

Pancreatitis Secondary to Bile Duct Cyst in a 36-year-old Pregnant Woman: Case Report

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Abstract

Pancreatitis in pregnancy has a prevalence of 1.5/1500-4500 cases, constituting one of the most common acute abdomen wedges, with biliary origin in 70% of cases, triglycerides in 20% and other causes in the remaining 10%, including choledochal cyst (CC) as a rare cause with three previous reports in literature, which may have a fatal outcome with fetal loss in some cases.

We report the case of a 25-year-old patient with 30.4 weeks of gestation (WOG) that arrived to emergency room with right upper quadrant and epigastric pain for the last 8 hours, associated with nausea and vomiting. No pathological background referred. At physical examination with jaundice, gravidic abdomen for 30.4 WOG pregnant, fetal movements presents, Murphy (+) and epigastric pain on deep palpation. Laboratories report total bilirubin (TB) 3.9 mg/dl and direct bilirubin (DB) 3.69 mg/dl Alkaline phosphatase (AP) 2038 IU/L Amylase 280 IU/L Lipase 1938 IU/L. Pancreatitis is confirmed and abdominal ultrasound (US) is requested to determine biliar origin. USG reports gallbladder of 9×4 cm, thin walls without filling defects, dilated intrahepatic bile duct and common bile duct cyst. Cholangiopancreatography Resonance Imaging (CPMR) concludes Todani I choledochal cyst of 17×9 cm, with displacement of duodenum, colon and páncreas. Due to gestation ongoing appropriate medical management with fluids and analgesics was started until remission of pancreatitis 72 hours later. After delivery at 34 WOG, cholecystectomy was performed with hepático-jejunum Roux-Y anastomosis successfully. Histopathologic analysis reports inespecific inflammation without displasia or metaplasia. At four months follow-up patient is asymptomatic.

Pancreatitis in pregnancy is a common cause of acute abdomen, rarely associated with choledochal cysts as the cause. Surgical resolution once pregnancy is over must be done as soon as possible by the high risk of adenocarcinoma degeneration and recurrent pancreatitis.

Keywords: Pancreatitis; Pregnancy; Bile duct cyst; Cholangitis

Abbreviations

WOG: Weeks of Gestation; CC: Choledochal Cyst; CT: Computed Tomography; TB: Total Bilirubin; AP: Alkaline Phosphatase; US: Ultrasound

Introduction

Acute pancreatitis is a relatively common disease in pregnant women (1/1500-4500 pregnancies), 70% secondary to gallstones and hypertriglyceridemia in 20%.

Other less common causes such as hyperparathyroidism, autoimmunity or toxics can trigger acute pancreatitis in pregnant women with fetal loss in up to 4.7% [1]. Choledochal cyst (CC) are rare congenital intra or extrahepatic biliary tract dilatations. The incidence in North America is 1: 150,000 live births. They may be responsible for complications such as biliary stasis, stone formation, cholangitis, pancreatitis and malignant degeneration [2]. The association of

pancreatitis secondary to choledochal cyst in pregnant woman has been reported only in other three cases [3-5].

Case Presentation

We report the case of a 25-year-old patient with 30.4 weeks of gestation (WOG) of her first pregnancy, without pathological background. She arrived to emergency room with right upper quadrant and epigastric pain for the last 8 hours, associated with nausea, vomiting and respiratory distress. The last 24 hours she noticed jaundice. At physical exam with 95 heartbeats per minute, 23 breaths per minute, arterial tension 130/80 mmHg, jaundice, globular abdomen at expense of pregnant uterus, absent peristalsis, Murphy sign and epigastric pain on deep palpation. Fetal wellbeing was confirmed by Obstetric Ultrasonography (US). Laboratories report total bilirubin 3.9 mg/dl and direct bilirubin 3.69 mg/dl alkaline phosphatase 2038 IU/L Amylase 280 IU/L Lipase 1938 IU/L. Pancreatitis is confirmed and abdominal US is requested to determine the probable biliar origin, reporting: gallbladder of 9×4 cm, thin walls without filling defects. Dilated intrahepatic bile duct and common bile

duct with cystic dilatation of 17×9 cm. Cholangiopancreatography Resonance Imaging (MRCP) is performed and reports Todani 1 choledochal cyst with displacement of adjacent structures, predominantly pancreas (Figures 1 and 2).



Figure 1: CPMR reconstruction showing Todani I CC.

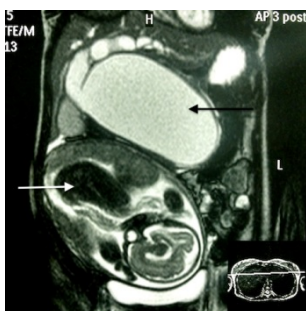


Figure 2: CPMR with CC (Black arrow) with displacement of adjacent structures and gravidic uterus (White arrow).



Figure 3: CC (Black arrow) with displacement of transverse colon (White arrow) and duodenum.

Ongoing pregnancy does not make it a candidate for cyst excision and biliodigestive bypass to the high risk of complications associated with pregnancy in the third trimester. For this reason medical management with fasting, fluids and paracetamol was established. The patient had adequate response to medical treatment, with pancreatitis referral within 72 hours. It is discharged pending resolution of pregnancy. After delivery at 34 WOG without negative effects or cesarean requirement, abdominal computed tomography (CT) scan is performed to anatomical delineation and surgical planning. Two months later cholecystectomy and total cyst excision is performed with

hepaticojejunal Roux-Y anastomosis (Figures 3-5), without complications and with uneventful postoperative course. Histopathology report Todani 1 cyst and inespecific inflammation. At four months follow-up patient is asymptomatic.

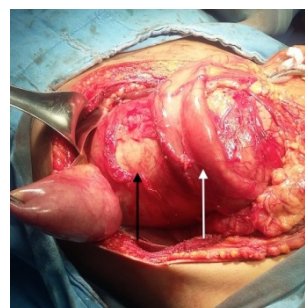


Figure 4: CC after dissection (Black arrow) of transverse colon.



Figure 5: Resected gallbladder (White arrow) and CC (Black arrow).

Discussion

Clinical evaluation of pregnant patients with acute abdominal pain is confounded by physiologic and anatomic changes related to pregnancy, representing a diagnostic and therapeutic challenge. Mild leukocytosis, anemia, and elevated alkaline phosphatase are considered normal during pregnancy [6]. Acute pancreatitis complicates approximately 1 of every 3300 pregnancies, most commonly occurring during the third trimester and postpartum; Pregnancy itself induces symptoms secondary to hormonal effects on biliary function, compression by the gravid uterus, and increased intra-abdominal pressure. During pregnancy CC are atypical and represent an additional challenge, including fetal cyst-related risk from complications [7]. The incidence of choledochal cysts is 1 in 150,000 live births, of which 20% is detected in adulthood, affecting women with a ratio of 4:11 [8]. CC is an expansion of the intrahepatic or extrahepatic biliary ducts, classified by Todani into five types, which can lead to complications like biliary stasis, stone formation, cholangitis, pancreatitis and malignant degeneration [1]. There are many theories about its origin, but the most accepted is an abnormal biliopancreatic duct connection, with retrograde flow, inflammation and pancreatic enzymes injury favoring cystic degeneration [9]. Patients usually present with pancreatic or biliary symptoms rather than cholangitis as seen in children [10]. US are used to detect cholelithiasis and bile duct dilatation but can't optimally evaluate the distal common bile duct and pancreas. When biliary obstruction is clinically suspected, MRCP can display the biliary anatomy and detect

small stones in the common bile duct with a high sensitivity and specificity. Rare causes for biliary obstruction, such as Mirizzi syndrome, CC or intrahepatic biliary stones can also be successfully diagnosed using MRCP [11]. Once the diagnosis is established, patients should be referred to specialized centers for treatment, bearing in mind maternal and fetal well-being, as well as the likelihood of cyst-related complications. The reported incidence of malignancy in choledochal cyst is age related, reported in 14.3% above the age of 20 [12]. For this reason the treatment consist in complete excision and Roux-en-Y hepaticojejunostomy anastomosis [13,14]. In pregnancy, however, a more conservative approach may have to be adopted, treating pancreatitis and planning surgery until the second trimester or after delivery, when the risk is lower. Care Checklist 2013.

Conclusion

Pancreatitis is a serious condition that occurs frequently in pregnant women and may represent a diagnostic and therapeutic challenge by physiological changes as well as the limitations in therapeutic resources due to ongoing pregnancy. For this reason a multidisciplinary approach should be established to ensure the best outcome for the fetus and mother. The presentation of choledochal cyst in adults and pregnancy is rare, so medical team need to know the best approach to provide optimal treatment, taking into account the stage of pregnancy and disease progression. Surgical treatment need to be performed as soon as possible without compromising maternal and fetal well-being by the risk of complications associated with choledochal cyst as repetitive pancreatitis and malignant degeneration. Once pancreatitis resolved, surgical resection must be performed. It should be performed in the second trimester or wait for pregnancy resolution depending on the patient status.

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