## CYSTIC ARTERY PSEUDOANEURYSM WITH CONCURRENT CHOLECYSTO-ENTERIC FISTULA: A RARE CASE REPORT

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**ABSTRACT:** Cystic artery pseudo aneurysm with cholecysto-enteric fistula represents two rare complications of gallstone disease. We report a case of 68 year old female who presented to the emergency department with pain in the right upper quadrant since 15days with presence of slight tenderness in this region and history of hematemesis. Ultrasound and MDCT was done, findings showed diffuse irregular GB wall thickening with large calculus in its lumen with focal defects in walls and loculated fluid with membranes in the pericholecystic region around body and fundus. A well-defined small pseudo aneurysm was seen projecting into lumen of gall bladder in left lateral aspect likely arising from cystic artery. Small focal communications were also evident between GB lumen and duodenum. This patient was operated, Partial cholecystectomy was performed and Choledochoduodenostomy was done.

**KEYWORDS:** Cystic artery pseudoaneurysm, cholecysto-enteric fistula, gallstone disease, subtotal/partial cholecystectomy.

**INTRODUCTION:** Rupture of Cystic artery pseudo aneurysms remain a rare cause of upper gastrointestinal bleeding.<sup>1, 2</sup> They develop primarily as a consequence of adventitial damage and thrombosis of the vasa vasorum, resulting in damage to the muscular and elastic components of the media and intima with ensuing extravasation of arterial blood, progressive enlargement and eventual rupture.<sup>2-5</sup> This can occur secondary to inflammatory conditions(e.g. cholecystitis, pancreatitis), malignancy, biliary tract manipulation or trauma. Formation may be further accelerated by patient factors, such as atherosclerosis, hypertension, bleeding dis- orders and vasculitis.<sup>1-8</sup> Cystic artery pseudo aneurysms tend to enlarge and erode into the gallbladder and adjacent biliary tree with approximately 45% bleeding into the biliary system(haemobilia).<sup>9</sup> The clinical presentation is that of biliary colic(70% of cases), obstructive jaundice(60%) and upper gastrointestinal bleeding(100%). 32–40% of patients will present with all three symptoms – Quincke's Triad.<sup>6-8, 10</sup>

**CASE PRESENTATION:** 68 yrs old female who presented to the emergency department with pain in the right upper quadrant since 15days with presence of slight tenderness in the region, with signs of jaundice. There was also history of hematemesis.

Ultrasound was done; findings showed a mildly increased liver size measuring approx. 16.2cm. Contracted gall bladder with large calculus of size 20mm in its lumen. GB was heterogenous and irregular in outline. A cystic structure that produced swirling flow signals on Doppler was seen in gall bladder.

CBD was dilated and showed multiple small echogenic foci were seen of size approx. 4-5mm. The above USG findings were suggestive of Choledocolithiasis with cystic artery aneurysm. Therefore CT scan was advised.

CECT scan of abdomen was performed, that showed enlarged GB and diffuse irregular GB wall thickening with focal defects in walls and loculated fluid with membranes in the pericholecystic region around body and fundus.

Intraluminal hyper attenuation (+64) was seen in lumen of GB on plain scan s/o hemorrhage. On post contrast scan a well-defined small pseudoaneurysm measuring 9mm x 9mm was seen projecting into lumen of gall bladder in left lateral aspect likely arising from cystic artery. Thickened GB wall abuts the walls of pyloro-duodenal canal with loss of fat planes. Small focal communications were also evident between GB lumen and duodenum. Compression of duodenal lumen was seen.

There was also loss of fat planes between thickened GB wall and liver parenchyma. CBD was dilated in entire extent measuring 11-12mm and showed intraluminal foci of soft tissue and mild hyper attenuation-?calculi. Mild dilatation of IHBR was seen, more in the central portion.

### The above CT findings Showed:

- 1. Perforated membranous/ gangrenous cholecystitis with pseudo aneurysm of cystic artery and suggestion of fistula between gall bladder lumen and pyloro-duodenal canal.
- 2. Dilated CBD with intraluminal calculi/ clots.

This patient was followed up and was managed Surgically: Subcostal incision was given. Stomach was found to be adherent to gall bladder. On separating them, aneurysm of cystic artery got teared. Heavy bleeding occurred this was controlled by pressure and figure of 8 sutures. Stomach was separated and a fistulous tract was found between anterior pyloric wall, 1st part of duodenum and gall bladder. Fundus of gall bladder was opened and a calculus of size 3cmx2cm was extracted along with clots.

**Partial or subtotal cholecystectomy** was performed. CBD was identified, stay suture taken over the CBD. Pylorus fistulous tract was separated in two layers, CBD was opened and explored and stone was removed.

**Choledocho-duodenostomy was done.** Feeding tube was placed in CBD. Taken out from GB stump. Drain placed in Morrissons pouch. Closure was done.



Fig. 1: USG-SHOWING PSEUDOANEURYSM



Fig. 2: USG SHOWING COLOR UPTAKE ON DOPPLER MODE



Fig. 3: MDCT Axial View



Fig. 4: MDCT Axial View



Fig. 5: Intraoperative finding



Fig. 6: Intraoperative finding

**DISCUSSION:** Rupture of Cystic artery pseudo aneurysms remain a rare cause of upper gastrointestinal bleeding, with only twenty-two documented cases in the English literature. They

develop primarily as a consequence of adventitial damage and thrombosis of the vasa vasorum, resulting in damage to the muscular and elastic components of the media and intima with ensuing extravasation of arterial blood, progressive enlargement and eventual rupture. This can occur secondary to inflammatory conditions (e.g. cholecystitis, pancreatitis), malignancy, biliary tract manipulation or trauma. Formation may be further accelerated by patient factors, such as atherosclerosis, hypertension, bleeding disorders and vasculitis. Cystic artery pseudo aneurysms tend to enlarge and erode into the gallbladder and adjacent biliary tree with approximately 45% bleeding into the biliary system (haemobilia). The clinical presentation is that of biliary colic (70% of cases), obstructive jaundice (60%) and upper gastrointestinal bleeding (100%). 32–40% of patients will present with all three symptoms.

Pseudo aneurysms are differentiated from true aneurysms by the presence of a known cause such as trauma or inflammation, and by radiologic findings of aneurysmal dilatation in the setting of otherwise normal artery. Causes of cystic artery aneurysms include abdominal trauma and intraabdominal inflammatory processes such as cholecystitis or pancreatitis.<sup>11</sup>

The exact mechanism to develop a pseudo aneurysm and the association between pseudo aneurysm and inflammatory process such as cholecystitis remains unclear. In the review of 16 cases of cystic artery pseudo aneurysm caused by cholecystitis, Akatsu et al. assumed that the acute inflammation of the gallbladder wall may cause both ulceration of serosa and partial erosion of the elastic and muscular components of the arterial wall, thus leading to the formation and rupture of the pseudo aneurysm. The presence of gall stone may accelerate the formation of the pseudo aneurysm formation and weakness of arterial wall due to arterial sclerosis may also accelerate the formation of the pseudo aneurysm. <sup>13</sup>

**CONCLUSION:** Cystic artery pseudo aneurysms remain a rare cause of upper gastrointestinal bleeding. The initial investigation for a patient presenting in this manner is Color Doppler Ultrasonography, which will demonstrate a pseudo aneurysm as an anechoic lesion with color flow through it. Contrast-enhanced CT scanning and 3-dimensional CT angiography should replace arteriography as a non-invasive method of establishing a definitive diagnosis. However, many sources state that, although invasive, selective hepatic artery angiography is the gold standard for diagnosis, with a sensitivity of 80% and the ability to define aneurysms <10 mm. Diagnostic angiography can then be combined with therapeutic arterial embolization with foam, thrombin or micro-coils, either as definitive management or as a method of hemostasis prior to cholecystectomy and aneurysm repair.

We have demonstrated the success of surgical management alone in the treatment of such a case but accept that, when available and clinically indicated, a combined therapeutic approach with angiography should be considered in the first instance.

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