



Document heading doi:

Successful pregnancy by IVF in a patient with congenital cervical atresia

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ARTICLE INFO

Article history:

Received 3 February 2015

Received in revised form 10 April 2015

Accepted 15 May 2015

Available online 20 September 2015

Keywords:

Pregnancy

IVF

Congenital cervical atresia

ABSTRACT

Congenital cervical atresia and hypoplasia are rare abnormalities that generally require reconstructive or extirpative procedures to relieve outflow tract obstruction. Infertility is a common sequel, and only four previous pregnancies have been reported. We report a case of successful pregnancy after *in-vitro* fertilization in a 32-year-old patient with congenital cervical atresia diagnosed at the age of 28 years. She was referred to our unit and had a successful pregnancy after *in-vitro* fertilization. Caesarean section was performed at 38 weeks gestation. A healthy male baby weighing 3 650 g was safely delivered.

1. Introduction

Congenital cervical atresia and hypoplasia are rare abnormalities caused by abnormal development of Mullerian system, they may arise as a result of abnormal fusion of the mullerian ducts with the urogenital sinus, imperfect canalization of the lower mullerian system, or segmental atrophy of a normally formed mullerian system[1]. They may occur in alone or in conjunction with other abnormalities, such as vaginal agenesis, bicornuate uterus, or didelphic uterus. A high incidence of endometriosis has also been associated with obstructive mullerian abnormalities, particularly if the obstruction is not relieved before age 20.

Primary amenorrhoea and cyclic abdominal pain related to haematometria and retrograde menstruation are the most common clinical presentations, usually occurring after menarche.

The aims of the treatment in this circumstance are mainly to relieve the symptoms, and to restore fertility and regular menstruation. Corrective surgical procedures have been performed to relieve the symptoms[2]. However, many complications such as intra-abdominal infection or re-stenosis of the neocanal can be observed, hysterectomy eventually cannot be avoided in most cases.

It seems that pregnancy is a dream for patients with partial or complete cervical atresia.

Successful pregnancy in those patients is a great challenge for assisted reproductive techniques and reproductive medicine.

2. Case report

In 2013, a 32-years-old woman was referred to our department of obstetrics and gynecology with the complaints of infertility for 4 years. Tracing back her past history, there had been a normal onset of the larche and pubarche at age 12.

Semen analysis for her husband was normal. Pelvic examination revealed a completely atresic cervix. Pelvic ultrasound and the hysterosalpingography had visualized a uterus with normal measurements and morphology. Both ovaries were normal and the bilateral Fallopian tubes were present.

The patient and her husband were elected to proceed with in-vitro fertilization(IVF) after 4 years of infertility.

An ovarian stimulation cycle was carried out using a long protocol with a total dose of 2025 UI of recombinant gonadotrophin (225UI/day during 9days). Transvaginal oocyte retrieval under ultrasound guidance was performed 36h after intra-muscular injection of recombinant human chronic gonadotrophin(r HCG).

Twelve oocytes were fertilized via IVF cycles before uterovaginal

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canalisation. After 48 h, 3 embryos at the 4 cell-stage were selected for transvaginal embryo transfer.

Seven weeks later, vaginal ultrasound found an intra-uterine gestational sac with fetal heartbeat.

The evolution of pregnancy had been simple without complications.

An elective caesarean delivery was performed at 38 weeks through a transverse lower uterine incision, resulting in a healthy male infant weighing 3 650 g.

3. Discussion

Congenital abnormalities of the cervix such as complete agenesis atresia and partial atresia (or hypoplasia) form a spectrum of related clinical disorders, differentiated by the presence of cervical stroma and epithelium[3].

Although 4.8% of women with an absent vagina may have a functioning uterus, the occurrence of a normal vagina and uterus in the presence of complete or partial cervical atresia appears to be rare, as less than 50% previous cases have been reported[3,5].

Women with a complete genital outflow tract obstruction characteristically present with primary amenorrhea and cyclic abdominal pain. The current case is unique, as it appears that with sufficient distending pressure, the incompletely canalized cervical canal ultimately perforated, leading to the first spontaneous menses at age 21 and abviating the need for surgical repair[6].

Various methods of cervical reconstruction have been described in an attempt to create an epithelialized uterovaginal fistula to allow cyclic menstruation. However many post-operative complications are observed: frequent reoperations, and high incidence of possible hysterectomy.

Several factors may contribute to infertility in women with corrected cervical atresia, including deficient cervical mucus production, absence of functioning endometrium, hematometria formation, severe progressive endometriosis, and postoperative adhesive formation following surgical correction of the disorders[7].

However, recent advances in assisted reproductive technologies may afford a better opportunity to achieve pregnancy in patients with complete or partial cervical atresia[9].

Nevertheless, both cases received cervical reconstruction operations before pregnancy. Here we present a case with complete cervical atresia that achieved pregnancy after IVF cycles before uterovaginal canalisation[7,8].

Once implantation occurs, cervical competency and the value of prophylactic cerclage remain to be determined[10].

Two of the five reported patients had an abdominal cerclage inserted at 11-12 weeks gestation because of marked cervical shortening or a palpable defect where the cervical canal had existed[4]. All five reported cases were delivered by elective cesarean. In our case, cervical integrity had not been compromised and reconstructive surgery had not been required.

Spontaneous conceptions rarely occur following correction

of outflow tract obstruction; however, judicious use of assisted reproductive techniques may facilitate the establishment of pregnancy in such women[9].

In summary, our case suggests that successful pregnancy in patients with congenital cervical atresia but functional uterus could be achieved by ART, no matter whether cervical reconstruction could be achieved or not. Hysterectomy is not the first option for managing these patients unless medical treatment and uterovaginal canalization have been unsuccessful.

With appropriate evaluation and an individualized management plan, we believe that a successful pregnancy outcome may be achieved in selected women with congenital cervical abnormalities.

Conflict of interest statement

We declare that we have no conflict of interest.

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