



## Case Report

# A Rare Case of Hepatic Artery Pseudoaneurysm by Penetrating Duodenal Ulcer in an Early Living Donor Liver Recipient

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### Abstract

Hepatic Artery Pseudo aneurysmal (HAP) bleed is a dreaded complication following living donor liver transplant. Most HAPs are associated with local milieu environment such as bile leakage, infected collection, pancreatitis and/or following percutaneous trans hepatic interventions. Penetrating duodenal ulcer causing hepatic artery pseudoaneurysm is a very rare cause of HAPs primarily reported in recipients with partial right graft. Herein we present a 59-year-old male who underwent a right lobe living donor liver transplant who presented during the 3rd post-operative week with melena and hematemesis. On endoscopy, an exposed artery segment with sutures was seen at the posterior duodenal bulb. An urgent angiography was done which revealed a hepatic artery pseudo aneurysmal bleed at the site of arterial anastomosis. Endovascular hepatic artery stenting was done across the pseudoaneurysm, which later got thrombosed. An attempt to thrombolysis failed to re canalize the stent graft following which he underwent an exploratory laparotomy to restore the graft perfusion with the extra anatomical splenic artery. He recovered without any ischemic sequelae and is doing well at 1-year follow-up.

**Keywords:** Living Donor Liver Transplantation; Hepatic Artery Pseudoaneurysm; Peptic Ulcer; Duodenal Perforation; Coronary Stent; Interventional Radiology

### Introduction

Hepatic artery pseudoaneurysm (HAP) is an uncommon but potentially life-threatening complication during the early liver transplantation period [1,2]. The time at which HAP appears is variable, but it usually occurs within the first few weeks' post-transplant [1,3]. The outcome is usually catastrophic if not

recognized early and managed accordingly. The location of HAP can be intrahepatic or extrahepatic; intrahepatic being generally secondary to percutaneous trans hepatic interventions [4]. The extrahepatic HAPs are preferentially detected at the site of hepatic artery anastomosis and are attributed to local inflammatory niche caused by bile fluid leakage, peritoneal sepsis (bacterial or fungal) and pancreatitis [1,4]. Primary sclerosing cholangitis and biliary reconstruction with Roux Limb are found to be associated with HAP [5]. The prevalence of Peptic Ulcer Disease (PUD) in cirrhotic is higher than in the general population [6]. Whether this pre-

existing peptic ulcer or stress induced development of ulcer in the early post-transplant period contributes to possible complications (bleeding/perforation) is not clearly known. We encountered a very rare complication of duodenal ulcer penetrating the hepatic artery resulting in hepatic artery pseudoaneurysm. To the best of our knowledge, only nine cases of HAPs attributing to duodenal ulcer bleed have been reported in the literature. Herein we report the diagnostic dilemma and the successful management of HAP presenting as peptic ulcer bleed in an early post-transplant living donor recipient along with the review of literature.

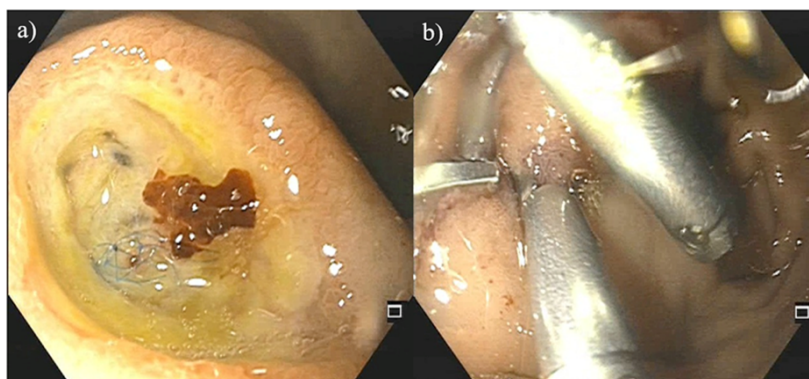
### Case Summary

A 59-year-old male underwent living Donor Liver Transplant (LDLT) with a modified right lobe graft for ethanol related chronic liver disease and underlying hepatocellular carcinoma (Within Milan Criteria) with a MELD sodium of 14. The graft weight was 949 gms with graft recipient weight ratio of 1.16. The segment 5 and segment 8-vein drainage of the anterior sector was reconstruction using Polytetrafluoroethylene graft (PTFEe). The implantation proceeded with the anastomosis of conjoined right hepatic vein and neo middle hepatic vein as a single orifice, end to side with the recipient's inferior vena cava. The portal vein anastomosis was done end to end using 6-0 polypropylene suture, the liver was then re perfused without any congestion of the graft. The graft's right hepatic artery (3.2 mm in diameter) was anastomosed to the recipient's right hepatic artery (5 mm in diameter) with interrupted 8-0 polypropylene in an end-to-end fashion and finally end-to-end bile duct interrupted anastomosis was done with 6-0 PDS. His intraoperative period was uneventful and Doppler ultrasonography at the end of the operation established the patency of the vascular anastomosis. He was extubated on table and immunosuppression was started with a tapering dose of methylprednisolone, tacrolimus being introduced on post-operative day (POD)-2 and myfortic on POD-4. Proton pump inhibitor (PPI) was prophylactically initiated as a mucosal protective agent for peptic ulcer bleeding. Doppler ultrasound was performed twice daily for 5 days followed by once daily for 1 week and showed good arterial flow. He did well in the postoperative period and was satisfactorily discharged on POD-15 without any bile leakage or intraabdominal infective complications. On his 1st follow-up visit he was clinically stable with normal liver functions. Subsequently on POD-22, he presented to the emergency department with complaints of hematemesis and melena for 2 days. On examination, there was pallor, temperature and oxygen saturation were normal. There was mild tachycardia (heart rate 100-120 beats per minute) with a low blood pressure, which responded to fluid resuscitation. Abdomen was soft with no distention. He was on tacrolimus, myfortic, oral prednisolone 20mg once daily and ecosprin 75mg once daily. Haemoglobin was 5.2gm/ dl on admission with normal liver function tests and a normal graft Doppler study without any suspicion of HAP. He

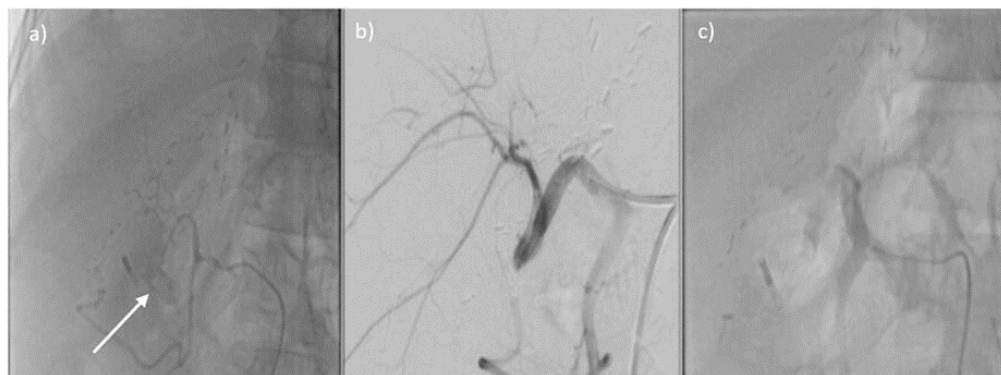
was admitted to the intensive care unit for observation; he was transfused with two units of blood. He was initiated on broad-spectrum antibiotics, PPI infusion and other supportive measures and did not require any inotropes. An urgent upper gastro intestinal endoscopy was done which showed a proximal posterior duodenal ulcer (approx. 2x2 cm size) with a clot and no active ooze.

Post endoscopy he was shifted to ICU for further monitoring. However, on serial laboratory examination there was a further fall in haemoglobin with persisting tachycardia. A triple phase CT angiogram showed normal graft vascularity except for an air-filled duodenal diverticulum. Hence, a repeat check endoscopy with the intent for possible endo therapy was done which detected an ooze from the site of previous duodenal ulcer for which hemoclips were applied. However, to our surprise during the process of hemoclip application polypropylene sutures were noted (Figure 1). This led us to suspect an occult vascular false aneurysm. Since he was hemodynamically stable, direct visceral angiography was done which detected an active contrast extravasation at the site of hepatic artery anastomosis (Figure 2a). After super selective catheterization of the hepatic artery proper, successful placement of an endovascular stent graft (Abbott graft master coronary stent, Abbott Vascular Inc. Santa Clara, CA, USA, 4.5mm/15mm) to cover the pseudoaneurysm was performed. Post procedure angiography showed successful occlusion of the pseudoaneurysm with a good flow to the graft with no contrast extravasation (Figure 2a, b). End procedure Doppler done showed satisfactory arterial wave front and patient was shifted to ICU with an indwelling femoral sheath. Since the repeat Doppler done after 2 hrs did not show any flow across the stent, a 5-Fr angiographic catheter was placed over the proximal part of thrombosed stent using the previously placed right femoral sheath and an intra-arterial catheter thrombolysis was attempted with intermittent reteplase infusion (18mg followed by same bolus dose). However recanalization could not be achieved after adequate cumulative dose (36mg) of reteplase (Figure 2c) and was taken up for an emergency laparotomy. Intraoperatively there were dense adhesions between the raw cut surface of the graft and supracolic compartment viscera, which required meticulous adhesiolysis. A full-thickness ulceration of the superior-posterior aspect of the duodenum adherent to the hepatic artery anastomotic site was noted. The duodenal perforation was repaired with interrupted PDS. The biliary anastomosis was dismantled and the thrombosed hepatic artery was localized with a stent in situ. A hemlok was applied just proximal to the indwelling stent and the distal donor hepatic artery was sharply divided to obtain a single arterial orifice. The available length of the proximal arterial branch was insufficient for tension free anastomosis, hence the decision for revascularization with extra-anatomical splenic artery was taken. The splenic artery was meticulously mobilized from the pancreatic bed with division of the small pancreatic branches and an adequate tension free splenic artery length was obtained. The

artery was swung behind the posterior retro gastric route and an end-to-end anastomosis was done with interrupted 8-0 polypropylene. Bile drainage was restored with Roux-en-Y hepatic jejunostomy. Post re laparotomy he was shifted to ICU and immunosuppressant were reintroduced and daily graft Doppler studies done were normal. He developed a splenic infarct, which was managed conservatively. During the entire course of the event, the peak aspartate aminotransferase was 130 U/L and alanine aminotransferase was 122 U/L. He was discharged on oral immunosuppressant and anticoagulants after 12 days of hospital stay. During the subsequent OPD visits, liver function tests were normal and there were no complications. Also, there was no tumour recurrence at 1 year of follow-up.



**Figure 1:** a. Repeat endoscopy showed bleeding from the duodenal bulb, blue coloured prolene sutures noted at the ulcer base b. Mucosal Hemoclips were applied for control of bleed.



**Figure 2:** a. extravasation contrast at site of hepatic anastomotic genu adjacent to hem clip (arrow) b. covered coronary stent deployed with no extravasation with good graft perfusion c. stent thrombosis after 3 hrs of stent failed thrombolysis.

## Discussion

Hepatic artery pseudoaneurysm revealing as peptic ulcer bleed is a very rare manifestation and the transplant team may be caught unaware by this surprising presentation. Over the decades, the microvascular technique in anastomosis has resulted in a decrease in the overall incidence of HAP from 2.6% (1985 to 1995) to 0.9% (1996 to 2005) [3]. Till February 2023, 4 out of 840 LDLT recipients had HAPs with an incidence of 0.47% which is similar to a recently reported incidence of 0.5% HAPs in LDLT [7]. Since the causative factors related to aneurysm formation such as intimal dissection, roux limb anastomosis, bile leakage and/or local inflammatory collection were not present in our recipient

we can conjecture that the peptic ulcer could have perforated the posterior duodenum causing erosion of the hepatic artery [5]. Whether the ulcer was recent or already pre-existing before the liver transplant could not be determined as no preoperative endoscopy was available in our case. We encountered a diagnostic dilemma as both Doppler and CT angio failed to detect the occult aneurysm. The second check endoscopy performed in view of persisting fall in haemoglobin revealed an ooze from the site of previous ulcer for which hemoclips were applied. The presence of prolene sutures within the ulcer bed during the clip application aroused a suspicion for a possible HAP. For both confirmation and with an intention to treat a direct visceral angiography was conducted which revealed HAP at the anastomotic site. Since the HAP was detected

during the early post-operative period with no major ongoing haemorrhage and without any bile leak or intraperitoneal infection, we preferred endovascular coronary stenting over hepatic artery embolization (HAE). However, despite successful technical deployment of coronary stent there was an early thrombosis of the stent, which could possibly be attributed to the discrepancy in diameters of the anastomotic artery conduits. Following a failed attempt to thrombolysed the stent block surgical revascularization of the graft was performed with the splenic artery. Various studies have reported splenic artery to be a safe and effective extra anatomical bail out technique for graft inflow in situations where anatomical arterial conduits were not present [8,9]. Since the bile duct is supplied solely from the hepatic artery branches, doing a duct-to-duct anastomosis could have jeopardized our recipient to biloma, bile leak and stricture [10]. Hence anticipating this potential risk, the biliary continuity was established with Roux-en-Y hepaticojejunostomy. The successful salvage of our graft without any significant ischemic hepatopathy could be attributed to the maintained hepatic flow except for the short duration of stent thrombosis prior to surgical revascularization. Nine cases of HAPs associated with penetrating DU are reported so far in literature out of which seven were with partial right lobe graft (6 LDLT + 1 Reduced right lobe) and 2 recipients received whole orthotopic deceased graft (Table 1). All the HAPs presented within 2 months of transplant except for one case described by Katlyn et al which presented at 18 months and was associated with prolonged NSAID use for trimalleolar fracture [11]. All the HAPs were extra hepatic in location and located at the site of anastomosis. The most common presentation was hematemesis (8/10 recipients). Hypotension was present in all recipients except in one case (M Kadohisa et al) which presented as anaemia with melena [7]. Only one case of HAP was discovered during exploratory laparotomy for thrombosed hepatic aneurysm while the others were detected at the time of endoscopy [4]. Endoscopic management techniques were varied and seemed only to tide over the initial crises and were later ineffective (Table 2). In 8/9 recipients direct visceral angiography was done for diagnosis and treatment, of which six underwent embolization (5 coil embolization and one glue embolization) while one recipient underwent successful endovascular coronary stent. In one recipient, following angiography TAE was abandoned in view of severe hypotension and was taken up for direct surgical revascularization. In one out of six recipients who underwent immediate surgical revascularization after initial haemorrhage control with embolization, the graft could be salvaged. None of the recipients

survived after embolization without either revascularization or retransplantation. This implies that embolization is only effective for initial control of haemorrhage. Seven recipients survived (one recipient with coronary artery stent, three with surgical revascularization and 3 needed re-transplantations) (Table 2). The poor outcome without revascularization could be due to less time from transplant for development of adequate collateral formation and due to sudden interruption of arterial supply to the graft (Table 1). Association of DU with HAPs was observed mostly in cases with right partial graft. This predilection of HAPs occurrence in the right graft could be due to the following reasons - dissection around the porta, anatomical proximity of arterial anastomosis to the 1st part of duodenum in right lobe graft and exposure of anastomotic site to caustic duodenal fluid contained by the supracolic post-operative adhesions.

The management strategies are quite heterogeneous in literature and clinical decision-making must be done on case-to-case basis [12]. Due to the unpredictable natural history, all HAPs require either a surgical and/or endovascular intervention irrespective of the symptoms or size of the aneurysm [3,5,12]. The decision with regards to management will depend on the following factors: stability of the recipient, presence of bile leak and/or local infections which may be associated with infective arteritis and the availability of interventional radiology (IVR) expertise. IVR, which is less invasive, is currently the preferred method of management of hepatic artery related complications [13]. In our case, we chose IVR due to hemodynamic stability after resuscitation and absence of arteritis, which would have required surgical resection before revascularization. The caveat in the management of HAP is to avoid prolonged hypotension, which could further compound graft ischemia. Earlier reports on HAP primarily focussed on stoppage of ongoing haemorrhage with surgical ligation or embolization of HAP. Since the expectant management strategies with the hope of collateral formation is unpredictable following occlusion of hepatic artery, more and more reports have shifted towards revascularization of the graft. Based on available literature, primary surgical intervention should be considered instead of endovascular stenting in cases of biliary leak and/or peritoneal infection as it could be associated with arteritis meriting resection of the infected arterial segment as well as local debridement. The causative factors for duodenal ulcer formation in the early post-transplant period needs further elucidation. Lack of more number of cases to analyse the contributing factors for DU was the limiting factor in our case review.

Studies	Cases	Age / sex	Aetiology	Symptoms	Hypotension	Type of biliary anastomosis	History of Complications before HAP	Bile leak	Local infection	Location Ulcer/ aneurysm
Present case	1-LDLT	59/M	Hepatitis B and solitary HCC	Hematemesis and melena	yes	D-D	None	no	no	Duodenum bulb (posterior)/ anastomosis
M Kadohisa (2022)[7]	Case 1-LDLT	49/M	Familial amyloid polyneuropathy	Hematemesis	yes	D-D	None	no	no	Duodenum bulb (posterior)/ anastomosis
	Case 2-LDLT	59/M	Hepatitis B and HCC	Melena and anaemia	no	D-D	None	no	no	Duodenal bulb(posterior)/ anastomosis
	Case 3-LDLT	62/M	Idiopathic portal hypertension	Hematemesis	yes	D-D	BAS- ERCP stenting	no	no	Duodenal bulb (anterior)/ bifurcation of anterior and posterior right hepatic artery
Votta K (2019) [11]	1-DDLT	54/M	Non-alcoholic steatohepatitis	Haematemesis and melena	yes	D-D	h/o prolonged use of Ibuprofen for trimalleolar left ankle fracture	no	no	Proximal duodenum/ anastomosis
Lee CC (2008) [14]	1-LDLT	54/M	Hepatitis -B	Hematemesis	yes	D-D	None	yes	no	Proximal duodenal /anastomosis
Slater K (2004) [15]	1-LDLT	38/F	Ethanol related cirrhosis	Hematemesis	yes	D-D	Yes (POD 1 bile leak) exploration convert R-Y	yes	yes	Second part of the duodenum/ anastomosis
MM Marshal (2001)[4]	1-DDLT (out of 13 recipients with HAPs)	NA	Extrahepatic biliary atresia	Melaena falling haemoglobin	yes	R-Y	Sepsis related to subhepatic collections	no	yes	Duodenum / anastomosis
Ruey-Shyang Soong (2009) [16]	Case 1-LDLT	53/M	Hepatitis B	Haematochezia	yes	NA	Preoperative treated with omeprazole for deep duodenal ulcer bulb	no	nil	Duodenal bulb (anterior)/ anastomosis
	Case 2-reduced-size right lobe graft	51/M	Ethanol related cirrhosis	Haematochezia	yes	NA	Gram neg sepsis	no	Yes	Superior wall of the 1st part of the duodenum / anastomosis

**Table 1:** Case summary of baseline characteristics of recipients with duodenal ulcer related HAP in literature.

Studies	Cases	Time of presentation	Location Ulcer/ aneurysm	Reported Endoscopic intervention	IR procedure	Surgical intervention	Reported Outcome
Present case	LDLT	POD 22	Duodenum bulb (posterior)/anastomosis	-Endoscopic clip	Endovascular Stent Stent	Re- vascularisation (splenic artery)	Alive
M Kadohisa (2022) [7]	Case1-LDLT	POD41	Duodenum bulb (posterior)/ anastomosis	-Endoscopic failure to control bleed	Coil embolization	-Interposition with recipient Inferior mesenteric artery	Alive
	Case 2-LDLT	POD 57	Duodenal bulb(posterior)/anastomosis	-Endoscopic control	TAE on POD 68(for rebleed)	none	Multiple liver abscess death
	Case 3-LDLT	POD19	Duodenal bulb (anterior)/bifurcation of anterior and posterior right hepatic artery	-Endoscopic failure to control bleed	Coil embolization	multiple liver abscesses Re-LDLT POD (multiple liver abscesses)	alive
K Votta K (2019) [11]	1-DDLT	19 months	Proximal duodenum/ anastomosis	Failed Endoscopy (endoclip+ epinephrine+ bipolar cautery)	TAE abandoned in view of thrombosed HAP	revascularization	alive
Lee CC (2008) [14]	1-LDLT	POD6	Proximal duodenal /proper hepatic artery	Heat probe Endoscopy failure	Coil embolization	retransplantation (30 months) - for ischemic cholangitis	Alive
Slatter K (2004) [15]	1-LDLT	2months	second part of the duodenum/ anastomosis	Endoscopy suspected aneurysm No intervention	Endovascular stent	none	Alive
MM Marshal (2001) [4]	1-DDLT	29 days post-OLT	Duodenum/ Anastomosis	none	TAE	retransplantation	alive
Ruey-Shyang Soong (2009) [16]	Case 1-LDLT	POD 14	Duodenal bulb (anterior)/anastomosis	Endoscopy failed to identify the bleeding site	Angiography TAE abandoned in view of hemodynamic instability	recipient's right gastric artery (1 mm) to graft's right hepatic artery (2 mm) by microscopic technique	Alive
	Case 2-reduced-size right lobe graft	POD 16	superior wall of the 1st portion of the duodenum/anastomosis	Hemoclip local adrenaline local injected	Embolization with n-butyl cyanoacrylate	graft's celiac artery and recipient's common hepatic artery	Graft failure – Death

LDLT: living Donor LIVER Transplant; DDLT: deceased donor liver transplant; HCC: hepatocellular carcinoma; POD: Post Operative Day; TAE: Trans arterial embolization; D-D: duct to duct; R-Y: Roux-en-Y hepaticojejunostomy; NA- not available

**Table 2:** Summary of Management strategies of recipients with duodenal ulcer related HAP in literature.

**Conclusion**

Recipients with partial right graft presenting with proximal duodenal ulcer bleed in the early post op period should be suspected to have hepatic artery pseudoaneurysms unless proven otherwise. Management should be aimed at early vascularization with either surgical or endovascular stent. Hepatic artery embolization can be used to tide over the active bleed albeit there can be high risk for graft loss and subsequent need for re-transplantation.

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**Conflict of Interest:** The authors of this manuscript have no conflicts of interest to disclose.

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