Heterotopic Caesarean scar pregnancy combined with intrauterine pregnancy successfully treated with embryo aspiration for selective embryo reduction: Case report

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Ectopic pregnancy situated in a Caesarean section scar is a rare but potentially life-threatening event. Because of its rarity, there are no universal treatment guidelines to manage this condition. We report a case of IVF-induced triplet heterotopic pregnancy of early gestational age that included one Caesarean scar pregnancy diagnosed as early as 6 weeks gestation. Treatment with embryo aspiration under vaginal ultrasonography for selective embryo reduction was given and the concurrent intrauterine twin pregnancy was preserved successfully.

Key words: Caesarean scar pregnancy/embryo aspiration/heterotopic pregnancy/selective embryo reduction

Introduction

Caesarean scar pregnancy (CSP) is a rare type of ectopic pregnancy (Herman et al., 1995; Ayoubi et al., 2001). If not diagnosed early and treated it may cause rupture and uncontrollable haemorrhage. The aetiology of this condition is not clear (Godin et al., 1997; Seow et al., 2000). Ultrasonography is used for the diagnosis and follow-up of the progression of the uterine sacculus until its disappearance. Because of the rarity of this event, treatment has not been standardized (Herman et al., 1995; Donald et al., 2002). Heterotopic CSP, a rarer event, causes a clinical dilemma while preservation of intrauterine pregnancy is desired. We report a case of triplet heterotopic pregnancy with one CSP and two intrauterine pregnancies in a patient with secondary infertility who received IVF. In order to preserve the intrauterine fetuses, we used a 16-gauge needle to aspirate the content of gestational sac for embryo reduction in the scar pregnancy without other invasive management and the twin pregnancy was successfully preserved.

Case report

A 38 year old woman, gravida 4, para 2, visited our clinic due to secondary infertility. She had undergone two lower segment transverse Caesarean deliveries due to breech presentation and repeated Caesarean section at 13 and 7 years previously, without any puerperal complication. Controlled ovarian stimulation was initiated with GnRH analogue down-regulation protocol. Thirty-six hours after hCG was administered, a total of 27 oocytes were retrieved. Three embryos were transferred into the uterus under abdominal ultrasound guidance. Pregnancy test was positive 14 days after embryo transfer. One week later, vaginal sonographic examination showed two intrauterine gestational sacs and the other gestational sac that was anterior to the uterine isthmus, with a thin myometrium between the sac and bladder wall (Figure 1), and she had no discomfort. A tentative diagnosis of heterotopic CSP was made. Follow-up study 7 days later was scheduled. Five days later, she visited our hospital due to vaginal bleeding and lower abdominal pain. Pelvic examination revealed a closed cervix and a small amount of blood in the vagina without any sign of active bleeding. Repeated vaginal ultrasound showed the pulsation of the fetus in the extrauterine gestational sac (Figure 2) and the colour Doppler ultrasound demonstrated proliferated growth of peritrophoblastic vessels distributed around the heterotopic gestational sac (Figure 3).

After extensive counselling with the patient about her condition, its risks and the treatment options, a conservative treatment strategy was adopted because of the patient’s strong desire to preserve the intrauterine pregnancy. Under i.v. general anaesthesia using propofol, the vagina was prepared with sterile saline solution, and a 16 gauge-needle (K-OPSD-1635-ET; Cook, Australia) was used to puncture through the vaginal fornix. After introduction of the needle into the gestational sac, we aspirated the contents of the gestational sac under vaginal sonography, resulting in the disappearance of the sac and ~5 ml serosanguious fluid was aspirated smoothly. The patient tolerated the procedure well. She was observed overnight and discharged the following day.
Ultrasound was repeated 2 weeks later and revealed that the gestational sac in the Caesarean scar had shrunk to a small echogenic area. Six weeks later, transvaginal ultrasound showed normal uterine anatomy with complete disappearance of the gestational sac. The two intrauterine pregnancies proceeded without further complication until the 32nd week gestational age, when emergency delivery was performed due to preterm labour.

Discussion
Heterotopic pregnancy is a rare entity which has increased in incidence with the widespread use of assisted reproduction techniques (Abusheikba et al., 2000). Heterotopic CSP is extremely rare, and its occurrence in conjunction with a viable intrauterine pregnancy and the patient’s desire to maintain the intrauterine pregnancy make management difficult. The diagnosis is sometimes not made until uterine rupture occurs and the patient develops haemoperitonium and hypovolaemic shock. Emergency hysterectomy may be the only effective treatment available in such cases. The availability of high-resolution ultrasound and magnetic resonance imaging has made earlier diagnosis of CSP possible, and, if it is treated more conservatively, fertility can be maintained (Godin et al., 1997). The sonographic criteria to diagnose the CSP included that the trophoblast be mainly located between the bladder and the uterine wall (Vial et al., 2000; Jurkovic et al., 2003). In this case, the diagnosis was further confirmed through the use of Doppler sonography, which revealed significant blood flow in the suspected CSP (Figure 3).

The first report of CSP was by Larsen and Solomon (1978); laparotomy was required to evacuate the gestational tissue. Medical treatment with systemic or local injection of methotrexate (MTX), potassium chloride (KCl) and hyperosmolar glucose has been reported (Godin et al., 1997; Ravhon et al., 1997; Roberts et al., 1998; Seow et al., 2000; Shufaro and Nadjari, 2001). Godin et al. (1997) reported successful transvaginal aspiration in a CSP with intrathoracic KCl followed by MTX. Recently, Nawroth et al. (2001) also described the combined effect of local and systemic MTX administration.

There are few case reports in the literature of heterotopic CSP. Some reports suggested that MTX treatment may be associated with spontaneous abortion and may cause congenital abnormality (Feldenkamp et al., 1993; Timor-Tritsch et al., 1998). Salomon et al. (2003) described a heterotopic CSP with one embryo implanted into the uterine cavity, and the other located in the anterior isthmus wall which was successfully managed with KCl injection into the area of the embryo to terminate the CSP at 8 weeks gestation. As we know, during injection of KCl, the fetus is sometimes pushed away from the needle and KCl diffuses into the amniotic sac, consequently it may diffuse to the adjacent sac. Toxic effects of KCl on the remaining fetuses have been reported by Tabsh et al. (1990) and Wapner et al. (1990). Our patient received IVF treatment and successfully conceived two intrauterine pregnancies coexisting with a CSP. This patient emphasized the need for preserving the intrauterine twin pregnancy and we opted for a
more conservative treatment approach. Because of the teratogenic effect of MTX and the potentially toxic effect of KCl on the remaining fetus, the treatment was more complicated. Ravhon et al. (1997) used i.m. MTX to treat a CSP at 8 weeks of gestation and aspiration of the fluid in the gestational sac was performed 9 weeks later after the MTX injection due to prolonged vaginal bleeding. They proposed that because the placenta is implanted mainly on fibrous tissue, absorption of the gestational sac is extremely slow and fine needle aspiration of the remaining fluid in the sac may be required. In this case, the CSP was in the 6th gestational week; the small embryonic size at this early stage of gestation makes the embryo more fragile, thus the chance of successful aspiration without the use of cardiotoxic agents may be higher. We used a technique of embryo aspiration of the CSP under vaginal sonography and successfully preserved the intrauterine twin pregnancy. If the treatment were delayed to later gestational age, it is hard to say if this technique would still work or alternatively cause a catastrophic event, because the risk of rupture and hemorrhage directly correlated with the duration of pregnancy (Lam et al., 2002). In conclusion, it is important to perform transvaginal ultrasound ~5–6 weeks gestation to rule out the possibility of an abnormal pregnancy for the patient.

In view of the increasing use of Caesarean section and of assisted reproduction techniques, obstetricians should be aware of the possibility of this type of heterotopic pregnancy. This case suggests that when the diagnosis of CSP with additional intrauterine pregnancies is made at an early gestational age, fetal reduction of the CSP by embryo aspiration without any other rescue may be an effective alternative which does not result in undue risk of trauma to the intrauterine pregnancy.

References

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