EARLY DIAGNOSTIS OF SPLENIC ECTOPIC PREGNANCY: A CASE REPORT AND REVIEW LITERATURE

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Splenic ectopic pregnancy is extremely rare but carries a high risk of life-threatening intraperitoneal bleeding. Here, we present a 40-year-old woman presenting with vaginal bleeding. Although the intrauterine device (IUD) had been in place for 6 years, urinary and serum tests were positive for pregnancy. However, transvaginal ultrasound showed an empty uterus, no apparent adnexal masses or free fluid. An abdominal ultrasound was subsequently performed, which revealed a viable gestational sac in the spleen. An MRI that did not induce ionizing radiation was also performed, confirming the diagnosis of this splenic pregnancy. The gastrointestinal surgeon completed a laparotomy which successfully removed the spleen's superior pole containing an ectopic pregnancy.

Keywords: Splenic pregnancy, Ectopic pregnancy, Ultrasound, MRI, Partial splenectomy .

I. INTRODUCTION

The most common site of ectopic implantation is within the fallopian tube, accounting for 95.5% of all ectopic pregnancy, the remaining extratubal ectopic pregnancy include ovaries and abdomen, accounting for 3.2% and 1.3%, respectively.[1] Abdominal pregnancies have been described in a diverse of extra-pelvic organs, with the spleen being one of the rarest sites. A Systematic review of the literature, Poole A et all. showed that in 225 abdominal ectopic pregnancies, splenic gestations accounted for only 5.3%. The remaining sites were divided into the following categories in order from most reported to least: pouches around uterus (24.4%), uterus-adnexa (24.0%), multiple abdominal organs (12.9%), omental (11.1%), bowel-appendix (6.7%), hepatic (5.8%), retroperitoneal (4.5%), abdominal wall (3.1%) and others (2.2%).[2]

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Primary splenic pregnancy was common at young maternal age (27.3-28.7), with few births (the average parity was 1.1).[2,3] As with other abdominal pregnancy sites, splenic pregnancy occurred with several risk factors, including a history of pelvic inflammatory disease, endometriosis, in vitro fertilization (IVF), previous pelvic surgery, previous ectopic pregnancy, uterotubal anomalies, and intrauterine device (IUD).[4,5] Most cases of symptomatic splenic pregnancy are diagnosed between 6th - 8th week of gestation, and tended to occur earlier than other sites in the abdomen, but later than tubal ectopic pregnancy.[1,2,3,6] Kalof et al postulated most splenic pregnancies at clinical presentation ranged in size from 2.0 to 3.5 cm, and suggested that the risk of rupture in ectopic gestation exceeds this size.[3]Herein, we would like to present a case preoperatively diagnosed by adbominal ultrasound and MRI as a splenic pregnancy with live embryos implanted at the superior pole of the spleen in a middle-aged woman who had an IUD in situ, successfully treated by partial splenectomy before its rupture.

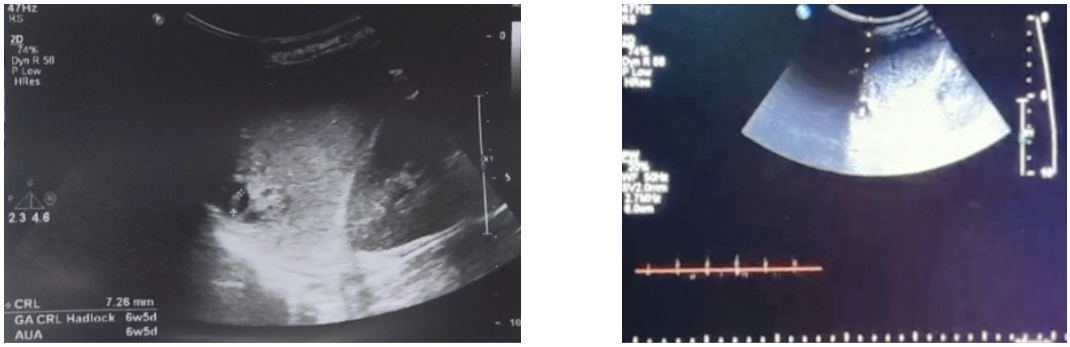
II. CASE REPORT

A 41-year-old woman, gravida 2 para 2, was presented to the Obstetrics and Gynecology Department due to intermittent vaginal bleeding and vague pain in the lower abdomen for several days. The individual has had a history of regular menses (28-days per period), and her last menstruation was 6 weeks prior. She denied a history of pelvic inflammatory disease, tubal surgery or previous ectopic pregnancy, except for a copper IUD in the community 6 years ago.

At admission, she had neither vomiting nor fever, physical examination showed no abdominal tenderness or rigidity and a blood pressure of 110/60 mm Hg. Bimanual examination indicated a normal cervix and minimal bleeding discharge with no tenderness of the uterus or adnexa.

Urine pregnancy test showed positive results and serum beta-human chorionic gonadotropin (βHCG) showed 34,279 IU/L (normal <5.3). Blood laboratory tests were unremarkable with a normal red blood cells count of 4.5 T/l (4.0-5.2). However, transvaginal ultrasound revealed a normal size uterus with 7.6mm thick endometrium but with an IUD, no identifiable intrauterine gestational sac and no sign of ectopic tubal pregnancy, normal bilateral ovaries and no free fluid in the pouch of Douglas.

After suspecting an extratubal pregnancy, further pan-abdominal ultrasound was performed to look for uncommon implantation sites. The splenic parenchyma at the superior aspect showed an ill-defined heterogeneous cystic mass measuring 36 x 47 mm, which contained an 11mm gestational sac containing an embryo with a crown-to-rump length of 7.2 mm (equivalent to 6 weeks 4 days fetus) and fetal heart movement confirmed at 145 beats/ min (Fig.1). Abdominal ultrasound also noted the absence of free fluid in the perisplenic space.



A **B**

Figure 1. Abdominal ultrasound at the superior splenic pole shows a gestational sac containing the embryos (A , arrows) in the presence of beating heart on pulsed Doppler (B, arrows)

Although abdominal ultrasound strongly identified a splenic pregnancy, an abdomino-pelvic MRI was performed to obtain additional information. This technique confirmed the existence of a mixed cystic lesion mesuring 28 x 37x 46mm accompanied by perilesional edema of the splenic parenchyma. It showed no other abnormality in the abdomen and pelvis (Fig.2). A diagnosis of primary splenic pregnancy was made.

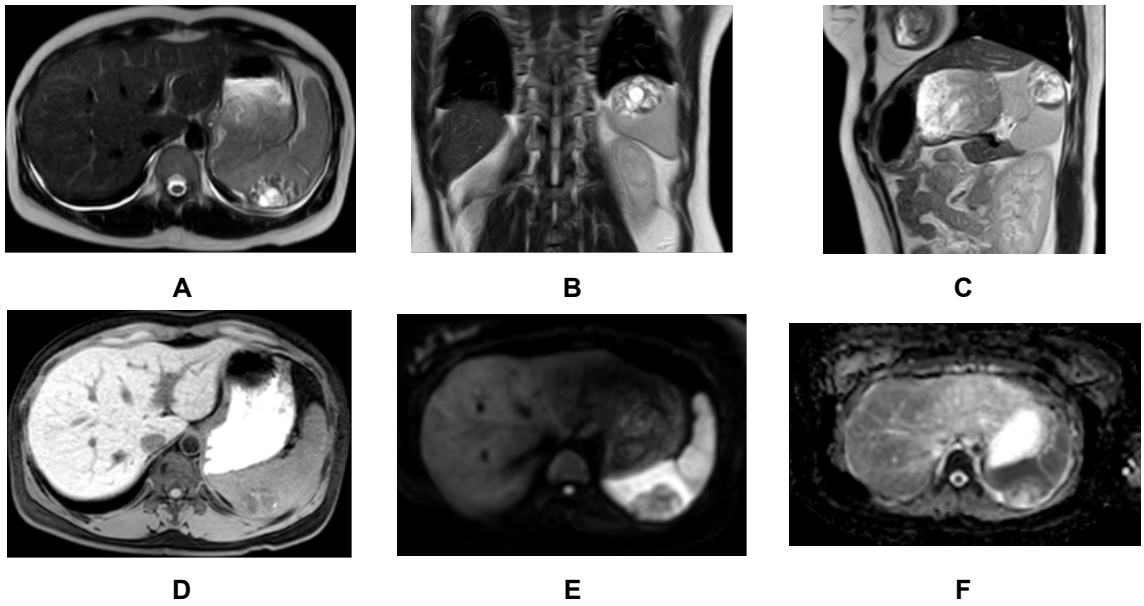
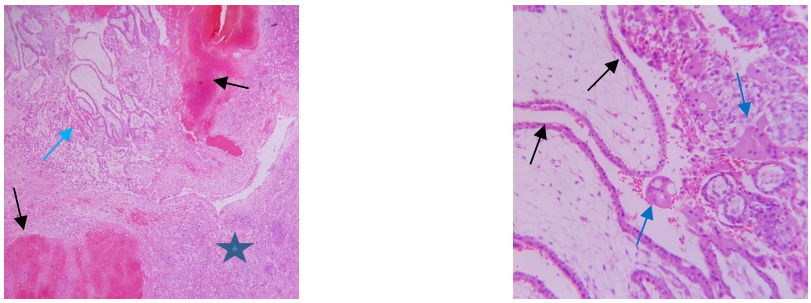


Figure 2. Abdomino-pelvic MRI. T2WI axial (A), coronal (B) and sagittal (C); T1WI VIBE FatSat (D); DWI b=800 (E) and ADC (F).

The images shows a mass located in the spleen adjacent to the left diaphragm, cystic component, well-margined, with many small septal in the periphery (A, B and C, arrows), interspersed with hyperintense foci of blood stasis (D, arrow), and restricted diffusion edema of peri-lesional splenic parenchyma (E and F, arrows)

The initial surgery was exploratory laparoscopic examination. It revealed a 3x4cm pregnant mass in the upper pole of the spleen with villous tissue deeply embedded in the spleen parenchyma and easy to bleed. The prognosis was difficult, therefore decision was made to converse from laparoscopic to open surgery by the xiphoid-umbilical incision. The spleen was released from the upper gastric pole, the tail of the pancreas, and the splenic flexure. the splenic artery branch to the upper pole was ligated, and s ubsequently, an anatomic resection of the upper pole of the spleen was performed and hemostasis with prolene 3.0 suture. Excellent hemostasis was achieved and the abdomen was closed up after being washout with insertion of a drain in the peri-splenic region. Postoperatively, the patient was transferred to the Intensive Care Unit for monitoring, before stepping down to the surgical ward 2 days later where the rest of her recovery was uneventful. ß-hCG levels fell to 908.83 IU/L 6 days after surgery. The pelvic drain was removed on day 3 and the patient was discharged on day 5 post-operation. She had an uneventful recovery at home and ß-hCG levels returned to normal 4 weeks after surgery.

Macroscopic pathology of the resected superior pole spleen revealed a protrusion with a generally thin and smooth surface. On sectioning in half, there was a corresponding oval mass with well-delineated but non-encapsulated measuring 20 x 25 x 35 mm, containing cystic and pinkish-white soft tissue mixed with dark red hemorrhages. Microscopic examination demonstrated numerous chorionic villi and intermediate trophoblasts invading the splenic parenchyma (Fig.3). No malignant cells were found.



A B

Figure 3. Histopathologic sample. A (H&E, ×50), chorionic villi (blue arrow) invading normal splenic parenchyma containg the lymphoid follicles (star) and adjacent areas of hemorrhage (black arrows). B (H&E, ×400), chorionic villi (blue arrows) and trophoblastic tissue (black arrows)

III. DISCUSSION

Due to strong peristalsis of the intestine and dynamics of intraperitoneal fluid flow, it is possible to carry fertilized ovum from the cul-de-sac to different abdominal cavities.[7,8] The spleen provides a smooth and flat surface of the thin capsule along with its abundant blood flow nature and accessibility in the supine position, making it a relatively sustainable site for the zygote implantation.[8,9,6,10] This explains most pregnant masses are implanted under the capsule, protruding outward and often beyond the border of the spleen, and their location can be anywhere from the upper pole to the lower pole and the hilum.[3]

Although the spleen is a favorable environment for embryonic growth, unfortunately the spleen parenchyma cannot distend to support the blastocyst growth and cannot accommodate placental attachment. Therefore, splenic pregnancy is rarely detected at the end of the first trimester.[6] In complicated cases, patients complain of acute severe abdominal cramping or typically presents as left upper abdominal pain radiating to the left shoulder, followed by signs of peritonitis and unstable hemodynamic status leading to urgent splenectomy.[2,3]

Nowadays, imaging modalities is always available to help early diagnostic identify the implantation site of an ectopic gestational sac. Once a woman with a missed period has abnormally elevated β-hCG and no intrauterine pregnancy on transvaginal ultrasound, the diagnosis of ectopic pregnancy can be established, even in the absence of histopathology of uterine curettage. Several cases have demonstrated that ultrasound combined with CT has a role in early diagnosis of splenic ectopic pregnancy.[4,11,12]

Ultrasound should be considered the standard imaging tool of first choice. Screening pan-abdominal ultrasound can reveal gestational sac-like echo image in the spleen, and color Doppler imaging can also show increased vascularity around the sac.[4,10] Rarely, embryos with a live fetal heart can be seen,[11,12] as was in our case. Wu et al published recently the first case of splenic pregnancy accurately diagnosed by ultrasound prior to treatment.[13]

To confirm the results of abdominal ultrasound, CT or MRI should be employed because they have an important role as diagnostic evidence and detailed assessment of ectopic gestations to help make management

decisions. Although abdominal CT provided accurate diagnosis in most of previous case reports, it carries the risk of radiation exposure, therefore, MRI can be considered.[5,6,12] Moreover, this method is increasingly available and useful in earlier or unruptured abdominal pregnancy. Our case was similar to that of Makrigiannakis et al, wherein both abdominal sonography and MRI verified the presence the embryonic sac in the splenic parenchyma.[6]

In unruptured splenic ectopic pregnancy cases, even if a patient with ruptured splenic pregnancy is hemodynamically stable or non-surgical candidate, splenic preservation should be considered when possible because of its functional benefit. Several studies have been published showing successful conservative splenic treatment by minimally invasive approaches and non-surgical management in combination with intramuscular methotrexate administration has been mentioned. As well as the laparoscopic injection of methotrexate in the embryonic sac,[14] CT-guided yolk sac aspiration with local injection of methotrexate, followed by ultrasound-guided percutaneous KCl injection,[12] ultrasound-guided methotrexate injection,[10] selective embolization of the splenic vessels feeding ectopic pregnancy by methotrexate,[6] and partial splenectomy or splenorrhaphy,[5] as well as our aforementioned case.

IV. CONCLUSION

In reproductive-age female with abnormally elevated β-hCG levels and no intrauterine or pelvic pregnancy revealing on transvaginal ultrasound, it is advisable to examine patients using other imaging modalities, such as abdominal sonography, CT or MRI, to detect any upper abdominal pregnancies. With rare abdominal ectopic gestations, early successful diagnosis is essential because of the high risk of uncontrollable life-threatening intraperitoneal bleeding. Partial splenectomy is the treatment of choice with the benefit of splenic preservation, especially in unruptured splenic pregnancy.

Conflict of Interest Statement

The authors declare no financial disclosures or conflicts of interest.

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