Successful pregnancy after uterovaginal anastomosis in patients with congenital atresia of cervix uteri

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Summary

We present a case of successful pregnancy after effective uterovaginal anastomosis in a 26-years-old patient with congenital atresia of the cervix uteri. She spontaneously achieved pregnancy after four years of uterovaginal anastomosis. Gestation was at the eighth lunar month and the delivery was done by cesarean section due to rapidly progressing fetal asphyxia. The patient gave birth to a live healthy male, weighing 1,950 g, with an Apgar score of 5 and 8 at 1 and 5 min, respectively. The postoperative course was uneventful, and leakage of lochia was normal.

Key words: Congenital cervical atresia; Uterovaginal anastomosis; Pregnancy.

Introduction

Congenital cervical atresia is a rare anomaly of the female genital tract. The incidence of Mullerian hypoplasia/agenesia-type I anomalies is approximately 0.1% in the general population, representing approximately 3% of all uterine anomalies [1]. In clinical practice, the aims of treatment of this condition are firstly to relieve the symptoms related to hematometra and retrograde menstruation, and secondly to restore fertility and regular bleeding. Conservative surgical treatment such as cervical canalization, cervical reconstruction and uterovaginal anastomosis of women with these anomalies has remained controversial [2]. Successful pregnancy in a patient with congenital cervical atresia is a great challenge with respect to techniques and reproductive medicine [3]. To our knowledge, reports of successful pregnancies achieved spontaneously after effective uterovaginal anastomosis are rare.

Case Report

The patient, a 21-year-old female, was admitted to the reproductive unit of our clinic with the diagnosis: primary amenorrhea, atresia of the uterine cervix, hematometra, and hematosalpinx, with virgo intacta. Based on this history, it was decided to proceed with surgery. In order to build a functional uterovaginal passage the operation was performed using combined transabdominal and transvaginal approaches. LCS scissors (i.e., involving the use of an ultrasound (US) harmonic scalpel) were used to resect the atretic uterine cervix. The resected part of the atretic cervix was sent for histopathological analysis. Anastomosis and sutures were carried out between the isthmic part of the uterus and vagina using single circle monocryl sutures. The uterine cavity was drained by a Foley catheter, which remained in the uterus for seven days. The surgical procedure was without complications. Four years later the patient presented again at the reproductive unit. She received estradiol valerate, levonorgestrel (Cyclo-Proginova 2 mg) tablets 21 days per month for six months each year. She visited the clinic because she was married and wished to become pregnant. Over the previous six months the patient had had regular menstrual cycles lasting 28 days. Repeated measurements of LH, FSH, estradiol, progesterone and prolactin concentrations were three times those measured during the previous menstrual cycle, on the 3rd, 11th and 21st days of the cycle. These results confirmed a normal ovulatory cycle, and her partner's semen analysis was normal. US examination performed on the 11th day of the menstrual cycle revealed a normal-sized uterus, a three-layer endometrium structure that was 11 mm in diameter, and normal ovaries. A pre-ovulatory follicle that was 25 mm in diameter was seen on the left side. Four months later, she presented at the clinic again because she had missed her menstrual period. US examination of the vaginal wall revealed an intrauterine gestational sac with embryonic echo 6.24 mm in diameter and presence of heartbeats. The age of pregnancy was six weeks and three days, which corresponded to the length of amenorrhea. Serum β-hCG concentration was 5588 U/l. For luteal support, 600 mg micronized progesterone (Utrogestan capsules, 100 mg) was administered daily, vaginally. Follow-up was scheduled at three-week intervals. On follow-up by clinical examination at 14 weeks of pregnancy, the cervix was fixed, closed and was palpable to a total length of approximately 0.5 cm. We concluded that cervical cerclage was not necessary. As there was a risk of premature delivery, maturation of the fetal lungs was induced using weekly dexamethasone 8 mg (dexametason) IM injections on three successive days and repeated again three weeks later. Then, the 26-year-old patient was admitted to our hospital at eight lunar months of pregnancy, exhibiting symptoms of premature membrane rupture. Upon examination, the uterus corresponded to the length of amenorrhea, with no contraction. Leakage of amniotic fluid and fetal heart tachycardia were present. The fetus was in the breech position. The vagina was 5 cm in length and the cervix was not palpable but was dilated to 1.5 cm. Based on US parameters, the pregnancy corresponded to 33.4 weeks of pregnancy. The patient was admitted to the high-risk pregnancy unit, and because of rapidly progressing fetal asphyxia, cesarean section was performed a few days later. The only difficulty during the operation was that the bladder was lifted high in front of the uterus. A healthy male weighing 1,950 g with an Apgar score of 5 and 8 at 1 and 5 min, respectively, was born. The postoperative course was uneventful, and leakage of lochia was normal.

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Discussion

Obstructive uterine anomalies usually present with abdominal pain and a pelvic mass at the time of menarche. The malformation described in our patient, according to the VCUAM classification, belongs to type C 2 a, including all cases of congenital hypoplasia/agenesia of the uterine cervix [4]. The diagnosis of a high barrier by clinical, transabdominal and transrectal US examination, magnetic resonance imaging and laparoscopy is important in the diagnosis of this type of anomaly [5-8]. Patients with cervical agenesis associated with or without vaginal aplasia can have serious health problems, and typically lack reproductive function [9, 10]. The literature contains multiple reports of various conservative surgical techniques that have been used to deal with these malformations. However, consensus regarding an optimal technique is still lacking. Cervical obstruction has been most frequently solved by forced formation of canals, uterovaginal canalization aided by intraoperative US or endoscopically monitored canalization, in order to maintain the connection between the cavity of the uterus and vagina [11-13]. The surgical approach leads to normal menstrual bleeding, resolution of cyclic pelvic pain, and frequently preserved fertility [14]. Preservation of the uterus provides the opportunity to achieve pregnancy spontaneously or by applying artificial reproductive technology techniques [15]. One of the operational techniques of establishing communication between the vagina and uterus is uterovaginal anastomosis. Uterovaginal anastomosis and cervical reconstruction have been proposed and described by several authors [3, 16, 17]. Often, very shortly upon this intervention, stricture of the newly formed canal occurs, and cervical permeability typically fails. Patients generally have severe complications because of infection, adhesions or retrograde menstruation, pelvic pain and endometriosis. Leakage of menstrual blood is rarely observed over the long-term, and spontaneous pregnancy rarely occurs [17]. Given these frequent complications of surgery, many authors agree that, total hysterectomy effectively addresses the issue [18]. In this type of surgery, the problem of sterility in these patients often remains unresolved. To preserve reproductive function, operations for the creation of neovaginal and uterovaginal anastomosis are recommended. Neovagina or uterovaginal anastomosis should be performed as soon as possible after puberty. Uterovaginal anastomosis involves securing the transience and functionality of the external genital tract. Our patient expressed a desire to have corrective surgery performed to preserve her reproductive capacity. In these clinical conditions, given that the length of her vagina was sufficient, we decided to perform uterovaginal anastomosis. This operational approach, and successfully completed pregnancy several years after the project, was our first experience with this condition. In short, the surgical technique consisted of the following: resection of the fibrous supravaginal part of the cervix uterus as well as anastomosis of the isthmic part of the uterus and vagina. Thus, the incised isthmicocervical part of the uterus was pushed through the open vaginal fornix and sutured by single-circle stitches with no tension. The advantage of this procedure is that instead of forced dilation through the fibrous supravaginal part of the cervix, amputation, creates a natural isthmicouterine canal, with no tendency to stricture formation. Thus, a permanent connection between the uterus and vagina is ensured. On follow-up, upon the first menstruation, through the speculum, a circular opening of the isthmicouterine canal was observed that was approximately 5-8 mm in diameter, from which mucous leaked. The patient was advised to take oral contraception in the six months after the operation to protect herself from pregnancy. After that time, the patient was advised to take estradiol valerate, levonorgestrel (Cyclo-Progynova 2 mg) tablets 21 days per month, for six to eight months every year. The patient was advised to attend gynecological, colposcopic and US follow-up every three months. The chance of a spontaneous pregnancy in these patients is reduced, even after a successful reconstruction of the genital tract, due to severe endometriosis, as well as cervical re-obstruction and tubal factors. Many authors have reported success with pregnancies that were spontaneously formed or aided by artificial reproductive technologies [19, 20]. We believe that in our case maintenance of the cervical canal without interrupting production and leakage of cervical mucus prevented infection, retrograde menstruation, and endometriosis. We also believe that the use of estradiol valerate, levonorgestrel (Cyclo Proginova 2 mg) helped to keep the neocervical canal open. Four years after successful uterovaginal anastomosis, our patient spontaneously became pregnant. Cesarean section was performed without any serious difficulties during the operation. The leakage of lochia was normal.

Conclusion

Our results show that the technique described for resection of the fibrous tissue of a supravaginal part of the uterine cervix and uterovaginal anastomosis has been proven effective in the treatment of congenital atresia of the uterine cervix. This technique allows preservation of reproductive ability and provides a chance for a subsequent desirable pregnancy.

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