ABSTRACT

Heterotopic cervical pregnancy is an uncommon condition, with a rising incidence due to the increasing number of pregnancies resulting from in-vitro fertilization (IVF). Although it is associated with maternal-fetal complications, there is no consensus in the literature about the best approach for this condition. This study aims to report a case of cervical heterotopic gestation after IVF in which the intrauterine pregnancy was preserved, with spontaneous elimination of the cervical gestational sac after patient sedation and introduction of the vaginal speculum. In addition, we reviewed the literature on the subject, which demonstrated that most cases have a favorable outcome, especially after treatment with surgical excision of the cervical pregnancy. The growing body of evidence is still scarce to define the best treatment for this condition.

Keywords: cervix uteri, in vitro fertilization, pregnancy, heterotopic

INTRODUCTION

Heterotopic pregnancy refers to the presence of simultaneous pregnancies at two different implantation sites, which commonly consists of an association between intrauterine (IU) and ectopic pregnancies. The uterine cervix represents the most rare site of implantation of an heterotopic pregnancy, with an estimated incidence of 1:30,000 pregnancies (Reece et al., 1983). However, this number is rising, which is thought to be associated with the growing popularity of assisted reproduction technology, reaching a frequency of 1% of the pregnancies resulting from these methods (Dibble & Lourenco, 2016).

Heterotopic cervical pregnancy is a rare event and its occurrence is related to a higher incidence of maternal-fetal complications, such as prematurity, severe bleeding and emergency hysterectomy (Molinaro & Barnhart, 2018; Kim et al., 2012; Tsakos et al., 2015). Considering that most of the affected patients were submitted to in vitro fertilization (IVF) due to infertility, treating cervical pregnancy maintaining the IU embryo and the patient’s fertility becomes a priority and a challenge to the practice of obstetrics.

The current literature on heterotopic cervical pregnancy is still scarce and there is no consensus about the best treatment for this condition. Therefore, this study aims to report a case of a post-IVF heterotopic cervical pregnancy whose treatment enabled the preservation of IU pregnancy. In addition, we present a review of the recent literature on the subject in order to help establish standards of treatment for this increasingly frequent condition in medical practice. About to our knowledge this is the first case described in Brazil.

CASE DESCRIPTION

A 39-year-old woman from Rio de Janeiro - Brazil sought specialized medical care early in 2015 due to secondary infertility. She reported a history of 5 previous spontaneous abortions with gestational age of around 5/6 weeks, including a tubal pregnancy (right tube), which was treated with methotrexate (MTX). In addition, she had once undergone a hysteroscopic myomectomy. The patient presented a transvaginal ultrasound (US) without significant alterations and a hysterosalpingography showing bilateral tube patency and mucosal thickening of the right tube infundibulum.

The initial diagnostic evaluation consisted of serologic tests (toxoplasmosis, rubella, cytomegalovirus, HIV, syphilis and viral hepatitis B and C), antithyroid antibodies (anti-TPO and anti-thyroglobulin), screening for thrombophilia, hysteroscopy and karyotype of the couple. None of the exams revealed clinically significant changes.

Between July 2015 and July 2016, the patient was submitted to three IVF procedures after ovulation induction with gonadotropins and none of them resulted in pregnancy. Therefore, oocyte donation therapy was proposed. In March 2017, two donated frozen embryos were transferred with no success. In October 2017, she was submitted to a new IVF cycle with oocytes from another donor and six embryos were obtained. The transfer of the first two embryos did not result in pregnancy. At this point, an Endometrial Receptivity Array (ERA) was performed, which showed a receptive endometrium. Then, two more embryos were transferred with no pregnancy outcome. Thereafter, it was decided to empirically use immunoglobulin during the transfer of the last two embryos and we finally had a pregnancy.

The first trimester transvaginal US showed an embryo compatible with 7 weeks and 5 days of gestational age, normoimplanted, with a heartbeat, associated to a hypoechogenic endocervical image. Since the patient was asymptomatic, we opted for expectant management, with a new US in 2 weeks. The following US (Figure 1) showed a 10-week intrauterine embryo and an endocervical hypervascularized echogenic image of 19 x 17 mm, compatible with a cervical gestation.

The proposed treatment would be the selective reduction of ectopic gestation with US guided Chloride Potassium (KCl) injection in the cervical gestational sac under general anesthesia. However, after anesthetic induction and introduction of the vaginal speculum to perform the procedure, there was spontaneous elimination of the cervical embryo (Figure 2), with preservation of the IU pregnancy. Discrete cervical curettage was performed for the excision.
Thus, we performed a review of the recent literature on cervical heterotopic gestation, aiming to analyze its epidemiology, the different forms of treatment and its relation with a favorable outcome. We used the PubMed database and searched for the terms “pregnancy, heterotopic” and “cervix uteri”, as well as later active search in the bibliographic references of the selected papers. We found 37 case reports of cervical heterotopic gestation, in the English and Spanish languages, in addition to the present report (Table 1). The reports vary from year 1989 to year 2018 and there was no case report from Brazil.

Most of the case reports (n=31; 81.6%) resulted from IVF, in comparison to a minority (n=6; 15.8%), which resulted from spontaneous conception, and in one case the form of conception was not specified. Maternal age at diagnosis ranged from 25 to 45 years, with a mean of 34.6 years. The gestational age at the time of diagnosis ranged from 5 to 12 weeks, and except for Bayati et al. (1989), in which the diagnosis was made by direct visualization during the surgical approach, all cases were diagnosed via the US. As for clinical presentation, a small percentage was asymptomatic (n=12, 31.6%), and most of the cases presented vaginal bleeding at the time of diagnosis.

Regarding the outcome (Graph 1), 29 cases aimed to maintain the IU pregnancy, opting for selective interruption of cervical pregnancy. Out of these, 26 resulted in live birth - nine of which were delivered in abortion; two resulted in abortion and in one case, the pregnancy was still ongoing at the time of reporting. In eight cases, the two pregnancies were intentionally interrupted, and one of the reports did not specify the clinical outcome. The main complications were prematurity (n=9, 23.7%), severe bleeding with the need for blood transfusion (n=6, 15.8%), and emergency hysterectomy (n=4, 10.5%), in addition to cervical hematoma, placenta accreta, chorioamnionitis, intrauterine growth restriction (IUGR), and disseminated intravascular coagulation (DIC).

Concerning the treatment, considering the cases which aimed to maintain the IU pregnancy (n=29) (Graph 2), the majority (n=23; 79.3%) preferred surgical excision of the cervical pregnancy. From most to least common, the surgical approaches included aspiration (n=12), extraction with forceps (n=6), cerclage (n=6), cervical curettage (n=5), Foley catheter insertion (n=4) and electrocauterization (n=3). In most cases, the procedures were guided by US, and in 10 cases, a combination of surgical treatments was performed. In eight cases, an association between surgical evacuation and intra-amniotic injection of KCl (n=4), MTX (n=2), hypertonic glucose (n=1) or sodium chloride (n=1) was used. In five cases, no surgical cervical evacuation was performed, with local injection of KCl (n=4) and MTX (n=1) - in one case there was an association with uterine artery embolization. It is noteworthy that the insertion of a Foley catheter, electrocautery and cervical injections of glucose and sodium chloride were never chosen as the single therapy, but rather combined with other methods. Of these 29 cases, in 27 the IU pregnancy was successfully maintained (Graph 2), and two cases resulted in abortion - one had been treated with an association of KCl injection and uterine artery embolization, and another with aspiration alone.

On the other hand, a minority of cases (n=8) opted for intentionally interrupting both cervical and IU pregnancies. Those were treated with systemic MTX (n=6), uterine artery embolization (n=3), uterine curettage (n=3), aspiration (n=2) and Foley catheter insertion (n=1). In most cases, a combined therapy was performed, and there was one report of exclusive treatment with systemic MTX.

In the current report, we present the first Brazilian report of cervical heterotopic pregnancy after IVF. The patient presented with secondary infertility, with history of
| Author (year)          | Maternal age | Method of conception | Gestational age at diagnosis | Treatment                                                                 | Pregnancy outcome | Presenting symptom / Complications                                                                 |
|-----------------------|--------------|----------------------|-----------------------------|----------------------------------------------------------------------------|------------------|---------------------------------------------------------------------------------------------------|
| Current paper 39      | 39           | IVF                  | 7w 5d                       | Spontaneous elimination of the CG after patient sedation, cervical (US guided) | C-section (39w)  | Asymptomatic                                                                                      |
| Saito et al., 2018    | 39           | IVF                  | 5w 2d                       | CG extraction with forceps (US guided)                                     | C-section (36w)  | Vaginal bleeding / Blood transfusion, total placenta accreta, hysterectomy                        |
| Punhani et al., 2016  | 34           | IVF                  | 8w                          | Local injection of KCI, uterine artery embolization, MTX IM, curettage      | Intentional interruption | Vaginal bleeding / Blood transfusion                                                               |
| Pinto et al., 2016    | 35           | IVF + ICSI           | 7w 2d                       | Cervical curettage (US guided)                                             | C-section (39w)  | Asymptomatic                                                                                      |
| Subedi et al., 2016   | 33           | IVF                  | 5w                          | Uterine artery embolization, hysteroscopic removal with forceps             | Intentional interruption | Vaginal bleeding                                                                                   |
| Elena et al., 2016    | 37           | IVF                  | 5w                          | Aspiration and cervical curettage, cerclage                                | Vaginal delivery (35.4w) | Asymptomatic / Bleeding, pelvic pain                                                              |
| Tsakos et al., 2015   | 41           | IVF                  | 5w 3d                       | CG aspiration, Foley catheter insertion, cerclage                           | C-section (38w)  | Vaginal bleeding, pelvic pain / Triplet gestation: association with tubal gestation                |
| Lin et al., 2013      | 32           | IVF                  | 8w                          | Cervical curettage, electrocauterization                                    | Maintenance of IU pregnancy (did not specify) | Vaginal bleeding, pelvic pain / Triplet gestation: association with tubal gestation                |
| Uysal & Uysal, 2013   | 31           | Spontaneous          | 6w                          | Local injection of KCI, CG aspiration, Foley catheter insertion, cerclage    | Delivery (38w)   | Vaginal bleeding                                                                                   |
| Moragianni et al., 2012 | 40             | IVF                  | 7w 3d                       | CG extraction with forceps, Foley catheter insertion, cerclage              | C-section (39w)  | Vaginal bleeding                                                                                   |
| Kim et al., 2012      | 36           | IVF                  | 5w 2d                       | CG extraction with forceps (US guided)                                     | C-section (40w 5d) | Vaginal bleeding                                                                                   |
| Deka et al., 2012     | 38           | IVF                  | 11w 1d                      | Local injection of KCl and MTX                                             | C-section (36w 4d) | Vaginal bleeding                                                                                   |
| Feaching bauer et al., 2011 | 25                | Spontaneous / Stimulation (clomiphene citrate) | 9w                          | CG aspiration (US guided), cerclage                                        | Vaginal delivery (39w 3d) | Vaginal bleeding                                                                                   |
| Sijanovic, et al., 2011 | 30             | Spontaneous          | 6-7w                        | Local injection of MTX                                                    | Vaginal delivery (39w) | Vaginal bleeding                                                                                   |
| Sánchez-Ferrer et al., 2011 | 33               | IVF                  | 6w 5d                       | Intra-arterial injection of MTX, uterine artery embolization                | Intentional interruption | Vaginal bleeding                                                                                   |
| Hafner et al., 2010   | 34           | IVF                  | 6w                          | Foley catheter insertion (US guided), cerclage, ligation of descending cervical branches of the uterine arteries, systemic MTX | Intentional interruption | Vaginal bleeding                                                                                   |
| Shah et al., 2009     | 34           | IVF + ICSI           | 7w                          | CG aspiration (US guided), prophylactic placement of hypogastric arteries occlusion balloons before delivery | C-section (37w)  | Vaginal bleeding / Bleeding after aspiration                                                      |
| Hoshino et al., 2009  | 37           | IVF                  | 6w                          | CG extraction with forceps and curettage (US guided)                       | C-section (38w)  | Vaginal bleeding                                                                                   |
| Kim et al., 2009      | 30           | Spontaneous          | 8w                          | CG aspiration (US guided), Foley catheter insertion                        | C-section (37w)  | Vaginal bleeding / Bleeding during aspiration, cervical hematoma                                  |
| Majumdar et al., 2009 | 36           | IVF                  | 7w 5d                       | Local injection of KCI (US guided)                                         | C-section (31w)  | Vaginal bleeding / IUGR, acute fetal distress                                                    |
| Nitke et al., 2007    | 45           | IVF                  |                              | Uterine arteries MTX injection                                             | Intentional interruption | Vaginal bleeding / Triplet gestation: 2 cervical gestational sacs                                 |
| Suzuki et al., 2007   | 35           | IVF                  | 5w 3d                       | CG aspiration and local injection of 33% glucose solution                  | C-section (34w)  | Asymptomatic / Bleeding, cervical hematoma                                                        |
| Prorocic & Vasiljevic, 2007 | 31            | IVF                  | 6w                          | CG aspiration and local injection of hypertonic solution of sodium chloride | Normal gestational course until 12w (time of report) | Vaginal bleeding / Triplet gestation: 2 IU embryos                                                |
| Honda et al., 2005    | 40           | IVF + ICSI           | 6w                          | Local injection of vasopressin, cervical curettage, local injection of MTX | Delivery (38w)   | Vaginal bleeding                                                                                   |
| Feinberg & Confino, 2004 | 35             | IVF                  |                              | CG electrocauterization (US guided) and extraction with forceps            | Term vaginal delivery | Vaginal bleeding                                                                                   |
| Gyamfi et al., 2004   | 34           | IVF                  | 6w                          | Local injection of KCI and aspiration                                       | C-section (31w)  | Vaginal bleeding, back pain / Bleeding, hysterectomy, blood transfusion                           |
| Author(s) and Year | Number of Cases | Type of Pregnancy | GA | Intervention | Outcome |
|--------------------|----------------|------------------|----|--------------|---------|
| Kumar et al., 2004 | 32             | Spontaneous      | 7w | Local injection of KCl (US guided) | C-section (35w) |
| Jozwiak et al., 2003 | 37             | IVF+ICSI         | 7w 4d | CG electro-cauterization (US guided), cerclage (12w) | C-section (38w) |
| Olah, 2003          | 34             | IVF              | 12w | Local injection of KCl | C-section (36w) |
| Porpora et al., 2003 | 29             | Spontaneous      | 6w  | CG aspiration (US guided) | Abortion (1d after aspiration) |
| (Seow et al., 2002) | 29             | IVF              | 5w  | CG extraction with forceps (US guided) | C-section (37w) |
| Mashiach et al., 2002 | 34           | IVF              | 8w 3d | Cerclage (Shirodkar) | Vaginal delivery (39w) |
| Chen et al., 2001   | 35             | IVF              | 7w 4d | CG aspiration (US guided), local injection of KCI, cerclage (10w) | C-section (38w) |
| Al-azemi et al., 1999 | 32         | IVF              | 6w  | Local injection of KCl and MTX (US guided), aspiration | C-section (30w) |
| Honey et al., 1999  | 37             | IVF              | 7w 4d | Uterine arteries embolization, local injection of KCI | Abortion |
| Bratta et al., 1996 | 30             | -                | 7w  | MTX IM (2x) | Intentional interruption |
| Peleg et al., 1994  | 35             | IVF              | 7w  | MTX IM, uterine arteries MTX injection, uterine curettage | Intentional interruption |
| Bayati et al., 1989 | 38             | IVF              | 11w 2d | Uterine aspiration and curettage, ligation of cervical artery | Intentional interruption |

CG=cervical gestation; DIC=disseminated intravascular coagulation; ICSI=intracytoplasmic sperm injection; IUGR=intrauterine growth restriction; IM=intramuscular; IU=intrauterine; IVF=in vitro fertilization; KCI=Chloride Potassium; MTX=methotrexate; US=ultrasound

Graph 1. Heterotopic cervical pregnancies outcome (n=38)

Repeated abortion, who underwent 6 cycles of IVF. Only after few transferred blastocysts, with a donor oocyte, the pregnancy was achieved. The patient was asymptomatic when we made the diagnosis of cervical heterotopic pregnancy, at the tenth week of gestational age. We chose to selectively reduce the cervical embryo with KCl injection guided by US, aiming to maintain the IU gestation. However, after patient sedation and introduction of the vaginal speculum, there was spontaneous elimination of the cervical embryo, with preservation of IU gestation.

Finally, according to the current literature, most cases of cervical heterotopic pregnancy have a favorable outcome, resulting in live birth and maintenance of maternal fertility after selective reduction of the cervical embryo.
However, it is possible that this analysis is overly optimistic, since experiences with an unfavorable outcome are more unlikely to be reported.

CONCLUSION

In conclusion, according to recent literature, it may be suggested that cervical heterotopic pregnancy should be treated through US-guided surgical excision associated or not with KCl intra-amniotic injection, once most of the favorable outcomes have been achieved in this manner. However, it is worth mentioning that the number of literature reports is still insufficient to safely establish the best treatment for this condition. Thus, the treatment should be individually chosen considering the patient’s will to maintain the IU pregnancy and the personal experience of the doctor in charge.

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest

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