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CLINICAL CASE

Choledochal cyst during pregnancy. Report of 3 cases and a literature review☆

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KEYWORDS
Choledochal cyst;
Pregnancy;
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Abstract
Background: Choledochal cysts are rare. They usually present during childhood in women, but it can also be seen during pregnancy. Clinical signs and symptoms are obscured during this time, thus it can complicate the diagnosis and represent a life threatening complication for both the mother and the child.
Objective: To communicate the case of 3 pregnant patients with choledochal cyst.
Clinical cases: Three pregnant women in which choledochal cyst were diagnosed. Two developed signs of cholangitis. The *first* one underwent a hepatic-jejunostomy, but had an abortion and died on postoperative day 10. The *second* one had a preterm caesarean operation due to foetal distress and underwent a hepatic-jejunostomy 4 weeks later; during her recovery she had a gastric perforation and died of septic complications. The *third* one did not develop cholangitis or jaundice. She had an uneventful pregnancy and had a hepatic-jejunostomy 4 weeks later with good results.
Conclusions: Management of choledochal cysts during pregnancy is related to the presence of cholangitis. When they do not respond to medical treatment, decompression of the biliary tree is indicated. Definitive treatment should be performed after resolution of the pregnancy.

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PALABRAS CLAVE
Quiste de colédoco;
Embarazo;
Complicaciones quirúrgicas

Quiste de colédoco y embarazo. Reporte de 3 casos y revisión de la bibliografía

Resumen
Antecedentes: Los quistes de colédoco son una enfermedad rara que afecta principalmente a mujeres en la infancia, pero que pueden presentarse durante el embarazo. Sus síntomas y signos están opacados por los cambios en esta fase, por lo que el diagnóstico puede ser tardío y sus complicaciones poner en peligro tanto a la madre como al producto.
Objetivo: Presentar 3 casos de quiste de colédoco en embarazadas, tratadas en un hospital de tercer nivel.
Casos clínicos: Presentamos 3 pacientes con diagnóstico de quiste de colédoco durante el embarazo. Dos desarrollaron datos de colangitis; la *primera* fue intervenida quirúrgicamente con anastomosis hepaticoyeyunal, pero abortó y falleció al décimo día del postoperatorio. La *segunda* tuvo una cesárea pretérmino por sufrimiento fetal; 4 semanas después fue intervenida con anastomosis hepaticoyeyunal, pero tuvo una perforación gástrica y falleció por complicaciones sépticas. La *tercera* no desarrolló colangitis, tuvo un embarazo sin complicaciones, se le realizó anastomosis hepaticoyeyunal 4 semanas después, con buenos resultados.
Conclusiones: El tratamiento de los quistes de colédoco durante el embarazo está relacionado con la presencia de colangitis. Cuando no responden al tratamiento médico, la descompresión de la vía biliar está indicada. El tratamiento definitivo debe realizarse una vez resuelto el embarazo.

Background

According to Angel and Nassar, et al. choledochal cysts were described by Vater in 1723.[1,2] Their aetiology is unknown, based on that published by Wu[3] and Gmijović et al.[4] in 1936 Yotsuyanagi posed the theory that they arise from inequality in the vacuolisation of the biliary tract in early embryonic life. Currently there are 2 theories; the *first* postulates that the distension or cyst forms secondary to congenital stenosis of the biliary tract; the *second*, proposed by Babbitt,[3,5] in 1969, is that they are the result of abnormal union between the bile duct and the pancreatic duct (greater than 15 mm), with pancreatic reflux, increasing the intraluminal pressure of the bile duct and its distension. The latter theory is the most widely accepted.

Martínez-Ordaz and Niño-Solís[6] contend that in 1959, Alonso-Lej proposed a classification that was modified by Todani in 1977. The symptoms are non-specific, principally jaundice without apparent cause during infancy and vague abdominal pain, and are even more evident in pregnant women. Pregnancy significantly alters the gallbladder, due to hormonal changes of oestrogen and progesterone levels. Oestrogens have been demonstrated to inhibit the motility of Oddi's sphincter. The gravid uterus can also accentuate or aggravate symptoms due to distal obstruction of the bile duct by compression.[2]

Objective

To present 3 cases of choledochal cysts in pregnant women, treated in a third level hospital and to compare them with cases reported in medical literature.

Clinical cases

Case 1

A 22 year-old primiparous woman, with an interuterine pregnancy of 9 weeks' gestation. She started to suffer abdominal pain that was treated with analgesics and antispasmodics. Then she experienced nausea and later vomiting, jaundice and choluria. On physical examination she was found to be conscious, cooperative and calm, adequately hydrated, with jaundiced skin and mucosa + cardiopulmonary examination showed no compromise; there was a palpable tumour in the hypochondrium and right flank of her abdomen, which was firm, mobile, painful, with no signs of peritoneal irritation. The laboratory results showed: direct bilirubin 3.54 mg/dl, indirect 0.22 mg/dl, total 3.76 mg/dl; with raised transaminases (alanine aminotransferase172 U/l, aspartate aminotransferase 89 U/l). An ultrasound was performed which reported an abdominal tumour dependent on the liver. There were no alterations obstetrically. Magnetic cholangioresonance reported a cystic lesion of 20 cm in cephalocaudal and 11 cm anteroposterior diameter, homogeneous, dependent on the extrahepatic bile duct (Fig. 1). The patient started with signs of cholangitis, which did not improve with medical treatment, and therefore underwent giant choledochal cyst resection, cholecystectomy and hepaticojejunal anastomosis. She was reoperated twice for a haemoperitoneum due to haemorrhage from the surgical site. As this developed absence of foetal heart rate presented, and therefore uterine curettage was performed. The outcome was poor, and the patient died due to multiple organ failure on the tenth day of the post-operative period.