Elif Gul Yapar Eyi*

Minimally invasive procedure for type II canal defect caesarean scar pregnancy with cardiac activity and high hCG titres at 8+2 weeks of gestation

Abstract: Type II caesarean scar pregnancy (CSP) not only poses important maternal hazards, such as severe bleeding, uterine rupture, disseminated intravascular coagulation and maternal death due to its abnormal location and invasive characteristics, but its surgical management may lead to operative complications and even loss of fertility. The sonographic and Doppler findings of a "canal defect CSP" that has previously been hypothesised, but not illustrated, are presented here. A minimally invasive approach was performed in the presence of a 38.3 mm gestational sac (GS) with a crown rump length of 11.3 mm embryo (8+2 weeks of gestation) and cardiac activity with high (118,839.2 mIU/mL) human chorionic gonadotropin (hCG) levels. A transabdominal intragestational sac injection of potassium chloride to stop cardiac activity, and consecutively, methotrexate (MTX) was given before systemic MTX therapy. Embryonic cardiac activity stopped. Systemic methotrexate was repeated 8 days after the procedure. While vaginal bleeding ceased in 3 weeks with gradual shrinkage of the GS, hCG fell to non-pregnant levels within 112 days (16 weeks); complete resolution of the ectopic mass required 8 months. This is the first report presenting the success of a minimally invasive procedure at a hCG level of 118,839.2 mIU/mL with embryonic cardiac activity in type II CSP.

Keywords: Cardiac activity; embryo; human chorionic gonadotropin; intraamniotic injection; methotrexate; scar pregnancy.

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Introduction

Despite efforts to decrease the number of caesarean sections (C/S) among European countries, Turkey has the highest C/S rate (42.7%) [1]. High C/S rates and the use of ultrasonography have contributed to increased reports from Turkey of the occurrence of caesarean scar pregnancy (CSP), the rarest form of ectopic pregnancies [2]. Of many theories that have been proposed to explain the occurrence of CSP, the most widely accepted is that the blastocyst enters the myometrial wall through a microscopic dehiscent canal that may have been created through a trauma, mostly associated with C/S or another uterine surgical procedure [3]. To our knowledge, this theoretical canal has not been depicted sonographically and it has not been added to the sonographic criteria of CSP. Herein, the sonographic and Doppler findings of a "canal defect type II CSP" suggesting the possibility of a minimally invasive procedure at 8+2 weeks of gestation with previously defined high-risk criteria are presented.

Case

A 29-year-old G2P1 woman who had undergone C/S 8 years ago was evaluated for vaginal bleeding at 7 weeks of gestation. The uterine scar was highly prominent and consistent with the previously established criteria of a CSP according to the following findings [3–5]: (i) empty uterine cavity and cervical canal (Figure 1A); (ii) implantation of the gestational sac (GS) in the anterior uterine wall at the isthmus (Figure 1A and B); (iii) presence of a rounded GS with a maximum longitudinal diameter of 38.3 mm, including a yolk sac with a maximum outer to outer diameter of 4.6 mm, and an embryo with a crown-rump length (CRL) of 11.3 mm having cardiac activity (Figure 1B); (iv) the presence of an area of increased peritrophoblastic vascularity revealed by a colour Doppler

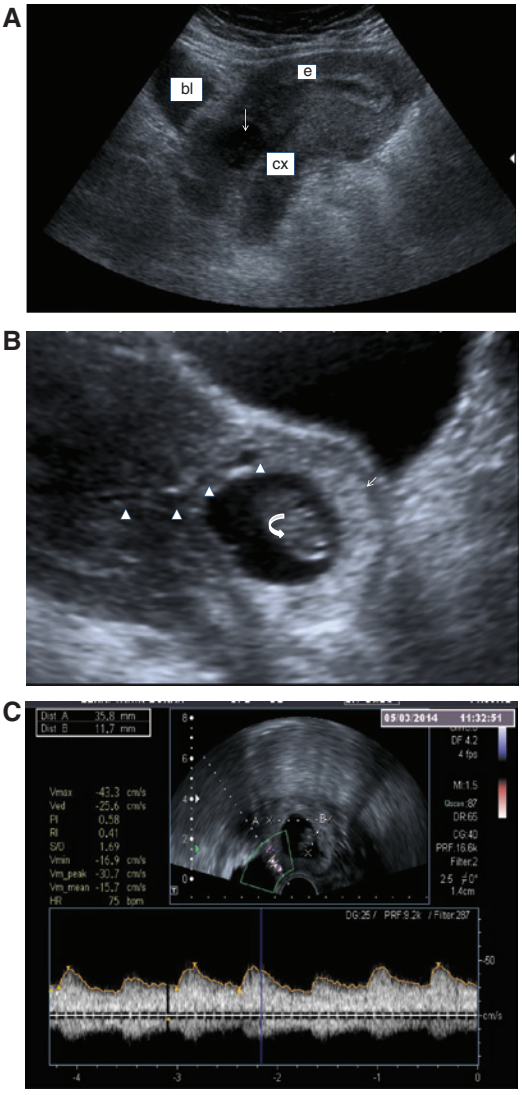

Figure 1: (A) Transabdominal ultrasound image showing the uterus in retroflexion with empty endometrial cavity (e) and echogenic cervical canal (cx). Development of the gestational sac (arrow) in the anterior uterine wall at the isthmus (presumed site of the previous lower segment) behind the bladder (bl) is seen. (B) Transabdominal oblique ultrasound image at $8^{2/7}$ weeks of pregnancy. Bulging of the 38 mm gestational sac (GS) into the bladder is seen. Outer margin of the sac is just 2.3 mm away from the bladder margin. The dehiscent tract generated between the type II scar pregnancy and the endometrial cavity is delineated by the consecutive arrow heads. The distal end of the tract ends at the outer margin of the ectopic GS. CRL: 11.3 mm embryo is shown by the curved arrow. (C) Transvaginal color Doppler ultrasound image (sagittal view). Evidence of functional trophoblastic circulation, defined by the presence of an area of increased peritrophoblastic vascularity; that is high velocity, low impedance trophoblastic flow with a peak systolic velocity: 43.1cm/s, Pulsatility Index: 0.52, Resistance Index: 1.38, systolic to diastolic ratio: 1.61 has been shown.

examination (Figure 1C); (v) the absence of healthy myometrium between the bladder and GS, allowing differentiation from the cervico-isthmic implantation (Figure 1A and B). The distance between the bladder and ectopic sac was measured as 2.3 mm. Moreover, the dehiscent canal at the boundary can be seen (Figure 1B).

The basal human chorionic gonadotropin (hCG) level was 118839.2 mIU/mL (ADVIA Centaur assay manual total hCG 1/1200609933 Rev. D, 2007-03 ThCG; Siemens Medical Solutions Diagnostics, Malvern, PA, USA).

Due to the high risk of uterine rupture and the risk of profuse haemorrhage, and even death, the patient was counselled about the risks of continuing the pregnancy. The patient declined surgical intervention and opted to continue with the pregnancy. At 8+2 weeks of gestation with a GS measurement of 38.3 mm and CRL of 11.3 mm, the patient declared that she was ready for a minimally invasive procedure and gave written informed consent. She was prepared for a usual transabdominal amniocentesis procedure. After confirming the placement of the disposable echo tip amniocentesis needle (JDAN 2020010, 20 cm, 20 gauges; Cook Medical, Limerick, Ireland) in the ectopic GS, initially 2 ml of 10% potassium chloride was injected into the site of embryo. Cardiac activity ceased, then, with another injector, inserted into the same entry site, 2.5 ml 25 mg methotrexate (MTX 50 mg/5 ml flacon; Koçak, Tekirdag, Turkey) was injected into the GS and the needle was withdrawn. The procedure lasted 3 min without anaesthesia and analgesia. Meantime, the patient received (1 mg/kg) 75 mg intramuscular (i.m.) MTX. The sonographic examination was repeated to confirm the absence of a heartbeat and the patient was discharged within 24 h and asked to return after 24 h for follow-up scans and hCG measurements. After the demise of the embryo, though hCG levels fell dramatically (Figure 2), together with the decrease in size of the GS, systemic i.m. MTX (75 mg) was repeated on the 8^{th} day after the procedure due to the high initial levels. The shrinkage of the GS led to a complex heterogeneous mass 6 weeks later (Figure 3A). The hCG levels fell to non-pregnant levels within 112 days (16 weeks); however, a complete resolution of the heterogeneous mass was only achieved after 8 months (Figure 3B).

Discussion

CSP has been reported to have two different types [5]. In type I, the amniotic sac is implanted on the scar with progression towards either the uterine cavity or the cervico-isthmic junction. Type II is characterised by a deep infiltration into the uterine myometrium and bulging from the uterine serosal surface, creating predisposition

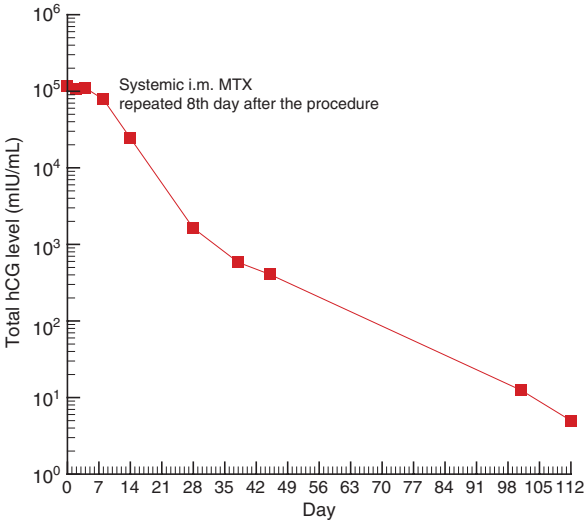

Figure 2: Logarithmic decline in human chorionic gonadotropin (hCG) levels after minimally invasive procedure for type II canal defect caesarean scar pregnancy. i.m.=intramuscular, MTX=methotrexate.

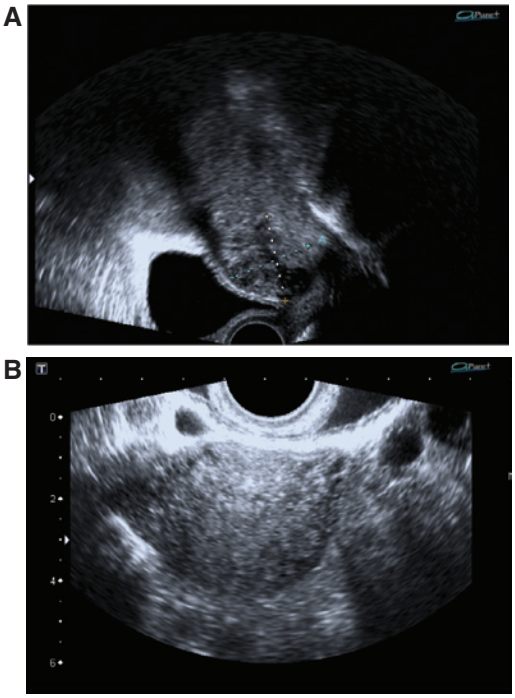

Figure 3: (A) Transvaginal ultrasound image depicting the heterogenous mass after combined therapies. Total hCG was 546mIU/mL in the sixth week after the initial therapy. Trophoblastic flow was still present. (B) Transvaginal ultrasound image demonstrating the resolution of the complex heterogenous mass after 8 months.

to disastrous outcomes, such as uterine rupture, disseminated intravascular coagulation and maternal death if unrecognized and not treated. As women with type II CSP are in a reproductive age, the aims of the therapy include prevention of uterine rupture, prevention of haemorrhage and preservation of fertility. However, there is lack of evidence concerning the treatment approaches. Previously defined criteria, such as lesion diameter, hCG level, peritrophoblastic flow and myometrial thickness anterior to the CSP, may neither be useful in selecting the type of treatment nor of value in predicting the success of the treatment [6]. On the contrary, the individualisation of treatment according to the localisation of the lesion determined by ultrasonography with the most simple procedure and follow-up may be more important to achieve success.

Surgical treatment options are excision of gestational tissues by laparotomy, laparoscopy, hysterectomy, laparoscopic bilateral uterine artery ligation and resection of the scar with gestational tissue and wound repair, laparoscopic bilateral uterine artery ligation and transvaginal resection of the C/S with gestational tissue and wound and robotic assisted laparoscopic repair appear to be complicated and potentially provoking iatrogenic complications compared with the minimally invasive procedure. Complications, in terms of major haemorrhage, uterine rupture and loss of fertility occurred in 62.1% (87/54) of the patients treated with MTX alone, whereas the same complications occurred in 46.9% (64/30) of the patients treated with uterine artery embolization [7]. A mean blood loss of 250±221.4 ml and an average operation time of 85.5±17.5 min have been reported in an analysis of 11 cases of laparoscopic management or laparoscopy combined with transvaginal management of type II CSP [8]. Hysteroscopic resection, and dilatation and curettage, are not suitable for type II CSP because of the localization of the lesion. As the surgical treatment of CSP may be associated with significant complications, a minimally invasive procedure combined with systemic MTX may be an initial choice, even with a hCG level of 118839.2 mIU/mL in the management of the hazardous type II CSP. The necessity of additional systemic MTX on day 8 may be controversial due to the potential side effects and cumulative toxicity associated with the drug, as a decline of more than 15% from the day 4 hCG level had already been achieved on day 8 according to the previously established management criteria for tubal ectopic pregnancy. However, as MTX therapy was not administered to any patient with an initial level of 118839.2 before, a second dose of systemic MTX was considered.

As the rate of C/S increases all over the world, complications associated with C/S scar pregnancy will also increase unless the sonographic diagnosis of C/S scar pregnancy and differentiation of type I and type II scar

pregnancies at first trimester become widely possible. To accomplish this goal, sonographic examination at early first trimester is essential for ascertaining the localization of every pregnancy after C/S, especially for countries with high C/S rates. As there is not a single and uniform management for C/S scar pregnancies, individualisation of a rationalised treatment according to patient prerequisites with a minimally invasive approach under close follow-up is necessary to achieve the best outcome.

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