Pseudoaneurysm of the Cystic Artery in Acalculous Cholecystitis Successfully Treated by Transcatheter Arterial Embolization: a case report

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Pseudoaneurysms of the cystic artery are rare complications related to cholecystitis. A 74-year-old male visited our Emergency Department (ED) for abdominal pain. He was febrile and jaundiced. However, no hematemesis or melena was noted. Physical examination showed no Murphy sign. Computer Tomography (CT) showed a pseudoaneurysm near the gallbladder fossa. The patient was managed with transcatheter arterial embolization (TAE) due to his poor condition. Scheduled cholecystectomy three weeks later showed residual chronic inflammatory process without cholelithiasis. This is the first documented case of a pseudoaneurysm caused by acalculous cholecystitis. The case also represents a rare instance in which a cystic artery pseudoaneurysm was successfully managed by TAE in the English literature.

Pseudoaneurysm of the cystic artery is a rare phenomenon, which occurs as a post-operative complication, a result of traumatic injuries or, more infrequently, a result of calculous cholecystitis [1]. It carries the potential to cause tremendous hemorrhagic shock, and thus should be detected earlier for better prognosis [2]. Pseudoaneurysm of the visceral organs can be well-demonstrated by color Doppler ultrasound, contrast-enhanced computed tomography (CT) and angiography. In the case reported here, the pseudoaneurysm was not caused by cholelithiasis according to operative findings. It was successfully embolized with microcoils.

CASE REPORT

A 74-year-old male patient who had experienced symptoms including abdominal pain, aggravated nausea and vomiting for a week visited our emergency department (ED). Initial laboratory data showed: leukocytosis (white blood cell: 16400/ul) with dominant neutrophils (91%), hemoglobin 9.4 g/ml, elevated bilirubin (total/direct: 6.2/4.8mg/dl), and elevated C-reactive protein (12.99mg/dl), while other data revealed unremarkable findings. Clinical impression of obstructive jaundice of an indeterminate level was highly suggested and thus abdominal CT was performed to delineate the obstruction level. CT scan (HiSpeed CT/I, General Electric Healthcare, U.S.A.) showed a distended gall bladder (GB) with marked wall thickening and peri-cholecystic fat stranding, favoring a diagnosis of cholecystitis. A strong-enhancing nodule with vascular attenuation embedded in the high-attenuation area in the submucosal layer of the GB (Fig. 1) was also found. Pseudoaneurysm within the gallbladder wall complicated with hematoma formation was considered. The initial diagnostic angiography was performed...
with 4.1 French Rosch Left Gastric catheter (Cook Bloomington, IN) and showed a pseudoaneurysm in celiac angiogram (Fig. 2a). A superselective cystic artery angiogram with a 3 French microcatheter (Renegade, Boston Scientific Corporation, Natick, MA) showed dilated distal branches of cystic artery with contrast stasis and extravasation (Fig. 2b). These findings confirmed pseudoaneurysm of the cystic artery. Due to the patient’s unstable vital signs, an emergency operation was not recommended, and TAE was therefore arranged. Embolization was initially planned with placement of two fibered platinum microcoils (2 mm in diameter; 23 mm in length) (Boston Scientific International, La Garenne Colombes, Cedex) at the parent artery of the pseudoaneurysm to lower the risk of GB infarction. However, follow-up angiogram showed residual aneurysm from collaterals. Therefore we placed additional two microcoils (2 mm in diameter, 41 mm in length) at the proximal part of the cystic artery (Fig. 2c) to achieve complete occlusion of the cystic artery. Percutaneous transhepatic gallbladder drainage (PTGBD) was recommended after TAE but the patient refused the treatment. Therapeutic aspiration of GB of 60 ml turbid fluid was performed once. The patient finally agreed to surgery three weeks later for persistent abdominal pain. Open cholecystectomy revealed empyema of GB and hematoma within the GB wall. A thrombosed pseudoaneurysm in the submucosal layer of the GB was depicted. No dilatation of common bile duct or intrahepatic duct was depicted.

**DISCUSSION**

Cystic artery pseudoaneurysm is a rare entity, with only 18 cases reported in the English literature. The majorities are complications of laparoscopic cholecystectomy [3]; while the rest are complications of acute or chronic cholecystitis [4]. The rarity of non-traumatic pseudoaneurysm of the cystic artery.

**DISCUSSION**

Cystic artery pseudoaneurysm is a rare entity, with only 18 cases reported in the English literature. The majority are complications of laparoscopic cholecystectomy [3]; while the rest are complications of acute or chronic cholecystitis [4]. The rarity of non-traumatic pseudoaneurysm of the cystic artery.

**Figure 1.** CT scan showing a distended gallbladder with marked wall thickening, peri-cholecystic fat stranding. There is a strong-enhancing nodule identical to vascular attenuation embedded in the high-attenuation area in the submucosal layer of the gallbladder wall.

**Figure 2.** a. Digital subtraction angiography (DSA) of the celiac axis in this patient revealed a pseudoaneurysm of a branch of the cystic artery (white arrow) from proper hepatic artery (black arrow) bordering the gallbladder. b. The tip of microcatheter (black arrowhead) was placed in the branch of the cystic artery. DSA showed detailed distal branches with active extravasation (star) and pseudoaneurysm (white arrowhead). c. After embolization with four microcoils, the controlled DSA showed absence of the pseudoaneurysm and cystic artery.
may reflect early thrombosis of the cystic artery as a reaction to the inflammation [4, 5]. Although cholecystitis is a common disease in daily practice, the formation of a pseudoaneurysm and subsequent rupture into the gall bladder or intraperitoneum is infrequent. The Quincke’s triad of hemobilia consists of RUQ pain, jaundice and gastrointestinal bleeding, either as hematemesis or melena. However each symptom presents in about two-thirds of cases [6, 7]. Unrecognized or delayed diagnosis can lead to severe complications such as hypovolemic shock due to uncontrollable intra-peritoneal bleeding and the mortality rate can be as high as 50% [2, 3, 8].

Understanding of the development of the pseudoaneurysm remains speculative. It is believed to develop from the erosion of an inflamed gall bladder into the wall of the cystic artery. Unlike most previous cases, no melena was observed in our patient. We assumed that the hemorrhage of the ruptured pseudoaneurysm was confined within the wall of GB. Other authors have also suggested that the intact sphincter of the cystic duct and common bile duct might obscure melena [1]. The role of cholelithiasis in pseudoaneurysm formation is not clear [2, 9]. Limited information on this from pre-treatment imaging and post-operative findings has appeared in the previous literature. Our case is the first one of acalculous cholecystitis complicated with pseudoaneurysm of cystic artery in the English literature.

The diagnosis can be accurately made by ultrasound, CT, or arteriography. The “yin-yang” sign may be of diagnostic value for such cases in Doppler ultrasonography [6, 9]. CT is a highly accurate method of investigating the hepato-biliary system and can clearly delineate the vascular lesion in the upper abdomen. The diagnostic value of arteriography is enhanced by the possibility of performing a therapeutic procedure. In our case, the initial angiography was for diagnostic purpose, and thus embolization was performed with the consent of the surgeon and the radiologist on duty because the patient’s vital signs were unstable.

TAE is recommended for the appropriate management of pseudoaneurysm of the cystic artery in cholecystitis because during surgery, identification and ligation of the aneurysmal cystic artery in an inflammatory territory may be difficult, especially if bleeding is serious and uncontrolled. Whenever the diagnosis is revealed by arteriography, TAE should be attempted because it can always achieve hemo-

stasis and potentially successful total obliteration of the aneurysm in unstable patients with hypovolemic shock or sepsis [7, 10]. The effect of PTGBD is not clear since there is no report in the English literature in which treatment with TAE was followed by PTGBD. However, PTGBD or aspiration should be attempted to release the distended and infected gall-bladder to speed recovery.

**CONCLUSION**

This is the first case of pseudo-aneurysm of the cystic artery caused by acalculus cholecystitis in the English literature. Accurate diagnosis is potentially challenging as the symptoms and signs could easily be mistaken for those of simple acute cholecystitis or unspecified gastrointestinal bleeding without detailed imaging. Early detection by CT and angiography facilitates timely and proper management with TAE or surgery.

**REFERENCE**

非膽結石膽囊炎造成膽囊動脈的假性動脈瘤成功地
由經皮動脈栓塞術治療：病例報告

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膽囊炎造成在膽囊動脈的假性動脈瘤並不常見。有一位七十四歲的男性病患因為右上腹痛
而來我們急診求診。他有發燒和黃疸的症狀，但是並沒有吐血或血便的症狀。因疑似膽囊炎而
作的電腦斷層發現在膽囊附近的假性動脈瘤。因為病人狀況不佳所以採取經皮動脈栓塞術治療。
三個星期後開刀看到慢性發炎的痕跡但卻找不到膽結石。這是第一個在英文文獻有具體證明的
非膽結石膽囊炎造成在膽囊動脈的假性動脈瘤，並成功地由經皮動脈栓塞術治療。