

Review Article

Cystic Artery Pseudoaneurysm: Current Review of Aetiology, Presentation, and Management

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Received 18 May 2021; Revised 28 October 2021; Accepted 5 November 2021; Published 24 November 2021

Academic Editor: Miltiadis I. Matsagkas

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Background. Cystic artery pseudoaneurysms are rare. Most commonly, they occur secondary to acute cholecystitis or after a cholecystectomy. Complications include haemobilia, biliary obstruction, and haemorrhage. Given the rarity and associated morbidity, a high index of suspicion is required. This article reviews the current literature on cystic artery pseudoaneurysms to investigate its aetiology, clinical presentation, and management options. **Methods.** A broad search of the Medline and PubMed databases was carried through. All peer reviewed literatures published in the English language between 1991 and 2020 with keywords “cystic” and “artery” and “pseudoaneurysm” in the title were selected for review. No further exclusion criteria; all studies yielded from the search were included in the results of this review. Additionally, we present a case of cystic artery pseudoaneurysm treated at our centre and included this in our analysis. **Results.** Sixty-seven case reports were found between 1991 and 2020. Aetiologies: Aetiology of cystic artery pseudoaneurysm was found to be cholecystitis in 41 instances (61.2%), cholecystectomy in 18 instances (26.8%), idiopathic in 6 instances (8.9%) cholelithiasis in 1 instance (1.5%), and pancreatitis in 1 instance (1.5%). Complications: Fifty-two cases were complicated by haemobilia (77.6%), 36 by anaemia (53.7%), 25 by biliary obstruction (37.3%), 13 by haemodynamic shock (19.4%), 9 by haemoperitoneum (13.4%), and 6 by contained rupture (8.9%). Most commonly, patients had two or more of these complications. Management: Forty-four patients were managed with endovascular embolisation (65.7%), 21 with endoscopic intervention (31.3%), 18 with open cholecystectomy (26.9%), 13 with laparoscopic cholecystectomy (19.4%), and 6 with pseudoaneurysm ligation (9%). Delayed presentation postcholecystectomy ranged from 8 days to 3 years. **Conclusions.** Cystic artery pseudoaneurysms are rare complications of a common operation. The most common clinical presentation is haemobilia, which can be difficult to diagnose clinically. A high index of suspicion and prompt investigation with targeted imaging and intervention is required. This is especially pertinent in gastrointestinal bleeding postlaparoscopic cholecystectomy as a missed diagnosis could cause significant morbidity.

1. Background

Laparoscopic cholecystectomy (LC) is a routine surgical procedure for the treatment of gallbladder pathology. In Australia, there are an approximate 50,000 annual hospitalisations for LC [1]. The incidence of intraabdominal vascular complications from LC is uncertain. Published data report incidences as low as 0.25% and as high as 7%, although there is variation in the patient population and investigation methods between studies [2, 3]. Cystic artery

pseudoaneurysm (CAP) is a rare vascular complication of LC. Other aetiologies for CAP include cholecystitis, cholelithiasis, and idiopathic CAP [4–6]. The complications of CAP include haemobilia, anaemia, biliary obstruction, and haemoperitoneum and haemodynamic shock [3]. The classical clinical syndrome of haemobilia is described by Quinke’s triad: jaundice, right upper quadrant abdominal pain, and upper gastrointestinal bleeding, present in approximately 22–35% of cases [6]. In this article, we present a thorough review of the current published literature to

identify the aetiology, clinical presentation, and management of CAP. We also present a case of CAP rupture postcholecystectomy managed at our centre and included this in our data analysis.

2. Methods

A broad search of the Medline and PubMed databases was carried out. All peer reviewed literatures published in the English language between 1991 and 2020 with the keywords “cystic” and “artery” and “pseudoaneurysm” in title were selected for review. No further exclusion criteria; all studies yielded from our search were ultimately included in the results of this review. Additionally, we present, with written consent from the patient, a case of cystic artery pseudoaneurysm treated at our centre and include this in our analysis.

3. Results

3.1. Case Report. A 29-year-old male underwent an uneventful laparoscopic cholecystectomy for recurrent biliary colic. There was no aberrant anatomy noted, and intraoperative cholangiography was normal. On postoperative day 8, he represented to the emergency department with increasing abdominal pain. On examination, he was tachycardic with a distended tender abdomen, and a periumbilical haematoma was noted. Initial investigation showed a haemoglobin of 136 g/L, a white cell count of $16.5 \times 10^9/L$, and normal liver function and coagulation studies. Initial contrast CT of the abdomen (Figure 1) showed a gas and fluid collection at the gallbladder fossa and large volume haemoperitoneum. No active contrast extravasation was detected. The patient proceeded to laparoscopic washout, and 1.5 L of blood and clot was evacuated from all quadrants. It was noted that the cystic artery and duct clips were intact. No obvious bleeding point was identified. Two 19F Blake drains were placed in the gallbladder fossa and pelvis. The patient recovered uneventfully postoperatively, and the drains were removed on postoperative day 4. Interval CT angiography was performed on postoperative day 5 to further investigate the cause of delayed bleeding. This showed resolution of haemoperitoneum but revealed a 3 mm arterial hyperenhancement in the gallbladder fossa consistent with a pseudoaneurysm of the cystic artery (Figure 2). Follow-up digital subtraction angiography showed a cystic artery pseudoaneurysm and another pseudoaneurysm in the right hepatic artery branch to segment 4a. Both pseudoaneurysms were successfully embolised with a combination of Tornado® Coils (Cook Medical, Ireland) and Gelfoam® (Pfizer, USA). The patient was discharged on postoperative day 9 and was well at two-month follow-up. No further surveillance was required.

3.2. Literature Review. Sixty-seven case reports of CAP were identified between 1991 and 2020, including the patient treated at our centre. The mean age at diagnosis was 60 years, with a range of 23–91 years. Forty-five of 67 patients (67%) were male.

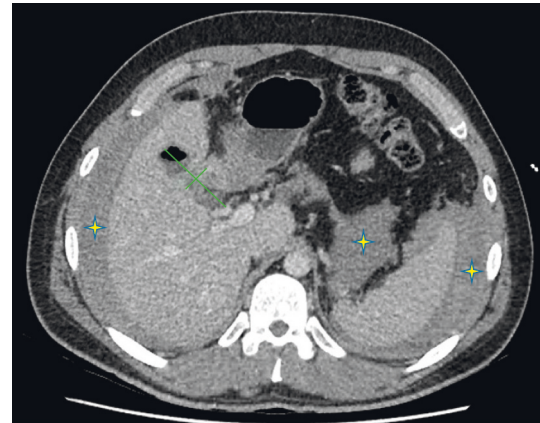


FIGURE 1: Computed tomography scan of the abdomen in the axial plane showing a gallbladder fossa collection (green marks), retroperitoneal, perihepatic, and perisplenic haematomas (yellow stars).

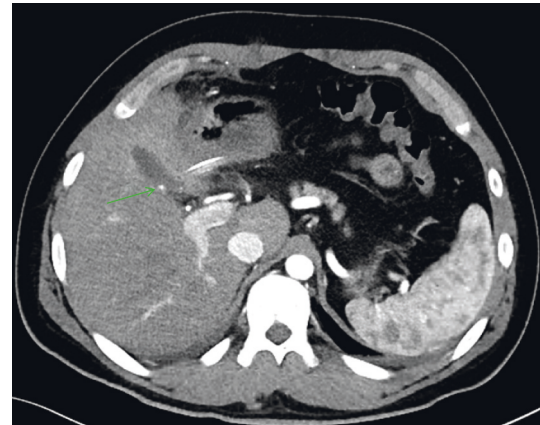


FIGURE 2: Contrast-enhanced computed tomography scan of the abdomen in the axial plane showing a cystic artery pseudoaneurysm (green arrow).

3.2.1. Aetiologies. Of the 67 cases of CAP reported, 4 cases were secondary to cholecystitis (61.2%) [7–44], 18 followed cholecystectomy (26.8%) [45–61], 6 were idiopathic (8.9%) [5, 62–65], 1 correlated with cholelithiasis (1.5%) [4], and 1 with pancreatitis (1.5%) [66]. Of the patients with CAP postcholecystectomy, the median postoperative time to clinical presentation was 50 days, with a range of eight days (in our patient) to three years [52].

3.2.2. Clinical Presentations. Fifty-eight of 67 patients (86.5%) presented with right upper quadrant pain. Fifty-two presented with haemobilia (77.6%). Seventy-seven percent of the patients who presented with haemobilia ($n = 40/52$) had clinical upper gastrointestinal bleeding (haematemesis or melena) and subsequent endoscopy confirming bleeding from the ampulla of Vater. The remaining 23% of patients with haemobilia ($n = 12/52$) were diagnosed on endoscopic retrograde cholangiopancreatography (ERCP), intraoperatively during laparoscopy or laparotomy, or on

insertion of a percutaneous cholecystostomy tube as part of management for cholecystitis.

Twenty-five of 67 patients (32.8%) presented with obstructive derangement of liver function tests. Of these, 22 were secondary to haemobilia and 3 had biliary obstruction due to direct compression of the bile duct by CAP. Nineteen of 22 patients with biliary obstruction due to haemobilia had clinical jaundice. All 3 of the patients with biliary obstruction due to direct compression of the bile duct were jaundiced. Overall, Quinke's triad was present in 11 of 52 (21.2%) patients with haemobilia.

Fifteen of the 67 (22.4%) patients had a ruptured CAP at clinical presentation, 6 of whom had a contained rupture and 9 who had haemoperitoneum. Overall, 36 of 67 patients (53.7%) were anaemic and 13 of 67 patients (19.4%) had haemodynamic shock at clinical presentation. The median and mode number of the aforementioned presenting problems in each of the 67 patients was 3, with a range of 0–6.

3.2.3. Diagnostic Modalities. Forty-six of 67 patients (68.6%) were diagnosed with CAP based on the results of arterial phase contrast-enhanced computed tomography (CT) scans. Thirteen patients (19.4%) were diagnosed with digital subtraction angiography. Colour Doppler ultrasound and magnetic resonance imaging were each used to reach diagnosis in three patients (4.4%). Two patients (3%) had an intraoperative finding of CAP at cholecystectomy for cholecystitis.

3.2.4. Management Options. Forty-four patients were treated with endovascular embolisation of CAP (65.7%). Of these 44 procedures, 4 were unsuccessful, with 3 of these proceeding to surgery and one being managed successfully with a reattempted endovascular approach. Including the 3 patients for whom endovascular embolisation was unsuccessful, a total of 6 patients (9%) were managed with laparotomy and ligation of the cystic artery proximal to the aneurysm. Two patients were treated with percutaneous thrombin injection (3%). Of these, one procedure was unsuccessful and was subsequently successfully managed with endovascular embolisation. 21 patients (31.3%) required ERCP. 31 patients underwent cholecystectomy as part of their treatment (46.3%). Of the patients who had a cholecystectomy, 12 were following successful endovascular embolisation of the pseudoaneurysms and 19 had cholecystectomy with proximal ligation or clipping of the cystic artery as the only management modality. No mortalities were reported.

Table 1 provides the aetiologies, diagnostic imaging modalities, clinical presentations, and management of CAP in the cases reported.

4. Discussion

CAP is a rare phenomenon with significant risk of morbidity in the form of biliary obstruction and bleeding. Most commonly, it occurs secondary to acute cholecystitis or

after cholecystectomy. The pathophysiology of CAP in cholecystitis is not clear, though it is likely related to inflammatory damage and weakening of the adventitia with subsequent pseudoaneurysm formation [3, 9]. In addition to this, surgery may contribute further to the development of CAP due to vascular erosion from manipulation, clip application, or thermal injury, although the incidence of vascular injury in cholecystectomy is low (0.2–0.5%) [52, 56]. In our reported case, we suspect that the right hepatic artery pseudoaneurysm is explained by some degree of regional inflammation secondary to cholelithiasis, and the CAP is explained by the same in addition to the effect of surgery.

Haemobilia is by far the most common clinical presentation of CAP, with the most common symptom of this being clinical upper gastrointestinal bleeding in the form of haematemesis or melena. Haemobilia can be difficult to diagnose clinically without a high index of suspicion and prompt investigation with targeted imaging. The reported incidence of Quinke's triad in patients with haemobilia is 22–35% [6]. Our review supports this statistic as the triad was present in 21.2% of the cases of CAP with haemobilia. Furthermore, there is a significant variance in the time from cholecystectomy to clinical presentation with a symptomatic CAP in patients where cholecystectomy is the aetiology. This makes a high index of suspicion particularly pertinent in the postcholecystectomy cohort. It is also noteworthy that a symptomatic CAP has been reported up to three years postcholecystectomy [52].

Overall, the morbid load of CAP when symptomatic is high, with most patients suffering at least three of the following problems at clinical presentation: haemobilia, biliary obstruction, anaemia, and haemoperitoneum or haemodynamic shock. It is important to note, despite its morbid load, that CAP is uncommon, and therefore, the astute clinician must pay mind to assessing the patient with due attention to differential causes of these findings.

The most common diagnostic modality for CAP is an arterial phase contrast-enhanced CT scan, which is a readily available imaging modality in most treatment centres. Colour Doppler ultrasound and MRI imaging are options for patients with contraindications to the use of intravenous contrast agents.

Endovascular embolisation was the most common choice of controlling CAP in our review, with an adequate rate of success on the first attempt in the majority of patients. Surgery in the form of cholecystectomy or vessel ligation would be most likely indicated in cases where the endovascular approach is unsuccessful.

Though the findings are of interest in light of the rarity of this pathology, our review is limited by its retrospective nature and broad inclusion criteria. There is no clear cause and effect relationship demonstrated by the aetiologies listed, and the therapeutic benefit of the management modalities compared to one another cannot be assessed. Perhaps, the strongest correlation that can be made is in regard to the way CAP clinically presents, as the clinical presentations can be directly corroborated with gold-standard confirmatory diagnostic imaging.

TABLE 1: A summary of the aetiologies, diagnostic imaging modalities, presentations, and management of CAP.

| Aetiology | Diagnostic imaging modality | | Complications | Management | | |
|-----------------|-----------------------------|--------------------------------|--|---------------|---------------------------------|---------------|
| Cholecystitis | 41 (61.2%) | CT [†] 46 (68.6%) | Haemobilia | 52 (77.6%) | Endovascular embolisation | 44 (65.7%) |
| Cholecystectomy | 18 (26.8%) | DSA [‡] 13 (19.4%) | Haemobilia + Quinke's triad* | 11 (16.5%) | Percutaneous thrombin injection | 2 (3%) |
| Idiopathic | 6 (8.9%) | US [§] 3 (4.4%) | Haemobilia with haematemesis or melena | 39 (58.3%) | ERC [¶] | 21 (31.3%) |
| Cholelithiasis | 1 (1.5%) | MRI 3 (4.4%) | Haemobilia with biliary obstruction | 22 (32.8%) | Cholecystectomy | 31 (46.3%) |
| Pancreatitis | 1 (1.5%) | | Direct biliary obstruction | 3 (4.5%) | Laparotomy, vessel ligation | 6 (9%) |
| | | | Contained rupture | 6 (8.9%) | | |
| | | | Rupture with haemoperitoneum | 9 (13.4%) | | |
| | | | Haemodynamic shock | 13 (19.4%) | | |

The number of patients in each category is presented, with a percentage in relation to the total of 67 reported cases of CAP in brackets. [†]Contrast-enhanced computed tomography. [‡]Digital subtraction angiography. [§]Colour Doppler ultrasound. ^{||}Magnetic resonance imaging. [¶]Endoscopic retrograde cholangiopancreatography. *Haemobilia + haematemesis or melena + biliary obstruction.

5. Conclusion

Although CAP is a rare phenomenon, it occurs as a complication of one of the most common surgical procedures performed worldwide, and it carries with it significant associated morbidity. It is therefore pertinent to the surgeon to be aware of its clinical presentation, diagnosis, and management options. A high index of suspicion and prompt investigation is suggested in patients with cholecystitis or previous cholecystectomy who present with upper gastrointestinal bleeding, anaemia, and biliary obstruction.

Data Availability

The data used to support the findings of this study are included within the article and the references provided.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

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