CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 78 (2021) 223–227

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

# Acute severe cholecystitis with empyema presenting as a gallbladder mass, jaundice and Mirizzi Syndrome: A case report

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ARTICLE INFO

*Article history:*
Received 19 November 2020
Received in revised form 9 December 2020
Accepted 11 December 2020
Available online 16 December 2020

*Keywords:*
Acute cholecystitis
Mirizzi Syndrome
Xanthogranulomatous cholecystitis
Gallbladder carcinoma

ABSTRACT

*INTRODUCTION:* Acute severe cholecystitis with empyema presenting as a gallbladder mass, jaundice and Mirizzi Syndrome (MS) is a complex surgical problem both diagnostically and in terms of management as it mimics both xanthogranulomatous cholecystitis (XGC) and gallbladder carcinoma.
*PRESENTATION OF CASE:* A 48-year-old gentleman was referred to us with biliary colic and weight loss with ultrasound findings of gallstones. At subsequent follow-up he became deeply jaundiced with deranged liver function and a CT showing a gallbladder mass and dilated biliary tree. Follow-up MRCP suggested XGC and concomitant MS, but a malignant process could not be excluded. Pre-operative fine needle aspiration cytology (FNAC) at the time of percutaneous biliary drainage for his jaundice demonstrated XGC with no evidence of malignancy. Given the dense inflammation and a tense empyema at laparoscopy, he underwent a subtotal fenestrating cholecystectomy. The final histopathological diagnosis was acute cholecystitis.
*DISCUSSION:* Our patient likely had unrecognised acute cholecystitis which progressed to a complex mass with empyema and type I Mirizzi Syndrome, ultimately resulting in severe obstructive jaundice mimicking gallbladder carcinoma. Given that a laparoscopic total cholecystectomy is dangerous in these cases of severe inflammation, a laparoscopic subtotal cholecystectomy has been shown to be a safe alternative to more invasive strategies and was successfully utilised in our patient.
*CONCLUSION:* Acute severe cholecystitis with empyema presenting as a gallbladder mass, jaundice and Mirizzi Syndrome is a rare manifestation that requires adequate pre-operative work-up to exclude malignancy. Subtotal fenestrating cholecystectomy is a safe and effective alternative to open surgery in these cases of complex inflammation.


## 1. Introduction

Acute severe cholecystitis with empyema presenting as a gallbladder mass, jaundice and Mirizzi Syndrome (MS) is a complex surgical problem both diagnostically and in terms of management as it mimics both xanthogranulomatous cholecystitis (XGC) and gallbladder carcinoma. We present this interesting and unusual case of a 48-year-old gentleman who presented with pain and weight loss and subsequently developed jaundice. We describe the diagnostic challenges in differentiating this benign but complicated disease process from a malignant aetiology and discuss our subsequent management.

This case report has been reported in line with the SCARE criteria [14].

## 2. Presentation of case

A 48-year-old Korean gentleman was referred to our surgical outpatient department with recurrent upper abdominal pain in the context of gallstones. Unusually, he also complained of intermittent dark urine and 7 kg of weight loss. There was no significant medical, surgical, pharmacological, psychosocial or family history of note apart from social alcohol consumption. He had a benign abdominal examination, with no appreciable masses. His full blood count and liver function tests were also unremarkable.

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https://doi.org/10.1016/j.ijscr.2020.12.035

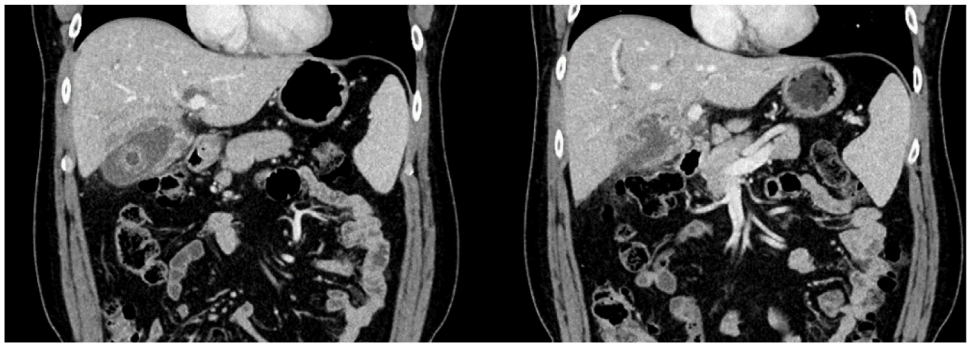

**Fig. 1.** CT demonstrating an irregular circumferential gallbladder mass contiguous with the adjacent liver parenchyma, involving the primary biliary confluence.

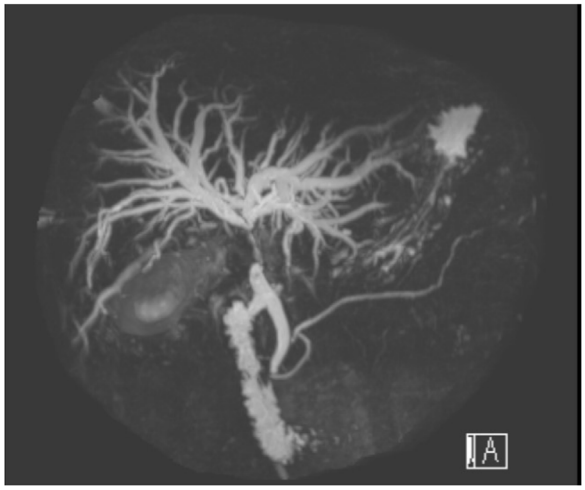

**Fig. 2.** MRCP demonstrating a 25 mm long stricture of the common hepatic duct involving the primary biliary confluence with moderate upstream bi-lobar intrahepatic bile duct dilatation.

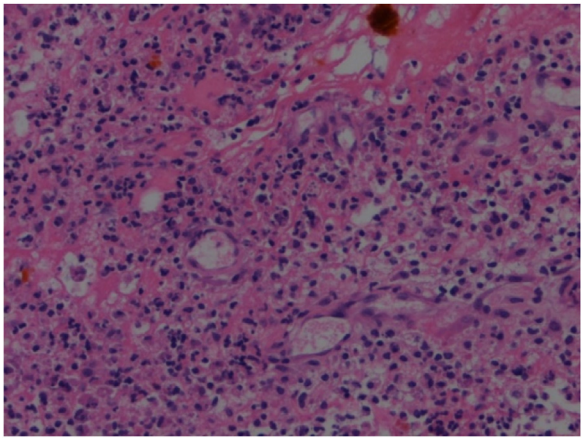

**Fig. 3.** Pre-operative frozen section demonstrating accumulation of lipid-laden macrophages, fibrous tissue and inflammatory cell infiltrate, suggestive of XGC (haematoxylin and eosin stained section – 400×).

Initial ultrasound organised by his referring general practitioner revealed gallstones and a non-dilated common bile duct. There was also significant irregular thickening of the gallbladder, with a poorly defined wall due to significant sonographic acoustic shadowing from the stone burden. This prompted a computed tomography (CT) of the abdomen.

On follow-up in the outpatient department 3 weeks later, the patient became deeply jaundiced, and repeat liver function tests were markedly deranged: total bilirubin 310 μmol/L (<20); conjugated bilirubin 184 μmol/L (<4); alkaline phosphatase 440 μmol/L (30–110); gamma-glutamyl transferase 513 U/L (<55); alanine transaminase 518 U/L (<45) and aspartate transaminase 221 U/L (<35). CA 19.9 was also elevated at 210 kU/L (<35), which may have been related to his jaundice.

The CT demonstrated a gallbladder mass which exhibited irregular circumferential thickening contiguous with the adjacent liver parenchyma and also appeared to involve the biliary bifurcation, concerning of a locally advanced malignancy (Fig. 1).

Further investigation with a magnetic resonance cholangiopancreatography (MRCP) revealed irregular circumferential thickening of the gallbladder wall measuring 16 mm, with a 24 mm impacted stone in Hartmann's pouch. At the region of impaction, there was a 25 mm stricture of the common hepatic duct involving the primary biliary confluence with moderate upstream bi-lobar intrahepatic biliary dilatation (Fig. 2).

The MRCP findings were suggestive of xanthogranulomatous cholecystitis (XGC) and concomitant Mirizzi Syndrome (MS), but given the mass like appearance and biliary stricture, malignancy could not be excluded.

Features characteristic of XGC were the variability of signal intensity and presence of intra-mural nodules demonstrating reduced intensity on 'out-phase' imaging. This is due to foamy histiocytes, lymphocytes, polymorphonuclear leucocytes, fibrosis, micro-abscesses and necrosis within these nodules.

Given the patient was deeply jaundiced, percutaneous transhepatic cholangiography (PTC) and biliary drainage was performed with an internal-external drain. Whilst an endoscopic retrograde cholangiopancreatography (ERCP) was considered, the concerns with this approach was the length of the biliary stricture involving the bifurcation, and concern that the ERCP could fail in providing adequate drainage. Should the ERCP fail to adequately drain both the right and left systems, the biliary instrumentation would likely precipitate cholangitis and further complicate his management.

A cholangiogram performed during the PTC demonstrated a tight stricture at the confluence of the hepatic ducts extending to the proximal common hepatic duct. Contrast injected in the right hepatic duct was seen in the left intrahepatic biliary system, confirming patency across the confluence. An internal-external biliary drainage catheter was placed traversing the stricture across the common hepatic duct terminating in the duodenum. Bile was sent for cytology and was negative for malignant cells. There were no post-procedural complications and his bilirubin successfully decreased.

A fine needle aspiration cytology (FNAC) and frozen section of the gallbladder taken at time of PTC identified no evidence of malignancy and suggested XGC (Fig. 3).

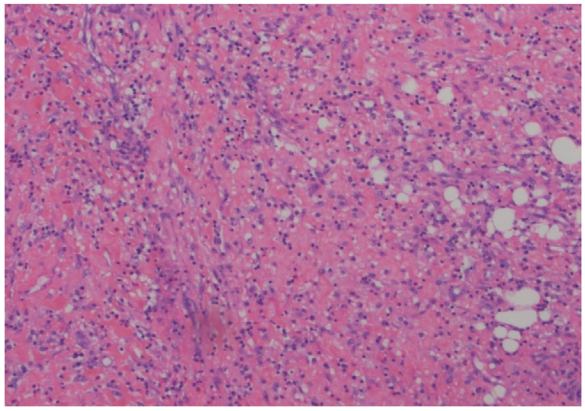

**Fig. 4.** Histology demonstrating a thickened gallbladder wall with mixed inflammatory infiltrate showing prominence of neutrophils, in keeping with a final diagnosis of acute cholecystitis (haematoxylin and eosin stained section – 100×).

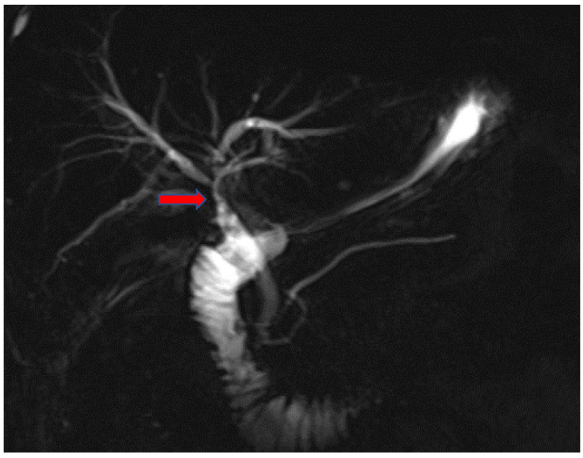

**Fig. 5.** Post-operative MRCP demonstrating improved appearance of the stricture (red arrow).

A fludeoxyglucose-positron emission tomography (FDG-PET) study revealed a non-contiguous but intensely avid uptake with a thickened gallbladder wall. This was a non-specific finding that could relate to either a malignant or inflammatory process.

The overall presentation and investigations were in favour of an inflammatory gallbladder mass secondary to an impacted stone with MS. Thus, the patient was scheduled for a laparoscopy with a view to perform a cholecystectomy and remove the large impacted gallbladder stone which was presumed to be the cause for his extrinsic biliary compression and stricturing. The operation was performed by an experienced consultant general surgeon subspecialised in hepato-pancreato-biliary (HPB) surgery.

The patient had an empyema of the gallbladder at surgery and had dense gallbladder adhesions. The gallbladder wall was fragile on handling and the hepatocystic triangle was frozen from gross inflammation. Given the degree of inflammation and difficulty in delineating the anatomy clearly, we proceeded to perform a subtotal fenestrating cholecystectomy.

The gallbladder empyema was drained and the anterior-mid gallbladder wall and fundus were deroofed. Multiple stones were extracted from Hartmann's pouch and the impacted gallbladder neck stone causing extrinsic biliary compression was also removed. There was no evidence of a cholecysto-choledochal fistula making this a type I MS. Cholangiogram was attempted, however was unsuccessful as it was not possible to establish a seal around the opened and distended Hartmann's remnant. A drain was left within the subhepatic fossa.

Final histology revealed acute cholecystitis without evidence of dysplasia or malignancy (Fig. 4). Interestingly, there were no features of XGC.

The patient tolerated the procedure well and had a good post-operative recovery on the ward. He underwent daily bloods which showed marked improvement in his liver function: total bilirubin 32 μmol/L (<20); conjugated bilirubin 14 μmol/L (<4) gamma-glutamyl transferase 50 U/L (<55); alanine transaminase 136 U/L (<45) and aspartate transaminase 40 U/L (<35). His subhepatic drain was then removed and he was subsequently discharged.

Repeat MRCP 13-days post-operative demonstrated a reduction in gallbladder wall thickening to 12 mm and significant improvement of the common hepatic duct stricture (Fig. 5). At 22-days post-operative the internal-external biliary drainage catheter was removed. He was reviewed as an outpatient one-month post-operative where he remained symptom free with no concerns and completely normal liver function tests. Given this, he was discharged from our clinic back into the care of his general practitioner.

## 3. Discussion

This patient represents an interesting and unusual case of severe acute cholecystitis, and highlights the complications that follow unrecognised acute cholecystitis. It is likely that our patient had acute cholecystitis for some time which ultimately resulted in a complicated gallbladder mass and empyema. The ongoing inflammation from the impacted gallstone then resulted in a type I Mirizzi Syndrome (MS), which presented with severe obstructive jaundice from extrinsic compression of the biliary tree.

MS is characterised by gallstone impaction within the cystic duct or gallbladder neck that results in mechanical obstruction of the common hepatic duct, manifesting as right upper quadrant pain, jaundice and fever. This impaction can be complicated further by formation of a cholecysto-choledochal fistula, which forms the basis on the classification of MS described by Csendes et al. [1–3]. Type I lesions involve external compression of the common hepatic duct only. Type II and III lesions encompass fistula formation, involving less than one third and involving between one-third and two-thirds of the circumference of the common bile duct respectively. Type IV lesions describe complete destruction of the common bile duct [3].

Even though the patient's presentation with a mass and jaundice and pre-operative work-up supported XGC, the final histological diagnosis disproved our suspicion. XGC is a rare, yet benign disease characterised by the accumulation of lipid-laden macrophages, fibrous tissue and an inflammatory cell infiltrate that results in destructive inflammatory process of the gallbladder. It may involve local invasion into the liver, common bile duct or colon, and therefore XGC is well-recognised to mimic that of both chronic cholecystitis and gallbladder carcinoma as was suspected in this case [4,5].

Pre-operative imaging often cannot definitively differentiate between these conditions as they share similar features including diffuse gallbladder wall thickening, hypoattenuated gallbladder nodules, invasion through cystic plate and portal lymphadenopathy. XGC therefore often requires pre-or-intra-operative frozen section to differentiate it from malignancy [6]. Studies suggest that 9.1%–32.5% of XGC also have concomitant MS [1,7–9].

With these pathologies superimposed, a laparoscopic total cholecystectomy in either chronic cholecystitis or XGC, with concomitant MS is dangerous as extensive inflammation will inevitably impair the ability to visualise and dissect the hepatocystic triangle safely, and increase the likelihood of vascular and bile duct injury [4].

Open cholecystectomy is often considered most appropriate for MS [10], however, studies have shown that a laparoscopic approach can be an effective alternative [10–12]. In a study by Gelbard et al. [11], 26/55 (47.3%) of patients with type I MS underwent a laparoscopic cholecystectomy, while the remainder had an open procedure, including the 13/55 (23.6%) type I MS patients converted from laparoscopic to an open. All 26 type I MS patients operated laparoscopically had a successful procedure, with no significant difference in complications and a shorter hospital stay compared to the open group.

Koike et al. [12] compared the outcomes of MS patients undergoing laparoscopic subtotal cholecestectomy (LSTC), where hepatocystic triangle dissection is avoided, versus non-MS patients undergoing routine laparoscopic cholecystectomy. Between the two groups there were no significant differences in operation time, blood loss, length of stay, bile leak and bile duct injury.

The terms 'subtotal' or 'partial' cholecystectomy are imprecise as they fail to stipulate whether the procedure results in a functional gallbladder remnant. Strasberg et al. [13] established that subtotal cholecystectomy can be classified into two subtypes, fenestrating or reconstituting which is differentiated by whether the lowest portion of the gallbladder is left open or closed respectively. In a fenestrating LSTC, Hartmann's pouch is opened, and the cystic duct lumen is closed or left open. In a reconstituting LSTC, Hartmann's pouch is closed by sutures or a stapler. Compared to a fenestrating LSTC, the reconstituting approach reduces the risk of postoperative fistula, but as it creates a gallbladder remnant it can result in a recurrence of symptomatic gallstones.

## 4. Conclusion

This case highlights the role of a fenestrating LSTC for the management of severe acute cholecystitis with an empyema and type I MS. It also highlights the importance of a multidisciplinary approach in managing a mass highly suspicious for malignancy, and successful pre-operative biliary drainage with a PTC.

## Declaration of Competing Interest

None.

## Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or non-for-profit sectors.

## Ethical approval

Case reports are exempt from ethical approval from our institution. We do not have ethics committee approval as this was not a study on multiple patients.

## 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's contribution

All authors have made substantial contributions to the conception and design of the study, acquisition of the data, analysis and interpretation of the data or drafting/critical revision. All authors have approved the version that is submitted.

## Registration of research studies

Not applicable.

## Guarantor

Derek Mao.

## Provenance and peer review

Not commissioned, externally peer-reviewed.

## Patient perspective

"I was relieved after realising my final diagnosis was not cancer, as I was told that my condition mimics it and could only be confirmed after surgery. I was very happy with my treatment and I am thankful to my surgeons to now be pain and symptom free."

## CRediT authorship contribution statement

**Derek Mao:** Conceptualization, Methodology, Writing - original draft, Writing - review & editing, Visualization, Project administration, Investigation. **Bishoy Mekaeil:** Writing - original draft, Writing - review & editing, Visualization, Project administration. **Matthew Lyon:** Writing - original draft, Writing - review & editing, Visualization, Project administration. **Harsh Kandpal:** Resources, Formal analysis, Writing - original draft. **Varghese Pynadath Joseph:** Resources, Formal analysis, Writing - original draft. **Shilpi Gupta:** Resources, Formal analysis, Writing - original draft. **Manju Dashini Chandrasegaram:** Conceptualization, Methodology, Resources, Writing - original draft, Writing - review & editing, Supervision, Project administration.

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