Conservative management of tubo-ovarian abscess as a complication of reconstructive surgery for congenital cervicovaginal agenesis: a case report and review of literature

Izat MohammadKhawajah¹, Sima Shamshiri Khamene¹, Zahra Rezaei¹, Fahimeh Azizinik¹, and Khadijeh Adabi¹

¹Tehran University of Medical Sciences

January 29, 2025

Title page

\received

DD MMMM YYYY \acceptedDD MMMM YYYYArticle title:

Conservative management of tubo-ovarian abscess as a complication of reconstructive surgery for congenital cervicovaginal agenesis: a case report and review of literature

Izat Mohammad Khawajah¹, Sima Shamshiri Khamene¹, Zahra Rezaei¹, Fahimeh Azizinik ², Khadijeh Adabi^{1, 3*}

¹ Division of Female Pelvic Medicine and Surgery, Department of Obstetrics and Gynecology, Yas Hospital complex, Tehran University of Medical Sciences, Tehran, Iran

²Advanced Diagnostic and Interventional Radiology Research Center (ADIR), Department of Radiology, Yas Hospital and Amiràlam Hospital Complex, Tehran University of Medical Sciences, Tehran, Iran.

³ Vali-E-Asr Reproductive Health Research Center, Family Health Research Institute, Tehran

University of Medical Sciences, Tehran, Iran

*Corresponding author:

Khadijeh Adabi (Prof)

Division of Female Pelvic Medicine and Surgery, Department of Obstetrics and Gynecology, Yas Hospital complex, Tehran University of Medical Sciences, Tehran, Iran

E-mail: khadabi88@gmail.com

Tell: 00989125477050

Abstract

Objectives: Cervical agenesis, a rare birth defect affecting the female reproductive system, is often accompanied by vaginal agenesis in only 39% of cases. In the past, the standard treatment for this condition was hysterectomy. However, as medical technology and techniques continue to advance, more conservative surgeries and assisted reproductive methods are now being considered as the primary treatment options. However, complications such as restenosis, pelvic abscess, and recurrent pelvic endometriosis should always be considered.

we report a case of cervicovaginal agenesis complication after reconstructive surgery in a 16-year-old adolescent girl. a conservative surgical approach to cervical malformations may cause complications that can be prevented or managed by regular follow-ups.

Keywords: Tubo-ovarian abscess, congenital cervicovaginal agenesis, reconstructive surgery

Main text

\received

DD MMMM YYYY \acceptedDD MMMM YYYY**Title**

Conservative management of tubo-ovarian abscess following rectovaginal fistula as a complication of reconstructive surgery for congenital cervicovaginal agenesis: a case report and review of literature

Introduction

The prevalence of congenital uterine anomalies vary from 1-10 %(1), Congenital cervical agenesis or dysgenesis is rare with a prevalence ranging from 1 in 80,000 to 1 in 100,000. (2)

Conservative surgery can lead to serious complications such as endometritis, pelvic inflammatory disease, persistent pelvic pain, bowel or bladder injury, repeat surgery, and death. (3) Consequently, many experts advocate hysterectomy as the treatment of choice for these patients due to the risk of reoperation or the serious complications described above, as well as the potential for death from reconstructive surgery. (4)

Demolitive treatment is generally reserved for patients with repeated failures of conservative therapy or postoperative complications (such as infections or cervical restenosis) (5)

This case report discusses the conservative management of complications following reconstructive surgery in congenital agenesis of the cervix and vagina, along with a review of articles related to conservative surgical restoration of the reproductive tract in women with congenital cervical agenesis.

Case history

A 16-year-old adolescent girl presented with persistent fever (up to 40 °C) with abdominopelvic pain, anorexia, and nausea. Laboratory data showed a white blood cell (WBC) count of $15.7 \times 10^3 / \mu$ L, 86%neutrophil count, a hemoglobin level of 10.5 g/dL, a platelet count of $285 \times 10^3 / \mu$ L and a C-reactive protein (CRP) level of approximately 15.2 mg/dL. The fever persisted despite the antibiotic treatment, which lasted for two weeks. Blood cultures performed on the day of referral were negative. She was a known case of cervicovaginal agenesis. She had undergone laparoscopic uterovaginal anastomosis with the placement of a polytetrafluoroethylene stent for the reconstruction of cervical agenesis and concomitant modified McIndoe vaginoplasty at the age of 11 years. One cm rectal injury occurred during vaginoplasty and was repaired primarily. Her menstruation was regular after surgery. She was under scheduled medical follow-up.

Differential diagnosis

Mesenteric adenitis, diverticulitis, ovarian cyst

Investigations and treatment

Trans-abdominal ultrasound showed a pelvic thick-walled multilocular complicated cyst containing echogenic debris, measuring 70*6.8*56 cm in the left adnexa. Magnetic resonance image revealed a unilateral complex multilocular thick-walled fluid-filled ovarian mass with an irregular thick uniform enhancing wall and septa at the left side of the pelvic, causing pressure on the adjacent uterus associated with surrounding pelvic fat haziness. The above mentioned lesion in MRI demonstrated heterogeneous intermediate and hyper signal intensity on T2-weighted images, low signal intensity on T1-weighted images, high signal intensity on diffusion-weighted imaging (DWI), and low signal intensity on apparent diffusion coefficient (ADC) indicated restricted diffusion. These observations indicated the presence of tubo-ovarian abscess(Fig. 1).

Additionally, a high T2 signal intensity track with enhancement was seen between the left anterior wall of the lower rectum and the left posterior wall of the vagina with surrounding fat haziness, in favor of the rectovaginal fistula (Fig. 2).

Due to persistent fever, abscess drainage was performed under an ultrasound guide. Puncturing of the swollen left ovary revealed internal pus and pus was collected for bacterial culture and the abscess was excised without any substantial compromise to the ovary. The culture was positive for *Escherichia coli*. Clindamycin 900 mg IV every 12 hours and Gentamicin loading dose IV (2 mg/kg) followed by 1.5 mg/kg every 8 hours was administered.

The rectovaginal fistula was managed conservatively according to the colorectal surgeon's recommendation. She was discharged after 6 days; the CRP level on the day of discharge was 4.12 mg/dL. Oral Clindamycin at 900 mg/BID was continued for 14 days. Finally, imaging performed one month after the procedure showed no recurrence of ovarian abscess.

Written informed consent was obtained from the patient and her mother for the publication of the report.

\received

DD MMMM YYYY \acceptedDD MMMM YYYYConclusion

The conservative surgical treatment of cervical malformations is a promising option that can be provided with the primary purpose of preserving the uterus for future fertility. Complications following these procedures can also be managed conservatively. However, after the initial surgery, patients should be ready for a lengthy period of follow-up care.

Key clinical message

While most similar cases are treated with total hysterectomy, in this case, we demonstrated that conservative treatments can also be considered as an alternative to surgical treatments.

Discussion

Managing cervical abnormalities, whether with or without a normal vagina, can be both challenging and rewarding. It demands creativity in devising management approaches.

Patient participation in the decision-making process is critical, as interventions ultimately affect her life and are associated with irreversible loss or negative impact on reproductive function. (3)

Hysterectomy was traditionally primarily the treatment of cervical agenesis to avoid serious complications following reconstructive surgery. (4)

Various techniques have been reported in the literature describing procedures creation of a neocervix and a neovagina if needed, and restoration of the continuity of the genital tract. The choice of neovagina and neocervix method and technique of neocervix and neovagina anastomosis is important. (2) Dornelas, reported eleven patients with vaginal agenesis underwent Utero-neovagina anastomosis using a Silicon mold covered by oxidized cellulose. One major postoperative complication occurred, which culminated in death. (6) In a survey done by Rock et al. cervical reconstruction was performed in 11 patients, 6 eventually experienced hysterectomy after obstruction of the neocervical canal. Two cases with cervical agenesis underwent uterovaginal anastomosis. Both required hysterectomy because of pelvic infection due to re-obstruction. (4)

A variety of treatment options are available for pelvic inflammatory disease (PID), including conservative management with IV antibiotics, laparoscopic aspiration, image-guided aspiration or drainage, laparoscopic salpingostomy with saline irrigation, and salpingectomy.(7) Rupture of the abscess can be fatal as high as 5%–10% of cases even with advanced treatment and surgical intervention.(8) Immediate and aggressive treatment can lead to a favorable outcome.

When OA is suspected, quick treatment is required to prevent adverse outcomes. (9) The treatment recommended for managing infectious complications after reconstructive surgery is hysterectomy.(10) Further,

in another case series, 14 patients underwent laparoscopic-assisted uterovaginal anastomosis, nine of whom also underwent concomitant vaginoplasty. Among them, only one patient required a hysterectomy due to restenosis and infection. (11)

Kimble R et al. reported two patients with the combined congenital anomalies of complete vaginal agenesis and partial cervical agenesis presented difficulties encountered with the limitations of MRI in the accuracy of diagnosis and clinical correlation of imaging was not easy, as well as the development of life-threatening sepsis requiring hysterectomy and limited counseling by not being able to make an accurate diagnosis. Both patients were at first imaged as having enlarged endometrial cavities and cervical canals with what was thought to be an obstructive upper vaginal septum and an absent lower vagina. Both required initial neovagina creation, however, the cervixes were never clinically or surgically visualized. (5)

Three cases in the literature discussed sepsis-related deaths and obstruction secondary to cervical agenesis. Initially, these patients were thought to have a high transverse vaginal septum and were treated by creating a neovagina and establishing communication with the uterine cavity. (12), (13) Despite initially having normal periods, all patients later presented to hospitals with severe infections and obstruction requiring hysterectomy due to infectious morbidity. In one case, the patient continued to decline, developing multi-system organ failure, and ultimately died after a hysterectomy. (12) In all five cases, including the above cases, there was a delay in the accurate definite diagnosis of the abnormality resulting in non-definitive initial treatment. Unfortunately, it was this delay that allowed the development of complications that led to significant morbidity and mortality.

Tareq Maraqa et al. reported conservative management of bilateral recurrent pyosalpinx in a 12-year-old girl secondary to retrograde menstruation caused by obstructed hemivagina due to Mullerian duct anomaly. In addition to irrigation and drainage of the abdomen and pelvis, IV and oral antibiotics were sufficient to achieve complete resolution without the need for a salpingostomy or salpingectomy.

(14)

Tsuyoshi Murata et al. presented a case of ovarian abscess in a virginal adolescent girl without any mullerian anomaly who was treated by laparoscopically abscess drainage. In OA early diagnosis and treatment can remarkably decrease the risk of sepsis, torsion, and adverse effects. (15)

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's parent has consented to report images and other clinical information in the journal. The patient's parent understands that the names and initials will not be published and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed.

\received

DD MMMM YYYY \acceptedDD MMMM YYYYAcknowledgments

None.

Funding

None.

Author contribution

I.K and S.K were involved in the original drafting, writing, and reviewing of the article. I.K supervised the article. K.A, Z.R, and F.A were involved in data curation and supervision. K.A, was also involved in conceptualization and editing.

Conflicts of interest

There are no conflicts of interest.

References:

1. Saravelos SH, Cocksedge KA, Li TC. Prevalence and diagnosis of congenital uterine anomalies in women with reproductive failure: a critical appraisal. Hum Reprod Update. 2008;14(5):415–29. 2. Mikos T, Gordts S, Grimbizis GF. Current knowledge about the management of congenital cervical malformations: a literature review. Fertil Steril. 2020;113(4):723-32. 3. Ludwin A, Pfeifer SM. Reproductive surgery for müllerian anomalies: a review of progress in the last decade. Fertil Steril. 2019;112(3):408–16. 4. Rock JA, Roberts CP, Jones Jr HW. Congenital anomalies of the uterine cervix: lessons from 30 cases managed clinically by a common protocol. Fertil Steril. 2010;94(5):1858–63. 5. Kimble R, Molloy G, Sutton B. Partial cervical agenesis and complete vaginal atresia. J Pediatr Adolesc Gynecol. 2016;29(3):e43-7. 6. Dornelas J, Jármy-Di Bella ZIK, Heinke T, Kajikawa MM, Takano CC, Zucchi EVM, et al. Vaginoplasty with oxidized cellulose: anatomical, functional and histological evaluation. Eur J Obstet Gynecol Reprod Biol. 2012;163(2):204-9. 7. Agbor VN, Njim T, Aminde LN. Pyosalpinx causing acute appendicities in a 32-year-old Cameroonian female: a case report. BMC Res Notes. 2016;9:1–4. 8. Cho HW, Koo YJ, Min KJ, Hong JH, Lee JK. Pelvic inflammatory disease in virgin women with tubo-ovarian abscess: a single-center experience and literature review. J Pediatr Adolesc Gynecol. 2017;30(2):203-8. 9. Hakim J, Childress KJ, Hernandez AM, Bercaw-Pratt JL. Tubo-ovarian abscesses in nonsexually active adolescent females: a large case series. J Adolesc Heal. 2019;65(2):303-5. 10. Mikos T, Lantzanaki M, Anthoulakis C, Grimbizis GF. Functional and reproductive outcomes following surgical management of congenital anomalies of the cervix: a systematic review. J Minim Invasive Gynecol. 2021;28(8):1452–61. 11. Kriplani A, Kachhawa G, Awasthi D, Kulshrestha V. Laparoscopicassisted uterovaginal anastomosis in congenital atresia of uterine cervix: follow-up study. J Minim Invasive Gynecol. 2012;19(4):477–84. 12. Casey AC, Laufer MR. Cervical agenesis: septic death after surgery. Obstet Gynecol. 1997;90(4 Part 2):706–7. 13. Rock JT clinical management of congenital absence of the uterine cervixA, Schlaff WD, Zacur HA, Jones Jr HW. The clinical management of congenital absence of the uterine cervix. Int J Gynecol Obstet. 1984;22(3):231-5. 14. Maraqa T, Mohamed M, Coffey D, Sachwani-Daswani GR, Alvarez C, Mercer L. Bilateral recurrent pyosalpinx in a sexually inactive 12-year-old girl secondary to rare variant of Mullerian duct anomaly. Case Reports. 2017;2017:bcr-2016. 15. Murata T, Endo Y, Furukawa S, Ono A, Kiko Y, Soeda S, et al. Successful laparoscopic resection of ovarian abscess caused by Staphylococcus aureus in a 13-year-old girl: a case report and review of literature. BMC Womens Health. 2021;21(1):198.

Figures

Figure 1 : A complex multilocular thick-walled cystic ovarian mass with heterogenous high T2 signal (A) and irregular thick uniform enhancing wall and septa in post contrast T1 images (B) at left adnexa is noted with restricted diffusion in DWI sequence (not shown), causing pressure on the adjacent uterus associated with surrounding pelvic fat haziness suggestive for Tubo-Ovarian Abscess (TOA).

Figure 2 : A high T2 signal intensity track (A) is seen between the left anterior wall of the lower rectum and the left posterior wall of the vagina with surrounding fat haziness and increase enhancement in post-contrast T1 images (B), in favor of the rectovaginal fistula.

Hosted file

TOA figures.docx available at https://authorea.com/users/885630/articles/1263759conservative-management-of-tubo-ovarian-abscess-as-a-complication-of-reconstructivesurgery-for-congenital-cervicovaginal-agenesis-a-case-report-and-review-of-literature