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OF MEDICINE

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Paper Accepted*

ISSN Online 2406-0895

Case report / Приказ болесника

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Primary hepatic pregnancy

Примарна хепатична трудноћа

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Received: February 7, 2020

Revised: August 5, 2020

Accepted: September 9, 2020

Online First: September 14, 2020

DOI: <https://doi.org/10.2298/SARH200207071D>

* **Accepted papers** are articles in press that have gone through due peer review process and have been accepted for publication by the Editorial Board of the *Serbian Archives of Medicine*. They have not yet been copy-edited and/or formatted in the publication house style, and the text may be changed before the final publication.

Although accepted papers do not yet have all the accompanying bibliographic details available, they can already be cited using the year of online publication and the DOI, as follows: the author's last name and initial of the first name, article title, journal title, online first publication month and year, and the DOI; e.g.: Petrović P, Jovanović J. The title of the article. *Srp Arh Celok Lek*. Online First, February 2017.

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SUMMARY

Introduction Hepatic pregnancy is an extremely rare form of ectopic pregnancy, and represents a difficult challenge for both diagnostics and treatment.

Case outline A 40-year-old gravida 0 para 0 patient in 6+0 gestational weeks was admitted to hospital with lower abdominal pain, positive bHCG values, and presence of free intraabdominal fluid. She had a history of infertility, and also a previous surgery due to pelvic endometriosis. Urgent open surgery was performed due to signs of hypovolemic shock. We discovered rupture of left ovarian corpus luteum cyst. Bleeding management was achieved with preservation of ovarian tissue. Patient recovered, bHCG levels continued to rise, and five days after surgery free intraabdominal fluid reappeared with upper abdominal pain and tenderness. After transferring patient to abdominal surgery clinic, second surgery was performed, where we confirmed the presence of hepatic pregnancy. After this procedure, patient fully recovered.

Conclusion Method of choice for an ectopic pregnancy treatment is laparoscopic surgery, but when laparoscopy is not possible, the site of ectopic pregnancy could be difficult to diagnose. Prolonged time for making accurate diagnosis increases the risk of ectopic pregnancy complications.

Keywords: Hepatic pregnancy, Ectopic pregnancy, bHCG, acute abdomen

САЖЕТАК

Увод Хепатичка трудноћа је веома редак облик ектопичне трудноће и представља велики изазов за дијагностику и лечење.

Приказ случаја Четрдесетогодишња пацијенткиња (*para 0, gravida 0*), у 6+0 недељи трудноће, примљена је на клинику због болова у доњем делу абдомена, са позитивним вредностима *bHCG*-а и присутном слободном течности у абдомену. У анамнези се наводи присутан инфертилитет, као и претходна операција због ендометриозе. Због знакова хиповолемијског шока, пацијенткиња је хитно оперисана. Интраоперативно је констатована руптура цисте жутог тела левог јајника. Хемостаза је успостављена уз очување ткива јајника. Пацијенткиња се опоравила, али су вредности *bHCG* наставиле да расту и петог постоперативног дана долази до поновног акумулације слободне течности у трбуху са појавом болова и напетости у горњим делу абдомена. Након превођења пацијенткиња на одељење абдоминалне хирургије, где је поново оперисана. Том приликом потврђено је да се ради о хепатичној трудноћи. Након ове процедуре, пацијенткиња се у потпуности опоравила.

Закључак Метода избора за ектопичну трудноћу је лапароскопска хирургија, али када лапароскопија није могућа, место ектопичне трудноће није лако пронаћи. Продужено време у постављању прецизне дијагнозе повећава ризик од компликација ектопичне трудноће.

Кључне речи: Хепатична трудноћа, ектопична трудноћа, *bHCG*, акутни абдомен

INTRODUCTION

Ectopic pregnancy is a rare condition that occurs in 1% to 2% of all reported pregnancies [1]. There has been an increase in the incidence of ectopic pregnancies over the past few decades. This could be explained by three contributing factors: a greater use of assisted reproductive technologies, a high incidence of pelvic inflammatory diseases, and increased awareness of this condition. However, the incidence of abdominal pregnancy is extremely low, and it occurs in only 1% of all ectopic pregnancies [2].

Abdominal pregnancy refers to a pregnancy implanted in the peritoneal cavity, external to the uterine cavity and fallopian tubes. The placenta can be implanted in any of the abdominal organs and it can separate from the site of implantation at any time during

pregnancy, which leads to hemorrhagic shock. Abdominal pregnancy is a potentially life-threatening form of ectopic pregnancy because of difficult and often late diagnosis. It is associated with a wide range of signs and symptoms according to the location. The diagnosis of abdominal pregnancy is often missed during routine ultrasonography [3].

In contrast to tubal ectopic pregnancies, primary methotrexate therapy of early gestations in abdominal pregnancy has a high risk of failure due to more advanced gestational age at which these pregnancies are discovered [4].

CASE REPORT

A 40-year-old patient was admitted to hospital with lower abdominal pain. She had a history of infertility and had a conservative surgery due to endometriosis (done five years prior through lower transverse laparotomy). Amenorrhea was 6+0 weeks. The initial value of β -HCG was 621 mIU/mL. Ultrasonography (US) exam showed the presence of intra-abdominal free fluid and empty uterine cavity with no adnexal masses. Monitoring of vital signs showed systolic blood pressure (BP) below 90, pulse rate of 130 bpm, and diuresis below 30 ml/h (after receiving 500ml of 0.9% Sodium Chloride solution iv). Due to clinical findings of intra-abdominal bleeding and hypovolemic shock, the patient underwent an emergent surgical procedure by lower transverse laparotomy approach from previous surgery. Intraoperative finding was the rupture of the left ovarian corpus luteum cyst with heavy intra-abdominal bleeding (over 2L). Both Fallopian tubes and the right ovary were unaffected. Small bowels, caecum, rectosigmoid colon and infracolic part of omentum were explored and no sign of ectopic pregnancy was noticed. Histopathology report showed a corpus luteum cyst on the left ovary. After the surgery the patient fully recovered, but β -HCG levels continued to rise (1052 mIU/mL). Postoperative ultrasonography showed no signs of either

intrauterine or extrauterine pregnancy. At that time a small formation found in the liver, 37×25 mm in diameter, was considered clinically indistinctive (Figure 1).

On the third postoperative day we performed diagnostic dilatation and curettage of the uterine cavity. Histopathology exam indicated presence of an endometrial intraepithelial neoplasia (EIN), but no evidence of intrauterine pregnancy. After this intervention β -HCG levels dropped to 861 mIU/mL, but soon continued to rise to the level of 1132 mIU/mL. Since intrauterine pregnancy was excluded, we suspected the presence of ectopic abdominal pregnancy of unknown location and administered a single dose of Methotrexate (50mg).

The patient decided to leave the hospital against medical advice. She was suggested to repeat serum β -HCG values in three days. The values were higher than previous, 1668 mIU/mL. On the fifth day upon the administration of Methotrexate the patient checked into our hospital with pain in the upper parts of the abdomen. Levels of β -HCG were 2060 mIU/mL. On ultrasonography we discovered the presence of free fluid intra-abdominally (small quantities) and abnormal US imaging of the liver. The US exam revealed a hyperechogenic structure of 6cm in diameter in the left lobe near the hilum of the liver. The patient had tenderness in the upper parts of the abdomen.

All clinical findings suggested the presence of ectopic pregnancy in the liver. The patient was transported to abdominal surgery clinic for further evaluation and treatment. An urgent computed tomography (CT) revealed the presence of a mass of 6x5 cm in diameter in the left liver lobe (between the III and IV liver segment), with an oval central portion of lower density, and with the peripheral region of higher density in the contrast plan scan. There was a significant amount of fluid, which corresponds to the density of blood, below the left lobe of the liver and above the stomach (Figure 2).

After the urgent CT examination the patient underwent an open surgery with an upper medial laparotomy. We found around 500 ml of blood in the space between the left lobe of

the liver and the stomach. After the evacuation of a blood which was predominantly coagulated, ectopic pregnancy was detected below the falciform ligament in the third liver segment. Expulsion of the ectopic pregnancy was then performed from the existing liver cavity. Gauze with hypertonic solution of sodium chloride was placed in the cavity of the ectopic pregnancy. The surgical procedure was completed with a single liver suture, as well as hemostasis with a bipolar diathermy. More than 3l of isotonic solution of sodium chloride was placed in the abdominal cavity and two drains were placed: one under the liver, and the other in Douglas space (Figure 3).

The β -HCG value (460 mIU/mL) one day after the surgical procedure indicates a significantly lower value than before the surgery. Six days after the surgery the β -HCG level was 32 mIU/mL. Prior to the surgical procedure, hemoglobin value was 109g/L, and on the day of the patient's release it was 110g/L only with one blood transfusion (280ml) after the operation. The patient was discharged on the seventh day in excellent condition.

The histopathology exam confirmed diagnosis of ectopic pregnancy with presence of trophoblastic tissue found in liver.

DISCUSSION

Ectopic liver pregnancy is a very rare condition (1:15000 pregnancies), but also a very dangerous gynecological disease mostly because of life-threatening intra-abdominal bleeding especially when occurring in the liver or spleen [5, 6]. Although the mechanism of primary hepatic pregnancy is unknown, it can be assumed that the risk factors of its occurrence could be: ovarian cysts, adhesions, pelvic or tubal inflammatory diseases, as well as the specificity of the hepatic tissue characterized by huge blood supply, which could lead to a higher possibility of trophoblast implantation and development of gestation sac [7].

In our present case the patient suffered from endometriosis and an ovarian cyst, which has been proven by ultrasound examination and operatively as well. In our case the rise of β -HCG levels after the prior operation was an alarm for ectopic pregnancy progression. Further evaluation of endometrial changes (EIN) found after D&C procedure is required to confirm the diagnosis of a true premalignant endometrial lesion and exclude an associated endometrial carcinoma in this patient [8]. Inappropriately prepared, as well as pathologically altered endometrium could be one of the explanations for ectopic pregnancy in described case.

Abdominal pregnancy represents great diagnostic challenge. Common symptom for hepatic pregnancy is upper right abdominal tenderness. However, at first our patient did not show any symptoms regarding upper parts of abdomen, which made diagnostic process more difficult [3, 9].

It is important to emphasize the presence of two episodes of internal bleeding in this case. The reason for first episode bleeding was rupture of left ovarian corpus luteum cyst, while second episode occurred due to the bleeding from liver at the implantation site of trophoblastic tissue.

Preferred method of choice for treatment of ectopic pregnancy should be laparoscopy, where the whole abdominal cavity could be explored [10, 11]. Certain, uncomplicated cases of hepatic pregnancy could be treated with Methotrexate [9]. In cases of hepatic pregnancy medial laparotomy could have advantage in bleeding control, considering vascularisation of hepatic tissue [12]. Unfortunately, due to hypovolemic shock our patient underwent urgent open surgery. Laparoscopy should be performed whenever possible to avoid misdiagnosis of abdominal pregnancy and further complications.

NOTE

Ethical compliance statement: We confirm that we have read the journal's position on issues involving ethical publication and affirm that this work is consistent with those guidelines.

Ethical standards: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Written consent to publish all shown material has been obtained from the patient.

Conflict of interest: None declared

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Figure 1. Ultrasonography finding: At first no significance was given to formation found in liver near hilus; Differential diagnosis was liver hemangioma

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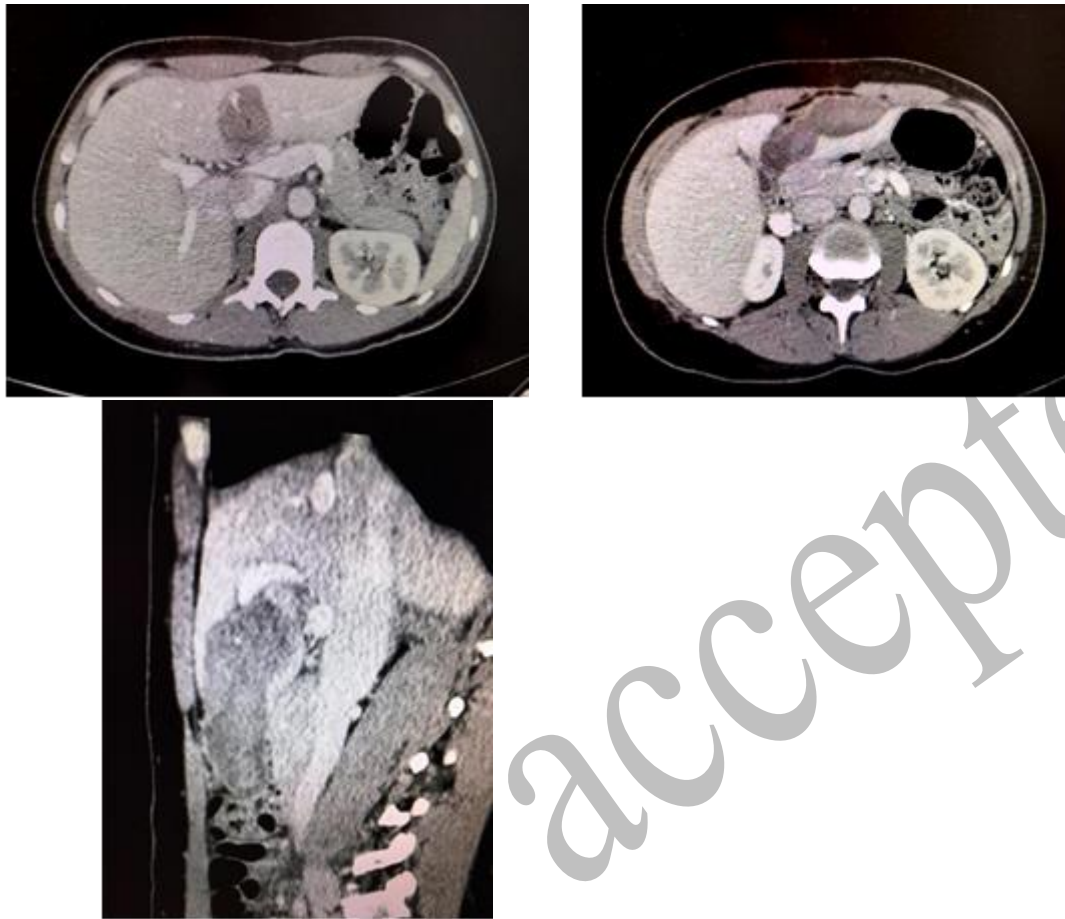


Figure 2. Computed tomography findings: (A) Computed tomography plan scan of the liver shows the presence of an oval mass in the III liver segment of 5×6 cm in diameter with central low-density and high-density periphery;(B) Computed tomography plan scan shows the presence of a large amount of blood in the space between the left lobe of the liver and the stomach; (C) Plan computed tomography longitudinal cross-section of the left lobe of the liver with the presence of blood in the space between the liver and the stomach

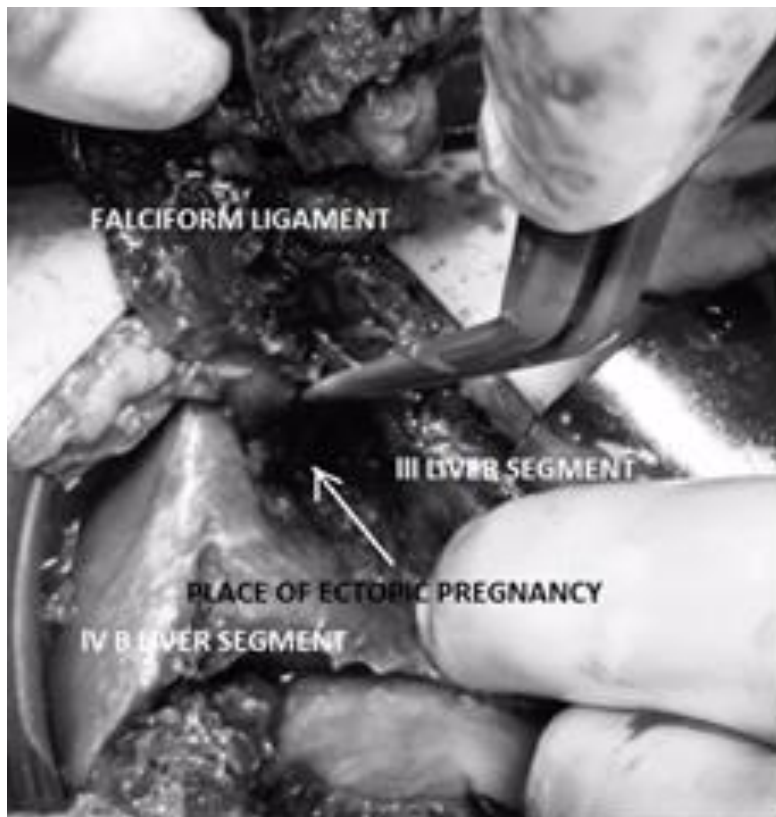


Figure 3. Intraoperative findings: The III segment of the liver and place of the ectopic pregnancy next to IV segment of the liver below the falciform ligament after the expulsion of the ectopic pregnancy from the liver cavity