Clinical Case Report

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Successful management of a spontaneous viable monochorionic diamniotic twin pregnancy on cesarean scar with systemic methotrexate

A case report

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Abstract

Rationale: Cesarean scar twin pregnancy is exceedingly rare and it remains to be a life-threatening condition even in the early weeks of gestation. Because of its rare occurrence, there is no consensus on the treatment modality of cesarean ectopic pregnancy.

Patient concerns: A 41-year-old, woman, gravida 7, para 6, with a history of low transverse cesarean section 2 years back presented with an estimated 6 weeks gestational age and viable spontaneous twin, monochorionic diamniotic gestation. The patient presented to the Emergency Department with complaints of vaginal bleeding and mild lower abdominal pain for 5 days.

Diagnoses: An ultrasound examination was performed demonstrating a single intrauterine gestational sac with 2 viable embryos (monochorionic diamniotic) implanted in the lower uterine segment at the level of the prior cesarean section scar. A diagnosis of viable cesarean scar twin pregnancy was made.

Interventions: A 2 doses of 50 mg/m^2 methotrexate (MTX) was given intramuscularly. The response to the treatment was monitored by serial beta-human chorionic gonadotropin (β-hCG) and ultrasound.

Outcomes: Patient was followed up with β-hCG weekly levels which became 0 after 68 days of treatment. During the follow-up, the patient was asymptomatic, no side effects of MTX were noticed.

Lessons: We reported a rare case of viable monochorionic diamniotic twin pregnancy on a cesarean scar that was successfully treated with systemic MTX without any additional therapy. Additionally, the decline of β-hCG in twin ectopic cesarean scar pregnancy appears to be similar to a singleton ectopic pregnancy.

Abbreviations: β-Hcg = beta-human chorionic gonadotropin, CRL = crown-rump length, MRI = magnetic resonance imaging, MTX = methotrexate, UAE = uterine artery embolization.

Keywords: cesarean scar, methotrexate, monochorionic diamniotic twin

1. Introduction

Cesarean scar twin pregnancy is exceedingly rare. Ectopic cesarean scar pregnancy is characterized by implantation of the fertilized ovum in the myometrium in the lower segment of the uterus on a previous cesarean scar and is considered to be the rarest form of ectopic pregnancy. It remains to be a life-threatening condition since it can cause uterine rupture and hemorrhage even in the early weeks of gestation.[1,2] With the increasing prevalence of cesarean section rate and use of ultrasonography in early gestation, the rate of cesarean scar pregnancy had been exponentially increasing. The incidence is reported to be extremely low, 1/1800 to 1/2216.[3,4] Because of its rare occurrence, there is no consensus on the treatment modality of ectopic pregnancy within a cesarean scar.

We report the occurrence of a rare case of a viable spontaneous twin, monochorionic diamniotic gestation which was diagnosed as a case of a cesarean scar ectopic pregnancy. It was successfully treated with systemic methotrexate (MTX) without any complication, thus preserving the uterus and the potential of future fertility.

2. Case report

A 41-year-old, woman, gravida 7, para 6, woman, with an estimated 6 weeks gestational age had a spontaneous twins pregnancy, monochorionic diamniotic gestation. The patient presented to the Emergency Department with a history of vaginal bleeding for 5 days. The bleeding was heavy for 2 days and it was associated with clots. She also complained of mild lower abdominal pain and nausea. There was no history of vomiting or other gastrointestinal symptoms, urinary symptoms, colicky

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Approval of ethics committee of the King Fahad Medical City was obtained.

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pain, and fainting attacks. The patient's obstetric history was significant for a prior one low transverse cesarean section due to breech presentation 2 years back, otherwise, she had 5 normal vaginal deliveries. She had no previous history of any illnesses or allergies. She denied the use of any medications. There was no significant family or psychosocial history. Her menarche commenced at the age of 11 years with subsequent regular cycles.

Her body weight was 70 kg, her height was 153 cm, and her BMI was 29.9 kg/m^2. Physical examination was done; she was afebrile with a pulse rate of 85 b/min, blood pressure of 110/73 mm Hg, and a respiratory rate of 19 cycles/min. There were no signs of anemia. Abdominal examination revealed tenderness to palpation in the lower abdomen, but there was no distention, guarding, or rebound. Intestinal sounds were normal. The external genital examination was normal. Bimanual pelvic examination demonstrated a slightly enlarged uterus with a normal uterine cervix and a small amount of vaginal bleeding from the uterus.

The concentration of the β-subunit of hCG was 7616 mIU/mL that was high. She had a hemoglobin of 11 g/dL and hematocrit of 33.6%. Other standard blood test results and urine analysis were in the normal range.

An ultrasound examination was performed demonstrating a single intrauterine gestational sac with 2 viable embryos implanted in the lower uterine segment at the level of the prior cesarean section scar (Fig. 1). The myometrium between the gestational sac and the bladder was very thin, with a thickness of 8 mm. Both embryos' crown-rump length (CRL) corresponded to 6 weeks with cardiac activity. Both the uterus and cervical canal were found to be empty. No free fluid was found in the abdominal cavity. Prominent peritrophoblastic blood flow was demonstrated by Doppler flow sonography. A diagnosis of cesarean scar ectopic pregnancy was made and the patient was counseled regarding different management options and the main goal to indicate the success of the treatment was to bring the levels of β-subunit of hCG to less than 5 mIU/mL.

As the patient was hemodynamically stable, a single dose of 50 mg/m^2 methotrexate was given intramuscularly in an inpatient environment, according to the local protocol. The response to the treatment was monitored by serial β-hCG and ultrasound. Within 7 days of the administration of methotrexate, both fetal hearts were negative but β-hCG was plateauing. Therefore, the second dose of 50 mg/m^2 methotrexate was given. Patient was followed up with β-hCG weekly levels which became zero after 68 days of treatment. During the follow-up, the patient was asymptomatic, no side effects of methotrexate were noticed. The patient went back home and was performing her normal daily activities. Consent for publication of the report and the image was obtained from the patient.

3. Discussion

Early diagnosis of ectopic cesarean scar pregnancy is essential to avoid the complications such as uterine rupture and life-threatening hemorrhage.[5] It is considered to be more aggressive than placenta previa because of its invasion in the myometrium in the first trimester of pregnancy.[6] Beside the fact that it is rare, this entity of ectopic singleton pregnancy in a Cesarean scar such as twin pregnancy carries a high risk of uterine rupture and uncontrollable hemorrhage. To our knowledge, only a few cases reported in the literature with spontaneous twin pregnancy on a cesarean scar. A high index of clinical suspicion is required to diagnose a cesarean scar pregnancy, since up to 40% of patients may be asymptomatic.[7] Alternatively, patients can present with vaginal bleeding, similar to our patient. Symptoms of acute abdominal pain with heavy vaginal bleeding should raise a concern for impending rupture.[8] Availability of transvaginal ultrasound combined with color and pulsed Doppler evaluation

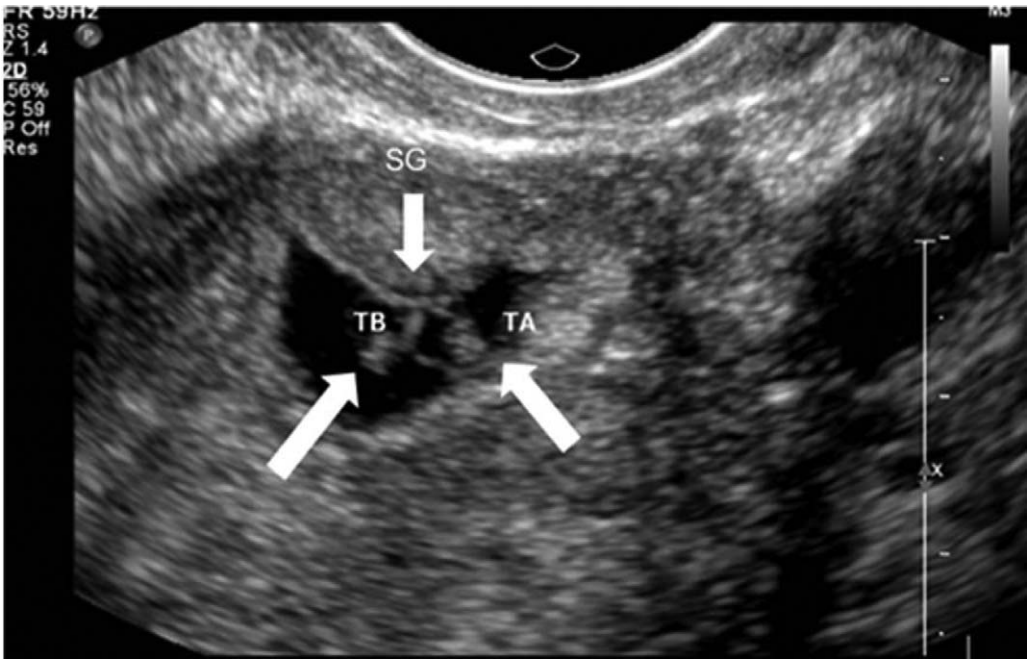

Figure 1. Transabdominal ultrasound showing a single intrauterine gestational sac (arrow head) with 2 viable embryos A and B (arrows) implanted in the lower uterine segment at the level of the prior cesarean section scar. *GS = gestational sac, TA = twin A, TB = twin B.

is a relatively reliable method to diagnose cesarean scar pregnancy. Most authors suggest limiting the use of magnetic resonance imaging (MRI) to cases in which transvaginal ultrasound findings are equivocal, or when the gynecologist requires additional information in preparation for surgical intervention.[8] Majority of scar ectopic pregnancies are case reports or small case series reported in the literature, with no consensus on the preferred mode of treatment. Generally, the patient's clinical symptoms and desire for future fertility, age and size of the gestation, and the clinician's experience dictate which treatment option is most appropriate. Both medical and surgical approaches have proven successful. Systemic and local injection of methotrexate has been described in published reports.[9] Other agents like potassium chloride can be injected directly into the fetus within a cesarean scar pregnancy.[10] Moreover, aspiration of gestational sac has proven successful.[11] Several other surgical approaches, including hysteroscopic approach [12] and suction curettage, have been successfully used. Uterine artery embolization (UAE) combined with intra-arterial MTX infusion[13] and bilateral uterine artery chemoembolization with methotrexate for cesarean scar pregnancy has also been described.[14] The challenge in our patient was that the twins were viable and therefore 2 doses of 50 mg/m^2 methotrexate 7 days apart had to be given. It was successful despite the presence of the fetal heart. Ultrasound within 7 days of the administration of methotrexate, both fetal hearts were negative. In addition, there were no side effects reported by the patient.

4. Conclusion

Systemic methotrexate administration was an effective treatment for our patient who had a viable twin cesarean scar pregnancy in the first trimester thus preserving the uterus and the potential future fertility. The decline of beta human chorionic gonadotropin in twin cesarean scar ectopic pregnancy appears to be similar to a singleton ectopic.

Author contributions

SB: wrote the case description, literature research, drafting of the manuscript, and corresponding author. FK: provided the description of the radiology images. DA-J: critically reviewed and revised the manuscript. All authors read and approved the final manuscript.

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