

## Three Episodes of Postcholecystectomy Syndrome Due to Remnant Cystic Duct with Prior Mirizzi Syndrome

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### Abstract

A 34-year-old female presented to an outside hospital with symptoms of severe, constant, sharp, right upper quadrant abdominal pain with band-like sensation across the epigastrium and left upper quadrant pain associated with nausea and vomiting. Six years ago, she received a cholecystectomy, but since, she demonstrated 3 episodes of Postcholecystectomy Syndrome (PCS) due to recurrent remnant cystic duct stones. Each occurrence was remedied by Endoscopic Retrograde Cholangiopancreatography (ERCP) with stone removal. She was asymptomatic post cholecystectomy for 5 years until 9 months prior and again 1 week prior to her current presentation with similar symptoms to her current episode. Multiple imaging modalities including ultrasound, computed tomography, and magnetic resonance imaging revealed a mass consistent with a stone with common bile duct dilation. Via ERCP, the stone was removed. She underwent laparoscopic hand port assisted remnant cholecystectomy with extraction of another endoluminal stone. Symptoms resolved with no reoccurrence for 1 month. This case demonstrates that postcholecystectomy syndrome and Mirizzi syndrome may present five years later with multiple episodes. Surgical referral is an important resource.

### Introduction

Postcholecystectomy Syndrome (PCS) is the persistence of symptoms of biliary colic after cholecystectomy. PCS is common with reports of 5 to 40% of patients experiencing the phenomenon [1,2]. However, of the multiple etiologies, stones in the remnant cystic duct caused only 2.5% of cases, thus a rare etiology. Mirizzi syndrome is a rare complication of gallstone disease with incidence of 2.8% [3]. It is defined as the extrinsic compression of the common hepatic duct from an impacted stone in gallbladder's Hartmann's pouch or cystic duct leading to obstruction. In this case, the patient suffered 3 episodes of recurrent remnant cystic duct stones starting 5 years after her cholecystectomy with each time requiring Endoscopic Retrograde Cholangiopancreatography (ERCP). One of her episodes caused Mirizzi syndrome 6 years after cholecystectomy. Surgical intervention was necessary to remove the remnant cystic duct, the source of her recurrent PCS.

### Case Presentation

A 34-year-old female presented to outside hospital with symptoms of severe, constant, sharp right upper quadrant abdominal pain with band-like sensation across the epigastrium, left up-

per quadrant pain, and associated with nausea and vomiting. The symptoms were exacerbated by food intake. Analgesics and vomiting relieved them. She was status post cholecystectomy 6 years prior and had 2 previous episodes of similar symptoms 9 months and 1 week apart from current presentation. Her diet did not consist of any fast food or alcohol. On presentation, physical examination revealed a soft abdomen with mild tenderness to palpation of the upper abdomen without guarding, rigidity, or a Murphy's sign. Laboratory tests showed elevated AST of 759, ALT of 650, ALP of 361, and total bilirubin of 2.4. Platelets were elevated to 497 bil/L, chloride was decreased to 94 mmol/L, and UA showed hematuria. Beta HCG test was negative. Lipase was normal at 19 IU/L.

Ultrasound imaging done one week prior to current presentation at the level of gallbladder fossa found hypoechoic materials within a dilated, sac-like structure measuring 1.9 centimeters (cm) along with a dilated common bile duct measuring >1 cm in diameter (Figures 1,2). Noncontrast Computed Tomography (CT), also obtained one-week prior, demonstrated a radiolucent, laminated stone measuring 1.4 centimeters in diameter with Hounsfield units of +1000 past the cholecystectomy surgical clips. Multiple small stones were also visible (Figures 3,4). During this time, Magnetic

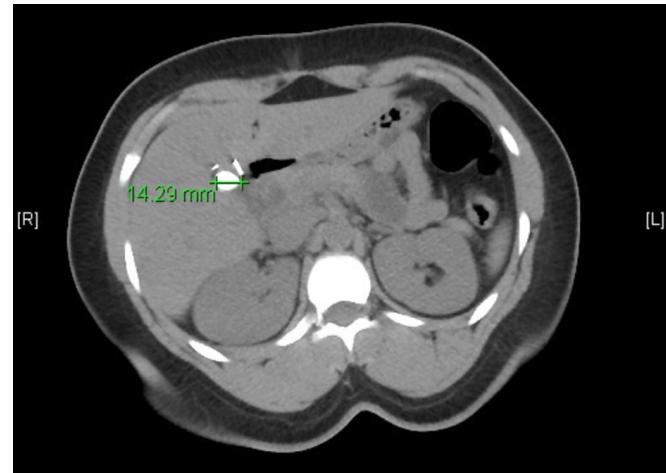
Resonance Imaging (MRI) performed showed a dilated common bile duct measuring 1.2 cm, an obstruction to distal duct by a 1 cm stone, and at least 50% of narrowing of the lumen. These findings were consistent with the Mirizzi syndrome.



**Figure 1:** Ultrasound in liver long view measuring a dilated cystic duct (1.90 cm) with hypoechoic structures within the sac.



**Figure 2:** Ultrasound in pancreas transverse view showing a dilated common bile duct measuring 1.40 cm in diameter.



**Figure 3:** Abdominal CT measuring a 1.42 cm of stone past the point of cholecystectomy surgical clips (arrow).



**Figure 4:** Abdominal CT showing multiple radiolucent structures, which is indicative of stones in an empty gallbladder fossa.

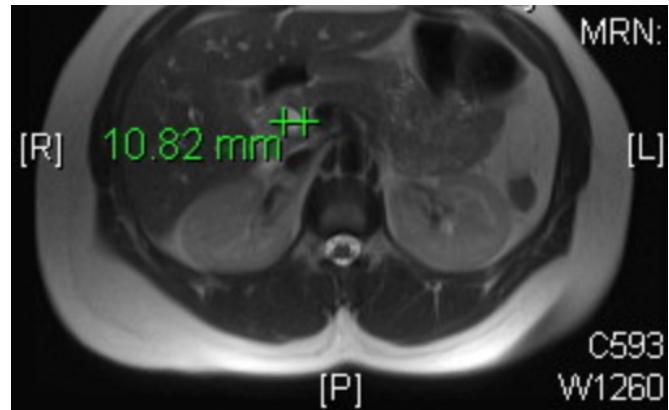
Magnetic Resonance Cholangiopancreatography (MRCP) captured on day of the case presentation showed dilated, remnant cystic duct with filling defect. Based on the patient's presenting symptoms, this was indicative of obstruction caused by stone (Figure 5). Comparison calculation resulted in measurement of cystic duct remnant as  $>4$  cm. However, comparison calculation was used with caution as it involved converting pixels into length.

Furthermore, the MRI captured the dilated remnant cystic duct with a  $>1$  cm stone surrounded by edema, a sign of inflammation. ERCP was performed with removal of that one stone, and the patient was transferred for surgical intervention for her recurrent episodes.



**Figure 5:** MRCP imaging demonstrating a remnant cystic duct with a filling defect (arrow), which is indicative of obstruction by a stone based on case presentation.

Upon transfer for surgery 2 days after her ERCP, patient presented in good health without abdominal pain, nausea, vomiting, nor anorexia. Physical examination was unremarkable without tenderness of the upper abdomen. Her Aspartate Aminotransferase (AST), Alanine Aminotransferase (ALT), Alkaline Phosphatase (ALP), and total bilirubin levels all down trended to 148, 320, 315, and 1.6, respectively. She underwent laparoscopic hand port assisted remnant cholecystectomy with stone extraction. Surgical findings noted remnant, cystic duct measuring 1.5 cm with an endoluminal stone. The mucosa of the enlarged cystic duct was cauterized. Surgical pathology report noted chronic inflammation of cystic duct but no signs of malignancy (Figure 6).



**Figure 6:** MRI measuring a  $>1$  cm stone in remnant duct with surrounding edema (arrow).

## Discussion

The cystic duct typically measures 2-4 cm in length, and formation of stone in the structure leads to symptoms of dyspepsia, nausea, vomiting, and possibly jaundice [4]. Cholecystectomy provides symptom relief to 85% of patients [5].

After cholecystectomy, an incomplete surgery can leave residual duct of greater than 1 cm in length, defined as a cystic duct remnant, or a gallbladder remnant [6]. Our patient had long residual cystic duct, and laparoscopic surgery led to finding of a remnant measuring 1.5 cm in length. In one study by Rogy et al., 10.8% of patients undergoing second bile duct operation post cholecystectomy had cystic duct stump of greater than 1.5 cm [7]. Formation of calculi in this structure can lead to postcholecystectomy syndrome (PCS) [8].

PCS is the presentation of biliary colic symptoms after cholecystectomy and can present as continuation of symptoms after the operation or interval of 2 days to 25 years after disease free period. PCS has been reported in 5 to 40% of patients post open and laparoscopic cholecystectomy [1,2,8]. Most common causes are extrabiliary disorders, such as, chronic pancreatitis. Biliary etiologies include eight different origins, such as strictures, sphincter of Oddi dysfunction or tumors, but the incidence of PCS due to cystic duct remnant calculi has been reported to be  $<2.5\%$  and typically presents less than 2 years after cholecystectomy [9,10].

Case series by Phillips et al from 2001 to 2012 at one center concluded unknown incidence of remnant cystic duct lithiasis [11]. Our patient initially presented with PCS 5 years later her cholecystectomy due to lithiasis. We were unable to obtain outside records, but she reported her first episode of PCS caused by retained stone in the cystic duct remnant took over 10 days to diagnose due to the anchoring heuristic of pancreatitis.

The diagnosis of cystic duct remnant stone can be difficult. In general, multiple imaging studies are available to depict the disease processes of the cystic duct. These include ultrasound, CT, ERCP, MRCP, MRI, and cholescintigraphy. Abdominal ultrasound is 50% less effective in diagnosing cystic duct stones whereas MRCP and ERCP have sensitivity 85-100% [12]. However, these modalities of imaging decrease in their ability to detect a remnant cystic duct stone. In one study, ERCP had specificity of 66.7% (4 out of 6 patients), and in another study, ERCP and MRCP failed to identify 2 out of 3 patients with stump stone [13,14]. For our patient, this was her third presentation of PCS. All the imaging modalities performed on her confirmed a remnant structure with calculi. However, this can be due to changes caused by the prior episodes of PCS leading to inflammation and structural alterations [15]. Our team had high degree of suspicion, but it is important to note the imaging modalities may not be as reliable in detecting stone in remnant cystic duct/gallbladder.

Surgical intervention for a “recholecystectomy” provides a definite for treatment stone in retained gallbladder or dilated cystic duct [6]. Our patient had experienced 3 episodes of PCS, each caused by stones in the remnant cystic duct. Each of the episodes was treated with ERCP, with improvement of symptoms followed by recurrence with decrease in time interval (5 years to 1 week apart).

## Conclusion

In internal medicine, it is important to have stone in retained cholecystectomy structures on differential for patients presenting with cholelithiasis/ cholecystitis symptoms even if the patient status post cholecystectomy. Laboratory tests would show derangements in similar pattern as cholelithiasis/cholecystitis. Imaging studies are not as reliable with remnant cystic duct lithiasis and cannot be used for rule out PCS due to retained stone. Surgical intervention is a definite treatment and delay in referral can cause multiple procedures for the patient as in our case.

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