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Case Report

Cystic artery pseudoaneurysm ☆

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ABSTRACT

Cystic artery pseudoaneurysm (CAP) is a rare entity, with just a few cases reported in the literature. The most common presentation of CAP is described by Quincke's triad of upper gastrointestinal bleeding, jaundice and right upper quadrant abdominal pain. We report the case of an 83-year-old male who presented to the adult emergency with a history of an acute cholecystitis 5 weeks prior for which responded to conservative management. Despite this patient not presenting with Quincke's triad, early suspicion of CAP was considered in light of his history of acute cholecystitis and a computed tomographic CT abdomen ordered promptly which showed a 6 mm cystic artery pseudoaneurysm and a thick-walled gallbladder with surrounding inflammatory changes. Management with an endovascular approach followed by an elective cholecystectomy was done.

Introduction

Cystic artery pseudoaneurysm (CAP) is a rare entity, with just a few cases reported in the literature. Most commonly, CAP cases have been reported to occur secondary to an inflammatory mediated process such as acute cholecystitis or pancreatitis or may occur post cholecystectomy or as a complication of biliary procedures. While its pathogenesis is not clearly understood, CAP formation is postulated to occur following pathological changes that result in the weakening and erosion of the muscular and elastic layers of the arterial wall.

Pseudoaneurysm rupture can result in significant intraperitoneal bleeding, upper gastrointestinal bleeding, hemorrhagic shock and an increased risk of patient mortality. The most common presentation of CAP is described by Quincke's triad of upper gastrointestinal bleeding, jaundice and right upper quadrant abdominal pain; however in the early stages, patient's may present with nausea, hematemesis, melena, vague abdominal pain or fever. Thus, it is imperative for clinician's

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to maintain a high degree of clinical suspicion in patients who may have atypical presentations as highlighted in this case report.

We report an interesting case of an 83 year old male with a history of acute cholecystitis who was found to have a 6 mm CAP on computed tomographic (CT) abdomen following presentation to the Emergency Department for an acute bright red per rectum (PR) bleed and syncope. He was managed with a coil embolization of the cystic artery with a view to perform an elective cholecystectomy at a later date. He demonstrated unremarkable recovery and was discharged 24 hours later with no further reports of PR bleeds.

This case highlights the importance of early clinical suspicion in an atypical presentation of CAP, prompt imaging and the role of endovascular treatment in the management of this case. It is of utmost importance for clinicians to be aware of the varied clinical presentation of CAP especially in the early stages in order to facilitate early clinical suspicion, prompt diagnosis and timely intervention to prevent pseudoaneurysm rupture and its associated life threatening complications.

Case presentation

We report the interesting case of an 83-year-old male who presented to the Adult Emergency Department for an acute PR bleed and syncope. The extent of blood loss could not be quantified by the patient; however, we suspect a massive PR bleed due to his syncopal episode following the PR bleed.

On physical examination, the patient was an elderly male of lean body habitus in no apparent pain or cardiopulmonary distress. His mucus membranes were pink and moist with no scleral icterus noted. He was afebrile and acyanotic with no obvious signs of jaundice. On abdominal examination, no right upper quadrant pain was elicited. His abdomen was not distended, soft, and nontender with no palpable masses. All other examinations were unremarkable.

His vitals were stable with a blood pressure of 134/82 mm Hg, pulse of 86 bpm, respiratory rate of 22 breaths/min and temperature of 36.3°C. Of note, he presented 5 weeks prior for an acute cholecystitis which responded to conservative management with a view to perform an elective cholecystectomy.

On admission, a complete blood count, renal function tests, liver function tests, electrolytes, clotting panel, D-dimer and Troponin were ordered. His CBC showed an initial Hb of 11.9 mg/dL which decreased to 10.2 mg/dL within 24 hours. All other labs requested were unremarkable. No further PR bleeds were noted on admission.

CT Abdomen done on the day of admission showed a 6 mm cystic artery pseudo aneurysm. The gallbladder was thick walled and the surrounding inflammatory changes makes the gallbladder inseparable from the hepatic flexure of the colon (Figs. 1 and 2).

A decision was made by the attending surgeon and interventional radiologist to utilize an endovascular approach with a view to perform an elective cholecystectomy another date in order to allow sufficient recovery from the acute presentation. The endovascular approach performed was a coil embolization of the cystic artery.

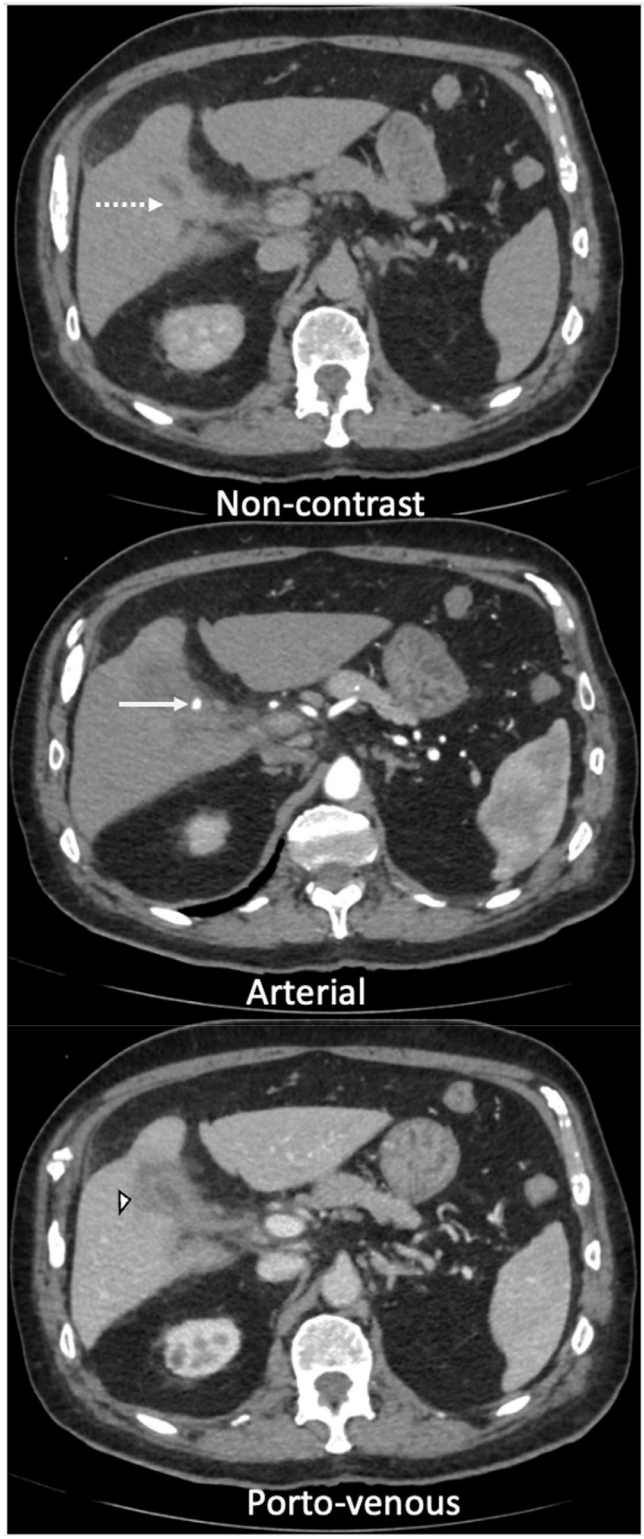

Fig. 1 – Axial reformatted noncontrast and postintravenous contrast computed tomography images in the noncontrast, arterial and portovenous phases with the dashed arrow demonstrating the gallbladder bed with no evidence of hyperdense focus, solid arrow demonstrating an enhancing focus representing a cystic artery pseudoaneurysm and arrow head demonstrating features gallbladder wall thickening with surrounding pericholecystic fluid and fat stranding.

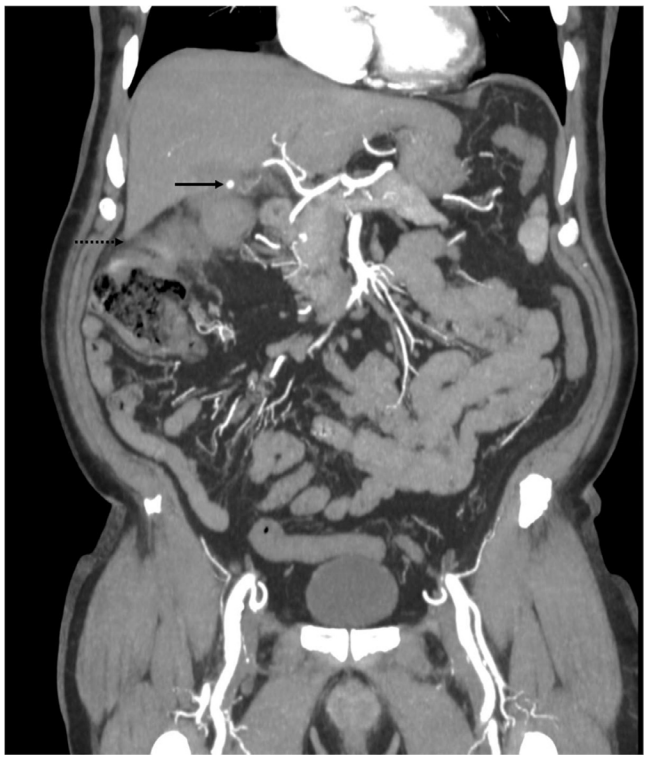

Fig. 2 – Coronal reformatted postintravenous contrast computed tomography images in the arterial phase with the arrow demonstrating an enhancing focus representing a cystic artery pseudoaneurysm and dotted arrow demonstrating surrounding inflammatory changes with the hepatic flexure and the gallbladder.

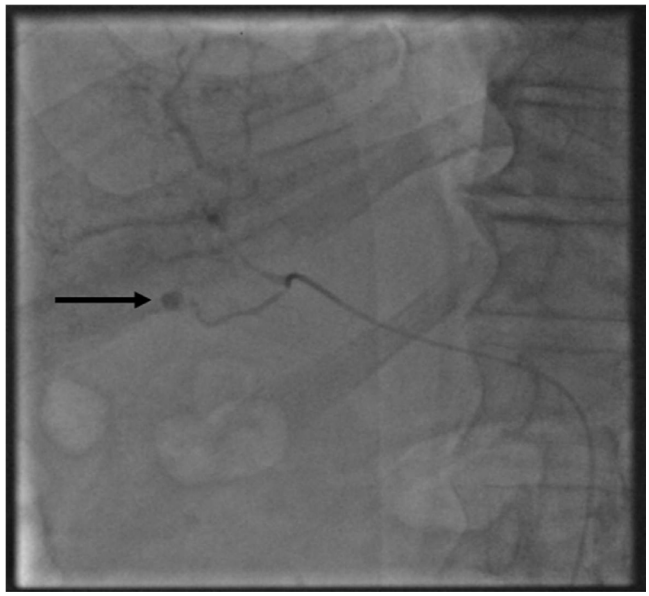

Fig. 3 – Angiographic image of cobra catheter within the right hepatic artery with arrow demonstrating a well-defined rounded opacity arising from the cystic artery consistent with a pseudoaneurysm .

Arterial embolization involved access at the right common femoral artery and cannulation of the right hepatic artery with a 4 Fr C2 Cobra catheter and 0.035 hydrophilic (Terumo) wire. A right hepatic artery angiogram outlined the cystic artery arising from the right hepatic artery and the aneurysm opacified at the distal tip of the cystic artery (Fig. 3). A RIM catheter was used to select the cystic artery and a Run-through NS 0.014 wire was advanced into the cystic artery and its tip coiled within the pseudo aneurysm.

The C2 (macrocatheter) was advanced beyond the midpoint of the cystic artery hence embolization was performed via this catheter without the need for a microcatheter. Two Cook Micronester 2-3 mm x 1.5 cm followed by one Cook Nester 4 mm x 4 cm coil were used. Approximately 1 cm of the 4 cm coil hangs into the right hepatic artery at the end of embolization. This was not removed as there was good flow into the right hepatic artery and the coil was securely anchored within the cystic artery. In addition, coiling of the right hepatic artery was considered if the cystic artery could not be selected.

The procedure was well tolerated and the patient had an unremarkable post procedure recovery. He remained pain free with no further PR bleeds during his hospital admission and was discharged home 24 hours post embolization. The following week at home was also unremarkable.

Follow up contrast enhanced CT was performed approximately 4 weeks later and showed resolution of pericholecystic inflammatory changes, nonopacification of the cystic artery (and pseudoaneurysm) and the coils in place (Fig. 5).

Discussion

CAP is a rare but life-threatening entity with just a few cases reported in the literature [1]. While it's pathogenesis is not well understood, CAP may result from any stimulus that weakens and erodes the elastic and muscular layers of the cystic artery [2]. Stimuli resulting in such damage can be categorized into inflammatory, traumatic/iatrogenic or idiopathic etiologies.

The most common etiologies of CAP reported in the literature are secondary to an inflammatory process such as cholecystitis and pancreatitis or postcholecystectomy/biliary intervention; however, cholecystitis is the predominant cause reported [1,3,4].

Between 1991 and 2020, 67 cases were reported which demonstrated varying etiologies for CAP. In 41 instances, CAP occurred as a result of acute cholecystitis accounting for 61.2% of the reported cases. CAP occurring secondary to cholecystectomy was reported in 18 instances, 6 instances were idiopathic and 1 case each resulted from cholelithiasis and pancreatitis [3,4].

During cholecystectomy, the cystic artery is a key anatomical structure that is dissected or ligated. As a result, the cystic artery may become damaged due to physical manipulation or thermal injury which subsequently weakens the arterial wall layers leading to CAP formation [2].

As stated previously, the most common reported etiology of CAP formation is secondary to an inflammatory mediated process predominantly cholecystitis. The surrounding inflammation leads to adventitial damage to the vasa vasorum subsequently compromising and weakening the structural architecture of the elastic and muscular components of the arterial wall leading to CAP formation [2].

In the reported case, our patient had a 5 week history of acute cholecystitis prior to his presentation to the Emergency Department for an acute PR bleed and syncope. His acute presentation and history of acute cholecystitis prompted an urgent CT abdomen which showed a 6 mm cystic artery pseudoaneurysm with surrounding inflammatory changes around the gallbladder. As a result, we suspect that a determinant factor in the development of this patient's CAP is likely his history of acute cholecystitis.

It is important to note that the clinical presentation in our reported case highlights an atypical or unusual presentation of CAP and there has only been 1 case of CAP presenting with a massive PR bleed documented in the literature by Carey et al. [5] in January 2020 entitled "Cystic Artery Pseudoaneurysm presenting as a massive per rectum bleed treated with percutaneous coil embolization."

CAP can often be challenging to detect clinically at presentation as patients may initially present with vomiting, nausea, melena, vague abdominal pain and fever which can represent a wider differential diagnosis. However, the classic clinical presentation of CAP is associated with its rupture and is most frequently represented by hemobilia known as Quincke's triad of upper gastrointestinal bleeding (45%), jaundice (60%) and right upper quadrant abdominal pain (70%) with approximately 40% of patients presenting with all 3 symptoms [2-4,6].

A literature review of 50 reported cases of CAP revealed that the median age of diagnosis was 68 years and most reported cases involved older patients. Contributing to this demographic were the presence of atherosclerosis, diabetes, hypertension, hypercholesterolemia and vasculitis [2].

Given the vague clinical presentation of CAP in the early stages, it is important for clinicians to maintain a high degree of clinical suspicion in patients who CAP is a likely diagnosis to facilitate prompt imaging, diagnosis and appropriate intervention to prevent CAP rupture, gastrointestinal bleeding, hemodynamic instability and patient death [2,3].

Our reported case bears many similarities to the case reported by Carey et al.[5] such that both patients who were elderly were noted to have an acute cholecystitis on CT abdomen and presented in a similar manner with massive per rectum bleeding. An endovascular approach of coil embolization of the cystic artery was employed in both cases with a view to perform an elective cholecystectomy later with both patients making a full recovery postembolization.

There were no other reported cases on CAP presenting with PR bleed for comparison and of the cases reported in the literature, the majority were secondary to acute cholecysitis or due to a complication of surgery most commonly cholecystectomy with clinical presentations of hemobilia.

Our case report aims to highlight the importance of early clinical suspicion in patients who may not present with the typical presentation of CAP as represented by Quincke' triad as early diagnosis and intervention is instrumental in preventing CAP rupture and its associated life threatening complications mentioned above.

The diagnosis of CAP relies on an appropriate imaging modality which provides a high degree of diagnostic detail. Modalities can be invasive such as conventional angiography or noninvasive such as duplex doppler ultrasonography (US), CT angiography and magnetic resonance (MR) angiography.

Conventional angiography, while an invasive procedure, is the gold standard method for diagnosis as it provides diagnostic detail with the ability to delineate lesions of < 10 mm in size. This modality may also be combined with endovascular treatment options such as embolization and stent-graft placement. However, due to its invasive nature, there are associated procedural risks such as the development of hematomas, pseudoaneurysms, AV fistulas, thrombosis, ischemia and intimal dissection [7,8].

Noninvasive imaging modalities mentioned above are also widely utilized and are effective with respect to diagnostic detail without the risks posed by conventional angiography.

Of the mentioned noninvasive modalities, duplex doppler ultrasound has the advantages of being cost effective and readily available; however, the quality of the imaging greatly depends on the skills and experience of the user. On ultrasound, an anechoic or hypo echoic sac is visualized adjacent to the damaged artery and the size and neck of the pseudoaneurysm can be measured; however, the communication between the originating artery and sac isn't always easily demonstrated [7,8]. On color duplex doppler US, arterial blood flow within the sac and neck is demonstrated by the characteristic "yin-yang" sign spectral waveforms show a "to and fro" flow pattern [4,9].

Similar to ultrasound, arterial phase contrast CT is also a noninvasive imaging modality of choice for diagnosing CAP; however, it is not user dependent unlike ultrasound and images can be generated within a few minutes making it useful in emergency settings. On arterial phase CT, the pseudoaneurysm may appear as a well circumscribed, contrast filled, oval or round collection and visualization of the aneurysm and its neck is permitted from various angles using multiplanar reconstruction (MPR). Multi organ screening is also provided which can aid in the determination of the cause of the pseudoaneurysm [7,8]. However, some patients may have contraindications to the use of contrast and thus color doppler ultrasound and MR angiography are viable options in these instances [1,7,8,10].

With respect to MR angiography, it may be advantageous in patients with renal failure or allergies to iodinated contrast and pseudoaneurysms can be demonstrated in axial, sagittal and coronal planes; however, disadvantages include cost and availability. The quality of images produced can also be disrupted due to the presence of artifacts caused by stents or surgical clips and MR angiography may not be compatible in patients who may have pacemakers [7,8].

In the reported case, the imaging modality chosen was a CT Abdomen which was done on the day of admission and showed a 6 mm cystic artery pseudoaneurysm with inflammatory changes around the gallbladder. The gallbladder was inseparable from the hepatic flexure of the colon. This pseudoaneurysm could have been easily missed in the absence of an arterial phase.

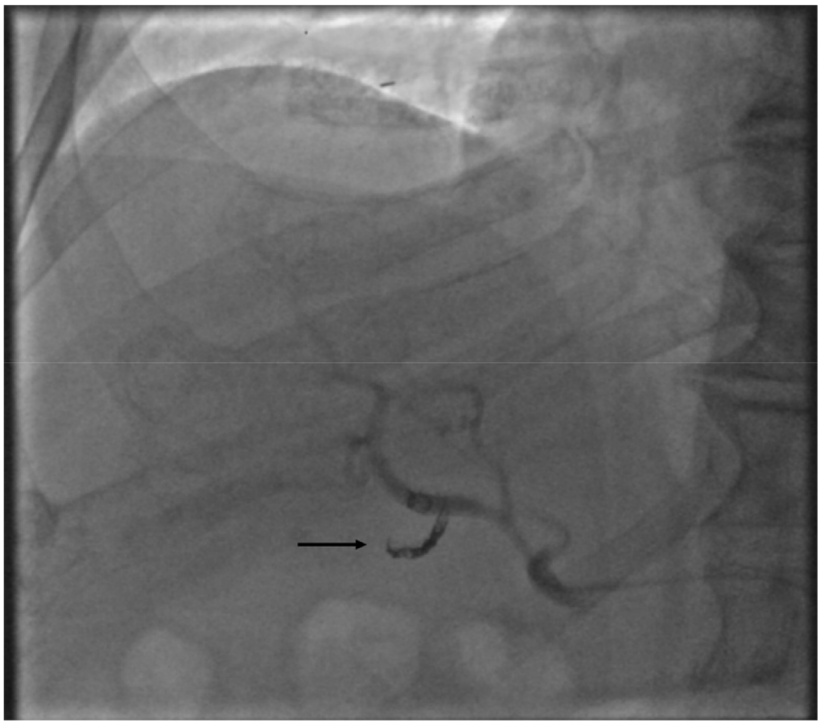

Fig. 4 – Angiographic image of cobra catheter within the right hepatic artery with arrow demonstrating a well-defined rounded opacity arising from the cystic artery consistent with a pseudoaneurysm .

Once an appropriate imaging modality is chosen and the diagnosis made, a decision is made as to whether an endovascular vs surgical approach will be utilized based on the patients clinical presentation. Traditionally, surgical repair was often the first choice of treatment for CAP; however, due to the risk of pseudoaneurysm rupture and bleeding, arterial embolization followed by elective cholecystectomy is preferred [3,7]. This also allows the patient sufficient time for recovery prior to cholecystectomy.

Endovascular management typically involves 2 methods, embolization, and stent placement.

Cystic artery embolization has been proven to be a safe procedure with less procedural risks when compared to a surgical approach with patients benefitting from a shorter hospital stay and recovery time and is often more cost effective [5,7,8].

Embolizing agents used include coils, detachable balloons, glue or gelfoam; however, coil embolization is often the preferred agent as a variety of vessels sizes can be targeted and there is low risk of increasing the pressure in the vascular lesion when compared to glue or gelfoam. Embolization may also be performed in cases where a patient is unfit for a surgical approach in the acute presentation due to hemodynamic instability or sepsis [5,7,8].

In the reported case, a decision was made by the attending surgeon and interventional radiologist to perform coil embolization of the cystic artery using 2 Cook Micronester 2-3 mm x 1.5 cm coils followed by 1 Cook Nester 4 mm x 4 cm coil with a view to perform an elective cholecystectomy at another date. This would facilitate the patients recovery from the acute presentation prior to cholecystectomy.

While there were no postprocedural complications noted, 2 main complications of cystic arterial embolization of concern include nontarget embolization and ischemia of the gallbladder.

In the reported case, the cystic artery was targeted for coil embolization and a right hepatic artery angiogram outlined the cystic artery arising from the right hepatic artery with the pseudoaneurysm opacified at the distal tip of the cystic artery (Fig. 3).

While the cystic artery was the target for coil embolization, 1 cm of the 4 cm coil was demonstrated within the right hepatic artery at the end of embolization demonstrating nontarget embolization. However, this coil was not removed as there was good blood flow into the right hepatic artery and the coil securely anchored within the cystic artery. If we were unable to select the cystic artery, coiling of the right hepatic artery was considered (Fig. 4).

Another important potential complication of utmost concern regarding cystic artery coil embolization is gall bladder ischemia. In the reported case, imaging demonstrated good blood flow to the gall bladder via the cystic artery intra-op and postprocedure; however, we were prepared to perform an emergency cholecystectomy if the blood supply to the gall bladder was comprised.

To our advantage and the patients, the procedure was well tolerated and the patient had an unremarkable post procedure recovery. He remained pain free with no further PR bleeds during his hospital admission and was discharged home 24 hours postembolization. The following week at home was also unremarkable.

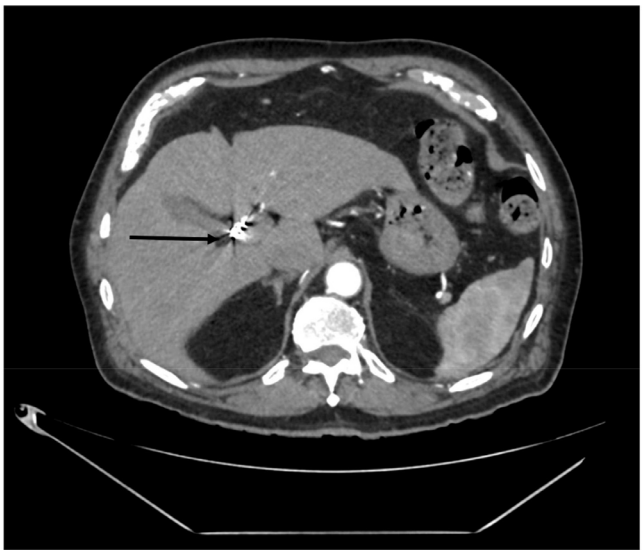

Fig. 5 – Axial reformatted postintravenous contrast computed tomography images in the arterial phase with the arrow demonstrating resolution of surrounding inflammatory changes with embolization coils within the gallbladder fossa.

Follow up contrast enhanced CT was performed approximately 4 weeks later and showed resolution of pericholecystic inflammatory changes, nonopacification of the cystic artery and psuedoaneuyrsm and the coils in place (Fig. 5).

Conclusion

CAP is a rare entity that may occur secondary to an inflammatory mediated process most commonly acute cholecystitis or postsurgical/biliary intervention most commonly postcholecystectomy. CAP typically presents as hemobilia in the form of Quincke's triad of jaundice, upper gastrointestinal bleeding and right upper quadrant abdominal pain; however, in the early stages patients may present with vague abdominal pain, vomiting, melena or fever which can represent a wider list of differential diagnoses.

As a result, a high degree of early clinical suspicion is warranted to facilitate prompt imaging, diagnosis and timely intervention to prevent CAP rupture and its associated complications such as significant intra peritoneal bleeding, gastrointestinal bleeding, hemorrhagic shock and patient mortality.

It is imperative that clinicians be not only aware of the typical clinical presentation of CAP but keep in mind that patients may present atypically as highlighted in this case report of CAP presenting as a bright red PR bleed and syncope.

Management includes endovascular or surgical methods with the definitive treatment being cholecystectomy; however arterial embolization of CAP with an elective cholecystectomy is a preferred pathway as embolization permits sufficient recovery time from the acute presentation until elective cholecystectomy is performed.

Patient consent

Informed consent was obtained from the patient.

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