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Cervical agenesis with a functioning uterus: Successful surgical treatment by Foley's catheter stent: A case report



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Abstract *Background:* The main objectives of treatment of cervical agenesis were symptom relief, achievement of regular menstruation and restoring fertility and management of these cases represents a challenge as in many cases treatment fails and hysterectomy may be needed. In this case report a simple method was used and proved effective. *Case report:* A size 16 Silicone Foley's catheter was used to create a uterovaginal tract through combined abdominal and vaginal approaches in 18 years old patient with the diagnosis of the uncommon cervical agenesis and hematometra of 24 weeks pregnant uterus size. Postoperatively three courses of cyclic estrogen/progestagen therapy were given for 20 days every month. Three days after the first course, the first menstruation occurred through the uterine catheter. The Foley's catheter was removed five weeks postoperatively. The patient was followed up for a year, and menstruation occurred at regular monthly intervals. *Conclusion:* The procedure promises to be a simple and reasonably effective method for the creation of a menstrual outflow tract for cases of cervical agenesis with functioning uterus.

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1. Introduction

Mullerian ducts are differentiated, fused and canalized to form fallopian tubes, uterus, cervix and upper vagina. These are fused with the urogenital sinus that forms the lower vagina. These processes start cranially and progress caudally. Arrest at any level leads to "localized gynatresia". Cervical agenesis

classified as type IB Mullerian anomaly, according to the American Fertility Society (1), results from abnormal fusion of the Mullerian ducts with the urogenital sinus, or atrophy of a segment of normally formed Mullerian system (2). The incidence of cervical agenesis is 0.01% in general population. It represents about 3% of all uterine anomalies (3).

Cervical agenesis is rarely associated with the presence of vagina and functioning uterus and if associated with functioning uterus, hematometra will occur (4,5). It is estimated that only 4.8% of women with cervical agenesis have a functioning uterus (6). These cases present with primary amenorrhea, well developed sexual characters and cyclic abdominal pain (7).

The main objectives of treatment were symptoms relief, achievement of regular menstruation and restoring fertility.

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Several methods of reconstructive surgery have been developed to create an epithelialized uterovaginal canal (7). To prevent closure of the surgically formed uterovaginal canal, it is recommended that a uterovaginal catheter stent is left for three to five weeks (8–10). Cyclic estrogen progestagen therapy such as combined contraceptive pills given postoperatively for 2–3 months promotes epithelialization of the surgically formed uterovaginal canal (8,9). However, frequently re-operations due to re-stenosis of the formed canal are necessary and in many cases hysterectomy cannot be avoided (7).

In this case a simple method was used and proved effective.

2. Case description

In 2014, an 18 years old newly married woman attended the outpatient clinic in Sohag Faculty of Medicine, presenting with primary amenorrhea and monthly colicky lower abdominal pain. Examination revealed well developed secondary sexual characters, a pelviabdominal mass of 24 week pregnant uterus size. Vaginal examination showed a blind vagina of 5 cm length, with no cervix felt.

Pelvic ultrasonography showed enlarged uterus filled with fluid content, suggestive of hematometra. Both ovaries were free. Intravenous pyelography revealed normal kidneys, and blood picture, renal function tests, blood sugar and urine analysis were free. Karyotype was 46XX.

After counseling and getting written consent, examination under anesthesia revealed a blind vagina of five cm length. A Foley's catheter was fixed in the urinary bladder under aseptic conditions. Vaginal exploration was done and a transverse incision two and half cm in the vaginal dome followed by deep dissection in the fibrous tissue. Simultaneously a transverse suprapubic incision for abdominal approach of the uterus was done. Identification of the pelvic organs showed uterus enlarged up to \pm 24 week pregnancy size, and both ovaries and tubes were free. The visceral peritoneum over the lower anterior wall of the uterus was dissected carefully downward to free the anterior wall of the uterus from the bladder. A transverse incision two cm in length was done to open the uterus, and evacuation of hematometra by suction was done followed by peritoneal toilet. A long artery forceps was put inside the uterus at the virtual level of internal os. The artery forceps was pushed gently downward till its tip appeared through vaginal incision where a Silicone Foley's catheter size 16 was placed in its tip; then, the artery forceps was gradually withdrawn until the tip of Foley's catheter appeared inside the uterus, and insufflation of its balloon by five ml of saline was done followed by closure of the uterine incision by interrupted sutures of polyglactin with careful hemostasis. Cystoscopy was done at the end of the operation to evaluate any possible bladder injuries.

Postoperatively, she received analgesics, antibiotics, and intravenous fluids. She recovered well and discharged on the sixth postoperative day. Starting from the fifth postoperative day three courses of cyclic estrogen/progestagen therapy (cycloprogynova-Sherring ® Egypt) were given for 20 days each month for three months. Three days after the first course, the first menstruation occurred and the menstrual blood got out through the uterine catheter. Hysterosalpingography was done after one month and showed intact uterine cavity and patent tubes. Leukocytic count estimations were done regu-

larly every 3 days while the uterine Foley's catheter was in situ to detect early indications of developing infection. With the end of the first course of cyclic estrogen/progestagen therapy when withdrawal bleeding was expected to occur, the patient received a course of broad spectrum antibiotic cephalexin 1 g/12 h orally till the end of withdrawal bleeding.

The uterine Foley's catheter was removed five weeks postoperatively. The patient was followed up for a year after the operation. Menstruation occurred at regular monthly intervals.

3. Discussion

The uterine cervix provides an outflow tract for the menstruation. Cervical agenesis hinders this function, affects the reproductive function and represents a therapeutic dilemma. Hysterectomy was the eventual treatment for cervical agenesis because of the common complications of recanalization of the cervix and the unlikelihood of a viable pregnancy (11). Recent advances led to conservative surgery as the first line treatment (8). However, hysterectomy might be necessary when the conservative treatment fails (7,12).

In this case, this conservative surgical treatment was tried putting in consideration that the simplicity of the technique might open the door for wider application of this therapeutic option with the possibility of restoration and maintenance of normal menstrual outflow tract, fertility preservation and also, improvement of the patient's psychic condition.

4. Conclusion

The procedure promises to be a simple and reasonably effective method for the creation of a menstrual outflow tract for cases of cervical agenesis with functioning uterus, however the procedure needs to be further evaluated in a case series studies and for various gynatresia levels.

Conflict of interests

The author has no conflict of interests relevant to this article.

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