



Gestational Trophoblastic Neoplasia Arising on Scar Pregnancy: A Case Report

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Abstract

Gestational Trophoblastic Disease (GTD) refers to a group of disorders including partial, complete and invasive molar pregnancy, choriocarcinoma and the less common placental site trophoblastic tumours. Persistent GTD with high β -hCG levels is called Gestational Trophoblastic Neoplasia. A case of complete molar pregnancy in a caesarean scar with complete myometrial infiltration is reported. This is the 4th case described in literature.

Introduction

Gestational Trophoblastic Neoplasia is a persistent Gestational Trophoblastic Disease (GTD), that invades locally or metastasizes, with persistent high level of β -hCG. Cesarean scar pregnancy is an abnormal implantation of pregnancy located in the low and anterior wall of the uterus on the scar of previous cesarean section. Cesarean scar pregnancy is a rare event, but scar molar pregnancy is even rarer. In this report we present a case of complete molar pregnancy in a caesarean scar with complete myometrial infiltration, in literature there are only other three cases described.

Materials and Methods

A 32 years old woman 3 para (2 spontaneous births, 1 caesarean) was admitted at the Emergency Department with abundant vaginal bleeding. She reported Dilatation and Curettage (D&C) about 10 days before for miscarriage. The first β -hCG dosage was 479,255.00 mUI/ml. 48 hours after D&C β -hCG decreased to 207,550 mUI/ml. New dosage, repeated in our Department, resulted 205,883.30 mUI/ml. Physical examination showed enlarged uterus, painless bimanual palpation and abundant vaginal bleeding. Vaginal ultrasound showed empty uterus, mildly thickened endometrial echoes in the fundus (10 mm) (Figure 1) and isthmic, very vascularised (Figure 2), vacuolated, in homogeneous echogenic mass (48 x 18 mm), located on the caesarean hysterotomy scar (Figure 3), with residual myometrial thickness <5 mm. Bladder did not appear involved and was separated from anterior uterine wall. NMR confirmed ultrasound results. Based on persistent bleeding, a new ultrasound-guided D&C was performed. Three days after operation β -hCG was 250,000 mUI/ml, Hb level decreased (9 gr/dL) and bleeding persisted. The patient referred severe abdominal-pelvic pain. Total laparotomy hysterectomy was performed. The bladder was not affected and final histological exam indicated invasive hydatidiform mole which infiltrated left parametrium and the isthmus wall of myometrium full thickness.

Results and Discussion

Notwithstanding Larsen and Solomon described it in 1978, Cesarean Scar Pregnancy (CSP) is still considered a new clinical entity [1]. Some Authors consider CSP a rare form of ectopic pregnancy, others affirm it cannot be defined as extrauterine pregnancy because trophoblastic tissue is in a surgical scar of the uterine cavity [2-6]. Cesarean deliveries increased drastically all over the world, but CSP has extremely low incidence: (1:2216). Out of total women with caesarean section, 0.15% have a CSP in following pregnancies and 6% of extrauterine pregnancies are CSP.

This disease causes morbidity and mortality in fertile women and determines about 9% of pregnancy-related deaths.

No relation was found between the number of caesarean sections and CSP risk. Etiopathogenesis is still unknown; different theories were proposed: the first one affirms that the product of conception reaches the myometrium through a microscopic scar dehiscence or a defective scar [4]. Kliman observed that the product of conception prefers exposed scar tissue without epithelium layer [7].

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Figure 1: Vaginal ultrasound showed empty uterus, mildly thickened endometrial echoes in the fundus (10mm).

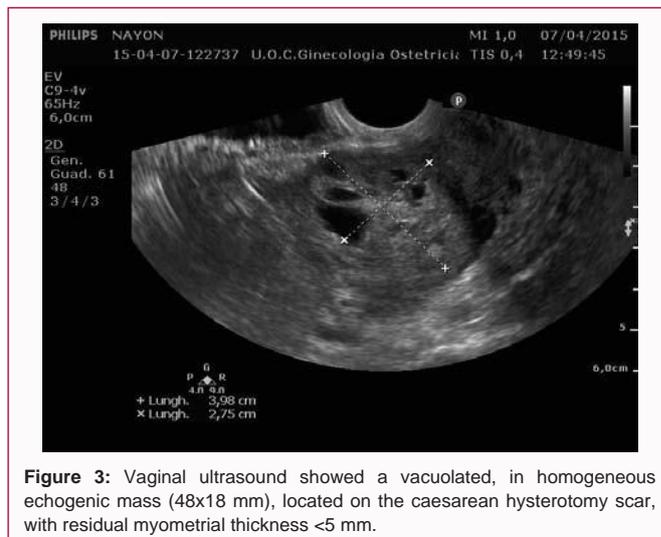


Figure 3: Vaginal ultrasound showed a vacuolated, in homogeneous echogenic mass (48x18 mm), located on the caesarean hysterotomy scar, with residual myometrial thickness <5 mm.

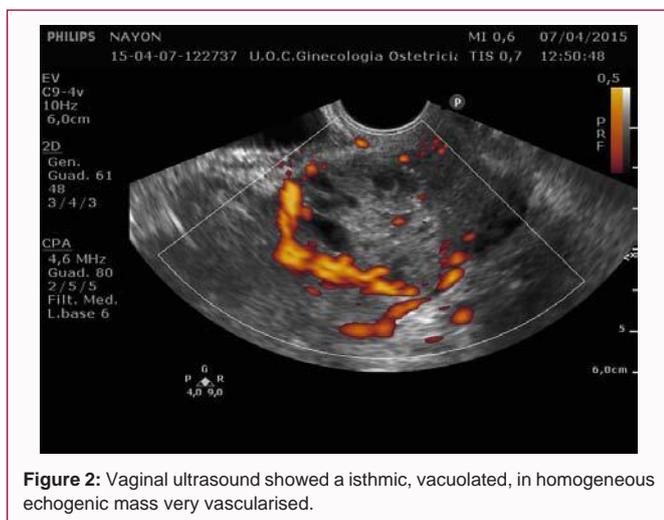


Figure 2: Vaginal ultrasound showed an isthmic, vacuolated, in homogeneous echogenic mass very vascularised.

CSP is rare, but scar molar pregnancy is even rarer. Only 3 cases were described in literature. The first one was a partial molar pregnancy in caesarean scar reported by Wu in 2006 [8]. About 7 days after first D&C, there were persistent vaginal bleeding, high β -hCG levels (30.756 UI/L) and vaginal ultrasound showed vascularised residual tissue on the caesarean scar. Second D&C was performed. Histological test confirmed the presence of chorionic villi and decidual material. β -hCG level gradually went back to normal and spotting disappeared in nine weeks without serious complications.

Michener C and Dickinson JE described in 2009 the second case of molar pregnancy on previous hysterotomy scar [9]. The Patient was initially treated with Methotrexate (MTX) injection into the gestational sac but, due to severe vaginal haemorrhage, urgent hysterectomy was performed. Histological test indicated molar pregnancy.

The third case was described in 2012 by Ko JK: first approach was ultrasound-guided D&C with femoral arteries preparation for surgery [10]. Histological test indicated partial hydatidiform mole.

Caesarean scar pregnancy should be suspected in all fertile women with variable vaginal bleeding with or without abdominal pain or recent D&C with persistent spotting and positive β -hCG levels. Uterine rupture, massive intra-abdominal haemorrhage and

hypovolemic shock are the most severe possible consequences if this disease is not diagnosed. Today, the only tool that allows early and accurate diagnosis vaginal ultrasound, with 85% sensitivity.

Ultrasound criteria for CSP diagnosis are :

- 1) Empty uterine cavity
- 2) Gestational sac located anteriorly at the level of the internal os covering the site of the previous lower uterine segment caesarean section scar
- 3) Evidence of functional trophoblastic/placental circulation on doppler examination
- 4) Negative 'sliding organs sign'

No standard treatment for this disease exists. In case of abundant and uncontrolled vaginal bleeding, hemodynamic instability, uterine rupture with massive hemoperitoneum emergency hysterectomy is certainly recommended. For patients with stable hemodynamic conditions, conservative medical and surgical treatments (alone or combined) were described: systemic and/or local administration of MTX; ultrasound guided D&C; hysterectomy; internal iliac artery ligation and UAE. A number of studies tried to define the best medical or surgical treatment for this rare disease [11-13]. Sole systemic MTX administration has high failure rates (about 57%), whereas UAE with local MTX administration is further more effective. Lian considers it a valid first-line treatment in stable CSP patients [14]. In a cohort study on 119 CSP patients, Gao L, et al. [15] compared the effectiveness of systemic MTX administration followed by D&C and UAE followed by D&C. He demonstrated that very high β -hCG levels do not exclude the possibility of medical treatment aimed at preserving fertility. According to Timor-Tritsch the lower complication rate was registered by medical local intragestational and intramuscular MTX treatment [16]. Chang Y Successfully performed vasopressin injection in the cervical stroma in two patients with subsequent hysteroscopy resection of trophoblastic tissue [17]. Laparotomy/laparoscopy with hysterectomy should be considered as a therapeutic option for all hemodynamically stable women not responding to medical treatments or less invasive surgery.

Conclusions

CSP incidence is rising due to the increase of caesarean sections.

This condition threatens woman's life: this is why the earlier it is diagnosed, the lower is complication rate (uterine rupture, massive haemorrhage and hemodynamic shock). Pelvic transvaginal ultrasound, today considered gold standard, should be recommended to all pregnant women with previous caesarean section for early assessment of gestational sac implantation site. No guidelines on the treatment of these patients exist, but sole systemic use of MTX or D&C does not always control haemorrhage. Waiting is not recommended due to high risk of uterine rupture. It was observed that UAE allows to control intra and postoperative bleeding and could therefore be adopted as therapeutic approach in organised centres. Hysterectomy is the only emergency way to stop bleeding.

The best treatment is the one allowing the removal of pregnancy tissue by preserving patient's fertility.

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