Case Report

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Laparoscopy in the management of emphysematous cholecystitis and secondary appendicitis in an 11-year-old child with insulin-dependent diabetes mellitus

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ABSTRACT

Acute abdomen in a diabetic child may cause diagnostic dilemma. Acalculous emphysematous cholecystitis (EC), although reported among critically ill or diabetic adults, is an uncommon occurrence in the children. It may complicate the presentation due to its varied manifestations; especially when associated with other concomitant intra-abdominal inflammatory pathologies. We encountered a rare concurrence of acute EC complicated with pericholecystic fluid collection and secondary appendicitis causing non-specific acute abdomen in an 11-year-old obese boy with insulin-dependent diabetes mellitus. Laparoscopy proved to be a highly useful tool in the diagnosis and treatment of this surgical dilemma.

Key words: Child, emphysematous cholecystitis, IDDM, laparoscopy, obese, secondary appendicitis

INTRODUCTION

Acalculous cholecystitis (ACC) is known to affect diabetic and critically ill adults[1-4] in their 50s or 70s. Emphysematous cholecystitis (EC) is a unique form of ACC associated with increased morbidity and mortality.[5-13] EC has rarely been reported in children, especially following torsion or trauma.[12] Acute abdomen due to concurrence of acute EC complicated by pericholecystic fluid collection, and secondary appendicitis posed diagnostic dilemma in an 11-year-old obese child with insulin-dependent diabetes mellitus (IDDM). Conventional radiological modalities failed to ascertain a definite diagnosis. Laparoscopy proved to be extremely useful in diagnosing and treating this surgical dilemma in the obese child with satisfactory outcome.

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CASE REPORT

An 11-years-old Kuwaiti boy, known case of IDDM, presented to paediatric emergency service with complaint of acute abdominal pain for 2 days; vomiting and high-grade fever for 1 day duration. There was no history of jaundice, change in bowel habit, dysuria or respiratory symptoms. He had an attack of right upper abdominal pain 3 weeks before in Kuwait which subsided on intake of antibiotics and opiod analgesics for 10 days duration.

Upon evaluation, child looked sick, dehydrated with high-grade fever (Temp. = 39°C) and tachycardia. There was no pallor, icterus or cyanosis. Chest and cardiovascular system were normal. Per abdomen examination revealed tenderness and guarding at right half of abdomen involving right hypochondrium (RHC), lumbar and right iliac fossa (RIF) with rebound tenderness in RIF; absent bowel sounds and no palpable organomegaly. Per rectally there was tender anterior bogginess.

Blood work up revealed leucocytosis (WBC=21.1x 10^3/ul, 84% segments), hyperglycemia (random blood sugar = 504 mg/dl), hyponatremia (Na=130 meq/l); and normal renal function, liver function, amylase and lipase. Plain X-ray abdomen showed radiolucent curvilinear line in RHC [Figure 1] with moderate gaseous distension of bowel (ileus). USG abdomen revealed free lumen, thickened heteroechoic GB wall (5 mm) with hyperechoic streak [Figure 2] and echogenic fluid collection around GB [Figure 3], in the right paracolic gutter, Morrison's pouch and pelvis. Aperistaltic thickened vermiform appendix was found

in the pelvis within the collection.

An impression of acute appendicitis with perforation and pericholecystic collection was made.

Patient was resuscitated with IV fluids and broad-spectrum antibiotics. Dehydration, hyponatremia and hyperglycemia were corrected and patient was taken for laparoscopy under general anaesthesia.

Diagnostic laparoscopy revealed a moderate pericholecystic fluid collection extending along lesser omentum and deep into Morrison's pouch with free fluid (light brownish yellow) in the pelvis. On laparoscopic exploration of pericholecystic collection, GB was found to be edematous with patchy brownish areas [Figure 4]. Appendix was turgid and edematous with thickened mesoappendix, serosal hyperaemia and exudative coating. Rest of the GIT was normal.

Exploration of the lesser sac revealed normal pancreas and retroduodenal area. Gall bladder and appendix were removed. Saline peritoneal lavage was given.

Postoperative course was uneventful with rapid resolution of fever, leucocytosis and abdominal symptoms. Patient regained full oral intake in 36 hrs and was discharged on 3rd postop day.

Histopathology confirmed the diagnosis of EC with impending perforation and acute appendicitis with transmural inflammation and mucosal ulceration. The bacteriology of peritoneal fluid and bile revealed *E*scherichia *coli* and *Enterococcus* sp., respectively, and both the organisms were sensitive to amoxicillin-clavulenic acid combination.

DISCUSSION

EC is a rare form of cholecystitis (constitutes < 1% of cholecystitis) due to gas-forming pathogens (including *Clostridium welchii, E. coli, Staphylococcus* or anaerobic

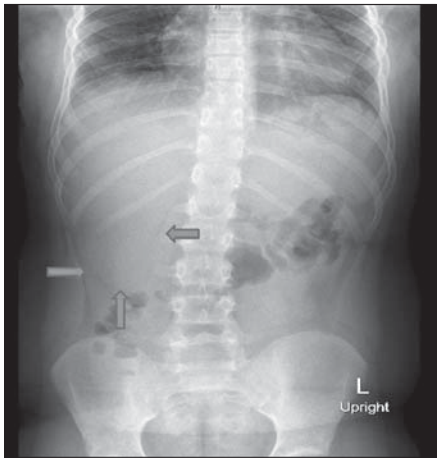

Figure 1: Plain X-ray abdomen showing radiolucent line in right hypochondrium (arrows)

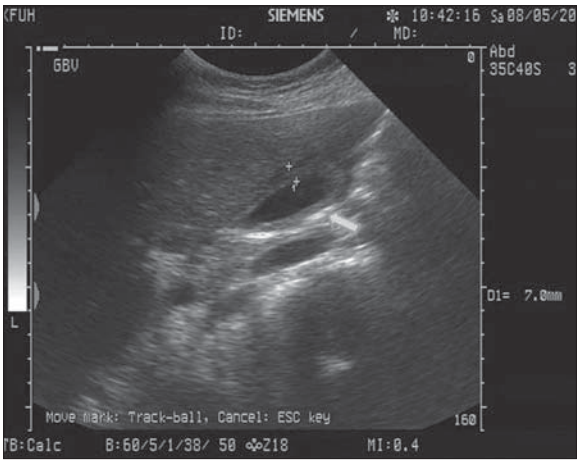

Figure 2: USG showing thickened wall of gall bladder with echogenic streaks due to air (arrow)

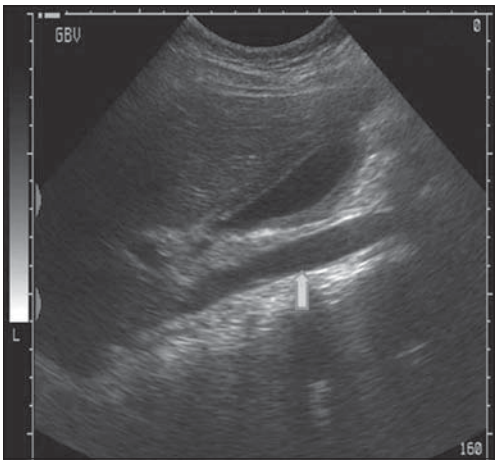

Figure 3: USG showing Type I pericholecystic collection (arrow)

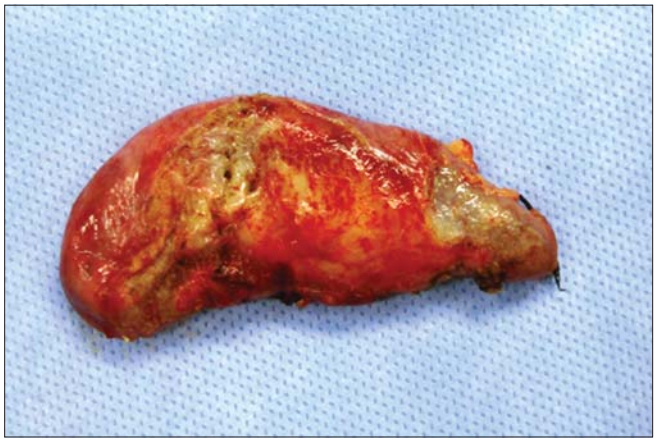

Figure 4: Gross view of gall bladder showing patchy brownish areas of ischaemia

streptococcus) invading an ischaemic gall bladder wall following thrombosis or occlusion of cystic artery.[1-5] Mostly affects the elderly men (50-70 years of age) with critical illness. Diabetes mellitus is present in 30-50% of cases, and cholelithiasis is present in up to 50%. The mortality rate is approximately 15%. Gangrenous changes occur in approximately 75% of cases, with progression to perforation in 20%.[6,7]

Clinical presentation is vague and varies from mild abdominal pain of simple acute cholecystitis to rapidly developing acute abdominal catastrophe leading to sepsis and shock.[1-7]

Diagnosis is based on clinical setting and demonstration of variable amount of gas in the wall of gall bladder or in the surrounding tissue. Rarely gas may escape into the lumen of GB manifesting as pneumobilia.[3] Plain X-ray abdomen may show a pear shaped translucency in the RHC in a supine film like in our case [Figure 2] or fine airfluid level on an erect film (intraluminal air in the GB).[4] Ultrasonography is fairly sensitive to demonstrate air in the edematous wall of GB as echogenic streaks which are positional (rise to the top with changing position) and very well differentiates from cholesterosis and calcification. Often accompanied with thin, anechoic crescent-shaped fluid collections adjacent to the gallbladder wall known as Type I collection as seen in our case [Figure 3] or have Type II fluid collections as large, round/irregular in shape and complex (thick walls, debris, septations); with indiscernable gall bladder.[10] Type I fluid collections were not typically associated with gallbladder perforation whereas type II fluid collections were nearly always associated with gallbladder wall perforation and abscess formation.

Pathophysiology of ACC including emphysematous type is a paradigm of complexity. However, in animal models, ACC has been shown to develop after administration of systemic mediators of inflammation. Small-vessel necrosis in the gallbladder serosa and muscularis has been demonstrated after activation of factor XII–dependent pathways, platelet activating factor, endotoxin or interleukin 2. This frequently is associated with gallbladder atony, which in turn predisposes patients to biliary stasis.[10,11] Biliary stasis results in more viscous bile, an increase in the concentration of the detergent bile salts, and sludge formation, which increases the bile histotoxicity to the gallbladder mucosa. Fasting, use of parenteral nutrition, use of narcotic analgesics and the postoperative state all predispose patients to biliary stasis and are commonly seen in patients with ACC.

Generalized or localized ischemia further predisposes patients to biliary stasis; it may result in gallbladder wall necrosis and perforation. Hypovolemic shock, cardiogenic shock and septic shock predispose patients to ischemia and are contributing factors. At times, ischemia is the primary cause; it may occur in the setting of small-vessel vasculitis or following therapeutic particulate embolization.

Individuals presenting with EC typically are older patients with microvascular disease and other comorbidities. Rarely EC has been reported in children due to torsion of GB, trauma and Henoch Schonlein Purpura.[12,13]

Our patient had an attack of RHC pain 3 weeks prior and had been treated with 10 days course of opioid analgesics. He did have uncontrolled IDDM as contributing factor; however, his overall general condition immediately preceding this illness was well preserved.

Although USG was showing some indicators of EC, it had actually made an impression of acute appendicitis with greater certainty and the pericholecystic and pelvic fluid collection were attributed secondary to perforated appendicitis. It was mostly retrospective confirmation of radiological findings of EC (line of lucency in plain X-ray and hyperechoic streaks in GB wall on USG) following diagnostic laparoscopy. CT might be the best technique for diagnosing EC because it shows the exact location of air, whether in the gallbladder wall, in the gallbladder lumen, or throughout the bile duct, but it is irrational to perform CT for all patients with vague abdominal symptoms,[5] particularly in children, due to the issue of radiation exposure which demands rationalization of the imaging modalities.

We, as matter of policy, had taken a decision to perform laparoscopic exploration in this child for therapeutic intent which eventually proved to be extremely useful in diagnosing two concomitant pathologies at two ends of the abdomen (EC and appendicitis) with benefits of therapeutic intervention. Nevertheless, the cosmetic outcome and rapidity of regaining normalcy were the obvious advantages of laparoscopic intervention of this potentially morbid pathology in an obese and poorly controlled diabetic child.

Although rarely described, secondary appendicitis is known to develop in conjunction with other intraabdominal inflammatory pathologies when appendix lies adjacent to the site of inflammation or

inflammatory collection.[14] However, there has been no mention of concurrence of EC and secondary appendicitis in children so far in the literature. Concomitant pathologies caused diagnostic dilemma; however, laparoscopy proved to be a safe and effective tool in diagnosis as well as treatment.

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