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Case Report

Neovagina creation using sigmoid colon in vaginal agenesis: a case report and review of literature

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ABSTRACT

Mullerian duct anomalies are rare and can present with abnormalities in upper vagina, cervix, uterus, and fallopian tubes. The exact incidence of cervicovaginal agenesis is unknown and there are very few cases recorded in the literature. Authors report a 12-year-old girl referred from gynaecologist with chronic cyclical abdominal pain with underdeveloped, geniatilia and developed secondary sexual characters. Radiological investigations showed hypoplastic cervix with hematometra and left hemato salpinx. A definitive repair with creation of a neovagina using a sigmoid colon segment was performed in a single stage. Many techniques are described for reconstruction of cervicovaginal canal. Use of colon in creation of a neovagina is described by authors. Here we report a case of complete vaginal agenesis presenting with Hematometra-Hematosalpinx where a neovagina was created using a segment of sigmoid colon. Single staged surgery for neovagina creation using segment of sigmoid colon offers a promising and safe alternative for cervicovaginal agenesis.

Keywords: Cervicovaginal agenesis, Mullerian duct anomalies, Neovagina, Vaginal agenesis

INTRODUCTION

Mullerian duct anomalies are rare and can present with abnormalities in upper vagina, cervix, uterus, and fallopian tubes.[1-3] The exact incidence of cervicovaginal agenesis is unknown and there are very few cases recorded in the literature.[1] Many techniques are described for reconstruction of cervicovaginal canal. Recently, as diagnostic and surgical options have expanded, preservation or improvement of reproductive potential is a primary goal. Accurate diagnosis of anatomical abnormality, evaluation of any associated abnormality, physiological and psychological preparation of the patient is necessary before undertaking any surgical management in such cases.[4] Use of colon in creation of a neovagina is described by authors.[5-7] Authors report a case of complete vaginal agenesis presenting with hematometra-hematosalpinx where a neovagina was created using a segment of sigmoid colon.

CASE REPORT

A 12-year-old young girl was referred from gynecologist for cyclical pain and tenderness in left iliac fossa region 3 times over a period of 6 months. She was previously investigated. Clinical examination showed under-developed external genitalia with absence of vagina and normal anal and urethral meatal opening. She had developed secondary sexual characters.

Ultrasonography revealed a rounded echo filled cystic space occupying lesion at pelvis just posterior to urinary bladder. Lesion was centrally placed with no solid or vascular component. Ovaries could not be visualized separately from

this lesion. Her MRI report showed collection in endometrial cavity (hematometra) with hypoplastic cervix, left hematosalpinx and absent vagina Uterus measured 5.6 × 4.4 × 3.6 cm with collection inside endometrial cavity and cervical canal with possibility of hematometra and hematosalpinx. She had not received any hormonal treatment in the past.

Examination under anaesthesia revealed underdeveloped external gentiles, no hymenal bulge and absent vagina. On per rectal examination vaginal tube was not palpable. Her anal opening and external urethral meatus were normal and urine output was obtained on catheterization with Foleys no 10. Clinical diagnosis of agenesis of vagina with Hematometra was kept.

Decision for elective operative intervention was taken. Patient was opened in a lithotomy position, with sub umbilical midline incision. Uterus was unicornuate, with hypoplastic cervix. Both ovaries were present. There were no features of endometriosis. Right fallopian tube was normal. Left fallopian tube had hematosalpinx which was drained. Space was created between rectum and urinary bladder. Perineal and pelvic dissection was done. Dilator was passed through the pelvic diaphragm. Approximately 15 cm long distal sigmoid colon segment was mobilized and isolated on marginal artery. Care was taken so that vascularity was not compromised at the ends of the segment (Figure 1).

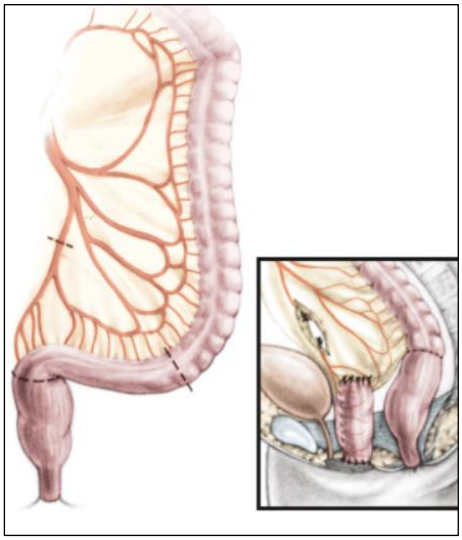

Figure 1: Diagrammatic representation of harvested segment of sigmoid colon.[17]

The harvested sigmoid colon segment was brought down at the introitus and using seromuscular anchoring sutures it was fixed to the introitus using vicryl. Upper end of the uterus was opened. Hematometra was drained which was thick dark blood. Upper end of sigmoid was anastomosed to the lower end of uterus in the same sitting. Bowel continuity was achieved by anastomosing proximal sigmoid colon with rectum in two layers. Drains were placed in pelvis and hemostasis was confirmed. Abdomen was closed in layers. Postoperative course was uneventful. On 8th postoperative day, neovagina admitted Hegars dilator no 16 and index finger comfortably. Wounds were healthy. She was discharged on post-operative day 21 with no complications. She had her first menstrual cycle 21 days after operation and is currently having irregular menstruation and is being regularly followed up (Figure 2 and 3).

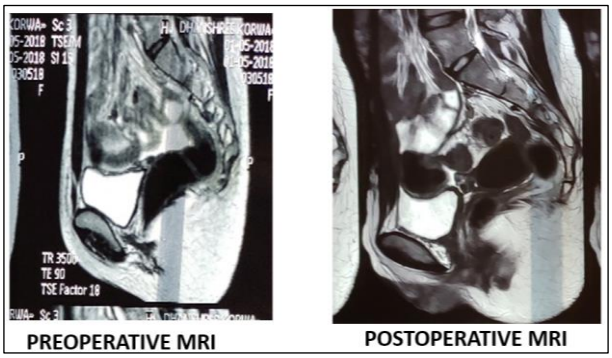

Figure 2: Pre- and post-operative MRI findings.

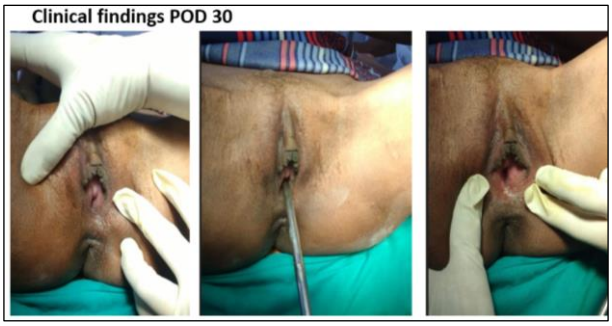

Figure 3: Clinical examination done postoperative day 30.

DISCUSSION

Definition and surgical anatomy

Cervicovaginal agenesis is a complex type of Mullerian ductal anomalies which present with functional ovaries, fallopian tubes and uterine fundus. The American Society of Reproductive Medicine (ASRM) has classified Mullerian duct anomalies into seven categories. Agenesis/hypoplasia of the vagina and cervix are categorized as 1A and 1B respectively. Embryologically there is failure or absence of Mullerian tubercle (vagina) and diverging portions of Mullerian ducts (cervical atresia). Class 1A includes cases of vaginal agenesis with functional uterus and endometrium, while cases of cervical agenesis are included in Class 1B (Figure 4).[8] Advantages of classification is uniform nomenclature and simplicity of description of anomalies, however there are certain unique anomalies which cannot be described using ASRM classification. As an alternative embryologic-clinical classification to consider all female

genitourinary malformations and their embryologic origin was suggested.[9] Another group described the vagina, cervix, uterus, adnexa, and associated malformations (VCUAM) system.[10] These two systems, although very comprehensive, are complex and not user friendly for practitioners, which limits their use. A recent systematic review indicates that current evidence favors continued use of the ASRM classification.[11]

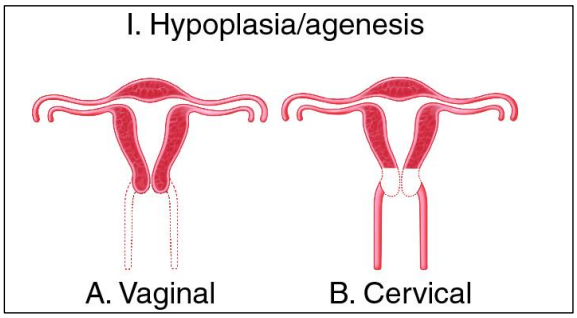

Figure 4: ASRM classification class 1A and 1 B.[8]

Embryology and anatomical variants

Proper development of the Müllerian ducts depends on the completion of three phases: organogenesis, fusion, and septal resorption. Failure in the organogenesis phase will develop uterine agenesis or hypoplasia. The failure of the distal Mullerian structures to develop may give rise to cervical and vaginal agenesis; these anomalies are closely related to a wide range of associated malformations of the genitourinary tract also.[3,8]

Incidence

The frequency of vaginal atresia is of 1 in 4000 to 1 in 5000 female subjects with abnormal development of the uterine corpus observed in >90% (Mayer-Rokitansky-Kuster-Hauser syndrome), Fujimoto et al, reported 50 patients in the literature, of them, 48% had a normal vagina, 42% with complete vaginal agenesis, and 10% had a shortened blind vaginal pouch.[12] Roberts et al, estimated that there were less than 200 cases of CVA published in the literature since 1942.[1] True cervicovaginal agenesis is rare and is associated with an obstructed single uterine canal, the absence of a patent cervix, and agenesis of the upper vagina. This has a poor prognosis for reconstruction. Hysterectomy is usually recommended, but creation of a vagino-uterine fistula has also been described using various modalities.[13]

Presentation

Most of the patients with cervicovaginal agenesis present in between the age group of 12-20 years with cyclical abdominal pain and amenorrhoea.[14] Clinical examination reveals normal secondary sexual characters. Obstruction of the menstrual outflow may give rise to hematometra or hematosalpinx. If left untreated this may give rise to endometriosis.[15]

Investigations

The primary workup for evaluation of patients with cervicovaginal agenesis, an abdominal ultrasonography is recommended. It helps in evaluation of uterus, kidneys and adnexal structures. Urological studies can be done to objectively rule out any associated urinary tract anamolies. Magnetic resonance imaging (MRI) of the pelvis is the best imaging modality for detailed anatomical evaluation of Mullerian duct anomalies, Imaging with the use of ultrasound, 3D ultrasound, and MRI has largely replaced the need for these diagnostic procedures.[11]

Management

The treatment of cervicovaginal agenesis has evolved over a period of time. It is largely dictated by the ASRM classification. Initially canalization procedures were performed which if unsuccessful were treated with hysterectomy Patients who have functional uterus, Colo vaginoplasty is also a safe procedure.[2,5,7,9,10] The creation of a neovagina and uterine opening anastomosis should be done at around age of 10-12 years to avoid endometriosis.[10]

Many surgical options for vaginal substitution have been described, with the split-thickness skin graft vaginoplasty for patients with agenesis. Materials such as amnion, peritoneum, and buccal mucosa have been described as an alternative to skin grafting. The use of sigmoid bowel pulled to the perineum for vaginal creation is less commonly utilized for this problem but has been associated with good long-term results.[14]

Henderen et al described the use of bowel segments in creation of neovagina.[5] Case series have been described where the sigmoid colon is anastomosed in an antiperistaltic fashion between the uterus and vagina. Kannaiyan et al, reported no requirement for any vaginal dilatations, and no indwelling moulds or stents were used. Stenosis of the neovaginal orifice was 9%, while complications such as endometritis or pelvic inflammatory disease were not seen. In contrast Kisku et al, documented leakage of the anastomosed colon in 5%, recurrence of the pain due to the recurrent chocolate cysts in 10%, mild stenosis of the neovagina in 10%, on daily self-dilatation as a precautionary measure in 15% of the patients.[2]

Complications

The use of sigmoid colon in creation of a neovagina poses challenges mainly related to the use of bowel loop. Tension over the harvested vascular pedicle, short mesentery, masculinized pelvis and obesity are some of the challenges. These can be overcome by using the techniques employed by Tu et al and Garcia-Roig et al. It also diminished the risk of endometriosis, neovaginal ischemia and anastomotic leakage. Furthermore, larger neovagina can be created using this technique. No

complications were reported in the study by Tu et al while Garcia-Roig et al had one patient with vaginal stenosis. It is important to mention that the series of Kannaiyan et al and, Kisku et al had 31 patients, and the Tu et al and, Garcia-Roig et al had ten patients in their study.[2,6,10] Thus more patients treated with this technique need to be evaluated for comparing immediate, late and long term outcomes.[16,17]

Authors created the neovagina using the sigmoid colon segment and anastomosed it in a single sitting, unlike the two staged technique as described by Yang-Monti. Patient denies any mucus discharge, with a bit irregular menstrual cycle without any vaginal stenosis and requirement of vaginal dilations and good cosmetic result. No significant complications have yet been reported in last 16-month follow-up period.

CONCLUSION

Mullerian duct anomalies presents with challenging scenarios. Identification of correct anatomy by detailed examination, proper classification, preparation of the patient with psychosocial counselling are fundamental requirements. As the surgeries for cervicovaginal agenesis are not yet standardized, the use of sigmoid colon in creation of a neovagina seems promising. The surgery can be done in a single step without any tension and anastomotic leak, avoiding the requirement of a second surgery. However, long term patient outcomes and complication rates in use of sigmoid colon in creation of neovagina need to be evaluated.

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REFERENCES

1. Roberts CP, Rock JA. Surgical methods in the treatment of congenital anomalies of the uterine cervix. Curr Opin Obstetr Gynecol. 2011;23(4):251-7.
2. Kisku S, Varghese L, Kekre A, Sen S, Karl S, Mathai J, et al. Cervicovaginal atresia with hematometra: restoring menstrual and sexual function by utero-coloneovaginoplasty. Pediatr Surg Inter. 2014;30(10):1051-60.
3. Chandler T, Machan LS, Cooperberg PL, Harris AC, Chang SD. Müllerian duct anomalies: from diagnosis to intervention. Bri J Radiol. 2009;82(984):1034-42.
4. Templeman CL, Lam AM, Hertweck SP. Surgical management of vaginal agenesis. Obstetr Gynecol Surv. 1999;54(9):583-91.
5. Hendren WH, Atala A. Use of bowel for vaginal reconstruction. J Urol. 1994;152(2):752-5.
6. Tu D, Badalato G, Reiley EA, Hensle TW. Colovaginoplasty using the Yang-Monti modification. J Pediatr Urol. 2012;8(5):477-80.
7. Garcia-Roig M, Castellan M, Gonzalez J, Gorin MA, Cruz-Diaz O, Labbie A, et al. Sigmoid vaginoplasty with a modified single Monti tube: a pediatric case series. J Urol. 2014;191(5):1537-42.
8. Society TA. The American Fertility Society classifications of adnexal adhesions, distal tubal occlusion, tubal occlusion secondary to tubal ligation, tubal pregnancies, Müllerian anomalies and intrauterine adhesions. Fertil Steri. 1988;49(6):944-55.
9. Acién P, Acién MI. The history of female genital tract malformation classifications and proposal of an updated system. Human Reprod Update. 2011;17(5):693-705.
10. Oppelt P, Renner SP, Brucker S, Strissel PL, Strick R, Oppelt PG, et al. The VCUAM (Vagina Cervix Uterus Adnex–associated Malformation) Classification: a new classification for genital malformations. Fertil Steri. 2005;84(5):1493-7.
11. Bhagavath B, Greiner E, Griffiths KM, Winter T, Alur-Gupta S, Richardson C, et al. Uterine malformations: an update of diagnosis, management, and outcomes. Obstetr Gynecol Surv. 2017;72(6):377-92.
12. Fujimoto VY, Miller JH, Klein NA, Soules MR. Congenital cervical atresia: report of seven cases and review of the literature. Am J Obstetr Gynecol. 1997;177(6):1419-25.
13. Castrillon DH. Development and Maldevelopment of the Female Reproductive System. Gynecol Obstetr Pathol. 2019;1:1-40.
14. Acién P, Acién MI, Quereda F, Santoyo T. Cervicovaginal agenesis: spontaneous gestation at term after previous reimplantation of the uterine corpus in a neovagina: case report. Human Reprod. 2008;23(3):548-53.
15. Breech LL, Laufer MR. Müllerian anomalies. Obstetr Gynecol Clin North Am. 2009;36(1):47-68.
16. Karateke A, Haliloglu B, Parlak O, Cam C, Coksuer H. Intestinal vaginoplasty: seven years' experience of a tertiary center. Fertil Steri. 2010;94(6):2312-5.
17. Coran AG, Caldamone A, Adzick NS, Krummel TM, Laberge JM, Shamberger R. Pediatric surgery E-book. Elsevier Health Sci; 2012.

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