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Case Report

Surgical Removal of Inadvertent Portal Thrombosis by Rescue-ALPPS for Perihilar Cholangiocarcinoma

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ABSTRACT

Introduction: Treatment of perihilar cholangiocarcinoma (PHC) usually requires extended resection after inducing hypertrophy of the future liver remnant (FLR). Associating liver partition and portal vein ligation for staged hepatectomy (ALPPS) can achieve rapid hypertrophy of the FLR. Though, due to significant morbidity and mortality, portal vein embolization (PVE) is considered gold standard. Despite remaining controversies, ALPPS might suit as reserve in patients who failed to achieve adequate hypertrophy of the FLR or suffered complications following PVE. We illustrate a rescue-ALPPS after inadvertent nontarget thrombosis of the FLR following PVE in a patient with PHC.

Presentation of Case: A 67-year-old patient requiring right trisectionectomy for PHC Bismuth type IV suffered inadvertent nontarget portal thrombosis of the FLR following PVE. Subsequently, insufficient FLR hypertrophy prevented the planned surgical resection. ALPPS procedure with concomitant thrombectomy of the left portal vein was used as a rescue strategy for this patient.

Discussion: Since ALPPS is associated with significant limitations, especially in patients with PHC, this approach remains controversial. However, surgery still remains the only curative option for patients with PHC and thus, in case of inadequate hypertrophy of the FLR or technical failure following PVE, these patients lack further treatment options. Recent technical refinements and methods of improved patient selection have the potential to emend outcomes of ALPPS in experienced centres.

Conclusion: ALPPS should be considered as reasonable rescue strategy not only in case of insufficient hypertrophy of the FLR but also in the event of technical failure or complications following PVE, even in patients with PHC.

Introduction

Regarding treatment of perihilar cholangiocarcinoma (PHC), surgery still represents the only curative option. Usually extended hepatectomy after inducing hypertrophy of the future liver remnant (FLR) is required [1]. Insufficient FLR is associated with a higher rate of post-hepatectomy liver failure (PHLF), which also significantly affects mortality [2, 3]. Hence, for patients undergoing major hepatic resection a minimum remnant liver volume to total liver volume ratio (RLV-TLV) of 30% respectively remnant liver volume to body weight ratio (RLV-BWR) of 0.5% is recommended to avoid PHLF [4]. Preoperative portal vein embolization (PVE) is considered the gold standard to mediate hypertrophy of the FLR and thereby reduce the risk of developing PHLF [5]. In case of inadvertent non-target thrombosis of the left portal vein during embolization, few treatment options remain for the patient aiming for right trisectionectomy [6]. We hereby present a case of surgical removal of inadvertent nontarget thrombosis of the FLR following PVE in terms of rescue-ALPPS in a patient with PHC in accordance with the SCARE and PROCESS criteria [7, 8].

ALPPS was introduced in 2012 as a new technique, which can achieve enhanced hypertrophy of the FLR prior to extended hepatic resection and thereby improves resectability [9]. Since ALPPS is associated with significant morbidity and mortality, especially in patients with PHC, this

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approach remains controversial [10-13]. The current case report provides a surgical solution treating PVE complications as well as enhancing FLR hypertrophy by in-situ splitting of the liver.

Presentation of Case

We report the case of a 67-year-old patient who initially presented with jaundice and pain in the right upper quadrant of the abdomen to a secondary care centre. He denied nausea, fever, weight loss and night sweat. Endoscopic retrograde cholangiography (ERCP) revealed a stenosis of the common hepatic duct extending a few millimetres into all branches of the biliary trifurcation. These findings supported the suspected diagnosis of perihilar cholangiocarcinoma Bismuth type IV. Brush cytology, however, showed no evidence of malignancy. Subsequently a pigtail stent was placed into the left hepatic duct (double pigtail endoprosthesis 8.5-French). Concerning the right hepatic duct, stenting remained unsuccessful due to configuration of the stenosis. After referral to our surgical department, computer tomography (CT) imaging provided no signs of distant metastases. First, with resolving cholestasis the patient was scheduled for open surgical exploration to establish definitive diagnosis and to evaluate the possibility of right trisectionectomy. Intraoperatively the suspected tumour appeared with a distinct right-sided predominance (segments IV, V to VIII). Histopathology confirmed the diagnosis of adenocarcinoma.

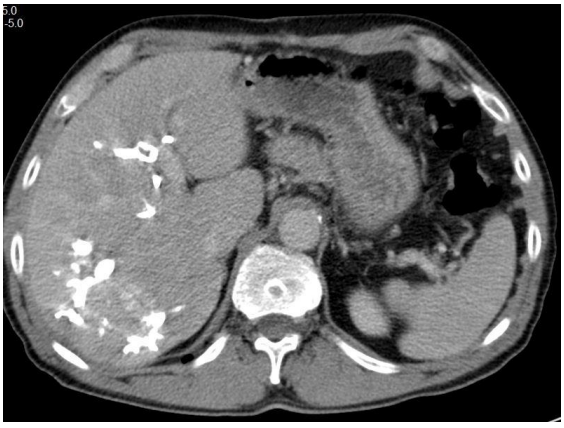

Figure 1: Initial CT volumetry of left-lateral liver segments II and III yielded 161 ml (RLV-BWR was 0.22%) prior to PVE

In view of the present findings and the insufficient volume of the FLR, it was decided to initially aim for augmentation by means of right portal vein embolization prior to right trisectionectomy. CT volumetry of left-lateral liver segments II and III yielded 161 ml (Figure 1). Taking into account the body weight of 72 kg, the RLV-BWR was 0.22% at this time. Due to the low volume of the FLR, a high risk of PHLF was assumed. In order to achieve the minimum RLV-BWR of at least 0.5%, a target volume of 360 ml was required. Subsequently, selective embolization of portal vein branches supplying liver segments IV to VIII succeeds using a histoacryl/lipiodol mixture at a 2:1 ratio. Postinterventional angiographic control indicates occlusion of the right portal vein, including segment IV branches and patent left portal vein. CT follow-up after 24 days showed a FLR volume of 282 ml (RLV-BWR 0.39%). Further imaging studies still resulted in marginal volume increase of FLR 45 (310ml; RLV-BWR 0.43%) and 63 (345 ml; RLV-BWR 0.48%) days after PVE (Figure 2). Inadvertent nontarget thrombus in the left branch of the portal vein caused by PVE was revealed in the CT scan. Partial occlusion of the left portal vein was supposed to be the reason for insufficient hypertrophy of the left lateral liver lobe even 9 weeks after PVE. We opt for proceeding with ALPPS procedure with concomitant thrombectomy of the left portal vein as a rescue strategy for this patient.

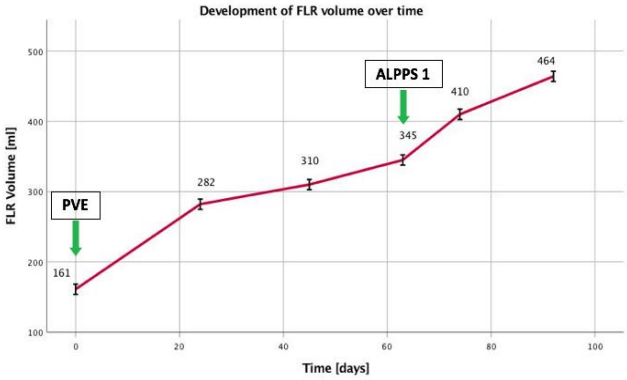

Figure 2: The depicted figure illustrates the development of the FLR volume and the calculated RLV-BWR over time

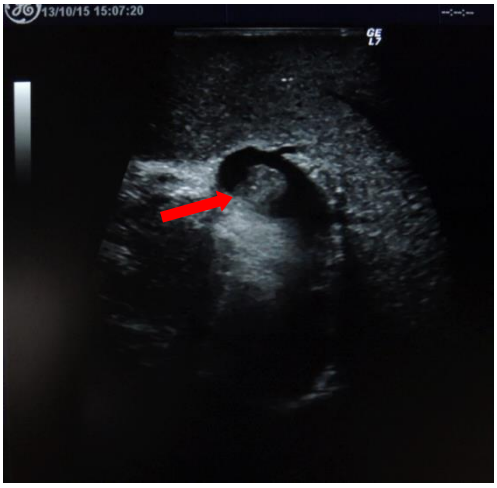

Figure 3: Intraoperative ultrasound imaging of inadvertent nontarget thrombus in the left branch of the portal vein following PVE (indicated by the red arrow)

Intraoperatively, the partial embolization of the left portal vein was confirmed by ultrasound examination (Figure 3). Intrahepatic portion of the left portal vein was exposed by partial transection of the liver parenchyma along the falciform ligament (Figure 4). Under inflow occlusion using Pringle manoeuvre, a venotomy was carried out at the ventral side of the intrahepatic left portal vein. Thrombus was completely removed and venotomy was closed with 6-0 prolene sutures. Ultrasound confirmed the patency of the portal vein. Postoperatively, the patient developed angina pectoris pain. Electrocardiogram (ECG) and coronary angiography diagnosed new-onset atrial fibrillation, Takotsubo cardiomyopathy and one-vessel coronary artery disease (left coronary artery, LAD). However, there was no indication for urgent coronary intervention. An antiplatelet therapy and anticoagulation (grade II according to the Clavien-Dindo) was initiated [14]. Further postoperative course was uneventful. Imaging controls showed slow but sufficient volume increase of FLR (464 ml; RLV-BWR 0.64%) after ALPPS stage one on postoperative day 28 (Figure 5). Finally,

completion of ALPPS stage two could successfully achieved on postoperative day 34 by means of right trisectionectomy including resection of the extrahepatic biliary tree and portal vein bifurcation as well as regional lymphadenectomy. No PHLF or portal vein thrombosis was observed during further course.

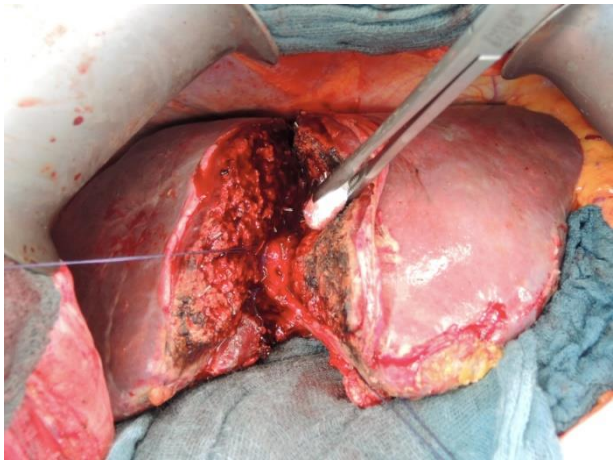

Figure 4: Exposition of the intrahepatic portion of the left portal vein by partial transection of the liver parenchyma along the falciform ligament

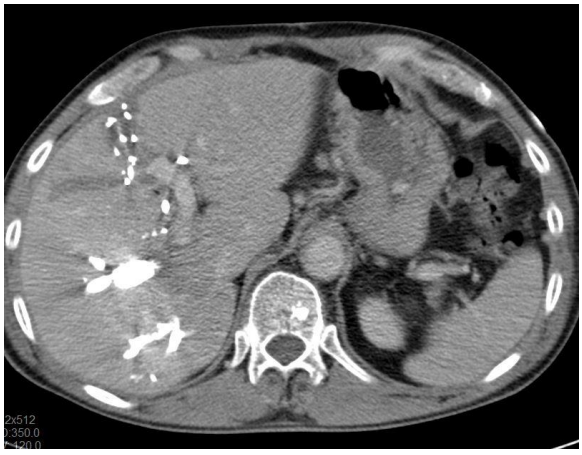

Figure 5: CT volumetry after ALPPS stage one shows sufficient volume increase of FLR (464 ml; RLV-BWR 0,64%) on postoperative day 28

Discussion

After initial enthusiasm driven by the perception of expanding resectability by rapid increase of FLR, ALPPS is appraised increasingly controversial, especially in patients with PHC, due to high morbidity and mortality rates in early reported series of ALPPS [10-13, 15]. PVE remains standard of care for treatment of PHC despite a known drop-out rate of 25% [16]. Migration of embolic material into the main portal or left portal vein is one reason for technical failure after PVE [6, 16]. In such a case, efficient treatment options are scarce [6]. The innovation of ALPPS provides a surgical strategy to handle such complications as demonstrated in the current case report. By splitting the liver parenchyma, the intrahepatic portion of the left portal vein was exposed. Under temporary occlusion of the inflow, the intraluminal thrombosis could be removed without pressure. Additionally, liver transection contributed to further hypertrophy of the FLR. Liver transection in combination of right PVE was reported as hybrid-ALPPS [17]. In-situ liver transection after insufficient volume increase post-PVE was also described as rescue-ALPPS [18, 19]. Both methods lead to adequate hypertrophy of the FLR. Superior surgical outcome of ALPPS in comparison to PVE has been confirmed by recent completed randomized trial [20]. Surgical refinement as well as proper patient selection keep on reducing the morbidity and mortality after ALPPS [13, 21-23]. Referring to this matter, diminution of surgical trauma especially regarding technically demanding first stage is considered key of the revised surgical strategy of ALPPS [24].

We presented the case of a Rescue-ALPPS procedure after inadvertent nontarget thrombosis of the future liver remnant following PVE in a patient with PHC. As previously demonstrated, ALPPS is a considerable rescue strategy in patients who failed to achieve adequate hypertrophy following PVE [18, 19]. Regarding the current report, ALPPS should be taken into account also in the event of technical failure or complications following PVE.

Conclusion

ALPPS should be considered as reasonable rescue strategy not only in case of insufficient hypertrophy of the FLR but also in the event of technical failure or complications following PVE. Particularly, regarding the significant technical refinements after overcoming early learning curve and improved patient selection. With regard to significant morbidity and mortality rates still reported in previous studies, this technically challenging approach remains reserved to experienced centres.

Conflicts of Interest

Jens Rolinger and Jun Li have no conflict of interest.

Funding

Jens Rolinger and Jun Li have no study sponsor.

Ethical Approval

Ethical approval was not necessary for this study.

Consent

We obtained written patient consent to publication.

Author Contribution

All authors designed the study, collected data and interpreted data. Jens Rolinger wrote the paper and Jun Li revised.

Guarantor

Jun Li.

Registration of research studies

Not applicable.

List of abbreviations

ALPPS	Associating liver partition and portal vein ligation
CT	Computer tomography
ECG	Electrocardiogram
ERCP	Endoscopic retrograde cholangiography
FLR	Future liver remnant
LAD	Left coronary artery
PHC	Perihilar cholangiocarcinoma
PVE	ßPortal vein embolization
RLV-BWR	Remnant liver volume to body weight ratio
RLV-TLV	Remnant liver volume to total liver volume ratio

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