

## HYDATID CYST LOCATED IN THE MAIN BILE DUCT-A RARE DIFFERENTIAL DIAGNOSIS OF OBSTRUCTIVE JAUNDICE IN PREGNANCY

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**ABSTRACT:**

*HEPATIC DISORDERS IN PREGNANCY MAY BE UNIQUE TO PREGNANCY, PREEXISTING, COINCIDENT WITH PREGNANCY AND POSSIBLY EXACERBATED BY THIS CONDITION. MOST COMMON NONOBSTETRIC CAUSES INCLUDE OBSTRUCTION BY GALLSTONES AND VIRAL HEPATITIS. TUMORS OR PARASITES ARE LESS FREQUENT. A DIFFERENTIAL DIAGNOSE MUST ALWAYS INCLUDE HYDATID DISEASE ESPECIALLY IN ENDEMIC COUNTRIES. PRIMARY EXTRAHEPATIC HYDATID CYSTS ARE RARE. WE PRESENT THE CASE OF A 33 WEEKS PREGNANT PATIENT WITH OBSTRUCTIVE JAUNDICE DUE TO A HYDATID CYST LOCATED IN THE MAIN BILE DUCT, WHICH IS A VERY UNUSUAL SITUATION.*

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**KEYWORDS:** OBSTRUCTIVE JAUNDICE, PREGNANCY, HYDATID CYST**INTRODUCTION:**

Jaundice during pregnancy is caused by a broad spectrum of pathologic conditions, some of them with no direct relation to gestation itself. The yellowish discoloration of the skin and mucous membranes caused by high bilirubin levels is visible when bilirubin exceeds 3 mg/dl.

In the general population, jaundice is classified as follows, according to the mechanism and level of production: pre-hepatic (hemolytic), hepatocellular and post-hepatic (cholestatic).

The main medical conditions representing possible causes of jaundice during pregnancy are (Houston *et al.* 2000): acute fatty liver of pregnancy, viral hepatitis, choledocholithiasis, HELLP syndrome, idiopathic cholestasis of pregnancy, severe preeclampsia.

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In the general population, post-hepatic jaundice is caused by a mechanic obstruction of the biliary system. The most common causes are gallstones, tumors of the pancreas and parasites.

Hydatid disease is an endemic zoonotic infection produced by larvae belonging to the genus *Echinococcus*. Hydatid cysts develop mostly inside the liver, but they can occur anywhere in the body. The parasite can travel through portal circulation, lymphatic system, biliary tract and also, by dissemination of daughter cysts in the peritoneal cavity or other cavities.

The occurrence of primary hydatid cysts in the gallbladder is extremely rare - only 6 cases reported in the English medical literature in 2010 (Krasniqi et al., 2010). The occurrence of primary hydatid cysts in the main biliary duct is extremely rare, and also, finding such pathologic condition during pregnancy makes the differential diagnosis for obstructive jaundice more challenging.

### **CASE REPORT**

We present the case of a 29 year-old woman, gravida 2, para 2, who at 33 weeks of gestation was admitted, in September 2014, to local district hospital for jaundice of the skin and mucous membranes lasting for 3 days. The abdominal ultrasound examination performed at the local district hospital suggested a large tumor or large gallstone located in the biliary tract and the patient was transferred to the Bucharest Emergency University Hospital.

At the admission, the patient had jaundice of the skin and mucous membranes, pain in the epigastrium, constipation for 3 days, dark urine, normal blood pressure and no other clinical symptoms. There was nothing significant to extract from her medical history and the possibility of viral hepatitis was excluded by rapid serologic testing. The patient was not taking any medication and had not ingested any toxins before the symptoms appeared.

The patient did not accuse any uterine contractions or any alarming symptoms related directly to pregnancy. Therefore, she was evaluated by the General Surgery Department, the Gastroenterology Department and another abdominal ultrasound was performed.

The General Surgery Department reported: yellow discolouration of the skin and sclera. Abdominal moderate distension and tenderness in the right upper quadrant. A mass was palpated in the right upper quadrant. Darkened urine and pale stools.

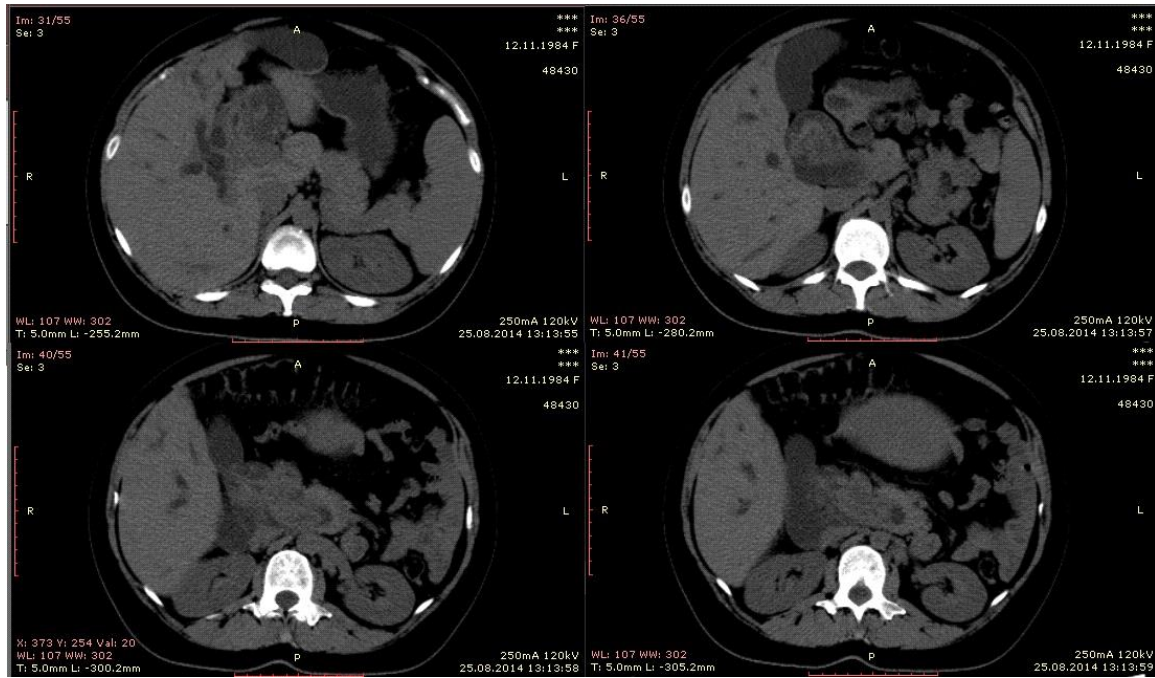
The abdominal ultrasound examination described: Distended Wirsung duct (up to 3 mm), distended intrahepatic bile ducts (up to 13 mm) and main bile duct (up to 16 mm), without images of stones or masses.

Laboratory investigations show leucocytosis with neutrophilia (Leu# = 13.130/mm<sup>3</sup>, Neu% = 90.5%), elevated levels of alanine aminotransferase (ALT = 75 U/L), aspartate aminotransferase (AST = 79 U/L) and gamma-glutamyl transferase (GGT = 141 U/L), total bilirubin and its fractions (total bilirubin = 8.01 mg/dL, direct bilirubin = 6.36 mg/dL), hypercholesterolemia (total cholesterol = 295 mg/dL) and hypertriglyceridemia (triglycerides = 476 mg/dL), without coagulopathies.

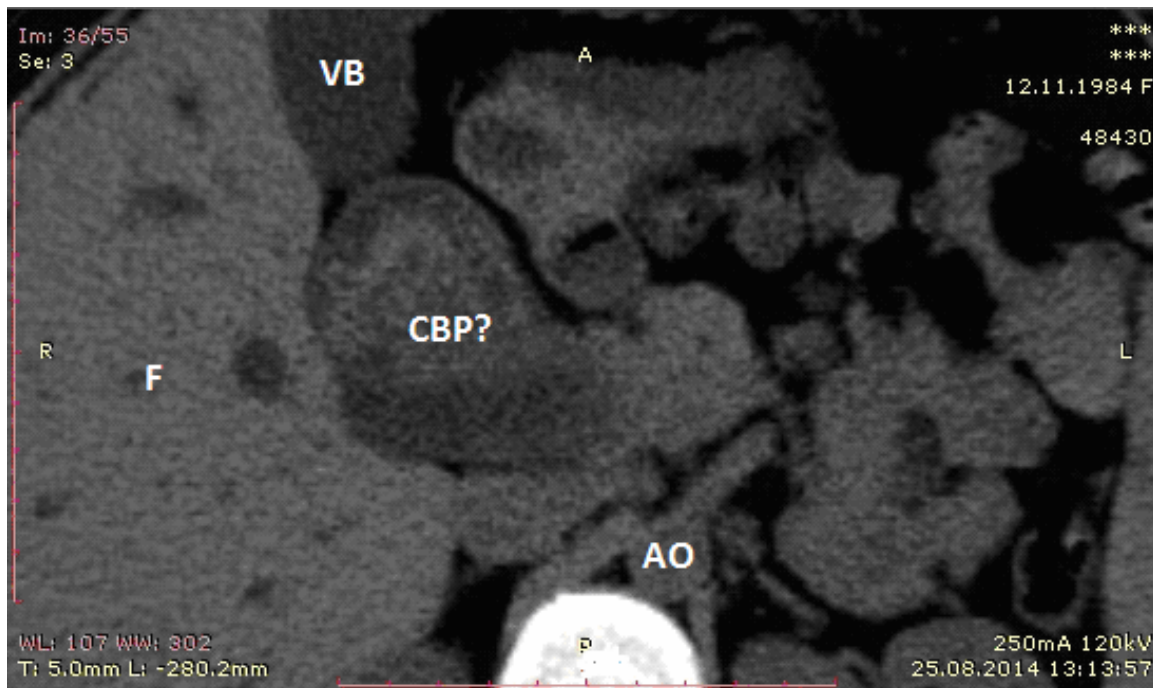
The patient was transferred to the Gastroenterology Department of Colentina Hospital for further investigations. The ultrasound examinations suggested a large dilated gallbladder, 9/3.4cm, double splitted, dilated intrahepatic biliary duct, main biliary duct of 2.3cm, dilated in its extrahepatic region. At the level of the hilum there was a clearly contoured but split mass of 4/8.8cm, surrounded by its own wall, having a mixed structure and no peristaltic movements. It was suspected to be a part of the duodenum.

Because no other investigations could reveal the cause of obstruction and both mother and foetus were at risk, an abdominal computer tomography was performed, with permission of the Gynecology Department and the patients' informed consent. CT examination

concluded that there was the possibility of a voluminous (55 mm x 110 mm), heterogeneous mass, externally obstructing the biliary duct, extended from visceral surface of liver and hepatic pedicle to cephalic portion of the pancreas (figure 1, figure 2).



**Figure 1.** CT-scan of the upper abdomen: dilated intrahepatic and extrahepatic bile ducts, heterogenous tissue mass between visceral liver surface, hepatic pedicle and cephalic portion of the pancreas.



**Figure 2.** CT-scan of the upper abdomen: liver (F), aorta (AO), gallbladder (VB), tumoral mass (probably biliary main duct - CBP).

Imagistic investigations were completed with cholangio-MRI. That non-invasive investigation confirmed biliary ducts and Wirsung duct dilatation (figure 3) but could not confirm the presence of a tumor described at CT-scan.



**Figure 3.** Cholangio-MRI sequence: dilated intrahepatic and extrahepatic bile ducts, dilated Wirsung duct.

The patient was transferred back to the Bucharest Emergency University Hospital where she received corticoid treatment for the prevention of prematurity related complications.

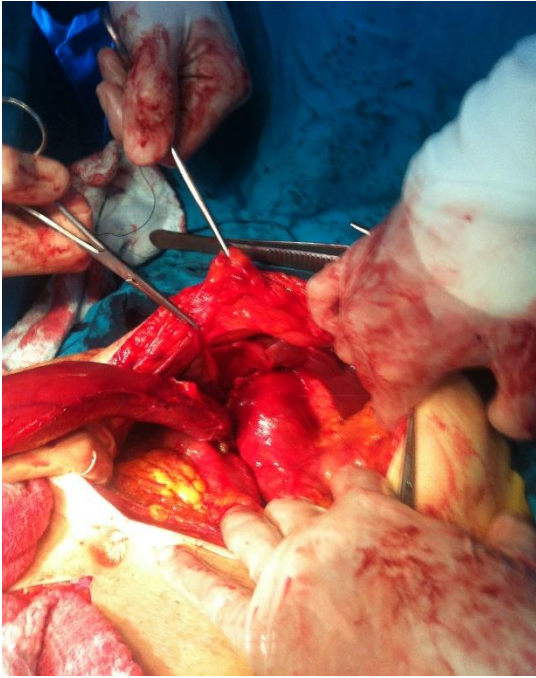
In the following days, the bilirubin levels continued to increase and an ERCP is performed. At cholangiography the main biliary duct appears horizontalized, 17 – 18 mm diameter, cystic duct is long and tortuous, but the gallbladder was not opacified. It drained bile and biliary sludge. A biliary 8.5 Fr stent was placed transpapillary.

Bilirubin levels dropped for two days, but continued to increase 3 days after the ERCP (TB=10.08 mg/dl, DB=8.2 mg/dl).

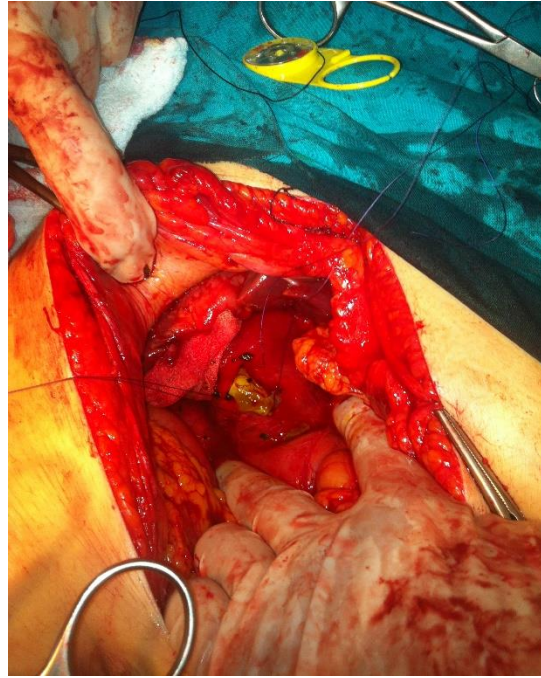
Facing the controversial results of investigations and clinic presentation, the medical team of general surgeons and obstetricians decide to perform a C-section to deliver the newborn safely and also to explore the abdominal cavity.

The obstetricians extracted a live 2350 g male newborn. The skin of the newborn, the placenta and the membranes appeared to be yellowish and impregnated with meconium. The baby was immediately transferred to the Neonatology Department and received Apgar Score 8.

The general surgeons continued to explore the abdominal cavity after the C-section delivery was completed, and found a very long (about 200 mm), tense gallbladder, main biliary duct about 18 mm diameter, without any palpable tumoral mass (figure 4). An anterograde cholecystectomy and supraduodenal longitudinal choledocotomy were performed (figure 5).



**Figure 4.** Intra-operative view: the gallbladder is pulled down. The dilated coledoc can be seen.



**Figure 5.** Intra-operator view: choledocotomy

Exploring the coledoc, the surgeons extract a few gelatinous membranes and biliary sludge (figure 6). Inside the coledoc, a Kehr tube is placed for external drainage of the bile. The peritoneal cavity is washed with saline solution. Multiple drainage of the peritoneal cavity is placed prior to closing the patient.

The hystopathological examination confirms the extracted membranes as hydatid cyst membranes.



**Figure 6.** Gelatinous membranes and biliary sludge extracted from coledoc.



**Figure 7.** Control-cholangiografy performed 12 days after surgery

The patient is transferred to the Intensive Care Unit where she recovered well.

The Kehr tube was removed 12 days after surgery, after control-cholangiography was performed (figure7).

The bilirubin levels decreased to normal and the patient was released from the hospital 13 days after the surgery.

The premature newborn had a total bilirubin level of 3.56 mg/dL in day 1 and developed neonatal jaundice with total bilirubin levels of 9.03 mg/dL in day 6 and needed specific therapy. He was released from hospital at 13 days after birth in good clinical condition, with no jaundice symptoms or other complications.

At discharge the patient received Albendazole p.o. therapy (800 mg/day).

## **DICUSSION**

Hydatid disease (HD), also known as cystic echinococcosis, is a zoonotic parasitic disease of human and mammals caused by the larval stage of tapeworm *Echinococcus granulosus*. *E. granulosus* normally exist among dogs and sheep and in its life cycle, humans sometimes become accidental intermediate host. Humans can orally ingest the *E. granulosus* eggs from infected excretions by handling the animal or egg-containing feces, plants, eating vegetables, uncooked fruits, or drinking contaminated water. Hydatid disease is widely spread in many endemic regions: North Africa, Turkey, South France and The Balkan Peninsula. It is a disease of rural areas where farming is practiced traditionally. The hydatid cyst can occur also in non-endemic countries because of the upsurge of emigration and trade.

The liver, followed by the lung are the most common sites of infection, but that disease can affect in 10% of cases other “atypical” sites, such as muscle, bone, brain, spleen. Rare locations, such as pancreatic, mammary, scrotal, adrenal, cardiac interventricular septum, ovarian were reported.

Hydatid disease is seen in subjects of any age and sex, although it is more common in those of 20 – 40 years old.

The clinical features of cystic echinococcosis are highly variable. The spectrum of symptoms depends on the involved organs, size of cysts and their sites within the affected organ or organs, interaction between the expanding and adjacent organ structures, particularly bile ducts and the vascular system of the liver, complications caused by rupture of cyst, bacterial infection of cysts and spread of protoscolices and larval material into bile ducts or blood vessels, immunologic reactions such as asthma, anaphylaxis, or membranous nephropathy secondary to release of antigenic material.

The rate of growth of cysts is variable depending on the strain differences and the organ involved. Typically measurements state that the average cyst growth is 1 to 1.5 cm per year.

Patients come to the clinician’s attention for variable reasons, such as when a large cyst has some mechanical effect on organ function or rupture of a cyst. The cyst may also be accidentally discovered, during imagistic examination.

The combination of imaging and serology usually enables diagnosis. The standard diagnostic approach for cystic echinococcosis involves imaging techniques, predominantly ultrasonography, computer tomography (CT), X-ray examinations, and confirmation by detection of specific serum antibodies by immunodiagnostic tests.

Serological tests are commonly employed to supplement the imaging data in the diagnosis of hydatid cyst. The current gold standard serology test for echinococcosis detects IgG antibodies to hydatid cyst fluid-derive native or recombinant antigen B subunits.

Sometimes cystic echinococcosis is difficult to treat. This disease can be medical or surgical managed. Clinical management includes albendazol or mebendazole therapy,

especially in small cysts. Larger cyst (>10 cm diameter) preferably undergo surgical resection.

We didn't found any report of intrabiliary main duct localization of the hydatid cyst, without other sites. The mechanism of cyst development at this level can only be supposed.

A possible scenario can be the intraluminal migration of the small hydatid cyst developed near the biliary intrahepatic system in the common biliary duct and then in the main biliary duct.

Another possible scenario is that the cyst was translocated in the main biliary duct by the pathological conditions that associate the presence of the bilirhagia.

The third option can be the evacuation of the hepatic, juxtaductal, hydatid cyst in the biliary tract. If one of them is blocked, the cyst will grow in the main biliary tract.

### **CONCLUSION**

Hydatid disease is a zoonotic parasitic disease of human and mammals caused by the larval stage of tapeworm *Echinococcus granulosus*.

The most common sites of localization are the liver and the lung.

The standard diagnostic approach for cystic echinococcosis involves imaging techniques, predominantly ultrasonography, computed tomography (CT), X-ray examinations, and confirmation by detection of specific serum antibodies by immunodiagnostic tests.

The diagnosis of unique hydatid cyst located at this level implies strong difficulties, especially when appropriate imagistic methods cannot be used.

The difficulty of the shown case was raised by the pregnancy, limiting other possible investigations (contrast administration, irradiation), the absence of other cyst in typical sites and a low grade of epidemiological suspicions.

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