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A pseudoaneurysm of the right hepatic artery treated successfully with a stent graft – A case report

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ABSTRACT

INTRODUCTION: Haemobilia caused by pseudoaneurysms of the right hepatic or cystic artery is rare. Haemobilia classically causes gastro-intestinal hemorrhage, jaundice and upper abdominal pain.

PRESENTATION OF CASE: A 76-year old female underwent laparoscopic cholecystectomy because of a severe acute on chronic cholecystitis. A massive arterial bleeding occurred during surgery, which was controlled with hemoclips. Approximately one week after surgery the patient developed severe colic pains and cholestatic liver enzyme alterations. Endo-ultrasound showed normal-width bile ducts, however during a subsequent ERCP haemobilia was observed. On computed tomography a pseudoaneurysm of the right hepatic artery was seen. Selective embolization was initially successful, however, a rebleed was observed two weeks later and a 6 × 50 mm Viabahn stent graft was placed in the right hepatic artery uneventfully. The patient remained free of complaints during 3-years of follow-up.

DISCUSSION: Pseudoaneurysms of the cystic or hepatic arteries are described to be cholecystectomy or cholecystitis related. The etiology of the pseudoaneurysm in this case can be inflammatory or iatrogenic. Embolization is the golden standard in pseudoaneurysm treatment. Stent graft implantation has not been frequently described as an alternative option to surgery after a failed attempt of embolization.

CONCLUSION: This case report presents a probable cholecystitis related pseudoaneurysm of the right hepatic artery, which caused haemobilia after cholecystectomy. The pseudoaneurysm was successfully eliminated with a stent graft after embolization had failed. Stent grafts should be considered a minimal invasive and effective alternative after failed embolization of a pseudoaneurysm.

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1. Introduction

Haemobilia caused by pseudoaneurysms of the right hepatic artery or the cystic artery is rare. The incidence has been reported up to 0.6% regarding isolated vascular damage without concomitant biliary injury [1,2]. Quincke's triad of symptoms in haemobilia consists of upper gastro-intestinal hemorrhage, jaundice and upper abdominal pain. However, the classic triad of symptoms is only present in 20–40% of the patients [2,3]. Described treatment methods of pseudoaneurysms causing haemobilia are embolization, implanting stent grafts, thrombin injections and surgery [2,4–6]. Mortality rates up to 2% have been reported [2]. This case-report describes a case of haemobilia caused by a pseudoaneurysm of the right hepatic artery and its therapeutic options. This work has been reported in line with the SCARE criteria [7].

2. Presentation of case

A 76-year old female was scheduled for laparoscopic cholecystectomy after visiting the outpatient clinic of our hospital with symptomatic cholelithiasis, confirmed by ultrasound. Her medical history reported hypertension, a slight aortic valve stenosis and bilateral breast cancer.

A laparoscopic cholecystectomy was performed by a gastrointestinal surgeon. During the procedure, a severe acute on chronic cholecystitis with many adhesions was observed. A spontaneous arterial rupture near the cystic artery caused a large intra-abdominal hemorrhage with a total blood loss of 900cc. After placement of multiple hemoclips the bleeding was controlled. After the content of the gallbladder was aspirated, some bile spill from this puncture site was observed, for which lavage was performed. A 28 French drain was left behind in the wound bed. The patient received intravenous antibiotics postoperatively.

Postoperatively the patient complained of persistent colic pains. During examination jaundice was observed. The laboratory results showed: total bilirubin 133 μmol/L, conjugated bilirubin 113 μmol/L, γ-glutamyltransferase 358 IU/L, aspartate aminotrans-

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Image 1. This Computer Tomography image shows the coils situated in the pseudoaneurysm of the right hepatic artery unsuccessfully eliminating the pseudoaneurysm causing a large intra-hepatic hematoma.

ferase 144 IU/L, alkaline phosphatase 214 IU/L, serum amylase 141 and serum C-reactive protein 254 mg/L. Endo- ultrasound showed normal-width bile ducts. The computed tomography (CT) showed a large amount of fluid in the gallbladder bed. An Endoscopic Retrograde Cholangiopancreatography (ERCP) was performed and reported haemobilia to cause the persistent colic pains and jaundice. A CT angiography showed a pseudoaneurysm of the right hepatic artery. The pseudoaneurysm was initially successfully embolized with coils by an experienced intervention radiologist. After this procedure the patient was free of pain, hemoglobin levels remained stable and liver function tests improved.

However, 2 weeks post-intervention the patient presented at the emergency department with recurrent colic pains in the right upper abdomen and jaundice. Laboratory results again showed increased liver function tests. A CT angiography showed an increase of the pseudoaneurysm leak (**Image 1**). An experienced intervention radiologist with over 8 years of experience was consulted and decided to exclude the pseudoaneurysm and simultaneously ensure vascularization of the right hepatic artery by implanting a stent graft. Introduction took place through the left brachial artery. The celiac trunk was catheterized with a 5 French catheter and advancement took place with a 7 French catheter. After successfully locating the catheter, a self-expandable Viabahn 6 × 50 mm stent was placed in the right hepatic artery over the bleed-

ing defect. **Image 2** shows how the pseudoaneurysm was occluded while the right hepatic artery remained open. The platelet aggregation inhibitor acetylsalicylic acid (ASA) 80 mg was initiated.

Follow-up of the patient took place by a duplex ultrasound initially and later annually by CT angiography. **Image 3** shows the location of the stent graft and the intact flow through the right hepatic artery after 3 years of follow-up. The Viabahn stent remained open and the patient is still free of complaints. Lifelong usage of ASA was indicated.

3. Discussion

This case report presents a successfully treated pseudoaneurysm of the right hepatic artery causing haemobilia in a female patient. Successful stent grafting of the right hepatic artery was performed after a failed attempt of embolization. This case reports a 3-year follow-up with no recurrence or growth of the pseudoaneurysm and persistent vascularization of the right hepatic artery.

Literature describes haemobilia to be associated with cholecystectomy and cholecystectomy-related-injury [2,8]. Inappropriate use of thermal or mechanical energy during dissection of Calot's triangle has been described as a possible explanation [9,10]. Another pathophysiologic process that has been described is the inflammation process in cholecystitis causing pseudoaneurysms to form

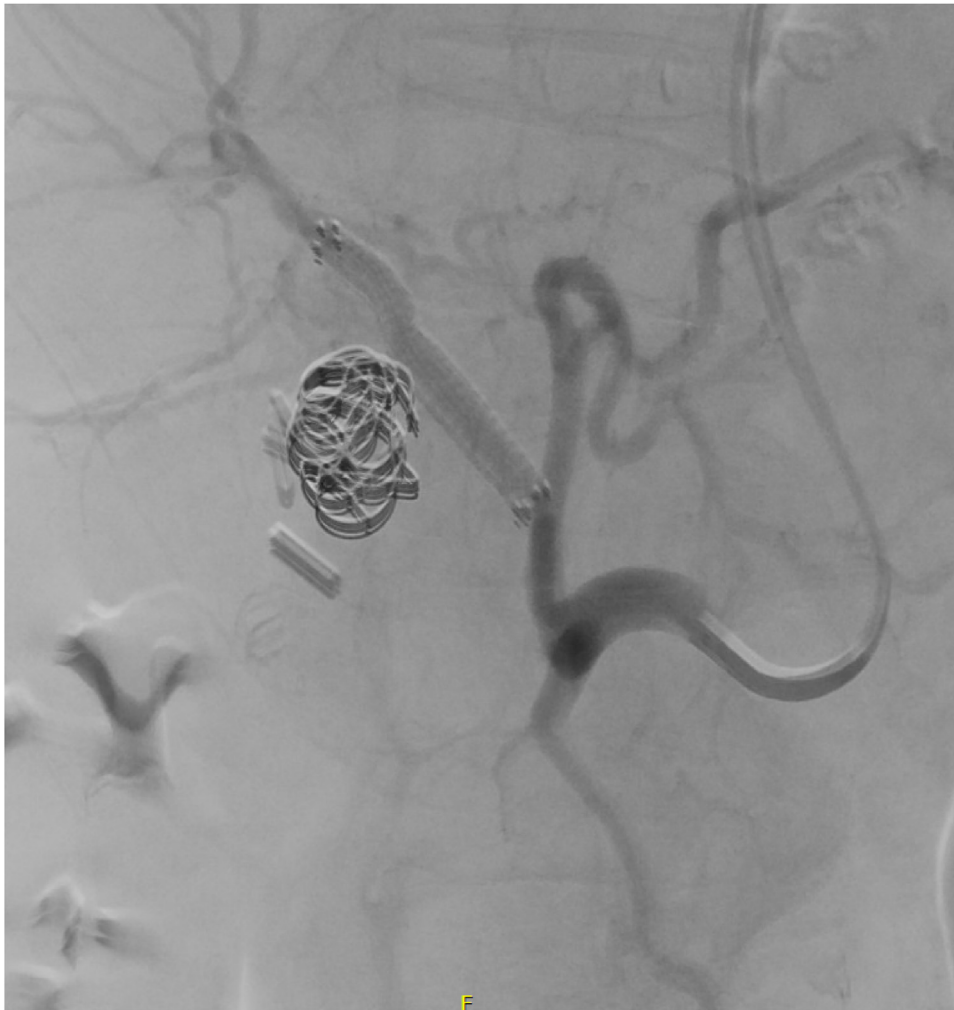


Image 2. This angiography image shows the stent graft positioned in the right hepatic artery. The coils that were placed during previous interventions are still in situ and indicate the location of the pseudoaneurysm.



Image 3. This CT-angiography Maximum Intensity Projection (MIP) presents the stent graft and the persistent flow through the right hepatic artery 2 years post intervention.

[11,12]. In this case-report it is likely that the cholecystitis caused the formation of a pseudoaneurysm, considering the finding of a highly inflamed gallbladder during surgery. Also, during the procedure a major and diffuse arterial bleeding occurred, while dissecting Calot's triangle close to the gallbladder and after the cystic artery was already identified. This can be an indication that the pseudoaneurysm already existed before surgery. Another possible explanation is a congenital anatomic variation [13].

The most frequently involved artery in the formation of pseudoaneurysms is the right hepatic artery, occurring in 87% of the cases. The cystic artery is involved in 7.9% of the patients [2]. The pathophysiologic mechanism causing the right hepatic artery to be most prone to form a pseudoaneurysm has not yet been clarified.

Literature describes multiple treatment options regarding pseudoaneurysms of which embolization is the golden standard [2]. Most patients (72.4%) are treated by embolization of the pseudoaneurysm by either occluding the sac or the feeding vessel with a variety of embolic agents or coils or by embolizing the vessel distal and proximal of the pseudoaneurysm. Sometimes, a second embolization attempt might be indicated to embolize the distal and proximal feeding vessel when occlusion of the pseudoaneurysm itself proved to be ineffective. Stent grafts were only used in 4%, possibly because the incidence of unsuccessfully treated pseudoaneurysms is low and success rates of embolization are high (94.5%) [2]. Two cases reported surgical ligation of the artery after failed embolization to be effective and one case reported successful thrombin injection. In one case anatomical challenges made embolization not to be effective and a stent graft was implanted successfully [14–16].

In this case the coiling procedure insufficiently packed the pseudoaneurysm causing the pseudoaneurysm and intrahepatic hematoma to increase. Sequentially, a different approach would potentially be more successful. Embolization of the distal and proximal feeding vessel of the pseudoaneurysm, in this case the right hepatic artery, can potentially cause bile duct necrosis. Similarly, ligation of the artery also potentially causes insufficient perfusion of the vascularized organ. Therefore, during the clinical decision-making process, the intervention radiologist decided to exclude the pseudoaneurysm and simultaneously ensure flow through the right hepatic artery by using a stent graft. However, when embolization fails perfusion is guaranteed, in contrast to a stent graft occlusion which will sequentially cause insufficient perfusion. Implanting stent grafts is considered a technically challenging procedure since it concerns a small vessel at a distant location and frequently concerns an altered anatomical situation. However, it should be considered a minimal invasive and effective alternative to surgery after failed embolization [16].

In this case a 3-year follow-up was performed without signs of growth or recurrence. Also, the stent remained open during the 3-year follow-up time. Pseudoaneurysms causing haemobilia have been described to present shortly after laparoscopic cholecystectomy or after cholecystitis [2]. Only one case reports a patient suffering from haemobilia 15 months after laparoscopic cholecystectomy [8]. Therefore, a 3-year follow-up should be considered a reasonable follow-up time since the chances of recurrence can be considered significantly small.

Overall, more research should be performed on stent grafts in treating pseudoaneurysms causing haemobilia. Additionally, research with a longer follow-up setting could reveal unknown long-term effects and complications of embolization or stent grafts.

4. Conclusion

Pseudoaneurysms of the right hepatic artery or cystic artery are rare but can be the source of jaundice, upper abdominal pain and

gastro-intestinal hemorrhage. Pseudoaneurysms can occur cholecystectomy or cholecystitis related. This case report presents a successfully treated pseudoaneurysm of the right hepatic artery by implanting a stent graft. Embolization is described as the golden standard. Stenting has been less thoroughly described in the literature but provides a method to occlude the pseudoaneurysm whilst ensuring vascularization of the artery. Therefore, stent grafts should be considered a minimal invasive and effective alternative after failed embolization of the pseudoaneurysm.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

This case report did not regard ethical approval. Written informed consent was obtained from the patient for publication of this case report.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Amber C Traa: Writing the paper, data analysis and interpretation.

Miriam L Hoven-Gondrie: Study concept, data collection and interpretation, supervision.

Arjen L Diederik: Study concept, supervision.

Registration of research studies

Not applicable since this is a case report and did not require a database.

Guarantor

The Guarantor's of this study were Amber C Traa and Miriam L Hoven-Gondrie.

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