

**Case Report**
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## Case Studies of Acute Cholecystitis Complicating Biliary Metal Stent-in-Stent after ERCP

Kevin Kyung Ho Choi<sup>1,3</sup>, Charlotte Knox<sup>2</sup>, Payal Saxena<sup>1,3</sup> and Arthur John Kaffes<sup>1,2,3\*</sup>

<sup>1</sup>Royal Prince Alfred Hospital – AW Morrow Gastroenterology and Liver Centre, Missenden Road, Camperdown, Sydney, New South Wales, 2050, Australia

<sup>2</sup>Prince of Wales Hospital – Gastroenterology Department, Barker Street, Randwick, Sydney, New South Wales, 2031, Australia

<sup>3</sup>The University of Sydney, Australia

**\*Corresponding author**

Arthur John Kaffes, Royal Prince Alfred Hospital – AW Morrow Gastroenterology and Liver Centre, Missenden Road, Camperdown, Sydney, New South Wales, 2050, Australia. Ph: +61295162033. E-mail: arthur@kaffes.com

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### Introduction

Palliative biliary stenting by endoscopic retrograde cholangiopancreatography (ERCP) is the primary modality for management of malignant biliary obstructions with reported efficacy exceeding 90% in expert hands [1-3]. Management of malignant hilar biliary obstructions are more challenging with reports of technical and clinical success rates ranging from 70% to 88.6% using self-expanding metal stents (SEMS) [4,5]. Despite the need for biliary drainage with ERCP, acute cholecystitis is a well-documented complication of SEMSs in malignant biliary obstructions which ranges from 0 – 10% [1,6-9]. Although there is theoretical concern for higher rates of cholecystitis with covered SEMS due to occlusion of the cystic duct, two meta-analyses failed to find a significant difference between covered and uncovered metal stents [1,9-11]. Thus, cholecystitis is a well-known complication after SEMS placement regardless of type, however there are no reports of stent-in-stent related cholecystitis in current literature. We describe the first 2 reported cases of stent-in-stent cholecystitis.

### Case 1

A 59-year-old woman presented following an ERCP with right upper quadrant pain. Past medical history consisted of cholangiocarcinoma with peritoneal metastases. Initially, the patient presented with painless jaundice and imaging confirmed the presence of a malignant appearing biliary stricture. An ERCP was undertaken which revealed a moderate malignant appearing biliary stricture (Bismuth IV), a sphincterotomy was performed and a 7Fr 12cm pigtail plastic stent each was placed in to the right and left hepatic duct. One month following the initial placement these stents were exchanged with bilateral uncovered 8mm self-expanding metal stents (SEMS) with lengths of 8cm on the right and 10cm on the left (Figure 1).

The patient represented 6 months later with right upper quadrant pain and fevers following last ERCP. Bilirubin (μmol/L) at the time was 12, WCC 15.8x10<sup>9</sup>/L and C-Reactive Protein (CRP, mg/L) was 122. An ERCP was performed revealing an atrophic

left hepatic duct whereas the right hepatic duct was dilated, both pre-existing uncovered SEMS were patent. A 7 Fr 12cm pigtail plastic stent was placed into the left SEMS and a 10mm by 8cm uncovered metal stent was placed into the right SEMS with good flow of bile at the end of the procedure. Following this procedure and a course of intravenous antibiotics the patient was discharged, free of pain.

Six days later the patient represented with severe right upper quadrant pain and on examination was murphy's positive. Bloods revealed a bilirubin of 14 and a CRP of 312. Abdominal ultrasound revealed no biliary dilatation but a thick-walled swollen gallbladder consistent with acute cholecystitis. Conservative management was initiated with antibiotics and analgesia. Due to worsening pain a further CT abdomen was done which revealed features consistent with gallbladder wall perforation with a deficient inferolateral wall and fluid contiguous with peritoneal fluid. The patient underwent an emergent laparoscopic exploration which was converted to a laparotomy with wash out and subtotal cholecystectomy. The patient recovered well following this.

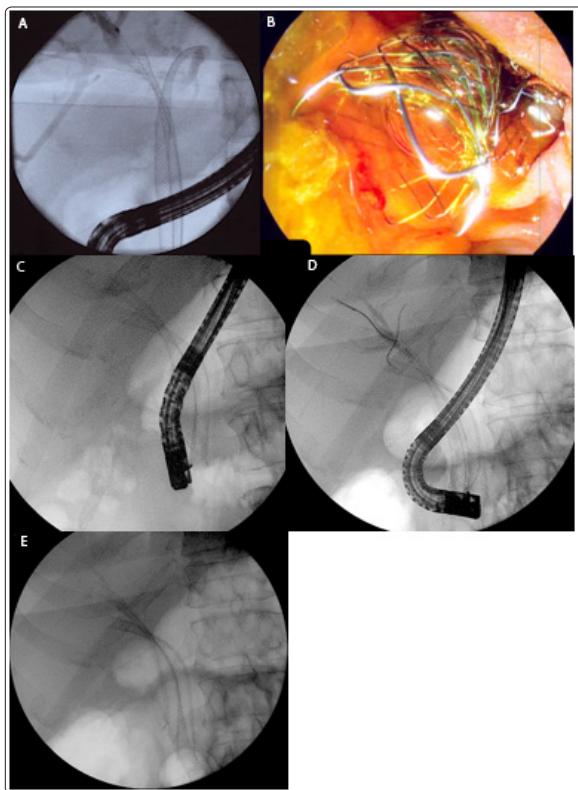
### Case 2

A 56-year-old gentleman initially presented with obstructive jaundice with an MRI strongly suggestive of a hilar malignant biliary tumor with portal vein involvement. A percutaneous transhepatic cholangiogram (PTC) approach was undertaken and palliative insertion of two uncovered SEMS into the left and right intra-hepatic ducts was performed without tissue diagnosis. A surgical referral was made for resection; however, concerns were raised regarding lack of definitive diagnosis.

A subsequent PET scan was performed 8 months later which revealed moderate increased metabolism outlining the stent position. The patient was asymptomatic and had cholestatic liver function tests (U/L), GGT 1299; ALP 670; ALT 111; AST 64 with a normal bilirubin of 12. CEA (μg/L) and CA19.9 (kU/L) were also mildly raised at 7 and 72 respectively. The patient underwent ERCP and cholangioscopy with a view to biopsy,

however histopathology was non-diagnostic. Within 4 weeks another ERCP was performed for staged stent removal with the stent-in-stent technique, to subsequently remove all stents over 2 sessions and biopsy underlying exposed bile duct tissue. A cholangiogram revealed a stenosis at the bifurcation. One 10mm x 8cm transpapillary covered metal stent was placed into the left hepatic duct, and one 10mm x 10cm transpapillary covered metal stent was placed into the right hepatic duct, with bile flowing well through both systems (Figure 2). Two months later a repeat ERCP was performed which revealed tissue in-growth and migration of the right hepatic duct stent and a wire was unable to be passed into the right system therefore a decision was made not to attempt stent removal. Repeat biopsy taken at the time ERCP did not reveal a diagnosis.

Soon after the patient presented with right upper quadrant pain and features of infection. Blood tests showed; CRP 318, WCC 14.1, bilirubin 21, GGT 529, ALP 407, ALT 22, AST 16. The patient underwent an urgent CT scan which revealed a ruptured gall bladder and surrounding free fluid. An emergent open subtotal cholecystectomy and abscess drainage was performed. The patient recovered well with an external drain.



**Figure 1:** Cholangiogram and endoscopic views

- A. Case 1** – Initial insertion of bilateral uncovered SEMS deployed in good position on cholangiogram
- B. Case 1** – Endoscopic view of Initial insertion of bilateral uncovered SEMS
- C. Case 2** – Left and right (most superior stent) uncovered metal stents in position from initial PTC
- D. Case 2** – Wire cannulation of both systems to place metal stents
- E. Case 2** – Successful placement of FCSEMS within uncovered metal stents in left and right intra-hepatic systems

## Discussion

Cholecystitis after SEMS placement is a well-documented adverse event. However, many studies that accurately report cholecystitis rates include a majority of patients with distal malignant biliary obstruction<sup>1, 6-9</sup>, whereas rates associated with hilar malignancies

are less reported. Although there is debate as to whether FCSEMSs causes higher rates of cholecystitis compared with USEMSs, our two cases show that cholecystitis occurs when FCSEMS are placed within stents in situ in hilar malignancies. This is explained by the interference of flow of the cystic orifice which is a known risk factor for acute cholecystitis<sup>7, 8</sup>. Biliary obstruction secondary to malignancy generates increased impedance compared to benign causes of obstruction; thus, those with malignant obstruction require drainage by ERCP and require stent placement to maintain biliary flow. We describe the first reported cases of acute cholecystitis following subsequent ERCP and placement of metal stent-in-stents in the biliary system.

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