# A Common Hepatic Artery Aneurysm Mimicking A Duodenal Submucosal Tumor Presenting as Upper Gastrointestinal Bleeding: A Case Report

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# Abstract

Hepatic artery aneurysm is an uncommon etiology of upper gastrointestinal bleeding. It can present as a duodenal submucosal tumor-like lesion due to extrinsic compression during endoscopic examination. Such aneurismal bleeding usually has abrupt onset and high risk of rebleeding. Hence, rapid diagnosis and early intervention is necessary. We presented a middle-aged man suffering from upper gastrointestinal bleeding due to a common hepatic artery aneurysm mimicking a duodenal submucosal tumor. After endoscopic hemostasis with hemoclip, the following computed tomography (CT) scan of abdomen showed a ruptured common hepatic artery aneurysm abutting the duodenal bulb. Recurrent gastrointestinal bleeding with hypovolemic shock complicated on the next day. This patient subsequently underwent surgical repair of aneurysm of common hepatic artery and recovered smoothly. No more recurrent bleeding was noted during six months' follow-up. In summary, vascular aneurismal lesion should be included in the differential diagnosis of a duodenal submucosal tumor-like lesion. Then, hemoclipping is a better and safer technique for temporal hemostasis to a suspected vascular lesion. Finally, further work-up like CT must be obtained as soon as possible for clarifying the submucosal tumor-like lesion and guiding further management. Surgical or radiological intervention is definite for aneurismal rupture-related gastrointestinal hemorrhage. (J Intern Med Taiwan 2010; 21: 56-61)

# Key Words : Duodenal submucosal tumor, Common hepatic artery aneurysm, Upper gastrointestinal bleeding

# Introduction

Hepatic artery aneurysm was firstly described at autopsy in 1809<sup>1</sup>. It can present as a duodenal submucosal lesion caused by extra-luminal compression. Most cases of common hepatic artery aneurysms with external compression of duodenum are asymptomatic until rupture with intestinal hemorrhage. Such cases had been reported with poor prognosis if diagnosis and treatment<sup>2</sup> were delayed. Hence, the management of this complication needs early diagnosis and timely intervention.

Here, we reported a rare case of common hepatic artery aneurysm mimicking a duodenal

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submucosal tumor causing upper gastrointestinal (GI) bleeding. After endoscopic hemostasis, computed tomography of abdominal scan was performed and disclosed a ruptured aneurysm. A surgeon was consulted. Recurrent GI bleeding with hypovolemic shock occurred one day later. Surgical intervention was performed and the patient had an uneventful recovery.

#### Case Report

A 46-year-old man presented to our emergency department with a 3-day history of hematemesis and melena. His past medical history was unremarkable. The blood pressure was 115/57 mmHg and pulse rate was 100 beats per minute. Physical examination revealed only tenderness in epigastric area. Hemogram showed slight anemia and leukocytosis with hemoglobin: 9.9 g/dL (normal range: M: 13.5-17.5, F: 14-16 g/dL) and white blood count:  $15.160 \times 10^3$  / mm<sup>3</sup> (normal range: M: 3.9-10.6, F: 3.5-11 x 10<sup>3</sup> / mm<sup>3</sup>). PT and APTT were normal. Blood biochemistry showed the following: BUN: 33 mg/dL (normal range: 7-22 mg/dL), creatinine: 1.2 mg/dL (normal range: 0.6-1.6 mg/dL), AST: 22 IU/L (normal range: 5-40 IU/L), ALT: 16 IU/L (normal range: 5-40 IU/L), and total bilirubin: 0.4 mg/dL (normal range: 0.2-1.5 mg/dL).

The patient underwent an upper gastrointestinal (GI) endoscopic examination, which revealed a 2.0 x 2.0 cm smooth duodenal submucosal tumor-like lesion at anterior wall of duodenal bulb with a small ulcer on its surface. There was a fibrin strand and some blood oozing in the ulcer. (Fig.1) Hemoclip was applied for hemostasis. Then, he was admitted.

On the 1<sup>st</sup> day of hospitalization, computed tomography (CT) of abdomen scan was arranged to evaluate the duodenal submucosal tumor-like lesion and disclosed a 5.8 x 2.2 x 2.6 cm heterogeneously enhancing mass lesion contacting the medial aspect of duodenal bulb with encasement of enlarged

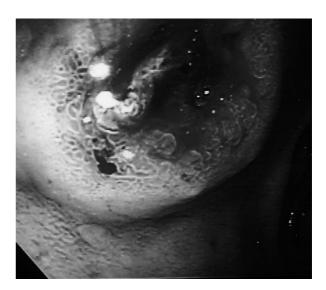


Fig.1. Endoscopic examination revealed a 2.0 x 2.0 cm smooth tumor-like lesion with a small ulcer on its surface, located at anterior wall of duodenal bulb. There was a fibrin strand and blood oozing noted from this ulcer.

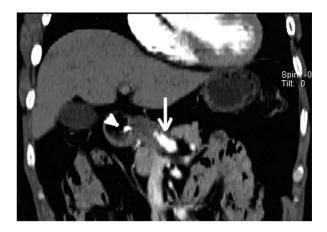


Fig.2. A reconstructive coronal view of CT scan with contrast showed a 5.8 x 2.2 x 2.6 cm heterogeneously enhancing mass lesion (arrow) contacting the medial aspect of duodenal bulb with encasement of enlarged hepatic artery. There was a residual hemoclip (smaller arrow head) in the duodenum.

hepatic artery. (Fig.2) A ruptured hepatic artery aneurysm (HAA) with hematoma or neoplasm with vessel encasement was diagnosed. Tumor markers were checked as following: carcinoembryonic antigen: 0.4 ng/mL (normal range: 0.0-3.0), alphafetoprotein: 2.37 ng/mL (normal range: 0.0-10.9), CA-199: 0 U/mL (normal range: < 37 U/ml). Then, we consulted a surgeon for further management.

On the 2<sup>nd</sup> day of hospitalization, upper GI bleeding recurred with massive melena and hypovolemic shock. After fluid resuscitation and blood component therapy, his hemodynamics were stabilized. Under the impression of HAA rupture into duodenum, the patient underwent emergent operation, including resection of ruptured aneurysm and repair of common hepatic artery by laparoscopic approach. The operative finding disclosed a 3 cm elastic submucosal cavitary mass communicating with common hepatic artery in the 1<sup>st</sup> portion of duodenum. The mass had perforated into duodenal lumen.

The pathology showed one hemorrhagic cavity of duodenum, measuring 3.0 x 2.5 x 1.2 cm in size with one fistulous tract on mucosal side. The serosal side of cavity showed one hemorrhagic aneurismal lesion. There was no evidence of malignancy in the whole surgical specimen. The patient was discharged on the 19<sup>th</sup> day after hospitalization. There was no recurrence of upper gastrointestinal bleeding after 6 months' follow-up.

### Discussion

A duodenal submucosal tumor-like lesion is usually an incidental finding during upper GI endoscopic examination. Most duodenal submucosal tumor-like lesions are asymptomatic until upper GI bleeding or mass effect with GI obstruction occurs. The duodenal submucosal tumor-like lesion can be a primary intramural lesion or secondary to extra-luminal compression. The intrinsic tumor can be classified according to benign or malignant histology. The benign lesions include cyst, heterotopic pancreas<sup>3</sup>, leiomyoma, lipoma, neurofibroma, schwannoma<sup>4</sup>, and Brunner's gland hyperplasia<sup>5-6</sup>. The malignant tumors have lymphoma, gastrointestinal stromal tumor (GIST), carcinoid tumor, and metastatic tumor. The external compression may be due to neoplasm, enlarged lymph node, vascular aneurysm or cystic lesions<sup>7</sup>.

Aneurysm of common hepatic artery is a rare etiology of extrinsic compression to duodenal wall and form a submucosal tumor. The pathogenesis of hepatic artery aneurysm can be mycotic aneurysm, atherosclerosis, surrounding organ inflammation (e.g. cholecystitis, pancreatitis), congenital fibromuscular dysplasia or blunt abdominal trauma<sup>8-12</sup>. When the aneurysm ruptured, the subsequent bleeding route is determined by anatomic relationships. The most catastrophic consequence is massive bleeding into peritoneum or GI tract with estimated mortality rate up to  $40\%^2$ . Some cases may develop hemobilia<sup>11</sup>, together with obstructive jaundice and colicky abdominal pain, constituting the Quincke's triad. For our patient, the common hepatic artery aneurysm might have bulged and eroded the duodenal wall gradually and created a fistula between aneurysm and duodenal bulb. When ruptured, a localized hematoma formed and resulted in bleeding into duodenal lumen.

It is advised to investigate duodenal submucosal tumor-like lesion with other imaging studies because endoscope cannot recognize its underlying composition. Endoscopic ultrasound (EUS) is a useful tool to evaluate the echo pattern of tumor or to detect the blood flow if equipped with color Doppler function<sup>13</sup>. In addition, it can also provide tissue sampling through echo-guided transmural aspiration biopsy. Other diagnostic modalities such as CT, magnetic resonance imaging (MRI) are also valuable if detailed anatomic information or preoperative staging for cancer is needed. Practically, the choice of diagnostic tool depends on urgency of symptoms and clinical stability of patient. EUS is not suitable for our case because of interference by metallic hemoclip and recent ulcer hemostasis.

The treatment of hepatic artery aneurysm depends on the number, size, location and patient's

hemodynamics. Based on physiologic rationale and limited case series experience, extrahepatic aneurysm benefits from aneurysmorrhaphy and arterial reconstruction<sup>15-17</sup>. On the contrary, intrahepatic aneurysm is preferentially managed by endovascular embolization or prosthetic stent<sup>18-20</sup>. However, embolization to common hepatic artery aneurysm bears the risk of hepatic ischemic injury especially when the patency between gastroduodenal and pancreaticoduodenal artery arcade was insufficient. In the review of medical literature, we found six cases of ruptured common hepatic artery aneurysm. Only one case received arterial embolization<sup>26</sup>. Other five cases all underwent surgical reconstruction<sup>14,23-25,27</sup>.

In our case, there are some diagnostic and therapeutic points deserving discussion. First, duodenal bulb is an unusual site for submucosal tumor and the differential diagnoses range from intrinsic tumor to extrinsic compression lesion. Endoscopically, this submucosal tumor-like lesion had a superficial ulcer with active oozing and fibrin strand, implicating the easy-bleeding character. In general, malignant lesions are more prone to bleed due to profuse neovascularization and cancer cell invasiveness. Therefore, malignant duodenal submucosal tumors such as leiomyosarcoma, lymphoma, GIST or metastatic cancer should be considered in priority<sup>21</sup>. For external compression lesion, there is wide variety of etiologies determined by adjacent viscera or structures. The first portion of duodenum can be compressed by hepatic tumor at caudate lobe, cholangiocarcinoma at hepatic hilum, gallbladder cancer, pancreatic head tumor, right renal tumor, aorta<sup>22</sup> or celiac trunk branches aneurysms.

Second, the consistency of this submucosal lesion was felt to be rather hard when applying hemoclip to it. This phenomenon probably reflected the already thrombosis of this aneurysm, which explained why a pulsatile feature was lacking. There are two major reasons to use hemoclips rather than other thermal or injectional device for hemostasis. First, mechanical hemostasis by clipping avoids the risk of tissue necrosis or perforation caused by electrical or chemical injury. This is of great concern because of unknown nature of this submucosal tumor. Second, the radio-opaque clip can provide a landmark during angiography if endovascular intervention is necessary.

In conclusion, the vascular lesion should be included in the differential diagnoses when encountering a bleeding duodenal submucosal tumor-like lesion. To achieve endoscopic hemostasis, hemoclip seems to be a more appropriate tool for a suspected vascular lesion. Then, further investigatory modality such as CT needs to be performed immediately to guide further management.

#### References

- 1.Guida PM, Moore SW. Aneurysm of the hepatic artery. Report of five cases with a brief review of previously reported cases. Surgery 1996; 60: 299-310.
- Abbas MA, Fowl RJ, Stone WM, Panneton JM, Oldenburg WA, Bower TC. Hepatic artery aneurysm: factors that predict complications. J Vasc Surg 2003; 38: 41-5.
- Thoeni RF, Gedgaudas RK. Ectopic pancreas: usual and unusual features. Gastrointest Radiol 1980; 5: 37-42.
- Orsenigo E, Di Palo S, Vignali A, Staudacher C. Laparoscopic excision of duodenal schwannoma. Surg Endosc 2007; 21: 1454-6.
- Inai M, Sakai M, Kajiyama T, et al. Endosonographic characterization of duodenal elevated lesions. Gastrointest Endosc 1996; 44: 714-9.
- 6. Tung CF, Chow WK, Peng YC, Chen GH, Yang DY, Kwan PC. Bleeding duodenal lipoma successfully treated with endoscopic polypectomy. Gastrointest Endosc 2001; 54: 116-7.
- Motoo Y, Okai T, Ohta H, et al. Endoscopic ultrasonography in the diagnosis of extraluminal compressions mimicking gastric submucosal tumors. Endoscopy 1994; 26: 239-42.
- Shanley CJ, Shah NL, Messina LM. Common splanchnic artery aneurysms: splenic hepatica and celiac. Ann Vasc Surg 1996; 10: 315-22.
- 9. Parmar H, Shah J, Shah B, Patkar D, Varma R. Imaging findings in a giant hepatic artery aneurysm. J Postgrad Med

2000; 46: 104-5.

- O'Driscoll D, Olliff SP, Olliff JFC. Hepatic artery aneurysm. Brit J Radiol 1999; 72: 1018-25.
- 11.Shussman N, Edden Y, Mintz Y, Verstandig A, Rivkind AI. Hemobilia due to hepatic artery aneurysm as the presenting sign of fibro-muscular dysplasia. World J Gastroenterol 2008; 14: 1797-9.
- 12.Mayo A, Aladgem D, Makrin V, Kluger Y. Traumatic hepatic artery pseudo-aneurysm with fistula to the hepatic vein. Isr Med Assoc J 2004; 6: 496-7.
- 13.Kawamoto K, Yamada Y, Utsunomiya T, et al. Gastrointestinal submucosal tumors: evaluation with endoscopic US. Radiology 1997; 205: 733-40.
- 14.Cimsit B, Ozden I, Emre AS. A rare intraabdominal tumor: giant hepatic artery aneurysm. J Med Invest 2006; 53: 174-6.
- 15.Pilleul F, Valette PJ. Management of aneurysms of hepatic artery. 15 patients. Presse Med 2001; 30: 1139-42.
- 16.Schick C, Ritter RG, Balzer JO, Thalhammer A, Vogl TJ. Hepatic artery aneurysm: treatment options. Eur Radiol 2004; 14: 157-9.
- 17.Tajima Y, Kuroki T, Tsutsumi R, Sakamoto I, Uetani M, Kanematsu T. Extrahepatic collaterals and liver damage in embolotherapy for ruptured hepatic artery pseudoaneurysm following hepatobiliary pancreatic surgery. World J Gastroenterol 2007; 13: 408-13.
- 18.Millonig G, Graziadei IW, Waldenberger P, Koenigsreiner A, Jaschke W, Vogel W. Percutaneous management of a hepatic artery aneurysm: bleeding after liver transplantation. Cardiovasc Intervent Radiol 2004; 27: 525-8.

- 19.Jung NY, Kim SK, Chung EC, Park H, Cho YK. Endovascular treatment for rupture of intrahepatic artery aneurysm in a patient with Behcet's syndrome. Am J Roentgenol 2007; 188: 400-2.
- 20.Park JY, Ryu H, Bang S, Song SY, Chung JB. Hepatic artery pseudoaneurysm associated with plastic biliary stent. Yonsei Med J 2007; 48: 546-8.
- 21.Zhou ZW, Wan DS, Chen G, Chen YB, Pan ZZ. Primary malignant tumor of the small intestine. World J Gastroenterol 1999; 5: 273-6.
- 22.Okano A, Takakuwa H, Matsubayashi Y. Aortoduodenal fistula resembling a submucosal tumor due to penetration of abdominal aortic aneurysm. Intern Med 2005; 44: 904-5.
- 23.Cohen J, Shapiro M, Grozovski E, Haddad M, Hananel N, Singer P. An unusual cause of intraabdominal hemorrhage: ruptured hepatic artery aneurysm. Isr Med Assoc J 2000; 2: 555-6.
- 24.Suat C, Ali K, Enver D. Hepatic Artery Aneurysms. Internet J Thorac Cardiovasc Surg 2005; 7: 1-2.
- 25. Vanessa WD, Karim S, Todd PC, Jennifer A, James SAM. Hepatic artery aneurysm secondary to epithelioid angiosarcoma. Can J Surg 2008; 51: 81-2.
- 26.Sotirios P, Lioudmila H, Edwin G, Leonard B, Elizabeth C. Hepatic artery aneurysm erosion into the stomach: an unusual cause of gastrointestinal bleeding. J Emerg Med 2007; 11: 112.
- 27.Jaunoo SS, Tang TY, Uzoigwe C, Walsh SR, Gaunt ME. Hepatic artery aneurysm repair: a case report. J Med Case Reports 2009; 3: 18-23.

# 總肝動脈動脈瘤以十二指腸黏膜下腫瘤表現 而導致上消化道出血—病例報告

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### 摘要

總肝動脈動脈瘤是上消化道出血的罕見病因。在内視鏡的檢查中,總肝動脈動脈瘤可以 因爲對十二指腸的外源性壓迫而以黏膜下腫瘤來呈現。這類動脈瘤的出血通常是突然發生的 而且再出血的風險很高。所以快速診斷與早期介入治療是必須的。我們報告一例中年男性患 有以總肝動脈動脈瘤所形成的十二指腸黏膜下腫瘤而導致的上消化道出血。經過內視鏡血管 夾止血,隨後的腹部電腦斷層掃描發現一個緊臨十二指腸球部的破裂之總肝動脈動脈瘤。在 隔日患者併發復發性的上消化道出血和低血容性休克。之後病患接受外科修補術而且獲得良 好的復原。在術後的六個月追蹤期間內病人不曾再有上消化道出血發生。我們歸納結論如下 ,血管動脈瘤性的病灶應該列入十二指腸黏膜下腫瘤的鑑別診斷。而血管夾對於疑似血管性 的病灶是一項較好較安全的技術以達暫時性的止血。之後進一步診斷如電腦斷層檢查必須儘 快獲得以便釐清黏膜下腫瘤的原因,並提供進一步治療的參考。對於血管瘤破裂所導致的消 化道出血,外科或放射科介入才是根本的治療方式。