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CASE REPORT**Cervical Agenesis**

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Abstract

Background : Cervical agenesis is an extremely rare congenital anomaly of female reproductive system with manifestation on the absence of uterine cervix with normal development of uterine and secondary sex signs. According to the American Fertility Society, cervical agenesis is classified as type IB Müllerian anomaly. Symptoms are include primary amenorrhea, cyclic abdominal lower pain and infertility.

Objective : To report the treatment of cervical agenesis.

Case Report : Reportedly, a woman, 28 years old, came with complaints of primary amenorrhea, cyclic abdominal lower pain and 3 years of primary infertility. From speculum and transvaginal ultrasonography examination, the uterine was slightly smaller than normal, without cervical uterine, found adhesion, both ovarian were normal. Laparoscopic utero-vaginal anastomosis was performed and catheter condom was maintained in the cervical canal for 3 months. Two weeks after catheter was removed, a hysteroscopy was performed, but the canal was found closed.

Conclusion : In this case, after the laparoscopic utero-vaginal anastomosis was performed, the cervical canal which has been made, is found closed. So, a further evaluation is needed to get more effective results.

Keywords: Type IB Müllerian anomaly, cervical agenesis, primary amenorrhea, infertility, laparoscopic utero-vaginal anastomosis

INTRODUCTION

Cervical agenesis is a congenital abnormality of the female reproductive system which is very rare with an incidence of about 1 per 80,000 -100,000 births.¹ Manifestations include non-formation of the uterine cervix with development of the uterus and normal secondary sex signs. Based on the American Fertility Society, cervical agenesis is grouped in IB type Müllerian anomalies.² Symptoms that appear are primary amenorrhoea, cyclic lower abdominal pain and infertility. Early diagnosis and treatment will provide a significant advantage in the management of patients with cervical agenesis.¹

CASE REPORT

Reportedly a woman, 28 years old, married, came to the gynecological clinic with complaint of cyclic lower abdominal pain felt since the past 1 year. The patient was known to have never

menstruated. The patient has been married for 3 years but has not had children. From the gynecological examination it was found the characteristics of normal secondary sex development, breast growth, axilla hair and Tanner 3-4 pubis. In external genitalia, labia majora and minora appeared normal. In the inspecular examination, the vagina was shorter than normal (3 cm), visualization of the external uterine portio and ostium was not obtained (Figure 1).



Figure 1. Speculum Examination

On transvaginal ultrasound examination obtained an anteflexive uterus measuring 4.9 x 3.08 cm, no cervical utery features detected (Figures 2 and 3). The right ovary was 2.42 x 2.16 cm and the left ovary was 3.29 x 1.90 cm (Figure 4).



Figure 2. Uterus-USG Transvaginal

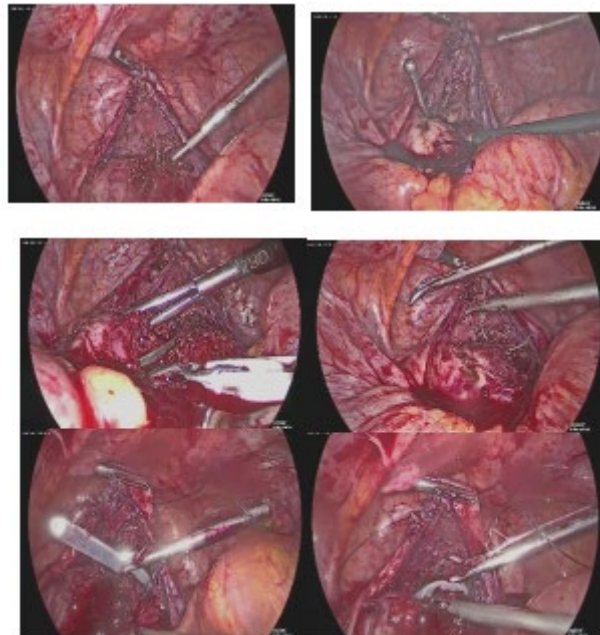


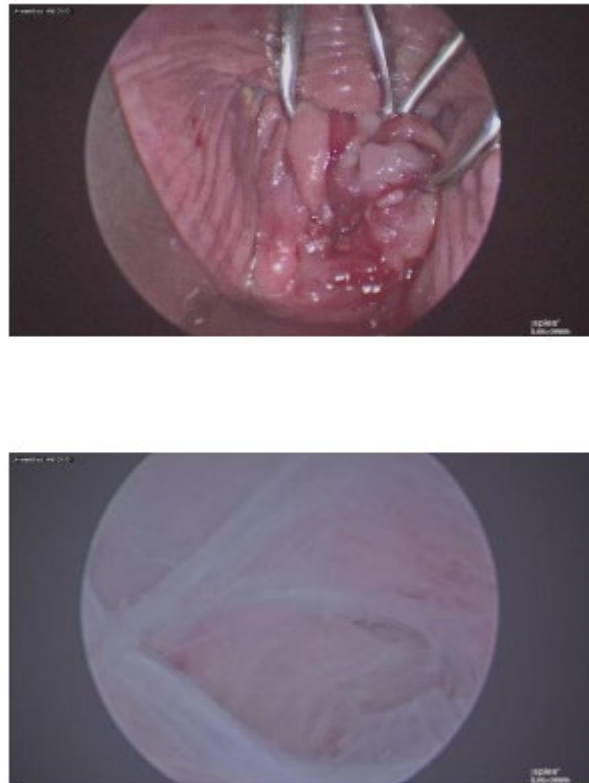
Figure 3. Uterus - USG Transvaginal

**Figure 4.** Right and Left Ovaries - Transvaginal Ultrasound

From laboratory tests the results obtained: FSH levels = 5.91 mIU/ ml (3-20 mIU/ ml), LH = 2.39 mIU/ ml (<7 mIU/ ml), TSH = 1.2 μ IU/ ml (0 , 4 - 4 μ IU/ ml), Prolactin = 9.7 ng/ ml (<24 ng/ ml).

Laparoscopic utero-vaginal anastomosis was performed. After the peritoneum was penetrated, the vesicouterine plica was separated into the lower uterine segment then an incision was made in the lower uterine segment until it penetrated the uterine cavity. Next an incision was made into the area of the vaginal crest, then a number 16 catheter condom was inserted until it reached the uterine cavity and a catheter balloon was developed. Catheter placement was maintained for 3 months then the catheter was removed (Figure 5). Two weeks after the catheter was removed, hysteroscopy was performed, it was found that the channel was made to close again (Figure 6).

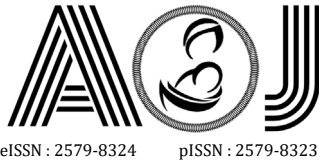
**Figure 5.** Laparoscopic Utero-vaginal Anastomosis

**Figure 6.** Hysteroscopy**DISCUSSION**

Based on the American Fertility Society, cervical agenesis was classified in the Müllerian type anomaly.² Cervical atresia has been further classified into the following types: (i) the cervix is still intact with cervical os obstruction, (ii) the cervix consists of fibrous tissue, (iii) Fragmented cervical section, (iv) Hypoplastic center of the cervix with rounded edges.³ Imaging modalities must be able to assess the condition of the cervix correctly because it will adapted with the method of operation or reconstruction. So far, MRI has been considered a consistently superior method for evaluating the anatomy of the vagina and cervix but ultrasound remains a routine and important examination.⁴

Obstructive uterine abnormalities will block normal menstrual flow resulting in primary amenorrhea. Any abdominal or pelvic pain in a woman who has entered puberty should consider the possibility of obstructive genital anomalies. Clinically it may appear with obstructive symptoms such as hematometra, hematocolpos, lower cyclic abdominal pain, or asymptomatic depending on the functional status of the endometrium. Endometriosis can develop from retrograde menstruation in such cases. Clinical examination helps identify the lower genital tract. Other anomalies such as imperforate hymen must be considered.^{5,6}

Imaging modalities such as ultrasound and MRI are very helpful especially correlated with careful pelvic examination findings. The initial suspicion of cervical agenesis can be easily



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done by ultrasonography. Three-dimensional ultrasonography can increase the ability to accurately describe cervical anatomy even though it requires an expert operator and special knowledge about anatomy.^{5,7}

At present the management of uterine cervical anomalies is controversial. It is advisable to carefully determine cervical anatomic abnormalities while considering the patient's wishes, after which a decision must be made for the final management. Making the endocervical canal for menstrual flow will allow to save the uterus.^{8,9} Success of cervical-uterine recanalization through laparotomy¹⁰ or laparoscopic assistance^{11,12} has been reported. Spontaneous pregnancy has been reported after conservative surgical therapy in cervical agenesis. Assisted reproductive technology can be considered in cases where future infertility is possible. Uterovaginal anastomosis itself has a high risk of secondary stenosis after the action which is 40-60%.⁶ The use of Small Intestinal Submucosa (SIS) graft has been proven to prevent secondary stenosis and increase the success rate of uterovaginal anastomosis.¹⁶

In the case presented, after uterovaginal anastomosis, secondary stenosis occurs during the evaluation of 2 weeks after the catheter was removed so an evaluation was required regarding more effective techniques and procedures.

CONCLUSION

Cervical agents are very rare congenital abnormalities of the female reproductive system that present challenges in establishing diagnosis and determining effective management. Conservative surgical treatment with uterovaginal anastomosis procedure aims to save the uterus by making the menstrual tract, relieve pain and maintain reproductive potential. A deeper evaluation of techniques and procedures is needed to get more effective results.

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