

Management of a Fulminant Upper Gastrointestinal Bleeding Exteriorized Through Hemobilia Due to Arterio biliary Fistula Between the Common Bile Duct and a Right Hepatic Artery Aneurysm – A Case Report

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Abstract. Right hepatic artery aneurysms are rare events that might remain asymptomatic for a long period of time. However, in cases presenting large lesions, symptoms might develop especially due to the association of compression of the surrounding elements. Most often these symptoms and signs include diffuse abdominal pain, jaundice or portal vein compression signs. In rare cases life-threatening complications might develop due to the aneurysmal erosion of the biliary duct, portal vein or due to the aneurysmal rupture in the peritoneal cavity. In all these cases emergency surgery is imposed. We present the case of a 66-year-old patient diagnosed with a partially thrombosed right hepatic artery aneurysm compressing the common bile duct who was initially submitted to a percutaneous arterial embolization of the aneurysm in association with an external biliary drainage; three weeks later the patient presented a fulminant upper gastrointestinal bleeding exteriorized through the external biliary drainage, hematemesis and hematochezia. The patient was successfully submitted to surgery, intraoperatively a synchronous rupture of the portal vein being revealed. The

right hepatic artery aneurysm was resected en bloc with common bile duct resection and segmental portal vein resection. The continuity of the portal vein was re-established through the interposition of a cadaveric allograft, the common bile duct was anastomosed with en Roux en Y limb while the right hepatic artery aneurysm was ligated and resected, the arterial vascularization of the liver being provided by the left hepatic artery.

Hepatic artery aneurysms represent uncommon lesions, accounting for up to 20% of all splanchnic artery aneurysms and are usually asymptomatic lesions, being discovered during various investigations for other pathologies (1). When it comes to the apparition of hepatic artery aneurysms, the main incriminated mechanism is the atherosclerotic one, followed by endocarditis, necrotizing vasculitis, Ehlers Danlos syndrome, Takayasu’s arteritis and post-traumatic procedures such as liver transplantation, percutaneous transhepatic cholangiography (2-4). In the study conducted by Stanley *et al.* on 162 patients diagnosed with various splanchnic aneurysms, the authors demonstrated that 63% of cases reported common and proper hepatic artery aneurysms, 28% of cases were diagnosed with right hepatic artery aneurysms, 5% of cases presented left hepatic artery aneurysms while 4% of cases presented left and right hepatic artery aneurysms (5). Increasing incidence of reported hepatic artery aneurysms which has been reported in the last decade is mainly explained through the improvement of the imagistic techniques, offering a higher rate of aneurysms’ detection especially among asymptomatic patients (6). Once hepatic aneurysms are diagnosed, performing an arteriography is

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Key Words: Right hepatic artery aneurysm, percutaneous arterial embolization, arterio biliary fistula, aneurysm resection, portal vein resection and reconstruction with cadaveric allograft.

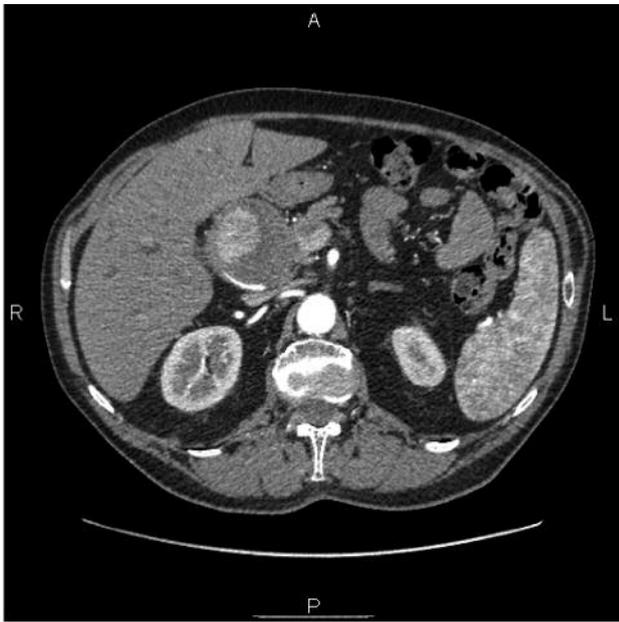


Figure 1. *Initial aspect – partially thrombosed aneurysm of the right hepatic artery compressing the common bile duct. Secondary dilatation of the intrahepatic bile ducts.*



Figure 2. *Initial aspect – the close relationship between the right hepatic artery and the portal vein.*

mandatory in order to get information regarding the presence of collateral vascularization; once this maneuver is performed, surgery might be scheduled. In certain cases aneurysmal rupture in the peritoneal cavity, biliary tree or portal vein might occur, all these events being life threatening complications, necessitating emergency surgery to solve the bleeding (1).

Case Report

A 66-year-old male patient presented for weight loss (7 kilograms during the last two months), asthenia, pruritus and painless sclero- cutaneous jaundice. At that moment the patient was submitted to abdominal computed tomography which revealed the presence of a partially thrombosed aneurysm of the right hepatic artery compressing the common bile duct and inducing an intrahepatic dilatation of the biliary ducts (Figures 1-3). In the meantime, the computed tomography revealed the presence of an accessory left hepatic artery originating from the left gastric artery. The patient was submitted to percutaneous embolization of the right hepatic artery aneurysm followed by an internalized external biliary drainage under tomographic control. The embolization was performed using polivinyl alcohol particles of 350-750 microns and gelspone in association with 5 mm detachable spirals, complete obstruction of the aneurysm being obtained (Figures 4-8).

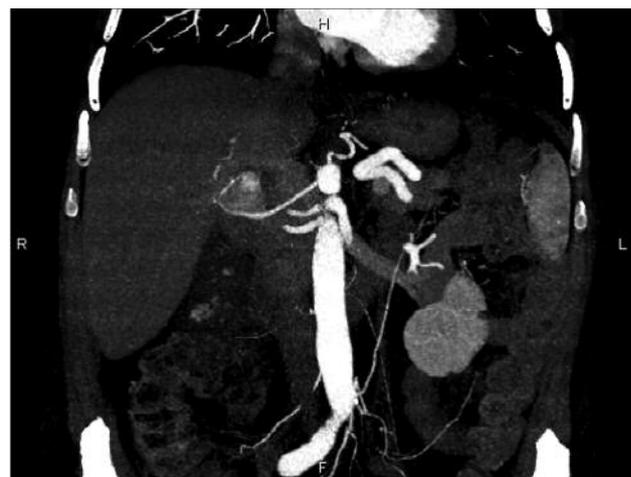


Figure 3. *Initial aspect – coronal reconstruction revealing the anatomical reports of the aneurysm.*

The post-interventional course was initially uneventful; however, at 24 hours after the second maneuver, the patient developed intense epigastric pain and nausea while the biochemical analysis revealed the presence of a pancreatic reaction (with a serum concentration of amylases of 736 U/L and a concentration of seric lipases of 4513 U/L). An emergency computed tomography was performed and an

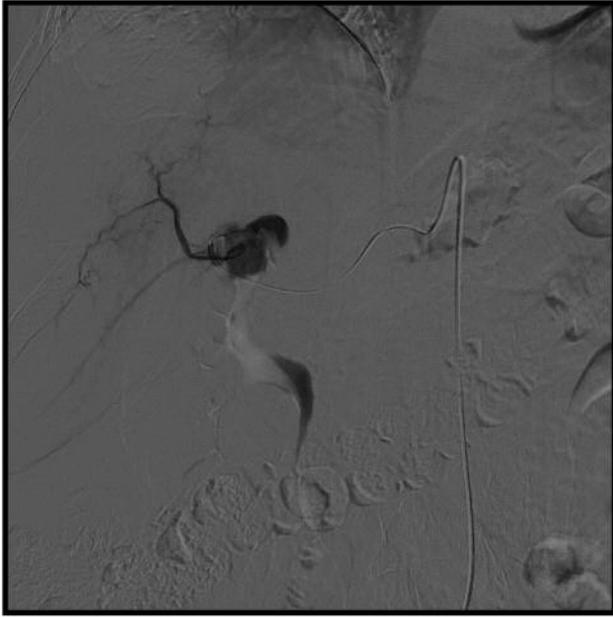


Figure 4. Percutaneous embolization of the aneurysm – catheterization of the right hepatic artery.

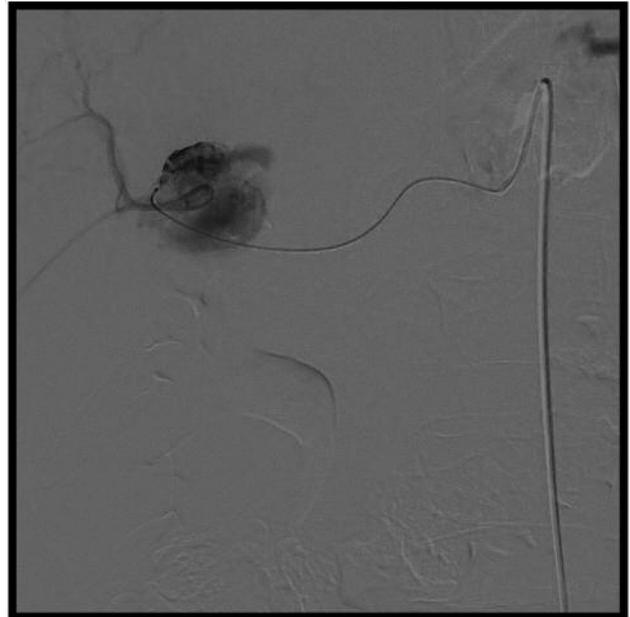


Figure 5. Embolization of the aneurysm with PVA 350-750 and gelspone.

acute edematous pancreatitis due to a transpapillary internalization of the external biliary drainage was revealed. A conservative treatment was instituted followed by the amelioration of the clinical symptoms and the normalization of the biochemical parameters. Seven days later a control arteriography was performed, showing the complete exclusion of the blood flow through the right hepatic artery; however, the aneurysm was alimented in a retrograde manner through the left hepatic artery (throughout the intrahepatic flow) (Figure 9). The patient was discharged with a good general and biological condition; however, three weeks later he presented a massive episode of an upper gastrointestinal bleeding exteriorized through hematemesis, hematochezia and hemobilia (exteriorization of blood through the external bile duct drainage tube). The upper digestive endoscopy revealed the presence of a choledocal – aneurysma fistula so the patient was submitted to emergency surgery. Intraoperatively a choledocal –aneurysmal fistula and portal vein rupture were found; the right hepatic artery aneurysm was resected *en bloc* with segmental resection of the portal vein and common bile duct resection (Figures 10-14). The continuity of the portal vein was re-established through the placement of a cadaveric portal vein allograft, the continuity of the bile duct was re-established through a Roux en Y hepatico-jejunal anastomosis while the right hepatic artery aneurysm was ligated and resected, the liver being vascularized exclusively through the patent left hepatic artery. The postoperative course was initially uneventful; however, in the eighth postoperative day

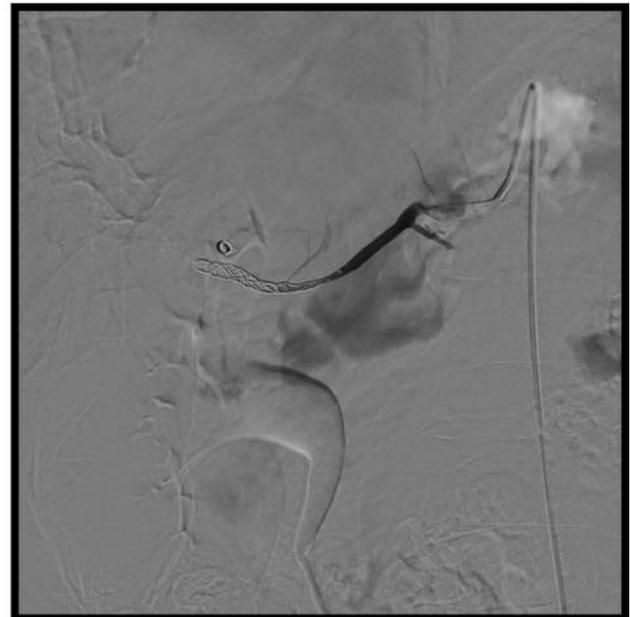


Figure 6. Placing the 5-mm detachable spirals.

the patient developed a febrile syndrome while the computed tomography revealed the presence of a subhepatic abscess due to the presence of a minimal biliary leak which was successfully drained percutaneously. Initially the debit of the



Figure 7. The final aspect after complete embolization of the aneurysm. No recanalization through the left hepatic artery was revealed.



Figure 8. Association of the internalized external biliary drainage.

biliary leak was of 300 ml/day and decreased to 0 ml/24 hours after two weeks, so the drainage tube was removed. At three months follow up the patient presented in a good clinical and biological condition while the control computed tomography did not reveal any pathological aspects.

Discussion

Hepatic artery aneurysms were first reported in 1819 while the first successfully treated such aneurysm was reported almost one century later, in 1903 (7); at that moment Kehr reported the case of a patient diagnosed with ruptured right hepatic artery communicating with the cystic duct which was successfully ligated and resected (8). Since then, almost 300 cases of hepatic artery aneurysms were reported, only 20% of them being successfully treated by surgery (9).

Most often patients with hepatic artery aneurysms are asymptomatic, although in certain cases symptoms such as right upper quadrant pain or jaundice mimicking cholelithiasis or common bile duct lithiasis might develop. The presence of painless jaundice as the first symptom leading to diagnosis is a rare event, only few cases being reported so far (6, 10).

In certain cases aneurysmal rupture in the biliary tree (leading to the apparition of hemobilia), in the portal vein, in the peritoneal cavity or in the gastroduodenal tract might occur (1). The risk of aneurysmal rupture has been estimated to range

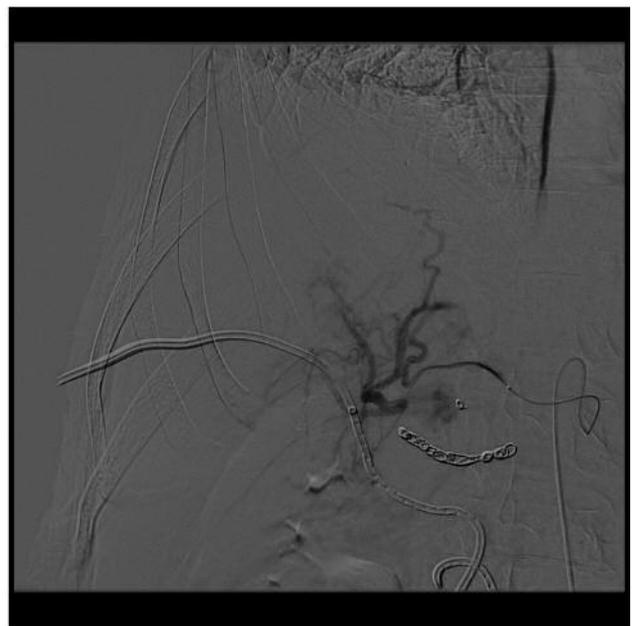


Figure 9. Recanalization of the aneurysm through the left hepatic artery.

between 20-80%, this event being associated with a mortality rate of up to 21% (11-13); however, this risk cannot be related to the dimension of the aneurysms, therefore is stated that all

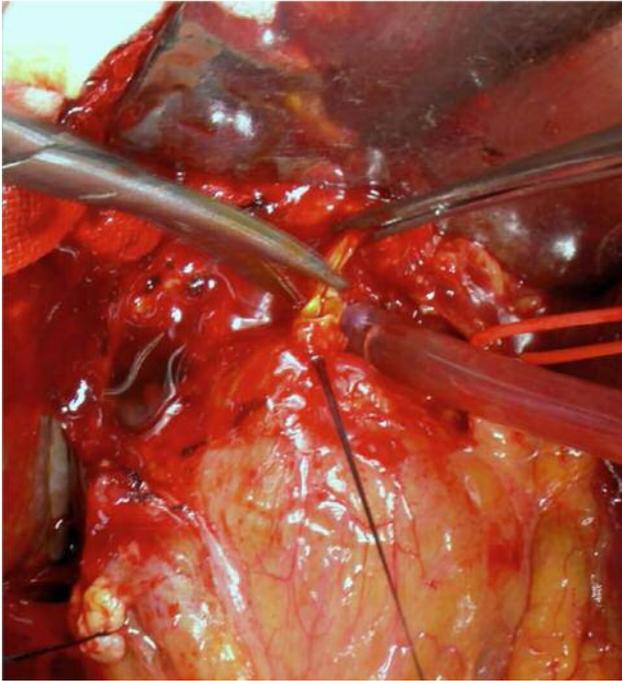


Figure 10. Sectioning the common biliary duct. The aspect after aneurysmal dissection.

lesions should be submitted to therapy (14). When it comes to the main therapeutic options of the uncomplicated aneurysms, percutaneous embolization or stenting have been successfully performed in the last decades (15, 16).

In cases presenting hepatic artery aneurysms and arterio-choledocal fistulas excision of the aneurysm *en bloc* with the affected segment of the common bile duct and bilio-digestive derivation remains the treatment of choice. Once the resection phase is ended, a close inspection of the macroscopic aspect of the liver is needed in order to observe if the remaining vascularization is able to provide a proper hepatic inflow. However, these modifications are rarely seen due to the fact that usually arterial aneurysms usually develop during a long period of time, being associated with the apparition of a well-organized collateral circulation (17). This mechanism was incriminated in our case too, the post-embolization arteriography which was performed three weeks before the development of the upper digestive tract hemorrhage revealing the occlusion of the right hepatic artery aneurysm in association with retrograde intrahepatic vascular inflow of the aneurysm.

When it comes to the main reconstructive options, the decision should be taken after analyzing the particularities of the hepatic vascularization. Patients presenting common hepatic artery aneurysms in the presence of a patent

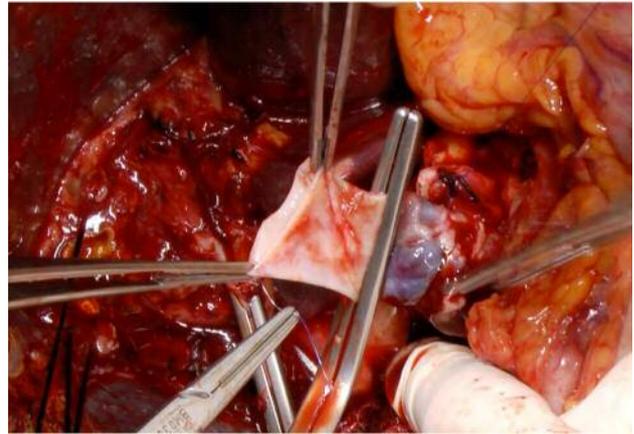


Figure 11. Reconstruction of the portal vein using the cadaveric graft.

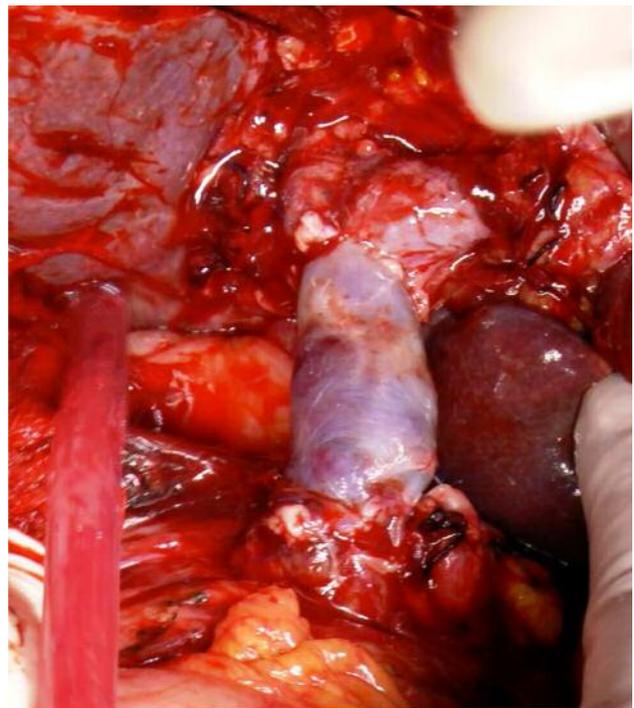


Figure 12. The final aspect after aneurysmectomy, arterial stump ligation and portal vein reconstruction.

gastroduodenal artery which could provide an adequate hepatic perfusion can be safely submitted to common hepatic artery ligation and aneurysmectomy (18). In cases presenting proper, right or left hepatic artery aneurysms, the presence of a collateral patent liver vascularization should be carefully



Figure 13. The specimen of aneurysectomy.

investigated before taking a surgical decision; whenever an adequate collateral vascularization cannot be demonstrated, arterial aneurysm ligation should be followed by reconstruction through an end to end anastomosis, graft interposition or splenohepatic anastomosis (1, 9, 19). In the eventuality in which the presence of collateral patent vascularization of the liver is revealed, aneurysmal resection without reconstruction might be proposed.

One of the largest studies regarding the role of surgery in treating hepatic artery aneurysms was the one conducted in Florence and Rome, Italy, and published in 2008. The study included 55 patients diagnosed with visceral arteries' aneurysms submitted to surgery between 1982 and 2007. Among these 55 cases there were seven patients diagnosed with common hepatic arteries' aneurysms which were treated by isolated aneurysectomy (in three cases), aneurysectomy and autologous venous graft (in one case), partial aneurysectomy with proximal and distal ligation of the common hepatic artery (in two cases) and aneurysmorrhaphy (in one case). In one of the patients submitted to partial aneurysectomy with proximal and distal ligation of the common hepatic artery there was no adequate flow from the gastroduodenal artery so a prosthetic splenohepatic bypass was associated. After a mean follow up period of 82.1 months no aneurysm related death was reported, demonstrating the benefits and the efficacy of the aneurysmal surgical repair (20).



Figure 14. The specimen of partially thrombosed, sectioned aneurysm of the right hepatic artery.

A similar case to the one that we have reported was presented in 2009 by Parmar *et al.* at Addenbrookes Hospital, Cambridge, United Kingdom. The patient, a 59-year-old man presented for painless jaundice and was diagnosed with a complex aneurysm of the right and common hepatic artery compressing the common bile duct and the portal vein. Intraoperatively a 9-cm complex aneurysm of the right hepatic artery compressing the portal vein and the common bile duct were revealed; when opening the right hepatic artery aneurysm minimal backflow bleeding from the liver was revealed. Due to this reason, in association with the presence of a patent accessory left hepatic artery arising from the left gastric artery the authors decided to tie off the aneurysm; this gesture did not induce any phenomena of liver ischemia. In the meantime, the common bile duct was divided during hepatic artery aneurysm resection, so a Roux en Y biliary reconstruction was associated (6).

Conclusion

Arterio-choledocal fistulas are rare complications that might develop in patients diagnosed with right hepatic artery aneurysm which can be successfully treated by vascular resection and biliodigestive anastomosis. Due to the fact that most cases have in fact a long history of aneurysmal development, a patent collateral hepatic circulation through the accessory vessels might be present; in these cases aneurysectomy with vascular stump ligation without vascular reconstruction is the option of choice. However, if an inadequate liver perfusion is observed, vascular reconstruction using grafts or spleno-hepatic bypasses have been reported with good results.

References

- 1 Wykypiel H, Pointner R, Bodner J and Oberwalder M: Venous graft for replacement of an aneurysm-bearing right hepatic artery. *Eur Surg* 36: 194-197, 2004.
- 2 Gorich J, Rilinger N, Brado M, Huppert P, Vogel J, Siech M, Sokiranski R, Ganzauge F, Beger HG and Brambs H: Non-operative management of arterial liver hemorrhages. *Eur Radiol* 9: 85-88, 1999.
- 3 Sieber CC and Jäger K: Duplex scanning – a useful tool for non-invasive assessment of visceral blood flow in man. *Vasc Med Rev* 3: 95-114, 1992.
- 4 Stack BH, Rankin JT and Bentley RJ: Hepatic artery aneurysm after staphylococcal endocarditis. *Br Med J* 3: 659-660, 1968.
- 5 Stanley JC, Thompson NW and Fry WJ: Splanchnic artery aneurysms. *Arch Surg* 101: 689-697, 1970.
- 6 Parmar J, Winterbottom A, Gordon E, Kathikesalingam A and Varty K: Hepatic Artery Aneurysm: A Rare Presentation as Painless Obstructive Jaundice. *EJVES Extra* 18: 47-50, 2009.
- 7 Wilson J: Lecture of the Royal College of Surgeons. London, 1819.
- 8 Kehr H: Der erste Fall von erfolgreicher Unterbindung der A. hepatica propria wegen Aneurysma. *Munch Med Wochenschr* 50: 1861-1867, 1903.
- 9 Lal RB, Strohl JA, Piazza S, Aslam M, Ball D and Patel K: Hepatic artery aneurysm. *J Cardiovasc Surg (Torino)* 30: 509-513, 1989.
- 10 Mazziotti S, Blandino A, Gaeta M, Lamberto S, Vinci V and Ascenti G: Hepatic artery aneurysm, an unusual cause of obstructive jaundice: MR cholangiography findings. *Abdom Imaging* 28: 835-837, 2003.
- 11 Stanziale SF, Wholey M and Makaroun MS: Large hepatic artery aneurysm with portal vein fistula. *J Vasc Surg* 43: 631, 2006.
- 12 Harlaftis NN and Akin JT: Hemobilia from ruptured hepatic artery aneurysm. Report of a case and review of the literature. *Am J Surg* 133: 229-232, 1977.
- 13 Routh WD, Keller FS, Cain WS and Royal SA: Transcatheter embolization of a high-flow congenital intrahepatic arterial-portal venous malformation in an infant. *J Pediatr Surg* 27: 511-514, 1992.
- 14 Chandramohan, Khan AN, Fitzgerald S, Sherlock D and Tam E: Sonographic diagnosis and follow-up of idiopathic hepatic artery aneurysm, an unusual cause of obstructive jaundice. *J Clin Ultrasound* 29: 466-471, 2001.
- 15 Guckel C and Steinbrich W: Arterial pseudoaneurysm as a complication of percutaneous transhepatic biliary drainage: treatment by embolization. *Z Gastroenterol* 33: 602-604, 1995.
- 16 Rosch J, Petersen BD, Hall LD and Ivancev K: Interventional treatment of hepatic arterial and venous pathology: a commentary. *Cardiovasc Intervent Radiol* 13: 183-188, 1990.
- 17 Gracey L: Jaundice due to hepatic artery aneurysm. *Proc R Soc Med* 63: 1311, 1970.
- 18 Busuttil RW and Brin BJ: The diagnosis and management of visceral artery aneurysms. *Surgery* 88: 619-624, 1980.
- 19 Brasoveanu V, Dumitrascu T, Bacalbasa N and Zamfir R: Splenic artery used for replaced common hepatic artery reconstruction during pancreatoduodenectomy – a case report. *Chirurgia (Bucur)* 104: 499-504, 2009.
- 20 Pulli R, Dorigo W, Troisi N, Pratesi G, Innocenti AA and Pratesi C: Surgical treatment of visceral artery aneurysms: A 25-year experience. *J Vasc Surg* 48: 334-342, 2008.

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