Case Report

Cystic Artery Pseudoaneurysm: A Rare but Serious Complication after Cholecystectomy
Report of Three cases

Talib A. Majid*,Raafat R. Ahmad*, Ahmed R. Jawad* , Akeil H. Eissa* , Akeel S. Mahmmod*

INTRODUCTION:

Cholecystectomy is the most common major abdominal procedure [(1)]. Pseudo-aneurysm of the cystic artery is a well documented rare but serious complication that might follow an open or laparoscopic cholecystectomy (LC). It may be seen in isolation or in association with bile duct injury [(2)]. It represents a technical failure and can be avoided by adopting a standardized procedure.

We present three cases of cystic artery pseudoaneurysm, one following an open and two following laparoscopic cholecystectomy. All of them presented with hemobilia but with different scenario. These patients required laparotomy to control their problem, two of them were emergencies and one was elective laparotomy.

Keyword: Pseudoaneurysm , Hemobilia , Post -cholecystectomy.

Case-1

A 55 year-old man presented to our emergency department at 10 p.m. complaining of severe agonizing continuous epigastric and precordial pain associated with sweating, of one-hour duration. His ECG showed changes suggestive of myocardial ischemia for which he was admitted to the coronary care unit. At 7 a.m. next day, the patient developed haematamesis. He gave a history of LC seven months ago. He was pale, sweaty, his pulse rate was 120/min and his blood pressure was 100/60 mmHg. He was faintly jaundiced. His abdomen was slightly distended, soft, and not tender with scars of previous laparoscopy. Hemoglobin was 10g/dl, white blood cell count was 7000/mm^3 and platelet count was 2.1 x 10^5/mm^3. The coagulation profiles were normal. Total bilirubin was 54mmol/l with direct bilirubin of 34mmol/l. Oesophago-gastro-duodenoscopy (OGD) revealed blood in the stomach and duodenum with no obvious source of bleeding. Ultrasonography (US) of the abdomen was normal.

The patient was resuscitated by administering intravenous crystalloid solution and he was kept under sedation and close observation.

At 9 p.m. of the same day, he collapsed suddenly with massive haematamesis and malena with a sense of impending death. He became severely pale, sweaty and thirsty. His pulse rate increased to 150/min, and his blood pressure dropped to 70/40 mmHg. He was rapidly resuscitated with fluid and blood transfusion, and then he was transferred to the operative theatre. At laparotomy; the stomach, duodenum, small bowel and colon were full of fresh and altered blood. There was a pulsating 3-cm mass at the portahepatis with a laparoscopic metal clip at one point of the mass.

A diagnosis of pseudoaneurysm of the cystic artery was therefore established.

After undertaking pressure below, behind and above with fingers and thumb of the left hand, the pseudoaneurysm was explored.

Lamellated clotted blood within an outer fibrous layer was evacuated to reveal a pulsatile bleeding cystic artery in the depth of pseudoaneurysm.

The bleeding artery was precisely controlled by transfixing ligature with 4/0 prolene.

The patient had a smooth postoperative recovery and discharged home nine days later without any problems on subsequent follow-up visits.

Case -2

A 45 year-old woman presented with recurrent upper abdominal pain associated with intermittent fever and jaundice of one-month duration. She had an open cholecystectomy for chronic calculus cholecystitis two months ago. She was jaundiced, her pulse rate was 76 beats/min, blood pressure was 130/70 mmHg and her temperature was 37.4 C. Abdominal examination revealed a healed right subcostal scar with some tenderness on deep palpation in the right upper quadrant.

Hemoglobin was 12 g/dl, white blood cell count 8500/mm^3, bilirubin 337 mmol/l with direct bilirubin 248 mmol/l and alkaline phsphatase was 483 iu/l. Abdominal US examination showed a well-defined vascular mass of 53x38x48 mm at the portahepatis. Doppler study confirmed the presence of pseudoaneurysm at the portahepatis with some mild dilatation of the biliary ducts.

*Department surgery, Gastrointestinal and Hepatology Teaching Hospital. Medical City - Baghdad.

Upper GI endoscopy identified blood in the lumen of the stomach and duodenum but failed to show blood flowing from the duodenal papilla.
During her stay in hospital, she bled massively so that she was transferred immediately to the operative theatre. At the laparotomy, there was a pulsating mass at the portahepatis with blood clots inside the bile ducts. With the help of Pringle's maneuver, the mass was explored with evacuation of hematoma and a bleeding cystic artery was controlled by transfixing ligature. The common bile duct was explored, blood clots evacuated and a T-tube inserted. She was discharged home at the 5th postoperative day. The T-tube was removed 20 days later after a normal T-tubogram.

Case-3

A 37 year-old man had a history of LC 6 months ago. One month after surgery, he started to complain of recurrent severe upper abdominal pain associated sometimes with haematamesis and malena requiring blood transfusions every now and then. The pain and bleeding episodes recurred for a period of nearly one month during which three OGDs failed to show the source of bleeding. Four months later an US examination on a follow-up visit revealed the presence of a vascular mass at the portahepatis. On examination, the patient looked healthy, slightly pale but not jaundiced. His vital signs were normal. Blood hematology and biochemistry were normal apart from hemoglobin of 11 g/dl. Doppler study and CT scan confirmed the presence of a pseudoaneurysm at the portahepatis, 45x36x25 mm in dimensions with mild intra-hepatic biliary tree dilatation and a normal common bile duct (figure 1, 2)

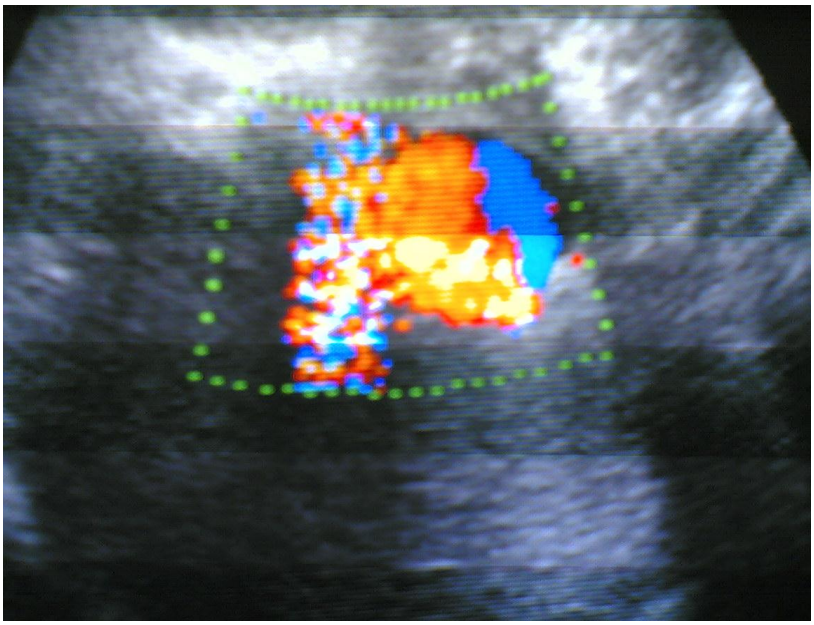

Figure 1: Doppler ultrasound showed cystic mass with arterial and venous signal

Figure 2: C.T. shows vascular mass at porta-hepatis

At elective laparotomy, the pseudoaneurysm was explored with the help of proximal, distal and direct control. After evacuation of the hematoma, there was a hole of 2 mm in the right hepatic artery that was sutured with a figure-of-eight fine prolene vascular suture (figure 3). The patient recovered smoothly and discharged home on the 4th postoperative day.

Figure 3: operative photograph of the pseudoaneurysm.

DISCUSSION:

Pseudoaneurysm is an expanding pulsating haematoma in continuity with an arterial lumen. It does have an outer fibrous layer, but it does not have an endothelial lining. Once an aneurysm is formed, its enlargement and eventual rupture is governed by Laplace law (T=RP). Cystic artery pseudoaneurysms are rare, and only few cases following cholecystectomy have been reported in the literatures [5, 6, 7, 8, and 9]. Some cases were reported associated with the inflammatory reaction seen with the acute cholecystitis [10,11,12,13,14], and one case is reported after liver transplantation [15].They have the tendency to rupture and bleed into the bile ducts producing the clinical syndrome of haemobilia consisting of a triad of upper gastrointestinal bleeding, abdominal pain and jaundice. It is found that in review 545 patients with hemobilia, aneurysm constitutes 7% of the

causes [4]. All the cases reported here were following cholecystectomy and they represent a technical failure in dealing with the cystic artery in the form of a slipped ligature, slipped clip, abuse of diathermy current and avulsion or laceration of the cystic artery. It is important to emphasize the need to apply a proper standard method in dealing with the cystic artery. The diagnosis of cystic artery pseudoaneurysm is based on clinical suspicion.

Upper GIT bleeding weeks or months after cholecystectomy is highly suspicious especially if accompanied by abdominal pain and jaundice i.e. hemobilia, but only about 40% of the patients presented with the entire triad [3].

Blood indices usually show elevation of alkaline phsphatase and direct serum bilirubin with anemia. Confirmation is established by upper GIT endoscopy; Side- viewing dudenoscope is best method to diagnose hemobilia by seeing fresh blood or clot oozing from ampulla [10]. Although selective hepatic arteriography is the procedure of choice for diagnosis [13], color-Doppler imaging is a favorable modality for the diagnosis [11] because it neither invasive nor required highly expertise personal. ERCP may be used in cases were patients presented with obstructive jaundice [10] and to exclude associated bile duct injury. C.T scan with intravenous contrast may identify the aneurysm by pooling of contrast material outside the blood vessels [10]. The treatment of this condition depend on if there is available expertise personal in interventional radiology, so this can be done by selective catheterization of hepatic artery with embolization of the feeding artery [5,6,7].

But this is not without complications and it may lead to rupture of the aneurysm, which requires emergency laparotomy [7]. If this modality were not available, the best treatment would be excision of the sac with oversewn of the feeding artery [8,15].

In case of pseudoaneurysm is duo to acute inflammatory reaction associated with a acute cholecystitis the treatment is either one step excision of the aneurysm and cholecystectomy [10,11,13,14], or two step modalities; which consist of embolization of the pseudoaneurysm followed by elective cholecystectomy ten days later [12].

The surgical interventions in our cases were very reliable in dealing with this lesion by applying the basic vascular technical steps of: proximal, distal and direct control of a bleeding artery by pringl's maneuver, and then the use of fine prolene to transfix and ligate a bleeding point. All patients passed uneventful recovery and they left hospital well in 4^{th}-9^{th} postoperative day.

REFERENCES:

1. Margret Oddsdottir and John Hunter. Gallbladder and the Extrahepatic Biliary System. In: F. Charles Bruicardi, Dana K. Andersen, Timothy R. Biliar, David L. Dunn, John J. Hunter, and Raphael E.Pollock (eds), Schwartz's Principles of Surgery. McGraw Hill. Philadelphia. 2004, 1187-1219
2. Yelle JD, Fairfull-Smith R, Rasuli P, Lorimer JW. Hemobilia complicating elective laparoscopic cholecystectomy: a case report. Can J Surg. 1996; 39: 240-2
3. Richard T. Schlinkert, Keith A.Kelly. Upper Gastrointestinal Bleeding. In: Douglas W. Wilmore, Laurency Y. Cheung, Alden H. Markan, James W. Holcroft, Jonthan L. Markins, Nathaniel J. Soper(Ed.). ACS Surgery, Principles and Practice. New York: WebMD. 2003; 280-288.
4. Ravi S.Chari, William C. Meyers. Hembilia. In: David C.Sabiston, H. Kim Lyerly (15^{th} Eds), *textbook of surgery: the biological basis of modern surgical practice*. Philadelphia: W.B. Saunders Company; 1997: 1084-1087.
5. Iannelli A, Karimdjee BS, Fabiani P, Benizri EI, Converset S, Medjoubi SA, Bornet P, Gugenheim J. Hemobilia due to pseudoaneurysm of the right hepatic artery following laparoscopic cholecystectomy. Gastroenterol Clin Biol. 2003; 27: 341-3.
6. Genyk YS, Keller FS, Halpern NB. Hepatic artery pseudoaneurysm and hemobilia following laser laparoscopic cholecystectomy. A case report. Surg Endosc. 1994; 8: 201-4
7. Pistorius G.A. Walter P. Hildebrandt U. Defreyne L. Pseudoaneurysm of Hepatic Artery. A rare Complication after Laparoscopic cholecystectomy. Langenbecks Arch Chir 1994; 379: 291-293.
8. Yelle JD, Fairfull-Smith R, Rasuli P, Lorimer JW. Hemobilia complicating elective laparoscopic cholecystectomy: a case report. Can J Surg. 1996; 39: 240-2.
9. Kapoor R, Agarwal S, Calton R, Pawar G. Hepatic artery pseudoaneurysm and hemobilia following laparoscopic cholecystectomy. Indian J Gastroenterol. 1997; 16: 32-3.
10. Lileswar Kaman, Santosh Kumar, Arunanshu Behera, Ranvindra Nath Katariya. Pseudoaneurysms of cystic artery: A Rarae Cause of Hemobilia. The American Journal of Gastroenterology 1988; 93:1535-1537.

11. Nakajima M, Hoshino H, Hayashi E, Nagano K, Nishimura D, Katada N, Sano H, Okamoto K, Kato K. Pseudoaneurysm of the cystic artery associated with upper gastrointestinal bleeding. J Gastroenterol. 1996; 31: 750-4.

12. Maeda A, Kunou T, Aono K, Murata T, Niinomi N, Yokoi S. Pseudoaneurysm of the cystic artery with hemobilia treated by arterial embolization and elective cholecystectomy. J Hepatobiliary Pancreat Surg. 2002; 9: 755-8.

13. Barba CA, Bret PM, Hinchey EJ. Pseudoaneurysm of the cystic artery: a rare cause of hemobilia. Can J Surg. 1994; 37: 64-6.

14. Lena Sibulesky, Mark Ridlen, Victor E. Pricolo, Hemobilia due to Cystic Artery Pseudoaneurysm. The American Journal of Surgery. 2006; 191: 797–798 .

15. Bussenius-Kammerer M, Ott R, Wutke R, Grunewald M, Hohenberger W, Reck T.Pseudoaneurysm of the hepatic artery--a rare complication after orthotopic liver transplantation. Chirurg. 2001; 72: 78-81.