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Case report

Extreme hyperbilirubinaemia associated with choledocholithiasis without ascending cholangitis

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Abstract

Introduction: Charcot's triad was traditionally used to diagnose ascending cholangitis. However it is already proven that only minority of patients with ascending cholangitis who fulfill the triad of fever, jaundice and right hypochondriac pain.

Aim: We would like to highlight the rarity of severe hyperbilirubinaemia secondary to benign cause as most of the incidence raised more suspicion for primary liver disease or malignancy.

Case study: We presented a case report of a 58-year-old male patient with no comorbid who presented to us with right hypochondriac pain and obstructive jaundice with severe hyperbilirubinaemia (total bilirubin 1025 μmol/L), without fever or leukocytosis.

Results and discussion: The patient was diagnosed to have cholelithiasis with choledocholithiasis causing biliary obstruction by ultrasound and CT scan. Therapeutic endoscopic retrograde cholangiopancreaticograhy (ERCP) was done and total of 7 stones extracted and biliary stent inserted. Liver enzyme returns to normal level post ERCP. Patient had an uneventful recovery period and was planned for cholecystectomy in later date.

Conclusions: Benign conditions such as common bile duct stones still can lead to severe hyperbilirubinaemia even though it is very rare. The usage of appropriate imaging is needed to exclude malignant causes.

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1. INTRODUCTION

Gallstone disease is common, with incidence of approximately 4%–20% in the adult population, and of those, 2%–4% become symptomatic annually.[1–3] It is estimated that 30% of all patients with symptomatic cholelithiasis may harbor concomitant common bile duct (CBD) stones, and 4%–20% will have CBD stone at the time of cholecystectomy. CBD stone usually presented with complications such as ascending cholangitis or pancreatitis, however, rarely patient may be asymptomatic presenting with only hyperbilirubinaemia with deranged liver enzymes. Multiple diagnostic imaging modalities available to diagnose CBD stone, however the commonest, cost effective, non-invasive imaging modality used, which is the abdominal ultrasound is unreliable for diagnosis of CBD stone due to its low sensitivity. Therefore, an adjunct to diagnosis of CBD stone such as biochemical parameters is paramount, in deciding further invasive or expensive imaging modality to look for presence of CBD stone in a patient at risk.

2. AIM

In this paper we are presenting the case of a male patient who presented with obstructive jaundice secondary to CBD stones without clinical sepsis even though the level of serum bilirubin was extremely high. We would like to highlight the rarity of severe hyperbilirubinaemia secondary to benign cause as most of the incidence raised more suspicion for primary liver disease or malignancy.

3. CASE STUDY

A 58-year-old male patient presented with right hypochondriac pain and obstructive jaundice with severe hyperbilirubinaemia (total bilirubin >1000 μmol/L), without fever

Figure 1. Ultrasound revealed choledocholithiasis causing biliary obstruction.

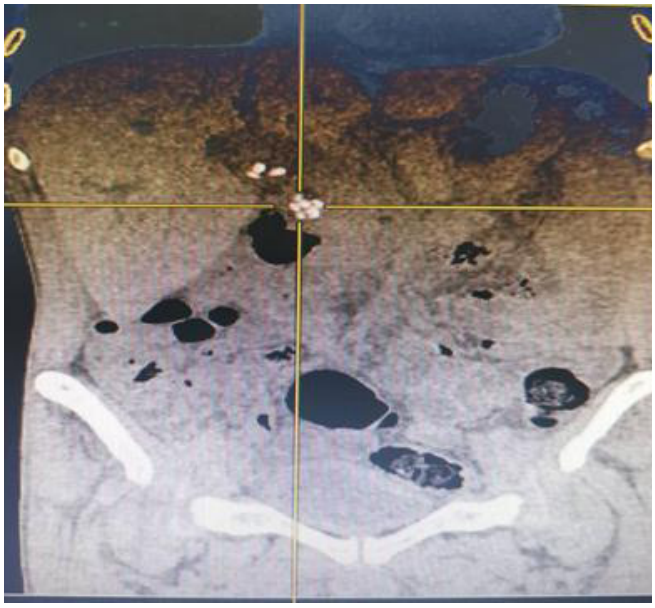

Figure 2. CT scan in coronal view showed cholelithiasis with choledocholithiasis.

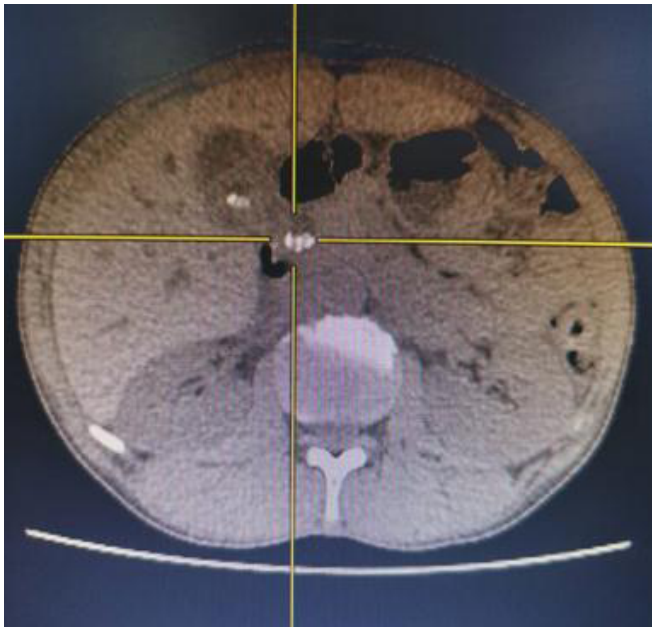

Figure 3. CT scan in axial view showed cholelithiasis with choledocholithiasis.

or leukocytosis. He was previously diagnosed to have symptomatic cholelithiasis, however not keen for surgical intervention. During the current presentation, he complained of right hypochondriac pain with deranged liver enzyme, however he was afebrile with negative Murphy's sign and septic parameters was not elevated. His liver enzymes were as stated with total bilirubin of 1025 μmol/L (direct 652 μmol/L and indirect 373 μmol/L) with ALP 419 U/L, AST 71 U/L and ALT 100 U/L

An urgent ultrasound of the hepatobiliary system revealed cholelithiasis with choledocholithiasis causing biliary obstruction (Figure 1). Computed tomography (CT) of the

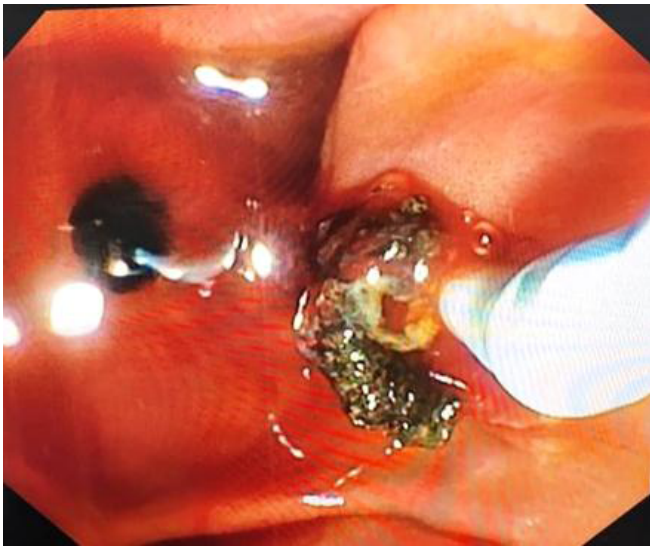

Figure 4. ERCP image showed successful stone extraction.

biliary system showed similar findings (Figures 2 and 3). Urgent therapeutic ERCP was performed and total of 7 CBD stones extracted and biliary stent inserted to decompress the obstructed biliary system (Figure 4). Liver enzyme returned to normal level post ERCP. Patient had an uneventful recovery period and was planned for cholecystectomy in later date.

4. RESULTS AND DISCUSSION

Gallstone disease is common, with an incidence of approximately 4%–20% in the adult population, and of those, 2%–4% become symptomatic from cholelithiasis annually.[1–3] It is estimated that 3%–33% of all patients with symptomatic cholelithiasis may present with concomitant CBD stones, and 4–20% will have CBD stone at the time of cholecystectomy. CBD stone usually presented with complications such as ascending cholangitis or pancreatitis, however, rarely patient may be asymptomatic presenting with only hyperbilirubinaemia with deranged liver enzymes. Extreme elevation of bilirubin levels raised concern for primary liver disease or malignancy, often leading to delays in diagnosis and extensive testing. Levels of bilirubin elevation in obstructive jaundice correlate with underlying pathology, and significant elevation (>188 μmol/L) may predict malignancy.[4]

Multiple imaging modalities available to diagnose CBD stone include abdominal ultrasound, ERCP, magnetic resonance cholangiopancreatography (MRCP), endoscopic ultrasound (EUS), and intraoperative cholangiography. Abdominal ultrasound has the advantage of immediate availability, non invasive and cost effective, however it is certainly not the most sensitive imaging modality for detecting bile duct stones with sensitivity ranging 13%–89%. It has limited visualization of intra-abdominal organs in obese patients, as well as interference with bowel gas, resulting in suboptimal visualization of intra-abdominal organs. It also offers limited visualization of the entire biliary tree, the distal CBD near the periampullary region in particular as this segment of the duct may be obscured by adjacent duodenal gas in the imaging field.[5] ERCP is the gold standard for diagnosis of CBD stone due to its high sensitivity, specificity, and accuracy. However, routine preoperative ERCP is low yield and carries associated risks given to its invasive nature.[6,7] MRCP and EUS are also highly sensitive and specific for diagnosis of CBD stone, however EUS is also invasive with risk and MRCP may not be immediately available and is slightly higher cost. Therefore, an adjunct to diagnosis of CBD stone such as biochemical parameters is paramount, in deciding further invasive or expensive imaging modality to look for presence of CBD stone in a patient at risk.

CBD stone typically follows a cholestatic pattern with mild to moderate elevation of aminotransferases. Few study has evaluated the use of biochemical marker trends to determine presence of CBD stone in a patient with known cholelithiasis. It is found that level of bilirubin at the time of presentation is suggestive of the presence of CBD stone.[8,9] The mechanism for accentuated transaminases in some patients with choledocholithiasis remains uncertain. The most plausible hypothesis is that higher biliary pressures lead to greater impairment of bile secretion and greater retention of bile acids with accompanying hepatocyte apoptosis, necrosis, or leakage of enzymes.

Utilizing serum markers in patient suspected to have CBD stone minimize the use of additional imaging while maximizing yield when imaging tests are performed. However, despite numerous studies evaluating the use of biochemical markers to assist in the diagnosis of CBDS, no consensus has been reached. Some studies found that GGT elevation was especially useful in evaluation for CBD stone, while others found that elevated alkaline phosphatase was suggestive of CBD stone. Other studies found that total bilirubin level at admission was an independent predictor of stone. Some authors concluded that biochemical markers alone were insufficient to predict stones, and suggest that a combination of radiographic studies and biochemical markers should be used to aid in decision making.[10,11]

5. CONCLUSIONS

Elevated AST/ALT and total bilirubin level can aid in diagnosis of CBD stone. The usage of biochemical parameters in adjunct to ultrasound of the biliary system in patient suspicious of CBD stone help to decide the need for more invasive or costly imaging, and avoid the unnecessary further investigation in patients without CBD stone. It also helps select patients to avoid non therapeutic ERCP which comes with associated complications.

Conflict of interest
No conflict of interest.

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