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Gallstone Ileus: A Rare and Often Disregarded Cause of Intestinal Obstruction: Case Report with Literature Review

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Gallstone ileus is very rare complication of cholelithiasis. As it is usually a disease of the elderly, the morbidity and mortality associated with it is high due to the presence of co-morbidities. This is additionally made worse by the delay encountered before reaching diagnosis which often demands a high index of suspicion. In Ethiopia, a country where intestinal obstruction is seen to be one of the common presentations in the emergency surgical department, gall stone has never been seen to be reported as a cause. Thus we present this rare case and discuss the mode of presentation, diagnostic difficulties and differing treatment options. We report the case of an 80-year old lady who presented with signs and symptoms of small intestinal obstruction. Upon exploration we were astonished to find the cause of the obstruction to be a gallstone which was removed by simple ileostomy. Post-operative course was uneventful.

Key words: gallstone ileus, intestinal obstruction, gallstone

Introduction

Gallstone ileus (GI) was first described by Bartolin in 1654 [1]. It accounts for 1-4% of all cases of small bowel obstruction and 25% of all cases of small bowel obstruction in those greater than 65 years of age[2, 3]. Even though GI has been reported in the age groups between 13-91years, the peak incidence is between65-75 years [2]. The problem starts when the stone passes through a cholecystoenteric fistula between gall bladder and duodenum (75%), gall bladder and colon (10-20%) or other types (15%), and is propelled by peristaltic waves to be expelled per rectum [3].But sometimes if the stone is large enough, at least 2-2.5cm in diameter, it can get stuck and cause intestinal obstruction [2]. At times, multiple stones may be found in 30-40% of cases [2].Usual sites of impaction are the ileocecal valve and terminal ileum [2, 3]. Diagnosis is often difficult and in about 50% of cases it is only made at laparotomy [2, 3].

Case Report

An 80-year old lady presented to our emergency surgical department with complaints of vomiting and abdominal cramp of one month duration. The vomiting was four to five times per day and initially consisted of ingested food which later became bilious. The abdominal cramp was gradually increasing in severity and was later accompanied by abdominal distension and failure to pass feces and flatus. Before her presentation to our hospital, she was admitted in a local hospital in her home town. The diagnosis made initially was cholelithiasis with chronic cholecystitis and later on intestinal obstruction was entertained. Since her general condition was weak she was referred to us for better evaluation and management. She had occasional bloating and fat intolerance since one

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year back and gave no past history of abdominal surgery. The patient didn't smoke and never consumed alcohol.

Physical examination findings were; blood pressure 80/50mmgh, pulse rate 120/min, respiratory rate 28/min and temperature 37C^{o}. Herconjuctivae were pink and had dry tongue and buccal mucosa. Abdominal examination showed a distended abdomen which was tender on both sides, with visible peristaltic waves and hyperactive bowel sounds. Hernial sites were normal and digital rectal examination revealed an empty rectum and no mass. Routine laboratory investigations done were; complete blood count: - WBC 7600/mm^3 and Hemoglobin 13.1g/dl. Urinalysis was normal. Serum electrolytes: potassium 2.8mmol/dl, sodium and chloride were normal. Renal function tests were in the normal range. Plain abdominal film showed distended small bowel loops with multiple air fluid levels.

She was resuscitated with normal saline and Ringer's lactate till blood pressure normalized and urine output was adequate. Potassium chloride was added to the intravenous fluids and nasogastric tube inserted and 500ml of bilious fluid recovered. After stabilizing the patient explorative laparotomy was decided and then performed. Intra-operative finding was, distended small bowel loops up to 20cm proximal to ileocecal valve where a hard mass was felt obstructing the lumen. Ileotomy was done along the longitudinal axis of the gut on the antimesenteric border. To our surprise, a 2.5cm stone was recovered. The defect was closed transversely. The rest of the abdomen was checked. Severe dense adhesions in the area of the gall bladder, duodenum and hepatic flexure of transverse colon found. Due to the frail general condition of the patient further procedure was deferred. Post-operative course was uneventful and patient was discharged on the 5th post- operative day. Follow up has continued till one year after the surgery and she reported no major complaints.

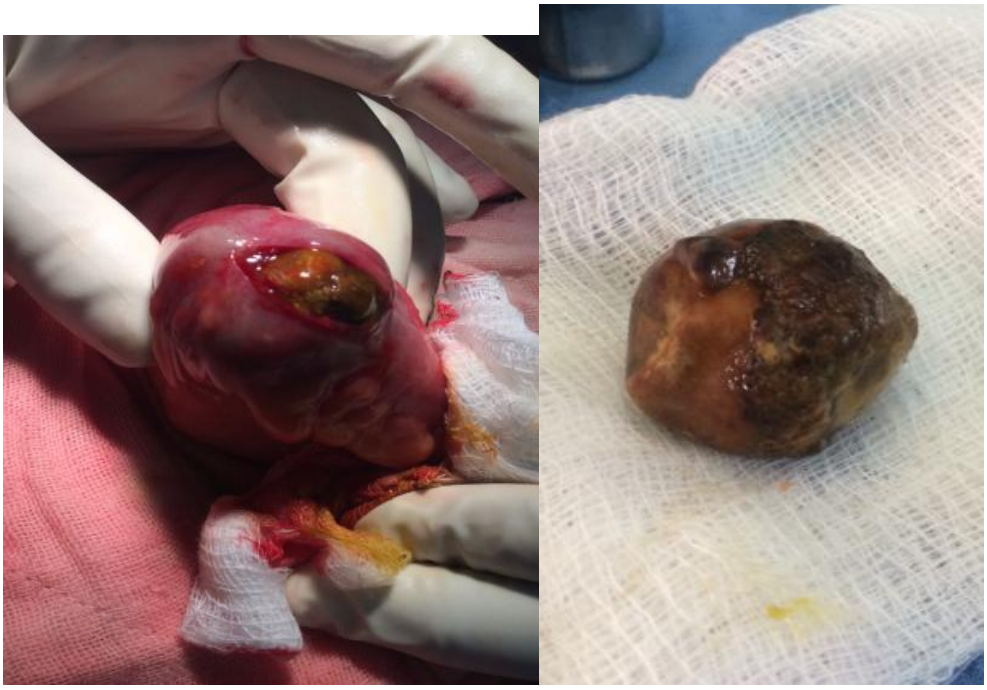

Figure 1.

Discussion

Cholelithiasisn is a common problem seen world-wide[4].The prevalence in Western Europe ranges between 5.9% and 21.9%; Norway, Sweden and Germany being the leaders.[4]Even though the most common symptom of gallstones is biliary colic, others may manifest due to complications like acute cholecystitis, acute pancreatitis, choledocholithiasis with or without cholangitis and gangrene of gall bladder[3]. Rare complications of gallstones include Mirizzi syndrome, cholecystocholedochal fistula and gallstone ileus [3].GI is a mechanical intestinal obstruction due to impaction of one or more large stones within the gastro intestinal tract [2].Pericholecystic inflammation due to acute cholecystitis leads to adhesion with the surrounding structures and the stone in the gall bladder causes pressure necrosis on the wall culminating in the formation of a fistula [5]. Less than 3% of cases of cholelithiasis complicate with fistula formation[2, 4] and less than 0.5% of patients with gallstones develop GI [6]. The most common site of impaction of gallstones is the terminal ileum [7].Jejunum, stomach, duodenum and colon are rare sites of impaction [7]. Bouveret syndrome is when the stone passes into the duodenum and causes gastric outlet obstruction [7].

Symptoms of GI are often non-specific like nausea, vomiting, abdominal pain and distension[5, 6]. More than a third of patients also have no previous biliary symptoms.[5] Plain abdominal X-ray may show the classic Rigler's triad; pneumobilia, small bowel obstruction, and ectoptic gallstones in less than 50% of cases [7, 8]. Moreover most gallstones are not calcified enough to be seen on x-rays, limiting the diagnostic accuracy[8]. Oral contrast radiographic examinations can be helpful to demonstrate the stone, site of obstruction, as well as the fistula especially when the obstruction is proximal as in Bouveretsyndrome [7]. Ultrasonography is used to confirm the presence of gallstones and sometimes may discover the fistula [7, 8].But its use is compromised by factors like anatomic alterations because of distension due to intestinal obstruction, collapse of gall bladder or presence of air in the gallbladder[7, 8]. The combined use of plain abdominal radiography and ultrasound can increase the diagnostic accuracy to as high as 74% according to Ripollesetal[8]. The investigative modality of choice for the diagnosis of GI is CT which has a high sensitivity and specificity, 93% and 100% respectively[9, 10].

In Ethiopia, small intestinal obstruction is seen to be one of the commonest illnesses patients present with to the emergency surgical department[11, 12]. Most frequently cited causes are volvulus and adhesions[12,13].GI has not been mentioned even as the "miscellaneous" causes [12, 13], which indicates the rarity of this entity. The main modality of treatment for GI is surgery. But great emphasis should be given to managing fluid and electrolyte disturbances as well as dealing with co-morbid conditions pre-operatively [2]. The choice between which surgical option is preferred is controversial [2, 3]. The first one is ileotomy alone with extraction of the stone, which may or may not be followed by cholecystectomy and fistula closure at a later date (two-stage) [2, 8].The second option is a one-stage procedure that includes enterotomy, stone extraction, cholecystectomy and fistula closure at one-go[2, 8].The trend has been initially in favor of ileotomy alone with extraction of the offending stone as it has been associated with lower morbidity and

mortality [8]. The biliary- enteric fistula is also seen to close spontaneously in up to 50% of such case [8].But less than 5 % of patients may develop recurrent GI [8].Ongoing biliary symptoms requiring surgery are seen in 10% of patients[2].Nowadays there are reports advocating the safety of a one-stage surgery [6, 14]. But this has to be backed up by a good pre-operative general condition of the patient and absence of dense inflammatory reaction at the site of fistula[14] unlike the situation in our patient. Other modalities of treatment that may have a role in the management of patients with gallstone ileus are laparoscopy and lithotripsy[15].

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Mirizzi Syndrome: A Case Report.

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Mirizzi syndrome is a rare disorder that usually presents with obstructive jaundice due to the presence of a gallstone impacted in the cystic duct, neck or infundibulum of the gallbladder. It is a complication of gallstone disease. The extrinsic compression of the common hepatic duct or the resultant inflammatory process due to the impacted stone results in the obstructive jaundice clinical picture. We report a case of a female patient who presented with this rare complication of gallstone disease. She had a 20mm stone that was impacted in the gallbladder neck and had extensive adhesions around the gallbladder. A retrograde cholecystectomy and Rouxe en Y hepaticojejunostomy was done.

Key words: obstructive jaundice, gallstone disease, Mirizzi syndrome

Introduction

The first case of benign extrinsic biliary obstruction due to a stone in the gallbladder was published by Kehr in 1905[1], although the condition only became known as Mirizzi syndrome in 1948 after an Argentinean surgeon Mirizzi reanalyzed and classified this condition[2]. Mirizzi syndrome is a rare complication of symptomatic gallstone disease with an incidence of less than 1% a year in the Western developed countries[3]. It appears that Mirizzi syndrome is a more common condition in underdeveloped countries; particularly Latin America where the reported incidence ranges from 4, 7% to 5, 7%[4, 5]. Obstructive jaundice and cholangitis are the common presentation features of this condition[6]. We report a case of a female patient with Mirizzi syndrome whom we successfully managed.

Case report

We present a case of a 40-year-old female patient, who had a month's history of right upper quadrant pain, one week history of yellowing eyes associated with generalized body itchiness, passing of dark colored urine and pale colored stool. The pain was of gradual onset and it would last for six to eight hours before subsiding. She did not have a fever. She did not have any co morbidities. On examination she was stable, jaundiced, normotensive with a blood pressure of 117/74 mm Hg and apyrexial with a temp of 36.4^0C. She had a positive Murphy's sign and the gallbladder was not palpable. She had pale stool on digital examination.

An abdominal ultrasound showed an elongated gallbladder with irregular mucosal thickening and a large 18mm stone at the cystic duct. There was significant inflammation at the porta hepatis. There was mild dilatation of the intrahepatic ducts

with the common bile duct and pancreatic duct being normal (Figure 1). Blood workup showed serum total bilirubin 235mg/dl, direct bilirubin 135mg/dl, alkaline phosphatase 325 international units per litre (IU/L), aspartate transferase 80 IU/L, and alkaline transferase 97IU/L. Full blood count and urea and electrolytes were normal. International normalized ratio was 0.99. Based on the history, physical examination, imaging and biochemical investigations a diagnosis of obstructive jaundice secondary to choledocholithiasis was made. The patient could not afford a computed tomography, magnetic resonance cholangiogram or endoscopic retrograde cholangiopancreaticograhy. She was then offered surgical exploration.

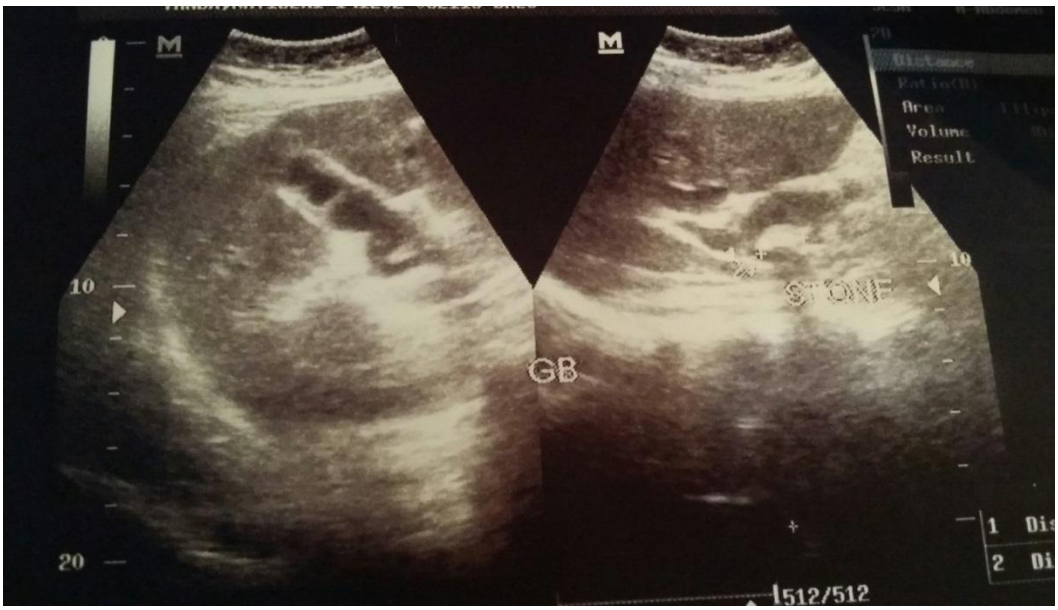

Figure 1. Abdominal Ultrasound showing a Gallbladder with Thick Walls and a Stone.

At laparotomy extensive adhesions were noted around the gallbladder.The distorted anatomy made it difficult to appreciate the cystic duct. A 20mm impacted stone was noted in the neck ofthe gallbladder causing external compression of the common hepatic duct. The gallbladder was opened from the fundus to the neck and the stone removed and retrograde cholecystectomy done. On removal of the stone bile flow was noted and there was a fistula communicating between the common hepatic duct and gallbladder, which made it a Type II (Beltran and Csendes classification) Mirizzi syndrome. A Rouxeen Y side to side hepaticojejunostomy was done. The patient had an uneventful post-operative recovery and was discharged a week after the operation. She was reviewed at 2 weeks with the jaundice having cleared with normalization of the liver function parameters and no complaints.

Discussion

The mean age of presentation of patients with Mirizzi syndrome varies from 53 to 70 years and are female in 70% of all cases[7, 8]. Our patient was a female at the age of 40years. Mirizzi syndrome can be due to an acute or chronic inflammatory disorder secondary to a single large gallstone or multiple small gallstones impacted in the Hartmann's pouch or in the gallbladder infundibulum and cystic duct[3,7,9, 10]. A stone of

20mm size was found in this case.Predisposing factors for the development of Mirizzi syndrome include; a long cystic duct, parallel to the bile duct and a low insertion of the cystic duct into the bile duct[6].

Preoperative diagnosis of Mirizzi syndrome is difficult and can be made in 8% to 62.5% of patients[6]. There is a 17% incidence of bile duct injuries in patients operated on with Mirizzi syndrome[6]. Ultrasound scan (USS), computed tomography, magnetic resonance and endoscopic retrograde cholangiopancreatography are diagnostic modalities that can be used in Mirizzi syndrome. Our patient could only afford an abdominal USS which suggested Mirizzi as a possible deferential diagnosis. The reported diagnostic accuracy of USS in Mirizzi syndrome is 29%, with a reported sensitivity varying from 8.3% to 27%[6]. Surgery is the treatment of Mirizzi syndrome. This could be either open or laparoscopic. Despite the feasibility of laparoscopic surgery in Mirizzi syndrome, forType II and aboveit is considered controversial and technically challenging, placing the patient at an unnecessary risk of bile duct injuries.As a result laparoscopic cholecystectomy for Mirizzi syndrome cannot be recommended as a standard procedure[6]. For open surgery, the surgical procedure is dependent on the type of Mirizzi syndrome. Our patient had a Type II Mirizzi syndrome. She had a cholecystectomy and a biliary enteric anastomosis (Rouxe en Y side to side hepaticojejunostomy) with good results.

On discharge the patient was showing clinical resolution of the obstructive jaundice. On review at 2 weeks, post operatively, the patient had no jaundice with normal liver function test. It has been reported that 6% to 28% of patients with preoperative diagnosis of Mirizzi syndrome actually had gallbladder cancer[9], hence during the operation, frozen section biopsy should be done in all patients with Mirizzi syndrome to definitely rule out gallbladder cancer[1]. Although there no clinical features to distinguish Mirizzi syndrome from gallbladder cancer, patients with gallbladder cancer are a decade older than patients with Mirizzi syndrome alone[6].

Conclusion

Mirizzi syndrome is a rare complication of gallstone disease. The diagnosis of Mirizzi syndrome preoperatively is very difficult. The presence of dense adhesions in the area of the Calot's triangle should raise a suspicion of intraoperative diagnosis of Mirizzi syndrome. Laparoscopic approach is not advisable. Definitive management of an associated fistula depends on the Csendes classification of the type of fistula. A Roux-en-Y hepaticojejunostomy, is a feasible surgical option for Mirizzi syndrome.

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