Abnormal placental findings in the case of a pregnant woman with biliary tract hydatid disease

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Abstract

We report the case of a 33 weeks pregnant woman who was admitted in our hospital for recent jaundice. The controversial results of medical investigations and the patient's clinical outcome indicated the necessity for immediate caesarian-section along with exploratory laparotomy. The surgeons extracted gelatinous membranes from the main biliary duct. At the Department of Pathology from University Emergency Hospital from Bucharest, we received the placental components, were we noted the presence of one true umbilical cord knot and meconium impregnation, a non-slatted gallbladder of 18 cm long and the numerous, heterogeneous membranes. On histopathological examination we found uteroplacental hypoxia-related features in the placenta together with retroplacental hematoma, umbilical cord edema, and inflammatory content of the gallbladder and hydatid cyst fragments on the common bile duct samples. The premature new-born survived and both the mother and the infant were discharged in good health condition after 2 weeks.
Keywords: *hydatid disease, uteroplacental hypoxia,true umbilical cord knot*

Introduction

Pregnancy imposes difficult medical management due to the limited number of investigatory and treatment possibilities when regarding acquired illnesses or primary associated conditions as infectious diseases or autoimmune-related pathologies. The feto-maternal barrier is an extremely sensitive area that exposes the fetus to maternal metabolic changes, infections and hemodynamic disturbances leading to various harmful effects on the infant.

Hydatid disease, produced by *Echinococcus* species, is a parasitic disease commonly found in developing countries as Romania. It affects mostly country-side individuals under promiscuityconditions, where dogs represent a major source of infection or in areas where cattle and sheep raising are important. It is rarely reported during pregnancy[1,2]. Abnormal placental findings indicating negative influence on the fetus are represented by accelerated villous development, increased fibrinoid deposition, retroplacental hematomas, and infarction areas. They are generally regarded as indicators of placental hypoxia[3]. Other finding was a true umbilical cord knot which is a rare feature in pregnancy. True umbilical cord knots occur in approximatively 0.3-2% of examined placentas and they can be loose or tight. The risk is that if they become very tight they produce compression of the umbilical vessels and the fetus might suffer hypoxic lesions and even exitus[3,4,5,6].

In this paper we report a case of a patient with these placental abnormalities observed after surgical management for biliary tract hydatidosis.

Case report

We report the case of a 29 year-old woman, gravida 2, para 2, residing in rural area, who at 33 weeks of gestation was admitted in the University Emergency Hospital Bucharest from Romania for recent jaundice and epigastric pain, associated with hyperchromatic urine.

During physical examination we detected a sensible voluminous mass in the upper right abdominal quadrate. Laboratory tests showed elevated levels of alanine aminotransferase (75 U/L), aspartate aminostrasferase (79 U/L) gamma - glutamyltransferase (141 U/L), total bilirubin and its fractions (TB = 8.01 mg/dL, DB = 6.36 mg/dL). The ultrasound examination encountered dilated gallbladder with distension of Wirsung's duct, intrahepatic bile ducts and main bile duct. The presence of a tumor located in the hepatic hilum was suspected. In order to clarify the diagnosis a computed tomography scan and cholangio-magnetic resonance imaging were performed. A voluminous mass of 110/55/43 mm, located in the hepatic hilum was detected. However the origin of this tumor could not be stated. Even with continuous medical support, the bilirubin fractions leveled up so an endoscopic retrograde cholangiopancreatography with stent placement was performed. The intervention was apparently successful - the bilirubin fractions decreased.

After two days bilirubin fractions leveled up again (TB=10.08 mg/dl, DB=8.2 mg/dl). Concerned for both the mother and the fetus's safety, the medical staff decided to perform exploratory laparotomy and cesarean section. A male newborn of 2350 g was extracted with Apgar Score of 8 with yellowish and meconium impregnated skin, placenta and membranes. An extremely dilated gallbladder and main duct (i.e. of approximatively 20 mm) were detected. An anterograde cholecystectomy and supra-duodenal longitudinal choledocotomy were performed. External drainage by a Kehr tube was effectuated. The gallbladder and gelatinous membranes fragments from the common bile duct were sent to the Department of Pathology of the same hospital,

together with the placental components: the placental disk, the umbilical cord and the fetal membranes.

We received an 18x3cm, non-slatted gallbladder. On sectioning we observed wall thickness of 0.2-0.3 cm and clear-green, mucous content. Numerous, millimeter gallstones imprinting the mucosa were noted. On microscopy we found intraluminal acute inflammatory content with neutrophils, pyocytes and lymphocytes admixed with calculi. The mucosa presented areas of exulceration, acute inflammatory infiltrate and hyperemia.

The histopathological examination of the numerous, heterogeneous membranes concluded hydatid cyst membranes, biliary sludge and lithiasis (Figures 1, 2, 3).

The Pathology Department received a discoid shaped placenta (Figure 4), measuring 18x12x2.5cm in greatest dimensions with a trimmed weight of 442 g and fetal membranes present. The fetal surface of the placental disk and the membranes had a green-black and glassy appearance and the maternal surface was disrupted with congested and friable soft spongy tissue areas. The trivascular umbilical cord had central insertion and measured 49 cm in length by 1 cm in diameter. It presented light green color, a left market twistand one slightly tight, true knot with congested vessels at this level.

Microscopic examination provided the following data of the placental disk: predominance of clustered, knotted terminal villi, with increased branching angiogenesis known as Tenney-Parker changes and suggestive of uteroplacental hypoxia, with a score of villi maturation of 33[4], increased intravillous and intervillous fibrinoid deposition as well as intervillous edema, also representing signs of malperfusion (Figures 5, 6, 7). Other findings included a retroplacental hematoma with surrounding areas of infarction and microcalcification (Figure 8). Also the umbilical cord presented edema (Figure 9) and no other pathologic features for the fetal membranes were noted. The absence of pigmented epithelial cells or macrophages indicated recentmeconium discharge.

Postoperative, the patient recovered well, the bilirubin levels decreased to normal. The Kehr tube was removed after control-cholangiography. She was discharged after 2 weeks and received albendazole oral therapy prescription. The premature newborn had total bilirubinemia of 3.56 mg/dl at birth and developed neonatal jaundice in day 6 (TB =9.03 mg/dl) for which he received specific therapy. He was serologic tested for *Echinococcus* at The National Institute for Infectious Diseases "Prof. Dr. Matei Bals" and the results were negative. He was also discharged after 2 weeks, in good health condition, without jaundice or complications.

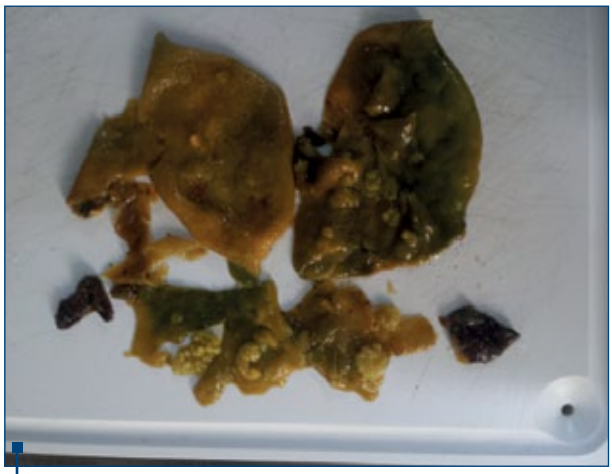

Figure 1. Macroscopic appearance of hydatid cyst membranes

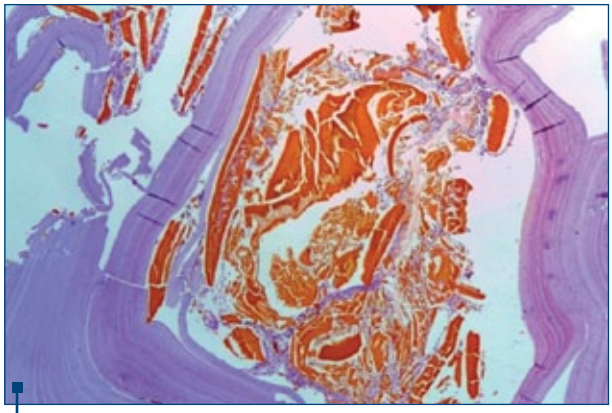

Figure 2. Hydatid cyst membranes, biliary sludge and lithiasis, H.E.x10

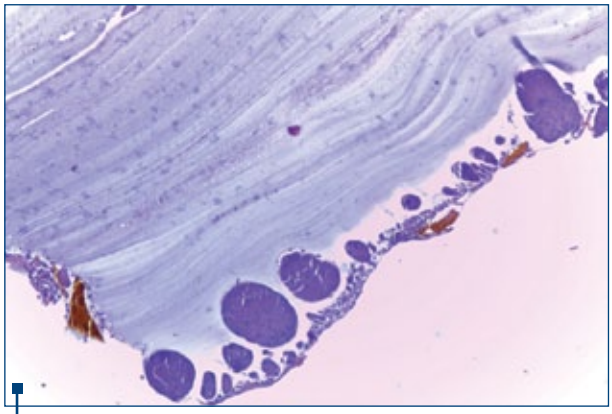

Figure 3. Hydatid cyst membranes, H.E.x10

Discussion

Disturbance of maternal blood flow or composition in pregnancy is known to be interfering with fetal development, but how specific or severe it interacts is still controversial and very difficult to establish[2,3].

Maternal jaundice can be caused by a wide spectrum of pathologic entities such as acute fatty liver disease of pregnancy, viral hepatitis, severe preeclampsia, biliary lithiasis, idiopathic cholestasis of pregnancy, hydatid disease or *Echinococcus*[7]. Regarding maternal hyperbilirubinemia, it is considered that only unconjugated bilirubin can harm the fetus and cause neurologic complications. Conjugated bilirubin does not cross the placenta. Normally, the fetus produces only unconjugated bilirubin because of the microsomal enzyme system immaturity, therefore the mother's liver conjugates it and excretes it. When there is a problem and the conjugating

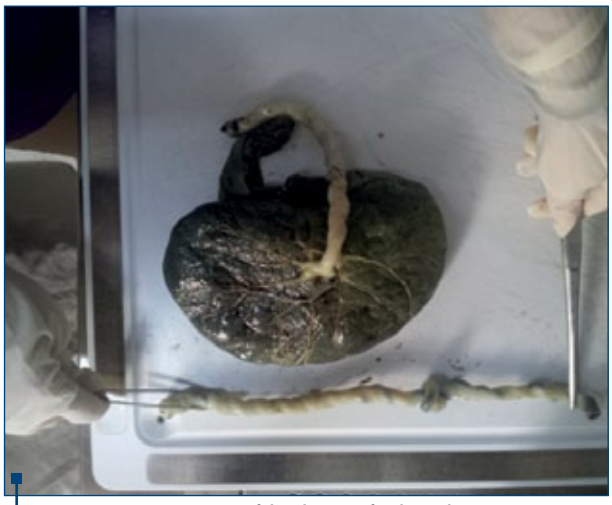

Figure 4. ***Gross appearance of the placenta, fetal membranes and the umbilical cord***

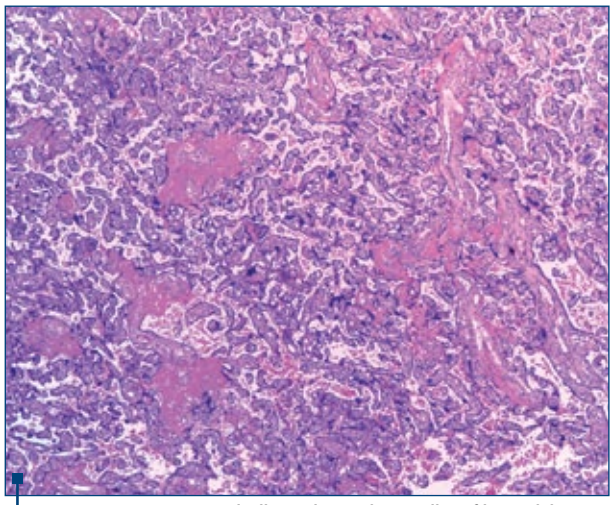

Figure 5. ***Increase terminal villi number and intravillous fibrinoid deposition, H.E.x4***

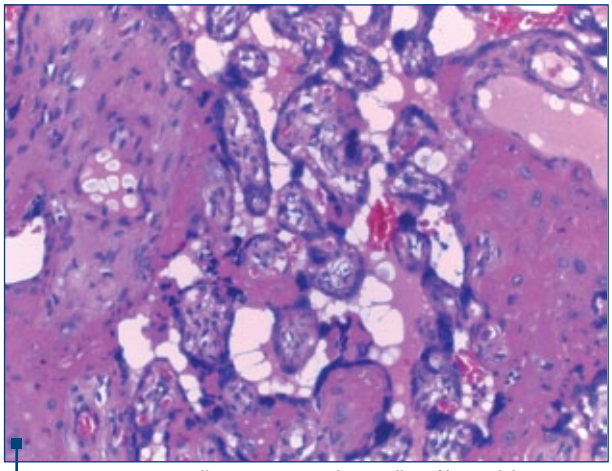

Figure 6. ***Premature villi maturation and intervillous fibrinoid deposition, H.E.x20***

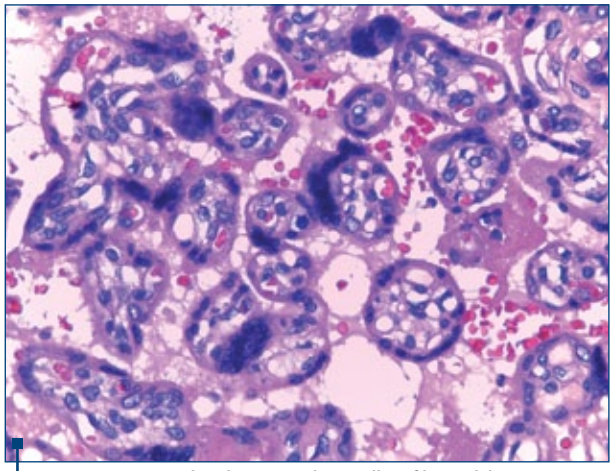

Figure 7. ***Tenney-Parker changes and intervillous fibrinoid deposition, H.E.x40***

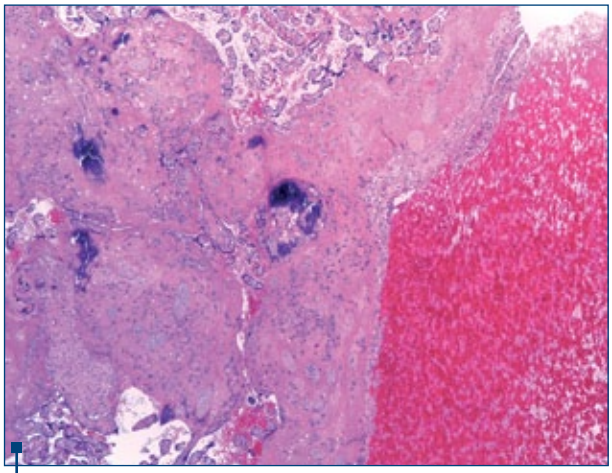

Figure 8. ***Retroplacental hematoma, surrounding infarction area and microcalcifications, H.E.x10***

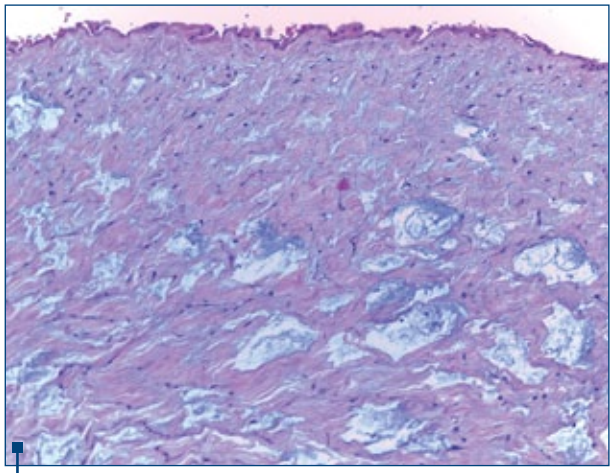

Figure 9. ***Umbilical cord edema, H.E.x10***

process is overwhelmed, the child can be born with jaundice[8,9]. Hydatid disease is rarely associated with pregnancy, with an incidence between 1:20000 to 1:30000[1]. This parasitic disease, presenting with cyst formation in humans, mostly with hepatic or pulmonary involvement, can be dangerous for pregnant women because it can rupture, cause anaphylaxis or any other obstruction-related symptoms or damage, such as: obstructive jaundice and high bilirubin levels, like in our case. It can determine preterm labor or intrauterine growth restriction due to large hepatomegaly[10-14]. A case of fetal malformation of *Spina bifida* and hydrocephalia in association with hepatic hydatic cyst was also reported[15]. We do not know if the placental abnormalities we found in our case are related to hydatid disease. Even if transplacental transmission for other parasites, such as *Plasmodium malariae* or *Toxoplasma* spp. is possible, and vertical transmission has been described for *Echinococcus* in calfs, we did not find any evidence in the literature of transplacental transmission of *Echinococcus* in humans[1,3,16]. For diagnosis, serologic and imaging tests can be used. Treatment depends on the cysts dimension and patient's health status and include surgery, medical treatment with albendazole[2,17,18].

Placental malperfusion is a term that describes modification of normal blood supply of placenta and fetus. This term includes placental underperfusion, uteroplacental malperfusion, uteroplacental underperfusion and they all have in common an abnormality in the uteroplacental vessels. The consequence of these pathologic entities is hypoxia, and we refer to it either as pre-placental hypoxia (uteroplacental hypoxia) or as post-placental hypoxia, depending on where the hypoxic cause appears. Also each type has its own characteristics[3]. Uteroplacental hypoxia means there is a maladaptation of the uteroplacental arteries only in the intrauterine compartment, therefore the placenta and the fetus may become hypoxic but the mother is generally normotoxic. This type of hypoxia can be found in maternal diseases complicating pregnancy such as: preeclampsia, diabetes, lupus erythematous or anticoagulant, tumors etc.

The histopathological findings are: premature villous maturation with increased branching angiogenesis, high degree of placental infarction, increased fibrinoid deposition[3,4].

The normal features of a 33 weeks placenta are represented by the predominance of mature intermediate villi, less common immature intermediate villi and few terminal villi and correspond to a score of villous maturation of 11. In our case we found premature villous maturation showing features of 38-41 weeks, term placenta, that in cause of normal oxygenation would receive Score 22, but the presence of highly branched angiogenesis, established a final Score of 33, which is characteristic of uteroplacental hypoxia. However, the fetus did not suffer intrauterine growth restriction (i.e. weight of 2350 g)[4]. Meconium discharge in premature is a sign of fetal distress, it can be a normal finding in term pregnancies explained by normal fetal intestinal motility and development although it can also be a hypoxic event indicator. Furthermore, it can cause no problems, though the risk of pulmonary complications arises when it is aspirated by the fetus[4]. True umbilical knots can lead to post-placental hypoxia if the knot is tight and the circulation in the umbilical vessels is affected, either with acute/chronic ischemia or with venous stasis and secondary thrombosis. In our case we found only a slightly tightenumbilical knot, with signs of some edema (macroscopically the umbilical cord measured 1 cm in diameter) and no signs of thrombosis. Of course, true umbilical knots represent a problem and lead to concerning reasons, even in the case of loosen ones, because there is always the risk that fetal movements tighten them and produce hypoxic damage and sometimes premature birth with low Apgar Score or severe hypoxia with intrauterine death and spontaneous abortion. It is associated with fetal mortality of 10%[4,19,20,21].

Conclusions

In the present case report, we found abnormal placental features of uteroplacental hypoxia, a retroplacental hematoma together with true knot related umbilical cord edema,in the case of a pregnant woman with biliary tract hydatid disease. Whether these findings are linked to hydatid disease or if they have unidentified cause, we do not know. Still, the overall maternal suffering, namely the infectious disease with its hemodynamic complications could be one of the causes leading at least partially to placental malperfusion. Even if interference of maternal hydatid disease with fetal development and association with serious malformations were reported in the literature, in our case the premature new born had no signs of maldevelopment and infection was serologically infirmed. ■

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