

Dual pathology—An unreported case

Darren Yap^{a,*}, Ashraf Rasheed^a, Majid Rashid^b

^a Department of Upper GI Surgery, Royal Gwent Hospital, Cardiff Road, Newport NP20 2UB, United Kingdom

^b Department of Histopathology, Royal Gwent Hospital, Cardiff Road, Newport NP20 2UB, United Kingdom

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ABSTRACT

INTRODUCTION: Symptomatic biliary disease in children and young adults requiring surgical intervention are uncommon. However even rarer is the occurrence of a spontaneous gallbladder necrosis in a child. We report a case of spontaneous necrosis in a child with no apparent causative factors.

CASE: Fit and well 16 year-old boy presented with acute generalized lower abdominal pain. Examination revealed mild epigastric pain with rebound tenderness and guarding of the right iliac fossa. Diagnostic laparoscopy showed a necrotic gallbladder and incidental finding of a Meckel's diverticulum. He had a cholecystectomy and Meckel's diverticulum resection. Patient recovered uneventfully and was discharged home. He was reviewed 2 months later and recovered well with no evidence of any post-operative complication. He was discharged without any further follow up.

DISCUSSION: Gall bladder necrosis is a rare cause of an acute abdomen. We present the first reported case of a spontaneous gallbladder necrosis with no apparent cause. Literature review showed various causes of gall bladder necrosis including trauma, acalculous cholecystitis, gallbladder torsion, gangrenous cholecystitis and etc.

CONCLUSION: We report a case of spontaneous gallbladder necrosis in a young healthy male with no family history of thrombotic disorders or any history of sepsis, intervention, trauma and no obvious underlying anatomical or histological abnormalities. This is an exceedingly rare pathology and one would be forgiven for not including it on the list of a differential diagnosis in such circumstance. However it is important to send tissue sample to exclude any underlying histological aetiological factors.

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1. Introduction

Although uncommon, children and young adults may occasionally present with symptomatic biliary disease requiring operative intervention. Even rarer is the occurrence of spontaneous gallbladder necrosis, and difficulty in diagnosing this uncommon occurrence can lead to operative delay. Common causes include cholecystitis, torsion, gallbladder torsion, infections and vasculitic diseases. Herein we present an unusual case of spontaneous gallbladder necrosis with no apparent causative factors.

2. Methods

This case report have been written in line with the CARE criteria [16].

* Corresponding author.

E-mail addresses: darrenyap.jx@gmail.com (D. Yap), ashraf.rasheed@wales.nhs.uk (A. Rasheed), majid.rashid@wales.nhs.uk (M. Rashid).

3. Case report

A 16 year old boy presented with a day history of sudden onset generalized lower abdominal pain. It is associated with nausea and vomiting. Patient denied any previous history of similar discomfort, change in bowel habits, trauma or urinary symptoms. Symptoms persisted despite an overnight treatment with paracetamol and ibuprofen and the pain localized to the right iliac fossa. He is otherwise a fit and healthy with no other significant past medical history and immunization up to date.

On examination, he was mildly tender in the epigastrium with marked right iliac fossa rebound tenderness, guarding and was Rovsing's Sign was positive.

His urine dipstick was negative, haematological investigations revealed a leucocytosis of 16.3, CRP of 4 with an isolated raised bilirubin of 65.

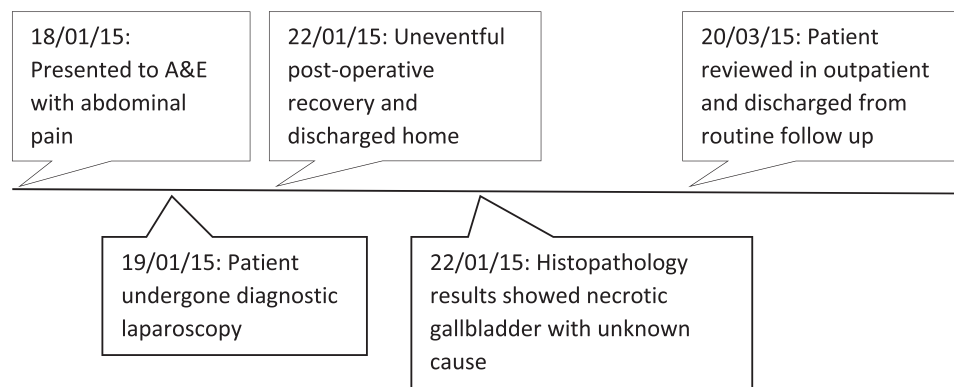
Our initial impression was that of an acute appendicitis hence proceeded to diagnostic laparoscopy noting a normal appendix with bile tinged peritoneal fluid secondary to a necrotic gallbladder; the gall bladder was of normal disposition i.e., untorted with an incidental finding of an ileal Meckel's diverticulum. We proceeded to laparoscopic cholecystectomy and pre-emptive laparoscopic Meckel's diverticulectomy in view of his young age. Both his blood



Fig. 1. Necrotic gallbladder with bile within peritoneum.

and peritoneal fluid did not isolate any organisms. Patient recovered uneventfully and was discharged home. The histology of the gallbladder confirms a spontaneous necrotic gallbladder with no evidence of any stones. Histologically there was no evidence of vasculitis, viral inclusion bodies or trauma. Patient followed up in outpatient in 2 months' time and have recovered uneventfully with no complication; hence he was discharged from routine follow up.

4. Timeline



5. Discussion

Gallbladder necrosis is a rare cause of acute abdomen. Even more unusual is to be unable to identify a specific cause. Various causes for gallbladder necrosis were cited before; the Journal of trauma reported one case of necrotic gallbladder secondary to hepatic trauma in the context of non-operative trauma management in a retrospective study of 185 paediatric patient [1]. There were 9 reported cases of acalculous cholecystitis secondary to non-typhoidal salmonella reported in the English literature up to present date [3]. The aetiology of acalculous cholecystitis is multifactorial and various risk factors including those resulting in decreased motility and bile stasis thought to contribute to the formation of “sludge,” a calcium bilirubinate mixture with an increased level of unconjugated bilirubin [14]. There was one reported candida peritonitis case in a patient with necrotising cholecystitis [4]. However primary fungal peritonitis remain rare with a reported 27 cases over the past 10 years. Emphysematous cholecystitis, a virulent form of acute cholecystitis secondary to gas-producing anaerobe infection of the gall bladder such as *Clostridium perfringens* or *Escherichia Coli* was also reported in the literature [5–7], mortality associated with latter has been reported

Table 1
Causes of gallbladder necrosis.

1. Spontaneous
2. Idiopathic
3. Secondary
 - a. Lithiasis/gallstone
 - b. Inflammation/infection
 - c. Torsion
 - d. Other: congenital, salmonellosis, anticoagulant
4. Traumatic
 - a. Blunt
 - b. Penetrating
5. Iatrogenic

to be 20–30% with a 30 fold increase risk of gangrene and 5 times risk of perforation [5–7] (Fig. 1).

Gangrenous cholecystitis has incidence ranging from 2 to 38% of all patients with acute cholecystitis and associated with major complication, increased morbidity and mortality [8–10] (Table 1).

Acute aortic dissection can cause malperfusion of end organs resulting in abdominal ischaemia. Gall bladder is especially vulnerable to ischemia due to the terminal nature of its blood supply. There is only one reported case of gall bladder perforation as an ischaemic complication of type A aortic dissection. It is a result from extension of dissection into the branch vessels or physical obstruction of the vessel orifice by the aortic flap [11] (Fig. 2).

Gallbladder torsion is a rare condition with only a few cases which have been reported in recent literature [2,15]. Incidence has been estimated at one in 365,000 hospital admissions [15]. Aetiology of gall bladder torsion is unknown however some case

reports suggest that it may be due to variation in the hepatobiliary anatomy [2,15]. With four main anatomic variation were described namely, the free-floating gallbladder suspended only by the cystic duct and artery with an absent gallbladder mesentery, due to abnormal migration of the pars cystica from the hepatic diverticulum during fourth to seventh weeks of embryological development; atrophy of the liver in combination with decreased elasticity of connective tissue in elderly patient with a normally formed mesentery leading to progressively mobile gallbladder; rotation of a portion of fundus when it is not fixed to the liver and finally a normal fixed gallbladder attached to a mobile hepatic lobe free from its coronary and triangular ligaments facilitating the torsion [15]. The clinical presentation of torsion cholecystitis is indistinguishable from the usual acute cholecystitis however it demands emergency surgery [2]. Lau et al., 1982 described a triad of triads of clinical features of gallbladder volvulus which are shown in Table 2 [2,15].

6. Conclusion

We report a case of spontaneous gallbladder necrosis in a young healthy male with no family history of thrombotic disorders or any

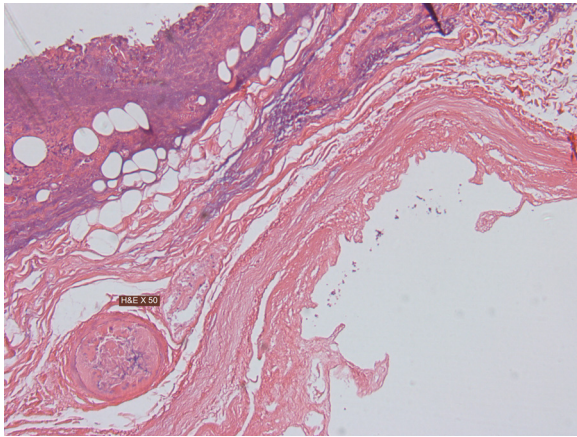


Fig. 2. Histology cross-sections showing transmural infarction of the gallbladder wall with mucosal necrosis and serosal acute inflammatory response at 50× magnification.

Table 2

Triad of triads of clinical features of gallbladder volvulus.

Triad 1 – Patient characteristics
- Thin
- Elderly
- Chronic chest disease or deformed spine
Triad 2 – Patient symptoms
- RUQ pain
- Early onset vomiting
- Short/acute history
Triad 3 – Patient physical signs
- Abdominal mass in RUQ
- Lack of jaundice/toxaemia
- Discrepancy in pulse and temperature

history of sepsis, intervention, trauma and no obvious underlying anatomical or histological abnormalities. And although gall bladder necrosis was not entertained at any stage pre-operatively; the early and rather serendipitous diagnosis with the timely treatment lead to a good outcome. This is an exceedingly rare pathology and one would be forgiven for not including it on the list of a differential diagnosis in such circumstance. It does however elicit a set of clinical signs that should force an emergent imaging or intervention that would eventually lead to the correct diagnosis, appropriate treatment and a good outcome. It is also important to send tissue sample to exclude any underlying histological aetiological factors.

Conflict of interest

No conflict of interest.

Sources of funding

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Ethical approval

No ethical approval required.

Consent

Patient consented for case report to be published including pictures taken.

Author contribution

Each author have contributed in the writing and editing of the case report.

Guarantor

Darren Yap.

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