

Case Report

Cystic artery pseudo aneurysm in a patient with xanthogranulomatous cholecystitis: a multi-disciplinary approach towards management

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ABSTRACT

Cystic artery pseudo aneurysm is a rare clinical entity, mostly occurring as a complication of laparoscopic cholecystectomy or following calculus cholecystitis. Patient presents with acute symptoms including upper gastro intestinal bleeding symptoms, abdominal pain and jaundice, the triad of symptoms termed as Quinke's triad, when there is a ruptured cystic artery pseudoaneurysm (CAP). Unruptured cases are mostly diagnosed incidentally. Here we present a 58-year-old patient presented to the emergency department with severe abdominal pain, malena and jaundice. CECT of abdomen was done which showed CAP. The patient was immediately taken up for radiological intervention and coiling of the pseudo aneurysm, which was unsuccessful. A covered stent was placed across the pseudoaneurysm in the right hepatic artery. Patient underwent Laparoscopic cholecystectomy and had an uneventful recovery. Owing to the fact that CAPs are rare entities, a high index of suspicion is required for diagnosis of the condition and when diagnosed, the patient can be managed by radiological intervention followed by definitive surgery which can be either open or laparoscopic.

Keywords: Pneumobilia, CAP, Quinke's triad, Xanthogranulomatous cholecystitis, Multi-disciplinary approach

INTRODUCTION

Cystic artery pseudoaneurysms (CAPs) are rare. Mostly, they occur secondary to acute cholecystitis/ as complication following laparoscopic cholecystectomy, and most common clinical presentation is hemobilia, which can be difficult to diagnose clinically. Complications include hemobilia, biliary obstruction and haemorrhage.¹

Most patients have h/o cholecystitis/have undergone a cholecystectomy and very rarely it was found to be associated with xanthogranulomatous cholecystitis.² Even though pathophysiology of CAP in cholecystitis is unclear, it is hypothesized to occur most likely due to inflammatory damage of the adventitia in cholecystitis.³

Given the rarity and associated morbidity, a high index of suspicion and prompt investigation with targeted imaging

and intervention is required. It is especially pertinent in gastrointestinal bleeding post laparoscopic cholecystectomy, as a missed diagnosis could cause significant morbidity.¹ Here we present a case of a 58-year-old gentleman who presented to our emergency department with severe abdominal pain, and upper Gastro Intestinal bleeding, later being diagnosed as CAP and was managed by a multidisciplinary approach.

CASE REPORT

A 58 years old gentleman, came to the emergency department in pre syncope, with complaints of sharp pain at right upper quadrant, associated with one episode of hemetemesis and malena. He was having on and off right upper abdominal non radiating pain associated with nausea and vomiting since past 1-2 months and was diagnosed as having gall bladder stones, one-month back

He is a known case of type II diabetes mellitus, systemic hypertension and hypothyroidism for which he is taking medications regularly.

On clinical examination, he was found to be severely pale and anicteric. His pulse rate was within normal range and he was hypotensive. Abdominal examination revealed mild tenderness at right hypochondrium without any distension, rigidity or guarding.

Basic blood investigations and imaging studies were done. His hemoglobin was found to be 5.6 mg/dL, total bilirubin being 1.6 and lactate levels of 1.15

CECT abdomen showed solitary large calculus measuring 3.8×2.2 cm within the lumen of gallbladder, gall bladder wall thickening (8 mm) and pericholecystic thin rim of fluid. There was a smooth well-defined 17×12 mm ovoid intensely enhancing lesion within the gall bladder closely abutting the right hepatic artery, likely arising from the cystic artery, representing pseudo aneurysm of cystic artery.



Figure 1: CECT abdomen showing the large solitary calculus with CAP in the arterial phase.

A multidisciplinary approach to the disease, was planned. The patient was assessed by Interventional Radiologist team, angiogram was done, which shows slow filling

aneurysm arising from the junction of cystic artery origin from hepatic artery. As cystic artery was inaccessible, a covered stent was deployed across the pseudoaneurysm. Post stenting angiogram was done, which showed non filling of the pseudoaneurysm and normal filling of hepatic artery branches.

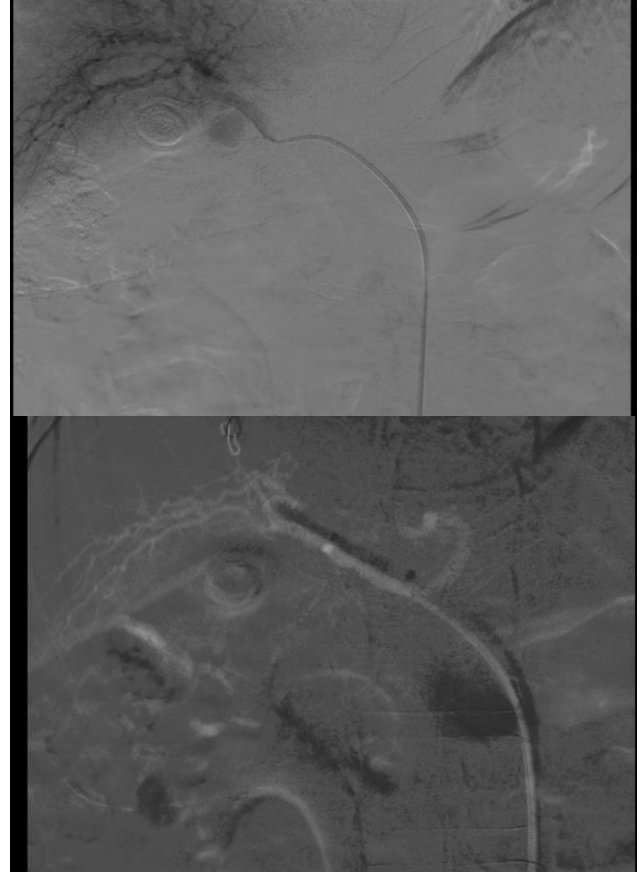


Figure 2: Angiogram images before and after the radiological intervention.

He was then shifted to ICU and managed with multiple blood transfusions, IV antibiotics, IV fluids, strict monitoring of vitals and Hemoglobin levels. Once the patient was stabilized clinically, ERCP was done by the medical gastrology team, which showed CBD sludge. They proceeded with biliary sphincterotomy and Biliary stenting.

Laparoscopic cholecystectomy was planned for him on the third day of admission. Under general anesthesia, using standard ports, right hypochondrium accessed. There were dense omental adhesions to the gall bladder, and colon was adherent to fundus of the gall bladder. Inflamed, thick-walled gall bladder with frozen calots. Thrombosed pseudoaneurysm with clot was found at the junction of cystic artery with the gall bladder, with solitary large calculus at neck of the gall bladder. The omental and colonic adhesions were gently released, calots was meticulously dissected, cystic artery was clipped without disrupting the aneurysm and cholecystectomy was done.

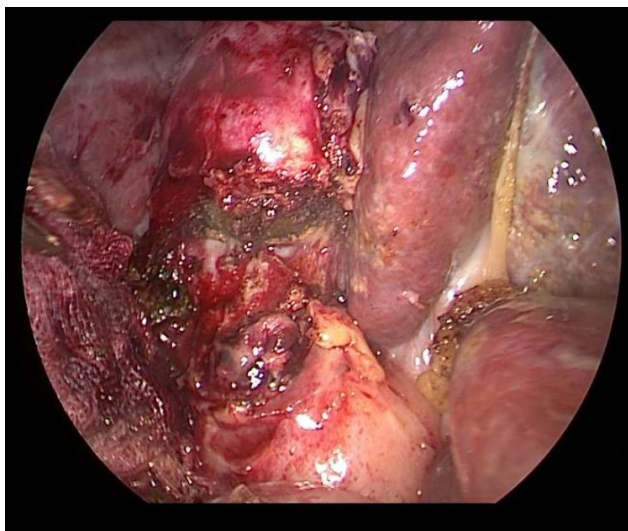


Figure 3: Intra operative image showing inflamed gall bladder with the thrombosed CAP.

Post operatively he was shifted to ICU, with regular monitoring of vitals and liver function tests and coagulation profile. He had an uneventful recovery and was discharged on Post-operative day third. His histopathology report showed gall bladder wall with surface ulceration and underlying pulsion diverticula, transmural chronic inflammation and fibrosis and focal sheets of histiocytic collection surrounded by dense chronic inflammatory infiltrate with luminal pigment concretions, the features which were consistent with chronic xanthogranulomatous cholecystitis.

On review, he was found to be comfortable and clinically stable with no recurrence of the symptoms. His liver function parameters were found to be within normal limits. He was advised regular ultrasound abdomen/Doppler imaging to monitor any re-canalization of the pseudo aneurysm

DISCUSSION

CAP is an uncommon complication of a very common clinical condition, i.e., Cholecystitis. It's also been postulated as a rare complication following cholecystectomy. Even though the etiopathogenesis is unclear, it is hypothesized based on recent literature as due to the inflammatory damage to the adventitia due to cholecystitis, and in post cholecystectomy patients, due to vascular erosion from manipulation, clip application or thermal injury.^{1,3}

A rare association of CAP with xanthogranulomatous cholecystitis has been described in literature. XGC is characterized by multiple yellow brown intramural nodular formation, severe proliferative fibrosis and foamy macrophages. Although the etiopathogenesis is not clearly explained, it is generally agreed that rupture and intramural extravasation of the inspissated bile and mucin from the occlusion of Rokitansky-Aschoff sinuses is the

main cause for the development of XGC. The other possible cause is gallstone with bile stasis and a chronic inflammatory reaction that provokes degeneration and necrosis of the gall bladder wall with subsequent intramural abscess formation. The intra mural abscesses are subsequently replaced by xanthogranuloma with foamy histiocytic foreign body giant cells. Pervascular inflammation causes thrombosis of vasa vasorum with damage to adventitia. These vessels are prone to rupture with the formation of pseudo aneurysms.³

In ruptured CAP, patients most commonly present with signs of shock and hemobilia, itself is noted to have a triad of symptoms, known as Quincke's triad, including jaundice, right upper quadrant pain and upper gastro intestinal haemorrhage.⁴ Unruptured CAPs are very rare.

The best diagnostic modality for the CAP is an arterial phase contrast- enhanced CT which is a readily available treatment modality in most of the critical care centres. Colour Doppler ultrasound and MRI are options for patients for whom IV contrast is contra indicated.¹

Owing to the fact that CAP is a rare clinical condition, there is no consensus on the clinical management of this disease. A variety of treatment strategies have been reported in the literature including radiological selective embolization and coiling, open cholecystectomy with ligation of the aneurysm, or 2-step approach involving radiological management of the pseudoaneurysm followed by an elective cholecystectomy.⁵ In hemodynamically unstable patients with ruptured pseudoaneurysms, angiographic embolization might be performed prior to definitive surgery.⁶ But the complications following trans arterial embolization are hepatobiliary necrosis, bleeding, abscess formation in the gall bladder, gall bladder necrosis, catheter related complications such as dissection, arterial-venous fistula formation, contrast related complications, etc. However, this offers a useful time-buying temporary measure for definitive management.⁷

In our case, we opted for a laparoscopic procedure, following an attempt at embolization and coiling. Even though he had inflamed and densely adherent gallbladder with a thrombosed CAP, we were able to achieve a better vision and plane of dissection laparoscopically, and successfully deploy the clip to artery followed by dissection of the gall bladder from the liver bed.

CONCLUSION

CAP is a rare complication of a very common disease - cholecystitis or it can develop in patients after cholecystectomy. Very few numbers of cases have reported. Most commonly, the patients present with hemobilia features of which include upper abdominal pain, jaundice and upper gastrointestinal bleed together known as Quincke's triad. Being a rare entity, high index of suspicion clinically, along with prompt investigations

and targeted imaging is required for the early diagnosis, as a delayed diagnosis results in high morbidity. The best investigation modalities include CECT abdomen, colour Doppler ultrasound and MRI. There have been no specific guidelines designed for CAP due to its rarity. The different treatment strategies reported include Radiological selective embolization and coiling, an open cholecystectomy and ligation of cystic artery or a combination of both as 2-step approach. But a radiological intervention if accessible followed by a laparoscopic approach to removal of the gall bladder with cystic artery ligation can be the safest option of management in expert hands.

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REFERENCES

1. Taghavi SMJ, Jaya KM, Damodharan PR, Puhalla H, Sommerville C. Cystic artery Pseudoaneurysm: Current Review of Aetiology, Presentation, and Management. Surg Res Pract. 2021;2021:4492206
2. Ahmed I, Tanveer UH, Sajjad Z, Munazza B, Azeem UD, Basit S. Cystic artery pseudo-aneurysm: a complication of xanthogranulomatous cholecystitis. Br J Radiol. 2010;83(992):e165-7.
3. Jaganathan S, Rangarajan M, Jyothirmayi V. Cystic artery pseudoaneurysm: an uncommon complication of a common disease. Int Surg J. 2022;9(11):1894-97.
4. Kelly S, Shah K, Motizada J, Soggiu F, Sheth H. Laparoscopic treatment of an unruptured cystic artery pseudo-pseudoaneurysm in the presence of calculous emphysematous cholecystitis. J Surgical Case Rep. 2022;1:1-3
5. Sofronis L, Asad A, Richard N, Krishna Kumar Singh, Laparoscopic management of a cystic artery pseudoaneurysm in a patient with calculous cholecystitis, International Journal of Surgery Case Reports. 2015;14:182-5.
6. Deniz Allis, Sina Ferahman, Suleyman Demiryas, Cesur Samanci, Fethi Emre Ustabasioglu, "Laparoscopic Management of a very Rare Case: Cystic Artery Pseudoaneurysm Secondary to Acute Cholecystitis. Case Repo Surg. 2016;1489013:1-3.
7. She WH, Tsang S, Poon R, Cheung TT. Gastrointestinal bleeding of obscured origin due to cystic artery pseudoaneurysm. Asian J Surg. 2010;40(4):320-3.

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