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Case Report

Management of cervical atresia and vaginal aplasia leading to hematometra and endometriosis in a young patient: a case report

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ABSTRACT

Cervicovaginal atresia is a rare congenital anomaly of the female reproductive system where there is a complete absence or severe underdevelopment of the cervix and /or vagina usually presenting in adolescence with severe abdominal pain and primary amenorrhoea which can further lead to endometriosis and pelvic mass. Hereby, we are reporting such a rare case of cervicovaginal atresia in 24 years female who was managed with hysterectomy. Surgery is not strictly required during menstruation, but if there is hematometra, the surgery should be performed as soon as possible to relieve the obstruction. This case reporting aims to offer insights and recommendations for future research on cervicovaginal atresia, ultimately striving to enhance the quality of life for affected individuals.

Keywords: Cervical atresia, Hematometra, Endometriosis, Hysterectomy

INTRODUCTION

Major part of the female genital tract develops from the Mullerian ducts. The duct forms one on each side as an ingrowth of coelomic epithelium in the lateral aspect of mesonephros at about 5-6 weeks. In the absence of androgen and anti-Mullerian hormone as in normal female, there is further growth and development of the Mullerian duct system with regression of the Wolffian ducts. Uterus, cervix, fallopian tube and upper two third of vagina are formed from Mullerian ducts, while the lower part of vagina develops from urogenital sinus.

Vaginal atresia is usually associated with uterine aplasia, Mayer-Rokitansky-Küster-Hauser syndrome. However, vaginal aplasia can occur in 9% of cases where the uterus is present.[1] Cervical agenesis is a rare congenital anomaly with an incidence of 1:80,000-1:100,000. Further, its association with vaginal agenesis is also rare.

Cervicovaginal agenesis usually presents around the age of menarche with complaints of primary amenorrhoea and cyclical abdominal pain. In these cases, endometrium is functioning normally. So menstrual blood having no passage to outside due to cervicovaginal atresia leads to formation of hematometra which further leads to hematosalpinx and endometriosis.

In such cases, although conservative management creating the uterocervico-vaginal passage should be the treatment of choice but sometimes due to anatomical difficulties there is need of hysterectomy to relieve the severe pain of patient.

Here, we present a clinical case of cervical atresia with vaginal aplasia and discuss the clinical and therapeutic aspects of this rare malformation.

CASE REPORT

Twenty-four years old unmarried female presented to gynae OPD with severe abdominal pain for 1 day which was not relieved with analgesics. She gave history of primary amenorrhea and cyclical severe abdominal pain

since 6 years which has worsened gradually. She also had difficulty in micturition cyclically. All milestones developed normally. General physical examination revealed well developed secondary sexual characters (breast, axillary and pubic hair). She had abdominal tenderness. On per speculum examination vaginal canal was short (about 3x2cm) and cervix looked atretic as cervical opening was not visible.

Since she had history of symptoms for the last 6 years and she was having treatment since then. She gave vague history about some procedure suggestive of vaginoplasty in 2019. CE MRI on 25/4/2024 was suggestive of hypoplastic cervix and poor visualization of cervical canal. Hypointense area was seen between urethra and rectum. Vaginal canal was not clearly visualized. After getting repeated consultations, she was not getting relief but it was getting worsened and it was leading to hamper her quality of life.

Then in December 2024, when she visited AMCH, Shahbad USG showed bulky uterus with dilated endometrial canal with hematometra with left paraovarian simple cyst measuring 49x34mm. ET was 43-44mm

MRI abdomen and pelvis was done on 13th December 2024 at AMCH, Shahbad was indicative of hypoplastic or rudimentary cervix with poorly visualized endocervical canal with hypoplastic and poorly visualized vagina with hematometra (collection measuring 5.9×4.2 cm) along with complex cystic tubo-ovarian mass with chronic blood products on right side suggestive of endometrioma and left ovarian cyst.

She was planned for diagnostic laparoscopy with examination under anaesthesia. Findings on diagnostic laparoscopy were suggestive of adherence of posterior surface of uterus to gut and omentum and anterior surface of uterus to bladder. Omentum was adherent to right fallopian tube and ovary also. There was right tuboovarian cyst of about 5×7 cm. Left fallopian tube and ovary were not visualized.

Later on, she was planned for exploratory laparotomy proceed reconstruction of cervicovaginal area with drainage of hematometra. Written and informed consent was taken about need of hysterectomy. Uterus was visualized after opening the abdomen. There was evidence of grade 3 endometriosis. Bilateral fallopian tubes were dilated. Gut was adherent to right side of uterus and fallopian tube. A par ovarian cyst of 4×5 cm was found on left side which was removed and sent for HPE. Meanwhile gap between lower end of uterus and upper part of vagina was assessed. Stab incision was given over the anterior lower end of uterus. About 80-100 ml of chocolate colored fluid was drained. A long straight artery forceps was introduced through the cavity towards vagina. Lower end of uterus was solid and fibrosed. There looked no communication and a gap of about 5-6 cm between both structures. It seemed difficult to create uterovaginal communication. So, decision for hysterectomy was taken. Postoperative period was uneventful. Histopathological examination revealed weakly proliferative endometrium. Lower uterine segment had areas of hemorrhage and hemosiderin laden macrophages. Left ovarian cyst had benign serous cystadenoma. Consent of publication was obtained from patient and family.

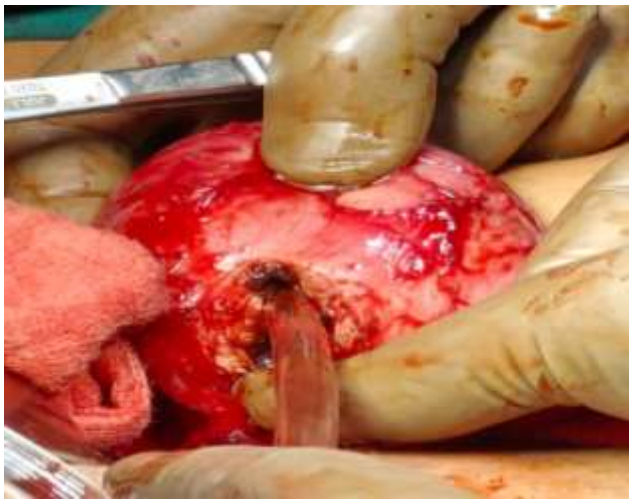

Figure 1: Incision over the lower part of uterus and drainage of hematometra.

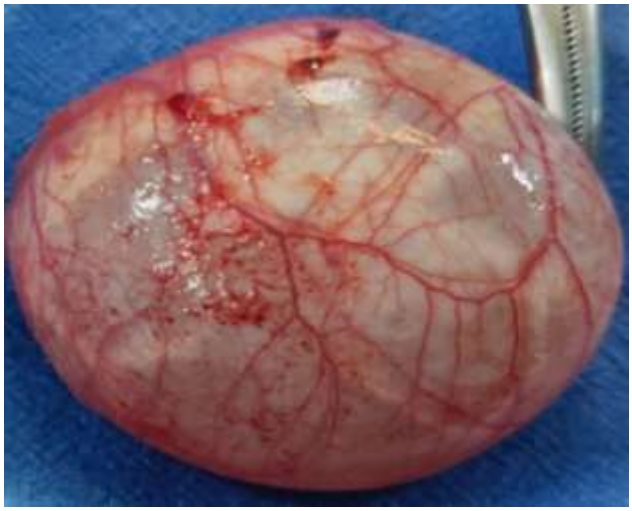

Figure 2: Left ovarian cyst.

DISCUSSION

Vaginal agenesis/cervical atresia arises from developmental defects of the Müllerian tubercle.[2]

Study by Mei et al found a mean age of diagnosis at 14 years, with the age range of patients being between 10 and 22 years.[3] Similarly, in the study by Zayed et al the age of patients ranged from 14 to 21 years.[4] In our case, patient presented at 24 years although her symptoms appeared at an age of 18 years. This age represents the time when patient starts with menstruation.

If retrograde menstruation is not detected in time, it can lead to endometriosis and pelvic mass. Pelvic mass is reported in 100 % of patients according to study by Mei et al and Xie et al.[3,5]

Very frequently, vaginal aphasias are accompanied by other malformations such as uterine malformations, uterine septum, renal agenesis, urogenital fistula, or vertebral malformation.[6] Our patient did not present with any of these anomalies.

MRI is considered the gold standard for the diagnosis and precise description of female genital tract anomalies.[5] It is the most sensitive examination allowing for a good visualization of complex malformations with menstrual retention and to determine the presence or absence of a uterovaginal axis.[7]

For all types of vaginal atresia, the primary goals of surgery are to alleviate the obstruction, restore anatomical function, and prevent recurrence. However, with varying degrees of uterine and cervical development in patients with different levels of vaginal atresia, as well as differing fertility requirements, specific treatment strategies-such as the choice of surgical approach and the material used to cover the newly created vaginal surface-may differ and often require individualized consideration.

Various surgical techniques are available for creating a neovagina, offering non-surgical techniques such as the Frank technique, which requires a depth greater than 2.5 cm of the vagina and patient compliance with dilator use. While this technique yields good anatomical and functional results, it is time-consuming, restrictive, and can be painful; however, in our case, the patient was not a candidate for this technique.[8]

Other techniques include McIndoe vaginoplasty: a complex surgical procedure involving artificial skin grafting, associated with numerous complications such as infection and obstruction. Moreover, it requires prolonged dilator use, which may adversely affect patient compliance and psychological well-being.[4]

Davydov technique utilizes peritoneum to create a neovagina, either with a segment of the digestive tract or bladder tissue. However, the use of these techniques has been associated with vesicovaginal fistulas.[8]

Vecchietti technique employs laparoscopy, sometimes with ultrasound or cystoscopy guidance, to create a neovagina from cervical tissue. This procedure is often painful due to suture tension and dilator use; in our case, the patient lacked cervical tissue required for this technique.[4]

Hysterectomy is opted for cases where reparative techniques are difficult due to insufficient cervicovaginal tissue as in our case. Although, it completely vanishes the reproductive life of patient but when symptoms disturb patient to the very extreme it becomes mandatory to make her relief of those intolerable symptoms which can worsen further if left uncared. Due to the rarity of this malformation, this case report aims to present a unique case of a young woman, which led to a hysterectomy to avoid the probable complications of conservative treatment.

CONCLUSION

Cervicovaginal agenesis is rare congenital anomaly of the female genital tract with only few cases being reported.

Usually, patient is a young girl presenting with primary amenorrhoea with cyclical abdominal pain and palpable mass per abdomen. Urinary symptoms can be present as in our case. Patient should be evaluated thoroughly with clinical history, examination and pelvic imaging. Ultrasound and MRI are useful. Differential diagnosis includes imperforate hymen, transverse vaginal septum, vaginal agenesis. Although conservative management like reconstructive surgery should be the first line of management but in extreme cases where procedure can lead to high morbidity, hysterectomy should be considered. As in our case we tried to do reconstructive surgery but due to anatomical difficulties it was not possible and hysterectomy was done after taking consent of patient and parents.

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