traumas during laparoscopic cholecystectomies were discussed in the light of the literature. The PE procedure was performed in the presence of three or more bile ducts that could not be merged. The classic Roux-en-Y style hepaticojejunostomy was performed to prevent postoperative ascending cholangitis. The ropeway system was used when sewing. 6-8 stitches were laid on the back or anterior wall and the sutures were tied on the outside. Thin-long silicone stents placed in the small diameter (2 mm) bile ducts coinciding with the anastomosis line were extended into the jejunum.

RESULTS: This study included six patients who underwent PE between 2015–2019. Five of the cases were male and one was female and the mean age was 70.33 years. Hepaticojejunostomy was performed in two of the four cases with biliary trauma, but the endoscopic and surgical revision was performed due to developing strictures and bile flow was corrected with stents. In these two cases coming from the external center, PE was applied to multiple bile ducts resulting from aggressive hilar dissection. In two patients who developed major biliary tract trauma (Strasberg-Bismuth-E4) at our hospital underwent PE in the same session. In the other two cases, PE was performed due to a large number of bile ducts caused by benign pathology-related complications (Mirizzi syndrome, Type 4). The mean follow-up period for six patients was 20.1 months (range 11 to 37 months).

CONCLUSION: Portoenterostomy can be performed as a salvage procedure in cases where multiple biliary tracts occur and hepaticojejunostomy is inadequate. PE can be safely used in selected cases that had benign pathologies, major bile duct trauma, in the presence of intense fibrosis, inflammation, very thin bile ducts and more fragile tissues in the liver hilum. PE should be performed in centers with surgeons experienced in hepatobiliary surgery. However, to better understand the efficacy of PE, large multicentric clinical series and patient follow-up are required.

Keywords: Benign biliary pathologies; multiple bile duct injuries; portoenterostomy; treatment.

INTRODUCTION

Portoenterostomy (PE) is the standard procedure for the treatment of biliary atresia.^[1,2] In addition, there are very few literature data on its use and other indications.^[3–6] It is a chal-

lenging and complex procedure to maintain hepaticojejunostomy (HJ) involving multiple subsegmental bile ducts damaged by benign pathologies or trauma. There are severe complications, such as the development of fistula, cholangitis, and stricture, as well as the technical difficulties during the treatment

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of major injuries involving biliary bifurcation.^[5,7,8] Endoscopic and interventional procedures to be applied in patients with bile duct stricture may adversely affect the comfort of life. The prolongation of the process may lead to sepsis, hepatolithiasis, biliary cirrhosis and ultimately cancer development due to cholangitis.^[7] Many patients are trying to survive with external and internal stents in this process. Here, we aimed to present our preliminary results of our PE experience between multiple subsegmental bile ducts and jejunum in cases where aggressive dissection is needed in hilar areas due to benign biliary pathologies and major biliary traumas.

MATERIALS AND METHODS

In the last four years, patients who were admitted to our hospital for bile duct trauma, benign pathologies were included in this study (Figs. 1a and 2a). Patients who underwent PE from patients who had biliary trauma in our hospital and were re-

ferred from other centers were included. As a routine procedure, written-signed informed consent form was obtained from all patients. İzmir Katip Çelebi University, Atatürk Training and Research Hospital, Human Research Ethics Committee approved this study: 2019-GOKAE-1086.

The PE procedure was performed in the presence of three or more bile ducts that could not be merged. The classic Roux-Y HJ was performed to prevent postoperative ascending cholangitis.^[1,2,9] The anastomosis was initiated by the portal vein side with interrupted sutures between the jejunum and the lateral wall of the bile duct (Fig. 2b). In the gaps between the stitches, the sutures passing through the hilar plate (liver tissue) and jejunum were used. The ropeway system was used when sewing. All the posterior wall sutures were first pre-placed then tied. 4/0 polydioxanone was used as the suture material (PDS® II, Ethicon, Edinburgh, UK). A total of 6–8

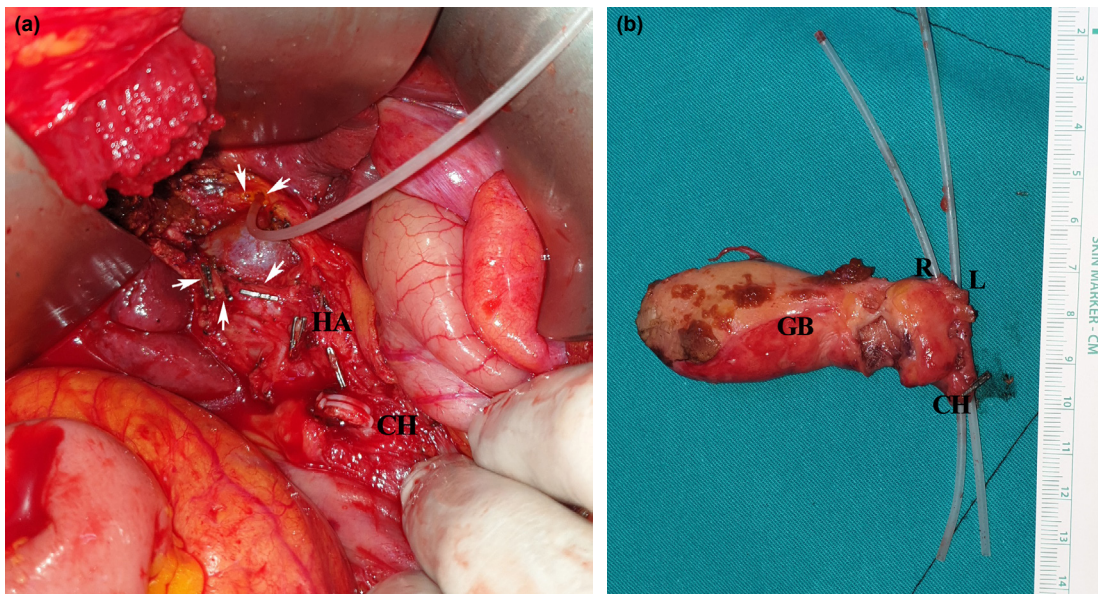


Figure 1. (a) Shows injured or clipped segmental bile ducts (Arrows) and right hepatic artery (HA). (b) Shows right (R) and left (L) hepatic ducts and choledochal end (CH) of the specimen.

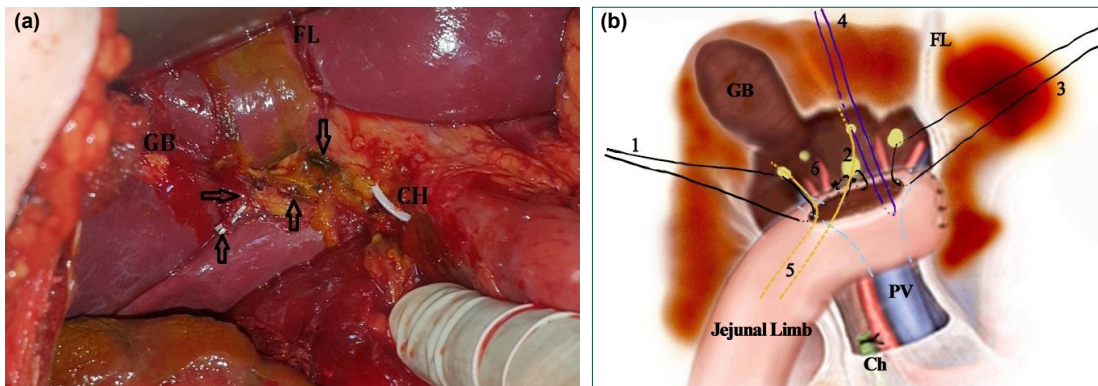


Figure 2. (a) Shows bile duct tips cut or clipped with a sealing device in another patient. (b) Shows the illustration of anastomotic sutures; Corner sutures (1, 3), posterior wall sutures (2), anterior wall suture between the jejunum and thin bile duct (4), and stents (5) for thin bile ducts of the anastomotic line, and free ducts in the center of the anastomosis (6). FL: Falciform ligament; GB: Gallbladder bed; PV: Portal vein and its projection.

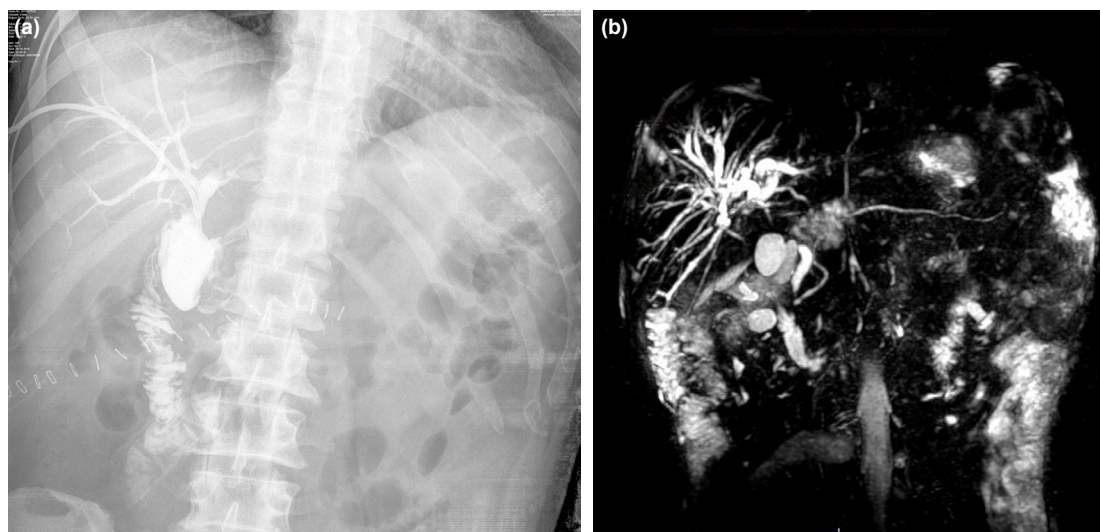


Figure 3. Cholangiography (a) taken postoperative 12th day with the PTC catheter shows the anastomotic patency. (b) Shows the image of a patient who underwent MRCP one year after surgery; anastomoses appear clear.

stitches were laid on the back wall and the sutures were tied on the outside. Care was taken to keep the jejunal mucosa inside. Thin-long silicone stents placed in the small diameter (2 mm) bile ducts coinciding with the anastomosis line were extended into the jejunum (Fig. 2b). The long part of the stents (2/3, approximately 8–10 cm) was placed in the jejunum as unbound, which would subsequently fall under the influence of bowel peristalsis. The bile ducts left in the middle of the anastomosis were released. A suture was passed through the outer walls of the small bile ducts, while the 2–3 sutures were crossed through the wide bile ducts that coincide with the corners of the anastomosis. More on the front, the remaining gaps between the sutures were closed with sutures passed through the liver tissue. After the end of the anastomosis, to decrease the tension, 2–3 approximation sutures were placed in the surrounding tissues. A Jackson-Pratt drain was placed in the operation area.

Patients who needed ventilator support postoperatively were followed up in the intensive care unit. Antibiotic, parenteral nutrition and early enteral nutritional support were routinely used. No additional procedure was taken for the bile drainage that was seen and lost in the first week. Stents, which were put into thin bile ducts, fell spontaneously. Anastomosis status and stents were checked with PTC or MRCP on the 10th–15th postoperative day (Fig. 3). In patients with PTC stent, the stents were removed first after control cholangiography and the drains inside the abdomen were taken one day later. Ursodeoxycholic acid (UDCA) was started for patients who have hepatolithiasis for 6–12 months period. The patients were monitored at 3–6 months intervals. In the patients who came to the control, bilirubin, alkaline phosphatase, liver enzymes, amylase, coagulation factors, CRP, leukocyte and routine biochemical parameters were evaluated. USG, ERCP, and MRCP were performed in patients with clinical complaints of pain, fever and jaundice.

RESULTS

Our hospital is 1100 beds and serves as a reference university and education hospital in the city and region. Approximately 1350–1400 laparoscopic cholecystectomies were yearly performed in our hospital, where 42 general surgeons were working. Surgical intervention was performed in 20 of 105 patients who were followed up with biliary tract trauma in the last four years. Most of the patients with biliary tract trauma were referred from the surrounding hospitals to our center. A total of six patients who underwent PE between the years 2015–2019 included in this study. One of the cases is female, five are male and the average age is 70.33 (range 43 to 86 years). A patient with multiple bile duct trauma (Strasberg-Bismuth-E4), right hepatic artery and right portal vein injury was not included in this study because right lobe hepatectomy and HJ were performed. Four of the patients for biliary tract trauma-related complications and two for benign pathologies (Mirizzi syndrome, Type 3–4) were operated on (Table 1).

Hepaticojejunostomy had been performed in two of the four cases with biliary trauma. A patient who underwent hepaticoduodenostomy and then HJ in other centers due to biliary tract trauma had suffered from anastomotic stricture. This lady had been tried to be treated with endoscopic and interventional methods and was finally replaced with a metal stent, but because of uncontrollable (management) bile duct stricture and accompanying pathologies (hepatolithiasis) was referred to our hospital (Table 1, Patient 2). Due to the severe fibrotic reaction around the metal stents, aggressive hilar dissection had to be performed and PE was added to the procedure. Another 83-year-old patient referred from the outer center also had bilateral hepatolithiasis and often had cholangitis attacks (Table 1, Patient 1). He had previously performed HJ due to biliary tract trauma but developed stenosis and could not be controlled by endoscopic and intervention-

Table 1. Demographic data of the patients

Pt	Sex/Age	History/Etiology	PTC/ERCP	Procedure	Results/Survey
1.	Male/82	Bile duct trauma (SBC-E2) + Roux-en-Y HJ (2012) + Stricture + Hepatolithiasis + Cholangitis	Repeated ERCP + Stone extractions + Stenting	Hilar dissection + EHBDR + Stone extractions + PE (2016)	Bile leakage (2 m), UDCA (1 year) Stable, 37 m
2.	Female/74	Bile duct trauma (SBC-E2), Repair + T-tube drainage (2007) Stricture (2008) + Cholangitis + Hepatolithiasis	ERCP (4) + Dilatation + Stenting (2008) + PTC + Internal-external and metal stenting (2015)	Hilar dissection + EHBDR + Stone/stent extractions + PE (2018)	Stabile, 20 m UDCA (6 m)
3.	Male/43	Bile duct trauma, (SBC-E3) (2017) + Stricture (2018)	PTC stenting (2018) Bil.:12.51	Hilar dissection + PE (2018)	Stabile, 19 m
4.	Male/69	Bile duct trauma (SBC-E4) + Hepatic artery injury (2018)	Intraoperative correction	PE (2018) with stenting	Stabile + CVO (2018) 19 m
5.	Male/68	Mirizzi syndrome (Type 3) + Cholangitis + Abscess (2018)	PTC stenting, Bil.:11.18	EHBDR+ PE (2018)	Stabile, 11 m
6.	Male/86	Mirizzi syndrome (Type 4), Hepatolithiasis?	ERCP stenting, Bil.:6.17	EHBDR+ PE (2018)	Stabile, 14 m

Bil: Bilirubin: mg/dL; COPD: Chronic obstructive pulmonary disease; CVO: Cerebrovascular obstruction; EHBDR-Extrahepatic bile duct (resection); ERCP: Endoscopic retrograde cholangiopancreatography; HJ: Hepaticojejunostomy; m: Month(s); PE: Portoenterostomy; PTC: Percutan transhepatic cholangiography; SBC: Strasberg-Bismuth Classification; UDCA: Ursodeoxycholic acid.

al procedures. Given that confluence is extremely fibrotic in the exploration of the main biliary tract and revision is not possible, dissection of the hilar region was decided. After the stones in the main biliary tract were also removed, the patient underwent PE for four bile duct ends. The fistula was closed due to a 2-month follow-up of the patient who developed a bile fistula in the postoperative period. UDCA is started for 12 months; the patient has been followed up for three years without any problems. In these two cases coming from the outer center, PE was applied to the multiple bile ducts, which had to be performed by aggressive hilar dissection. There was also hepatolithiasis in two patients who underwent surgery due to biliary tract trauma and stricture.

The other two patients who developed major biliary tract trauma at our hospital underwent PE in the same session. In both cases, it was seen that 4–6 bile ducts were clipped or cut during dissection with tissue sealing devices (Figs. 1 and 2, Strasberg-Bismuth-E4).

Two patients with complicated Mirizzi syndrome underwent PE for multiple bile duct injuries after aggressive hilar dissection. In one of these patients, the gallbladder was observed to combine with the confluence of the main bile ducts (Type 4). PE was applied after cholecystectomy and after the appearance of the anatomy since four channels were seen. In our other case, PE was applied to the three ducts resulting from the dissection made due to intense fibrosis and inflammation since the right hepatic duct was observed to be traumatized at the 2nd branch level and could not be combined.

The mean follow-up period for six patients who underwent PE for trauma and benign strictures was 20.1 months (range 11 to 37 months).

DISCUSSION

Major bile duct trauma is a rare but life-threatening condition during surgery for cholelithiasis or benign biliary tract pathologies. HJ is sufficient in most cases where the main biliary tract is cut, but it is challenging to perform HJ in cases with multiple biliary tract injuries and PE may be required. PE is the standard treatment for biliary atresia. There is little clinical information in the literature regarding the use of PE for other pathologies. It was not able to find a place except for salvage PE after major bile duct traumas. However, recently, cases and case series have been reported that the PE procedure has been applied in different hilar pathologies.^[3,4]

Hepaticojejunostomy or hepaticoduodenostomy is usually well tolerated by patients after bile duct injuries. Although hepaticoduodenostomy is easier, the high risk of cholangitis and bile reflux to the stomach is a significant disadvantage. HJ requires more processing, but it is a more preferred procedure because of the low risk of cholangitis and more suitable for hilar injuries.^[5,9] Moraca et al.^[10] used HJ or hepaticoduodenostomy to restore bile duct injuries following laparoscopic cholecystectomy in 27 cases. They followed duodenojejunostomy and HJ for an average of four years and showed that 25 of the cases (92%) biliary functions continue without problems. Pickleman et al.^[5] performed PE in five cases with Bismuth type 4 biliary tract injury and reported that PE could

be used safely in cases in which normal bile duct restoration could not be performed.

In recent years, after surgical treatment of biliary trauma, a consensus has been reached on the follow-up of the patients and the approach to the encountered morbidities. For this purpose, the standards for the degree of biliary trauma, procedures to be performed, whether anastomosis is open, identification of anastomotic stenosis (grading), and algorithms related to surgical or interventional treatments are presented. In short, the approaches are based on algorithms based on whether the biliary tract is open (anastomotic patency) and clinical findings.^[11] For this purpose, clinical, laboratory, endoscopic and radiological diagnostic tools were used in accordance with the proposed algorithms in our patients. Endoscopic and interventional radiologic tools have also been used for treatment.

The development of stricture in the anastomotic line after HJ (12–25%) is a severe problem.^[12–15] The tension in the anastomotic region, the suture material used, the ischemia due to aggressive dissection, the fibrosis and adhesions in the course of the development of the biliary fistula may lead to a biliary stricture in the later period. In cases of PE underwent for biliary atresia and developed an anastomotic stricture, revision results are not very good and transplantation is recommended. The development of stricture in the HJ line after liver transplantation is one of the most feared complications and up to 40% prevalence series have been reported.^[16] de Revuer et al.^[18] reported that they had to perform Roux-en-Y jejunal limb in almost all of the cases despite endoscopic and interventional treatments in patients with anastomotic stricture followed by biliary tract trauma.^[17] In addition, to increase the postoperative quality of life more effectively, they should be treated more aggressively in the early period instead of duct-to-duct anastomosis.^[18] Revisions further disrupt the anatomy of the hilar region and increase the development of fibrosis.^[19] PE appears to be less risky, especially in cases of anastomotic dehiscence and bile leakage.^[6] In our two cases, repeated endoscopic and percutaneous dilatations were made and plastic stents were placed to treat the stricture developed after HJ. However, follow-up and management of the patient after metal stenting in the benign strictures of the biliary tract are challenging. Depending on the stents, inflammation, dense fibrosis and mucosal ingrowth developed, and ultimately, stents became dysfunctional. In these two patients, the cut ends of the four and six subsegmental bile duct were revealed with the extraction of adjacent bile ducts along with the ingrowth metal stent. The stents that were placed with PTC were taken in the early postoperative 15th days after MRCP or PTC cholangiography (Fig. 3). However, a series of stents placed into the bile duct up to 9–25 months have also been reported.^[5] Mercado et al.^[20] reported that they performed HJ in 26 patients for major bile duct trauma and placed transanastomotic stents in nine patients for nine months. HJ was performed on 11 of the 20 patients who underwent surgery due to bile duct

injuries in our clinic. In four of these, minimally invasive procedures (balon dilatation and stenting) and surgical procedures were applied due to the anastomotic stenosis, and in the end PE was performed in two patients. Two patients with stenting who underwent HJ in other centers due to trauma underwent PE surgery (Table 1).

Bile leakage (0.4–8%) is a severe problem after HJ.^[9,12–14] Infection, ischemia, edema, applied techniques and anastomotic tension in the bile ducts that are dissected are the most important causes of bile fistula. We have two cases of biliary fistula, one of whom was seen in the early postoperative period and have a low flow of spontaneous healing in the first week. The other case had a biliary fistula which had spontaneous healing for two months due to a defect on the Roux-en-Y jejunal limb was present. The presence of a missed bile duct ends may also be a cause of persistent bile fistula. In these cases, there are studies suggesting re-anastomosis or ethanol injections.^[15] It may be an advantage if the missed bile ducts remain within the anastomosis area with the PE technique to be applied. Although the appearance of a bile fistula after PE is a severe cause of morbidity, we believe that there will be a problem-free recovery in cases where passage continuity is sufficient and anastomosis is not tense. We think that the silicone stents (Fig. 2b) that we placed in the thin channels during the PE also contributed to the anastomosis to remain open and to facilitate drainage. We recommend routinely placing drains in the anastomosis site. In cases with a PTC catheter, the catheter should be kept in place until control cholangiography is performed.

The suture technique used in an anastomosis is important. Some surgeons prefer interrupted sutures in the anterior wall and continuous suture in the posterior wall, as well as also some surgeons prefer interrupted sutures for the whole anastomosis. In a large survey study in Germany, it has been reported that both techniques are equally preferable when performing HJ and there is no difference in complications.^[12] We used the interrupted suture technique in all cases as described in the method section. Although there is no significant difference between the two techniques, we think that continuous sutures are easier to apply, but due to a technical error, the whole anastomosis will be more adversely affected. It involves more risk in the development of edema and stenosis. We believe that these risks will be less with the interrupted stitches.

Hepatoolithiasis may be seen in addition to persistent and recurrent cholangitis due to bile duct strictures. The most common cause of hepatoolithiasis is bile duct strictures.^[21] Secondary biliary cirrhosis is one of the most severe complications of long-term follow-up after biliary reconstructions and subsequent revisions.^[10] Hepatoolithiasis was observed in two of our patients who had strictures at the place of HJ (Table 1). UDCA was started after PE in both of our patients, and there was no problem in clinical follow-up.

Due to the high bilirubin values in five patients, the ERCP stenting was administered in three patients and PTC stenting in two patients. The reason for this is that the patients usually come with jaundice, and the process is prolonged due to diagnostic tests/procedures and pre-operative preparations. Bilirubin elevation and stent-dependent cholangitis cause delayed treatment and increased morbidity.^[22] Comorbidities also play an effective role in the survival of patients (Table 1).

In conclusion, PE can be performed as a salvage procedure in cases where multiple biliary tracts occur and HJ is inadequate. PE can be safely used in selected cases that have benign pathologies, major bile duct trauma, in the presence of intense fibrosis, inflammation, very thin bile ducts and more fragile tissues in the liver hilum. Less severe problems, such as biliary fistula and stricture in the early postoperative period of PE, appear to be a positive factor in the patient's recovery/resuscitation periods. The patient's escape from external drainage, shorter time, less and later drainage catheter application may be another advantage. PE should be performed in centers with surgeons experienced in hepatobiliary surgery. However, to better understand the efficacy of PE, large multicentric clinical series and patient follow-up are required.

Ethics Committee Approval: This study was approved by İzmir Katip Çelebi University, Atatürk Training and Research Hospital, Human Research Ethics Committee (2019-GO-KAE-I086).

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Peer-review: Internally peer-reviewed.

Authorship Contributions: Concept: O.N.D., A.A.; Design: T.A., F.G.; Supervision: O.N.D., H.B.; Resource: F.G., A.A.; Materials: O.N.D., FG; Data: O.N.D., T.A.; Analysis: A.A., O.N.D.; Literature search: H.B., F.G.; Writing: O.N.D., A.A.; Critical revision: O.N.D., T.A.

Conflict of Interest: None declared.

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OLGU SERİSİ - ÖZET

Çoklu safra yolu travması olan hastaların tedavisinde portoenterostominin yeri: Olgu serisi ve literatürün gözden geçirilmesi**Dr. Osman Nuri Dilek,¹ Dr. Feyyaz Güngör,² Dr. Halis Bağ,² Dr. Turan Acar,¹ Dr. Arif Atay²**¹İzmir Katip Çelebi Üniversitesi Tıp Fakültesi, Genel Cerrahi Anabilim Dalı, İzmir²İzmir Katip Çelebi Üniversitesi, Atatürk Eğitim ve Araştırma Hastanesi, Genel Cerrahi Kliniği, İzmir

AMAÇ: Benign patolojiler veya majör safra yolu yaralanması sonucu ortaya çıkan çoklu safra yolunun yaralandığı olgularda hepatikojejunostominin güvenle yapılması zor ve kompleks bir işlemdir.

GEREÇ VE YÖNTEM: Burada, benign hiler patolojiler nedeniyle yapılan agresif diseksiyonlar sonucunda ve laparoskopik kolesistektomiler sırasında majör safra yolu yaralanması gelişen olgularda uyguladığımız portoenterostomi (PE) olgularımızı literatür verileri ışığında tartışmayı amaçladık. Travmatize olmuş ve birleştirilemeyen üç veya daha fazla safra kanalı varlığında PE tekniğini uyguladık. Asendan kolanjit riskini azaltmak amacıyla klasik Roux-N-Y tipi hepatikojejunostomi tekniğini uyguladık. Anastomozun arka ve ön duvarını yaparken, ropeway (teleferik sistemi) tekniğiyle, tek-tek (separe) 6–8 dikiş kullanarak ve düğümü dışında bağlayarak işlem tamamlandı. İnce safra kanalları (>2 mm) içerisine ince-uzun silikon stentler konularak jejunuma uzatıldı.

BULGULAR: 2015–2019 yılları arasında altı hastamızda PE uygulandı. Hastaların beşi erkek, biri kadın olup yaş ortalaması 70.33 idi. Safra yolu travması gelişen dört olgumuzdan ikisinde hepatikojejunostomi yapıldığı, ancak gelişen striktürler sonucu endoskopik ve cerrahi revizyon yapıldığı ve stentlerle safra akımının düzeltilmeye çalışıldığı saptandı. Bu iki olgumuzda agresif hiler diseksiyon sonucu ortaya çıkan multiple safra kanallarına PE uygulandı. Diğer iki olguda ise majör yaralanma sonucu ortaya çıkan çok sayıda safra kanalı (Strasberg-Bismuth-E4) nedeniyle PE yapılmıştır. Mirizzi sendromu (Tip 4) nedeniyle yapılan agresif hiler diseksiyonlar sonucunda ortaya çıkan çoklu safra kanalları için iki olguda da PE yapılmıştır. Hastalarımızın ortalama izlem süresi 20.1 aydır (11–37 ay).

TARTIŞMA: Portoenterostomi işlemi çok sayıda safra yolunun ortaya çıktığı ve hepatikojejunostominin yetersiz kalacağı durumlarda, kurtarıcı bir işlem olarak uygulanabilir. PE işlemi özellikle benign hiler patolojilerde, majör safra yolu travmaları, yoğun fibrozis ve enflamasyonun olduğu durumlarda, frajil doku ve ince kanalların olduğu durumlarda uygulanabilir. PE işlemi hepatobiliyer cerrahi konusunda deneyimli cerrahların olduğu merkezlerde uygulanmalıdır. Bununla beraber, PE etkinliğini saptayabilmek için çok merkezli, daha geniş ve daha uzun süreli takibi yapılan çalışmalara ihtiyaç vardır.

Anahtar sözcükler: Benign biliyer patolojiler; majör safra yolu travmaları; portoenterostomi; tedavi.

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