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Case Report

Splenic hilum pregnancy with a live fetus: a case report and review of literature

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Abstract: Primary splenic pregnancy is a rare form of ectopic pregnancy and potentially life-threatening. We would like to present a case of a 27-year-old woman who had splenic hilum pregnancy with a viable fetus, successfully treated by splenectomy before its rupture.

Keywords: Splenic pregnancy, ectopic pregnancy, splenectomy

Introduction

Ectopic pregnancy is refers to the implantation of a fertilizedovum outside of the uterine cavity, and has an estimated incidence of 19.7/1000 pregnacies [1]. Abdominal pregnancy, a type of ectopic pregnancy, is rare and has an estimated incidence of 1/15000 pregnancies [2, 3]. It can be divided into two categories: primary abdominal pregnancy and secondary abdominal pregnancy. Primary abdominal pregnancy is extremely rare, which is an ovum implant within the peritoneal cavity directly; secondary abdominal pregnancy is because of tubal rupture.

Abdominal pregnancies are potentially life-threatening, particularly without an accurate preoperative diagnosis, and account for a maternal mortality rate about 5.1 per 1000 cases (7.7 times higher than other ectopic pregnancies and 90 times higher with respect to normal uterine pregnancies) [4]. Primary abdominal pregnancies have been described in a variety of sites, including the pouch of Douglas, the posterior uterine wall, the uterosacral ligaments, omentum, small and large intestines, liver and spleen [4, 6].

The spleen is one of the rarest sites of ectopic pregnancy. The highly vascular nature of the spleen makes it a desirable site of implantation, and because of its flat surface where the fertilized ovum easily reached [5].

We report a case of primary splenic hilum pregnancy with a viable fetus, successfully treated by splenectomy before its rupture, while most of the literatures in which patients commonly present with signs of shock and hemoperitoneum [2-7].

Case report

A27-year-old woman (gravida 3, para 2), who was pregnant as evidenced by high β-hCG levels, presented with a single episode of vaginal bleeding colporrhagia for half a month. The patient had a history of regular menses, she was expecting her menstrual period. Routine investigationsin community care facility showed a serum β-hCG of more than 10,000 mIU/ml. However, transvaginal sonography showed a normal-sized uterus and no identifiable intrauterine gestational sac. Physical examination was negative, in particular, revealed no abdominal tenderness or rigidity. Bimanual examination showed a normal cervix and no uterus or adnexa tenderness. The location of the pregnancy could not be found in the community care facility. Then they performed uterine dilatation and curettage, however, the subsequent histopathological examination did not reveal any evidence of chorionic villi or trophoblasts. A day later, quantitative assay of β-hCG still was more than 10,000 mIU/ml.

Therefore the patient was hospitalized at our hospital. Quantitative assay of β-hCG increased

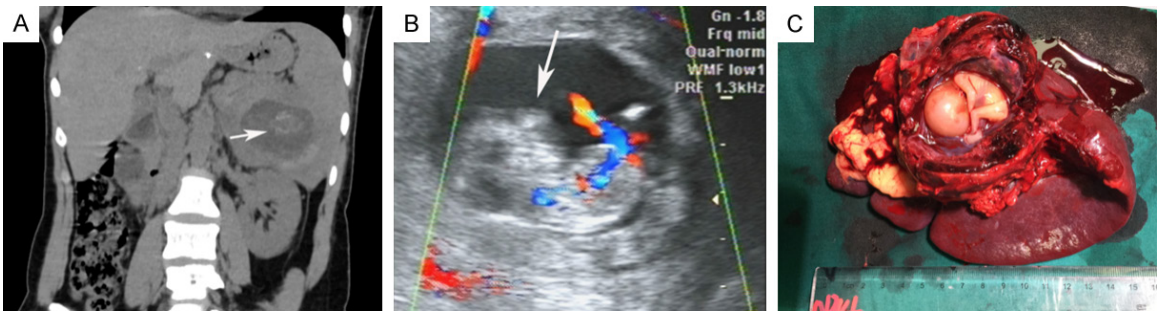

Figure 1. A. Arrow: Abdominal CT showed a well-defined heterogeneous mass on the splenic hilum; B. Arrow: Abdominopelvic ultrasound showed a live fetus; C. Excised spleen and the fetus.

from 68,764 mIU/ml to 711431 mIU/ml. Our hospital transvaginal sonography revealed a normal-sized uterus with a thinned 4.6 mm endometrium and noidentifiable intrauterine gestational sac; at the level of left tube, an image which was about 19 mm*13 mm*16 mm suspected for fallopian tube thickening or ectopic pregnancy was detected, with no fluid in the pouch of Douglas. To rule out the possibility of a β-hCG-secreting tumor, multi-slice CT of the chest was performed and a heterogeneous hypervascular masson the splenic hilum was detected. Then we decided to investigate with abdominal CT. This showed a well-defined 8.5 cm*6.2 cm heterogeneous mass on the splenic hilum (**Figure 1A**). An abdominopelvic ultrasound performed by an experienced ultrasonic-doctor confirmed these findings and a fetus (5.0 cm in length from head to hip) was seen and the fetus was alive (**Figure 1B**).

A splenectomy was performed owing to the attachment of the placenta to the splenic vein and thus the risk of its rupture, which revealed a well-developed 12-week male fetus with a crown-to-rump length of 5.0 cm (**Figure 1C**). The patient's recovery was uneventful: postoperative serum β-hCG levels decreased to 276 mIU/ml 9 days after surgery and returned to normal 16 days after surgery.

Discussion

Primary splenic pregnancy is a type of abdominal pregnancies. According to the criteria for primary abdominal pregnancy proposed by Studdiford [8], they are as follows: (1) fallopian tubes and ovaries are grossly normal and show noevidence of recent injury; (2) no evidence of uteroplacental fistula; (3) pregnancy of no more than 12-week gestation with trophoblastic elements related exclusively to a peritoneal surface. In our operation, there was no evidence of pregnancy outside the spleen, a well-developed 12-week male fetus with a crown-to-rump length of 5.0 cm was in the splenic hilum. Therefore our patient adheres to the criteria. Preoperative and postoperative diagnosis was consistent.

To our knowledge, there have been 18 splenic pregnancies published reports. In the previously reported cases, most of the patients presented with acute abdominal pain and hemorrhagic shock due to spleen rupture, received surgical intervention [2-7]; two of them were asymptomatic at presentation and successfully treated with laparoscopic methotrexate injection or methotrexate and KCl injection [9, 10].

Our case is more interesting because transvaginal sonography, curettage were not diagnostic, the patient was only presented with a single episode of colporrhagia and bimanual examination was normal. A raising blood level of β-hCG, from 68,764 mIU/ml to 711431 mIU/ml, suggesting the presence of ectopic pregnancy, was confirmed afterwards by abdominal CT and abdominopelvic ultrasound. So how to increase awareness of primary abdominal pregnancy and make early diagnosis is vital to avoid abdominal pregnancy-related deaths. The absence of an intrauterine gestational sac on transvaginal sonography, aside from a high level of β-hCG (>1500 U/L), is suggestive of an ectopic pregnancy, and the patient should be submitted to abdominal ultrasound and computerized tomography extended to the whole abdominal cavity in order to avoid misdiagnosis. Yagil et al. [11] described the role of ultrasonography and-computerized tomography in making an early diagnosis of abdominal ectopic pregnancy. Li Y et al. [12] analyzed the application of transabdominal sonography and transvaginal sonography in early diagnosis of pregnancy. They concluded that transabdominal sonography in combination with transvaginal sonography can

complement information, improve detection rate, and reduce or avoid misdiagnosis and missed diagnosis, which provides a scientific basis for the formulation of clinical treatment scheme. Si MJ et al. [13] concluded MRI plays an important role in the early diagnosis of tubalpregnancy and provides accurate evaluation of the lesion to help make management decisions. Although abdominal CT was diagnostic in the present case and other cases, it carries the risk of radiation exposure, and in future cases abdominal MRI can be considered [10].

The spleen is one of the rarest sites of ectopic pregnancy. We successfully treated the splenic hilum pregnancy with a well-developed 12-week male fetus by splenectomy before its rupture. The size (crown-to-rump length of 5.0 cm) of the spleen pregnancy is much more larger than those which reported in the published literatures. If the gestational sac is small, partial splenectomy is required. In our case, the fetus is so large and owing to the attachment of the placenta to the splenic vein, total splenectomy was performed.

Nowadays, the incidence of ectopic pregnancy has been steadily increasing, with the increased use of assisted reproductive technology (ART), more cases of ectopic pregnancy and primary abdominal pregnancy can be expected. One case presented simultaneous tubal-splenic pregnancy after ART [5]. Recognition of these rare forms of abdominal pregnancy is vital owing to the risk of hemorrhagic shock and death, and should be considered in the differential diagnosis of acute abdomen in women of reproductiveage.

Conclusion

We present a splenic hilum pregnancy case with a viable fetus, successfully treated by splenectomy before its rupture. If an ectopic pregnancy is suspected and no identifiable intrauterine sac could be detected by transvaginal sonography, abdominopelvic ultrasound and abdominal CT must be performed for early accurate diagnosis prior to rupture.

Disclosure of conflict of interest

None.

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References

[1] From the centers for disease control and prevention. Ectopic pregnancy--United States, 1990-1992. JAMA 1995; 273: 533.
[2] Zhang Y, Kang D, Zhang B, Yang L and Fan Z. Ectopic pregnancy causing splenic rupture. Am J Emerg Med 2016; 34: 1184, e1181-1182.
[3] Elghanmi A, Mohamed J and Khabouz S. Spontaneous splenic rupture in pregnancy. Pan Afr Med J 2015; 21: 312.
[4] Biolchini F, Giunta A, Bigi L, Bertellini C and Pedrazzoli C. Emergency laparoscopic splenectomy for haemoperitoneum because of ruptured primary splenic pregnancy: a case report and review of literature. ANZ J Surg 2010; 80: 55-57.
[5] Kitade M, Takeuchi H, Kikuchi I, Shimanuki H, Kumakiri J and Kinoshita K. A case of simultaneous tubal-splenic pregnancy after assisted reproductive technology. Fertil Steril 2005; 83: 1042.
[6] Kalof AN, Fuller B and Harmon M. Splenic pregnancy: a case report and review of the literature. Arch Pathol Lab Med 2004; 128: e146-148.
[7] Gao H, Yuan T, Ding Y, Lin X, Wang Q, Dai L, Jiang G and Chen J. Primary splenic pregnancy with hemorrhagic shock. J Obstet Gynaecol Res 2017; 43: 1342-1345.
[8] Elzey ND. Primary abdominal pregnancy in the lesser peritoneal cavity. West J Surg Obstet Gynecol 1948; 56: 410-413.
[9] Gang G, Yudong Y and Zhang G. Successful laparoscopic management of early splenic pregnancy: case report and review of literature. J Minim Invasive Gynecol 2010; 17: 794-797.
[10] Klang E, Keddel N, Inbar Y, Rimon U and Amitai M. Splenic pregnancy: a new minimally invasive approach to treatment. Cardiovasc Intervent Radiol 2016; 39: 1339-1342.
[11] Yagil Y, Beck-Razi N, Amit A, Kerner H and Gaitini D. Splenic pregnancy: the role of abdominal imaging. J Ultrasound Med 2007; 26: 1629-1632.
[12] Li Y, Feng T and Sun J. Evaluation of clinical effect of transabdominal sonography and transvaginal sonography in early diagnosis of ectopic gestation. Pak J Med Sci 2017; 33: 53-58.
[13] Si MJ, Gui S, Fan Q, Han HX, Zhao QQ, Li ZX and Zhao JM. Role of MRI in the early diagnosis of tubal ectopic pregnancy. Eur Radiol 2016; 26: 1971-1980.