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REVIEW ARTICLE

Successful Pregnancy in Post-traumatic Splenosis Woman: A rare condition

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ABSTRACT

Splenosis is a rare benign condition which was first described in 1929. Most of cases found in men following the splenic rupture without symptom. A successful pregnancy in a splenosis woman has never been reported in English literatures. Herein we experienced a first case of a successful pregnancy in splenosis woman. She was successfully delivered a healthy male infant by Cesarean delivery using a Pfannenstiel skin incision.

Keywords: splenosis, pregnancy, post-traumatic.

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Introduction

Splenosis is a rare benign condition caused by an ectopic autotransplantation of splenic tissue following a splenectomy or traumatic splenic rupture. It was first described by Buchbinder and Lipkoff in year 1929[1]. The incidence of post traumatic abdominal splenosis has ranged from 65-93% of splenic rupture cases[2]. Affected patients are mostly asymptomatic but some present with pain[3]. Most cases have been reported in men, which has been hypothesized to be related to a higher incidence of trauma in young men. A successful pregnancy in a woman with splenosis has never been reported in English literatures. Herein we describe a case of a successful pregnancy in a splenotic woman.

Case presentation

A 29-year-old, G2P1 Thai woman was referred to our hospital for first antenatal care at gestation age 8 weeks because of a history of multiple intra-abdominal operations. At her third year of life, she was in a car accident and later had a splenectomy via midline incision due to splenic rupture. Ten years ago (at 19 years of age) she had an appendectomy via a Lanz incision. Her first pregnancy 3 years ago was a term pregnancy delivered by low transverse cesarean

section via Pfannenstiel incision because of breech presentation at general provincial hospital. No intra-abdominal mass was reported. One year ago, two years after the cesarean section, she visited our hospital complaining of chronic pelvic pain at her left pelvis radiating to her back. Abdominal and gynecologic examinations found nothing remarkable other than the three previous operation scars. Transvaginal ultrasonography revealed normal uterus and ovaries. Computer tomography showed multiple arterial enhancing peritoneal nodule/masses scattered throughout the abdominal and pelvic cavities with subcentimeter lymph nodes at the paraaortic and mesenteric regions, size up to 1.0 cm. Three large lesions were documented: one 48 x 46 x 30 mm posterior to the liver, another 74 x 23 mm at the anterior of the abdomen and a third 38 x 17 mm at the lower posterior of the uterus. An ultrasonographic-guided biopsy was done to obtain a sample from the subhepatic area and the pathological examination reported post-traumatic splenosis. Supportive pain-control management was our first management. Three months later, she became spontaneously pregnant.

Her initial antenatal laboratory test showed a heterozygous hemoglobin constant spring without anemia, with other tests unremarkable. Thalassemia screening of her husband was negative, thus a non-couple at risk for fetal severe thalassemia was concluded. The first transabdominal ultrasonography showed a single intrauterine pregnancy, compatible with 8 weeks of gestation. The patient regularly attended our antenatal clinic, and standard ultrasonography showed a male baby without structural abnormalities at 20 weeks of gestation. At 32 weeks of gestation, an elective repeated cesarean section was planned at term pregnancy (38 weeks of gestation). Transabdominal and transvaginal ultrasonographies were done for preoperative evaluation and fetal growth monitoring. The fetal growth was normal. The transabdominal ultrasonography revealed multiple homogenous, well-defined border hypoechoic masses at the uterine fundus and subhepatic area, sizes from 2-3 centimeters. A mass was found at the anterior abdominal, measuring 29x18 mm (Fig. 1).

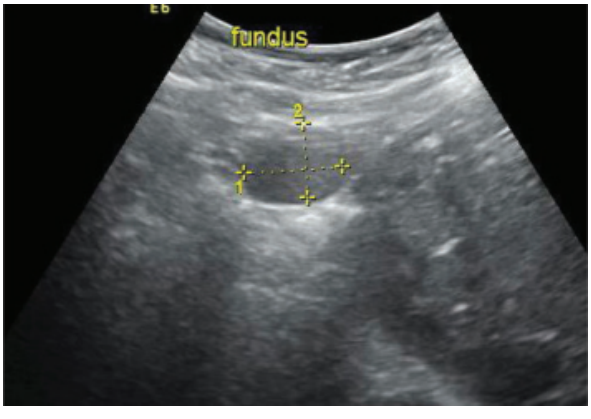

Fig. 1. Transabdominal ultrasonography revealed a well-defined border hypoechoic mass at anterior uterine wall, measuring 29 x 18 mm.

Transvaginal ultrasonography revealed two lobular homogenous hyperechoic, well-defined border masses at the cul-de-sac, measuring 38 x 17 mm. Doppler ultrasonography revealed a hypervascular signal compatible with a vascular mass (Fig. 2). A multidisciplinary team was put together for the cesarean delivery, including a gynecologic oncologist, a vascular surgeon, and a neonatologist. At 38 weeks and 4 days, an elective cesarean delivery through a Pfannenstiel skin incision with bilateral tubal salpingectomy was performed by the gynecologic oncologist. The salpingectomy was done under

individual opinion of oncologist to prevent future malignancy. A healthy male infant without abnormalities was delivered, weighing 3,020 g, with Apgar scores of 9 and 10. During the cesarean section, the uterus and both adnexae appeared normal. Multiple purple vascular mass like splenic tissue seedings were found in the upper abdominal cavity (Fig. 3). The cul-de-sac was not explored to avoid massive bleeding. No significant intra-abdominal adhesion was noted. There were no operative complications and the patient was discharged with her baby 5 days after the operation. She was scheduled for postpartum evaluation and to plan for long term pelvic pain treatment.

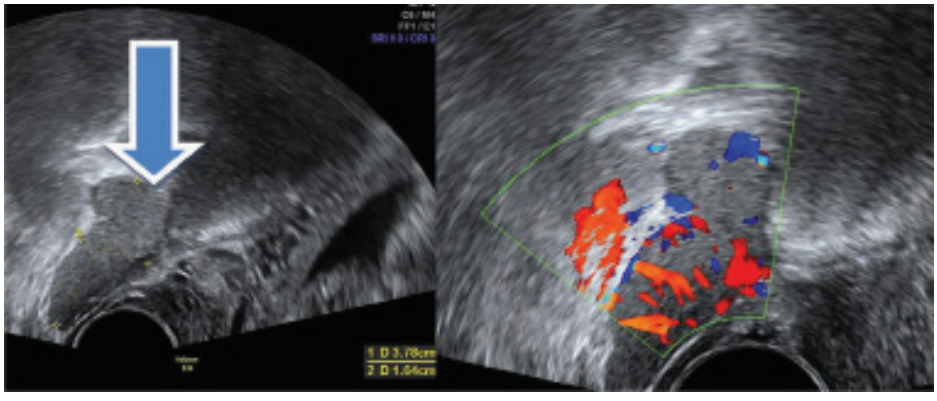

Fig. 2. Transvaginal ultrasonography revealed two lobular homogenous hyperechoic, well-defined border mass at cul-de-sac, measuring 38 x 17 mm (left). Doppler ultrasonography found a hypervascular signal compatible with vascular mass (right).

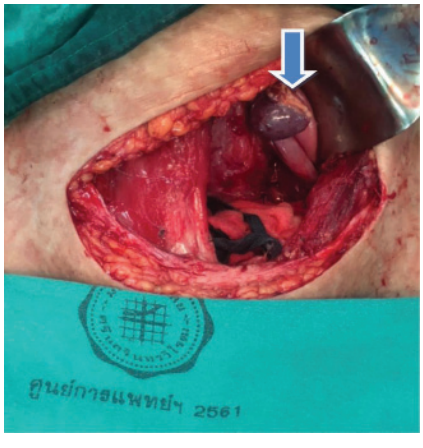

Fig. 3. Intraoperative gross finding shows an intraabdominal bluish-red soft solid tissue (arrow).

Discussion

Splenosis is an acquired condition which can occur after a rupture of the spleen. It is more common in men. A common presenting symptom in reproductive women is pelvic pain, as in our patient. Abnormal vaginal bleeding or dysmenorrhea have been reported with splenosis but these symptoms were later explained by other associated pathology such as a uterine mass[4-6]. Our patient is, to our knowledge, the first case of a successful

pregnancy in a post-traumatic splenotic woman. Thus, the natural progression of splenosis during pregnancy has never been presented. Previous studies from humans[7] and mice[8] did not show an enlarged spleen in a pregnant woman. Estrogen causes an increased in size and weight of the spleen in its normal location by increasing the number of erythroid cells and erythroblasts, particularly in the red pulp of the spleen. The white pulp did not show any significant changes. Progesterone has no influence upon the splenic pulp. This basic knowledge is contrast to the findings in our patient. During her pregnancy, we found that our patient's ectopic splenic mass did not significantly increase in size compared to in non-pregnant stage. Also, the mass at the anterior uterus reduced in size from 74 x 23 mm to 29 x 18 mm. We hypothesize that the vessel feeding the ectopic spleen at infrahepatic area came from adjacent organs but the splenic artery feeds the normal position spleen. Thus, a more limited blood supply may limit the growth of ectopic splenic tissue.

Concerning the diagnosis, our patient's splenosis was confirmed by direct biopsy before she became pregnant. Diagnosis confirmation during pregnancy is more difficult and complicated. Lack of Howell-Jolly bodies, reticulocytosis and other postsplenectomy cellular abnormalities on peripheral blood smears after splenectomy can lead the physician to assume a diagnosis of splenosis[2-5]. Additional imaging through ultrasonography, computer tomography with or without contrast and magnetic resonance imaging can be performed, but even then it can be difficult to differentiate splenosis from other possible malignancies. 5 mCi (185 MBq) Tc99m –tagged heat-damaged autologous red blood cells (RBCs) or indium 111 labeled platelets scintigraphy can be performed in non-pregnant patients suspected to have splenosis, as more sensitive and specific imaging modalities for diagnosis[9]. For these non-invasive nuclear scintigraphic studies, the fetal exposure to radiation is quite low and they are quite safe for both the fetus and the pregnant woman. However, they should be used only strong indication needed[10]. Finally, intraoperative gross and microscopic findings confirmed the diagnosis. Soft solid tissues with various colors (bluish-red, pink, dark pink and greenish black)[2] have been reported in another study.

In our literature review, we did not find any studies on adverse pregnancy outcomes such as diabetes arising in pregnancy, pregnancy- induced hypertension or preterm labor associated with this condition. However, because of a limitation of patient numbers, we could not definitely conclude that no adverse pregnancy outcomes are associated with splenosis during pregnancy. Given our experience, we would now suggest that if a case of splenosis in pregnancy is encountered that all stages of the pregnancy should be monitored as any high risk pregnancy. Fetal growth monitoring during late second or early third trimester of pregnancy should be done. Vaginal delivery should be the first choice, and cesarean section considered only following appropriate obstetric indications. For delivery, a joint operation with both an experienced obstetrician and a general surgeon is suggested, because the operation may prove to be difficult and massive bleeding may be encountered. The skin incision will depend on the patient's general presentation including factors such as the location of the ectopic spleen, fetal position, previous abdominal scars, etc. And finally, a known splenotic woman who is sexually active should be counseled concerning the dangers of pregnancy in her condition, and contraception and future pregnancy planning should be offered.

Conclusion

Splenosis is a rare condition, especially during pregnancy. Our patient is the first report of a pregnancy affected by post traumatic splenosis. We suggest conservatively continuing the pregnancy while offering the appropriate antenatal care as a high risk pregnancy. The decision concerning the route of delivery depends on the patient's condition and proper postpartum contraception and future pregnancy planning should be offered.

Acknowledgements

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Potential conflicts of interest

The authors declare no conflict of interest.

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