Full Length Research Paper

Abdominal surgical management of partial cervical agenesis in a virgin

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We present a 14-years old virgin with partial congenital cervical canal agenesis and hematometra. On examination, she was virgin with intact hymen and a 7 cm long vagina. A pelvic ultrasonography revealed hematometra and hematocervix. Surgical therapy included creation of a patent cervix by making a midline vertical cervical and upper vaginal incision with a subsequent excision of the distal fibrous tissue at the distal portion of the cervix. A stent was left in place for 6 weeks. Regular menstrual periods were observed for 6 months beginning at the first month. The follow-up pelvic ultrasonography was normal. We concluded that transabdominal partial cervical excision of the distal atretic portion permits the creation of a patent cervical canal in virgin and preserves the hymen intact.

Key words: Cervical agenesis, canalization, hematometra.

INTRODUCTION

Uterine cervix agenesis is a rare form of lack of development, regarding only a tract of the Müllerian ducts. Patients affected this "non by rare, communicating", abnormality have a functional uterus, but due to lack of cervix they get primary amenorrhea with cyclic pelvic pain, due to hematometra (Spence et al., 2003). Congenital agenesis or dysgenesis of the uterine cervix is a class IB in the American Fertility Society classification system (1998). The presence of a normal uterine corpus is a challenge for the clinician because a successful surgical repair could restore normal menses and potentially preserve the patient's fertility. However, current opinion in the literature considers complete agenesis to be the most difficult anatomic form of cervicouterine anomaly to correct. In this form, a total hysterectomy is recommended, because of the high incidence of complications or failure when attempting surgical correction (Rock et al., 1995).

We present a virgin of 14-year-old with partial congenital cervical canal agenesis and hematometra, and describe the surgical procedure to constitute a patent cervical canal.

MATERIALS AND METHODS

A 14-year-old virgin referred to the outpatient gynecology clinic of Tanta University Hospital in April 2011 with a history of primary amenorrhea and severe lower abdominal pain and vomiting occurring at irregular intervals over a period of 2 years. The medical and surgical history was normal. Physical examination revealed normal breast development and other sexual characters. The hymen was intact and had normal perforations. The vagina was 7 cm in length. The people of the patient were completely careful to maintain the hymen even at the expense of the uterus, because it is a respectable sign of chastity and virginity.

The abdominal pelvic ultrasound (Figure 1) showed a $10 \times 5.5 \times 4$ cm uterus. The uterine cavity was distended with blood (hematometra) and the cervix appeared ballooned with blood.

Magnetic resonance imaging (MRI) revealed the main abnormality as intracavitary blood (hematometra) with distended endometrial cavity and cervical canal (Figure 2). The vagina appeared collapsed.

An informed consent from the patient's family was obtained after verbal counseling and explanation of the potential risks of the surgical procedure and before surgical interference was done.

Laparotomy was performed with the patient under general anesthesia. On inspection, an enlarged uterus with bilateral normal ovaries was present. The visceral peritoneum between the bladder and uterus was incised transversely (Figure 3). The bladder flap was dissected away. The space between the bladder and cervix was dissected, and the proximal end of vagina was exposed. Midline incision lower part of the anterior uterine wall to reach the caudal portion of the endometrial cavity, cervical and upper part of the vaginal wall.

The accumulated blood was aspirated by suction. The external os

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Figure 1. Ultrasonographic image of the uterus and cervix. Notice that the uterus and cervix were distended with blood.



Figure 2. Initial preoperative middle sagittal T2 weighted magnetic resonance imaging showing the enlarged fluid-filled uterus and cervix.

was exposed through the incision, it was found closed. The cervical canal was opened and a Hegar dilator No. 4 was inserted in the cervical canal till its lower atretic portion (Figure 4). The portion of the cervix below the dilator was excised and dilator passed through it, this created a new ostium in the most caudal part of the cervix which was sutured circumferentially with no. 0 Vicryl. A 12 Fr Foley catheter was placed in the uterocervical canal for 6 weeks. Lastly,

we closed the longitudinal cervical and vaginal incisions, achieved hemostasis and finally closed the abdominal wall.

The patient's postoperative course was unremarkable, and menses was observed in the first month after the operation. She continued to have regular menses thereafter as we followed her for the next six months. The postoperative pelvic ultrasonographic examination was within normal limits.



Figure 3. The peritoneum between the cervix and urinary bladder was incised, the bladder dissected low down, the whole anterior wall of the cervix as well as the upper part of the anterior vaginal wall were exposed. A midline incision was done in the cervical canal, notice blood escaping from the incision.



Figure 4. After aspiration of hematometra a Hegar dilator No. 4 was introduced into the cervical canal to localized the plane of cervical obstruction (partial cervical atresia).

DISCUSSION

In the general female population, genital malformations occur with an incidence of 1 to 5% (Nahum, 1998). The congenital absence of the cervix is relatively infrequent and it occurs in 1 in 80,000 to 100,000 births (Suganuma et al., 2002). Jacob et al. (1989) made an extensive

review of the published cases of surgical reconstruction for correction of an atretic cervix, adding two cases of their own. They reviewed a total of 30 patients who underwent an attempt at permeation of the cervical canal, and found 17 successes. The factors that could have influenced the outcome could not be assessed because of missing or insufficient descriptions of the anatomy found at surgery and lack of precise descriptions of the procedures used. However, some important facts regarding the surgical therapy of the cervical agenesis have emerged and could be summarized as follows: (1) the successful establishment of a patent cervical canal has been possible in some cases, (2) partial or complete cervical atresia is often, but not always, associated with partial or complete vaginal atresia, (3) two reported deaths due to transgenital ascending infection with secondary peritonitis should prompt great attention to postoperative infection control, and (4) as stated in one editor's comment, further surgical innovations are necessary to improve the success rate (Bugmann et al., 2002).Rock et al. (1995) aided the matter by clearly defining the different anatomic findings that they had encountered. They classified the anatomic forms of their 21 cases into four categories: cervical agenesis (10 cases), cervical fragmentation (4 cases), cervical cord (3 cases), and cervical obstruction (4 cases).

Cervical agenesis or dysgenesis is an extremely rare congenital anomaly. Conservative surgical approach to these patients involves uterovaginal anastomosis, canalization, and cervical cervical reconstruction (1995) performed (Kidanto. 2011). Rock et al. reconstructive surgery in the defined categories clearly yielded differing prognoses. Their four attempts at surgical correction of the agenesis or fragmented dysgenesis type invariably failed, whereas five attempts on seven patients with the cord or obstruction dysgenesis type led to four successes with one pregnancy. They concluded that the preferential treatment for complete cervical agenesis or the fragmented dysgenesis type should be hysterectomy. Others concur with this opinion (Jacob and Griffin, 1989).

The best surgical procedure for cervical permeation is still a matter of debate. Several factors could possibly influence the surgical outcome: the size of the created channel, the amount of time the stent is left in place, the presence of residual endocervical glands in the proximity of the created channel, the presence or absence of an added epithelial lining, the presence of a normal vagina adjacent to the distal end of the created channel (which could favor epithelial cell ingrowth), or the number of menses allowed to pass through a stented new channel to seed a nonlined channel with endometrial cells. The importance of these various factors remains unclear, but our patient's case suggests that a lined neochannel allows a better outcome, reducing if not avoiding immediate stricture and/or stenosis (Bugmann et al., 2002).

The most available successful surgical methods employed to date for cervical agenesis have involved a transvaginal or transabdominal approach to create an ostium through the dense fibrous cavity with uterus and vagina by application of stents (Jacob and Griffin, 1989; Hovsepian et al., 1999; Olive and Henderson, 1987). However, restenosis of fibrous tissue, postoperative severe infection, or septicemia in a particular proportion of cases has resulted in the recommendation of hysterectomy for this group. Because these patients usually preserve their fertility, a procedure that sustains cyclic menses and does not allow restenosis would be the optimal treatment option (Rock et al., 1995). Magnetic resonance imaging may be valuable in the differential diagnosis of mullerian duct anomalies (Reinhold et al., 1997), but it may not be as accurate for diagnosing cervical aplasia, particularly when the vagina and uterus are normal. It was not used in our patients, and diagnosis reached by sonography and intraoperative was examination (Alborzi et al., 2005). In the current presented case MRI reported the possibility of high vaginal septum or external os obstruction.

Although current recommendations for the treatment of cervical atresia commonly call for hysterectomy, there is still hope that fertility can eventually be achieved in selected patients and the final decision depends upon intraoperative findings.

Lastly, we suggest that the transabdominal surgical approach may be a proper option for treatment of virgin in order to keep the hymen intact as it is a very important sign of chastity and virginity in Arabic and Islamic countries.

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