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Case Report: Unilateral Ovarian Absence (UOA) with Unilateral Ovarian Hypoplasia and Uterine Hypoplasia

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ABSTRACT

This case report concerns a woman diagnosed with unilateral ovarian absence (UOA) with right ovarian hypoplasia and uterine hypoplasia. She had not attained menarche till seventeen years of age. She was presently admitted to the hospital with complaints of primary amenorrhea with acute pelvic pain, which she had in the last two days before admission. Clinical examination revealed right-sided abdominal tenderness. Her vagina, and vulva were normal. Physical examination showed no axillary, pubic hair and no breast developments. Her height was 3 feet and body temperature were normal, there were no urinary or bowel symptoms. A trans-vaginal ultrasound scan revealed uterine hypoplasia and right-side ovarian hypoplasia. The left ovary could not be identified during the scan. After consultation, she consented to and underwent an emergency diagnostic laparoscopy. The laparoscopy confirmed the diagnosis of right ovarian hypoplasia, but also revealed absence of the left ovary, its associated ligaments, and the distal part of the ipsilateral fallopian tube. The patient was followed up with a whole-abdomen CT scan, which did not reveal urinary tract variations or malformations. Karyotyping was also done, result showed normal female karyotype (46, XX).

Keywords: Unilateral ovarian absence, Unilateral ovarian hypoplasia, Uterine hypoplasia

INTRODUCTION

A 17-year-old female patient was presented with primary amenorrhea with acute pelvic pain. Clinical examination revealed right-sided abdominal tenderness. Her vagina, and vulva were normal. Physical examination shows no axillary, pubic hair and no breast developments. Her height was 3 feet and body temperature were normal, there were no urinary or bowel symptoms. And there was no history of consanguineous marriage of parents and no family history of any abnormal pregnancies. Her history was unremarkable, and β-hCG test was negative. Laboratory tests revealed a serum follicle-stimulating hormone and estradiol levels of 2.50 mIU/ml and 42 pg/ml, respectively.

A trans-vaginal ultrasound scan revealed uterine hypoplasia measuring 1.0*0.7*2.8cm and right-side ovarian hypoplasia measuring 0.9*0.6cm. The left ovary could not be identified during the scan. After consultation, she consented to and underwent an emergency diagnostic laparoscopy. The laparoscopy confirmed the diagnosis of right ovarian hypoplasia, but also revealed absence of the left ovary, its associated ligaments, and the distal part of the ipsilateral fall opiantube. The patient was followed up with a whole-abdomen CT scan, which did not reveal urinary tract variations or malformations. Karyotyping was also done, result showed normal female karyo type (46, XX).

EPIDEMIOLOGY

Unilateral ovarian absence (UOA) is a rare event. It has a prevalence of one in 11,240 women [1]. Even rarer is the absence of both the ovary and distal portion of the ipsilateral fallopian tube. It has been stated that the incidence of UOA could be higher than that reported in 1986 and the ever-increasing number of cases in the literature supports this view. This could be because of either an unknown environmental factor or the more widespread use of laparoscopy, which has revealed cases that otherwise would have remained undiagnosed [2].

PATHOPHYSIOLOGY

It has been suggested that this phenomenon may be a result of a congenital malformation or an ischemic event due to mechanical alterations (e.g., torsion and subsequent resorption of the ovary) that occur during fetal life or childhood [3]. Women with mullerian-duct anomalies might

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be more prone to ovarian torsion due to abnormal anatomic connections between the ovary and the pelvic sidewall [4].

When only the distal part of the ipsilateral fallopian tube is missing along with UOA, the proposed mechanism is ovarian torsion with necrosis and resorption, which can occur either antenatally or postnatally. Adnexal torsion and infarction is usually associated with severe symptoms and is a well-documented surgical emergency in both adults and children. When the entire fallopian tube is missing, it is probably the result of a congenital malformation (agenesis or aplasia). This is explained embryologically by developmental alterations of the paramesonephric (Müllerian) ducts and unknown paracrine and autocrine signals, which result in agenesis of the associated gonad [4]. This condition potentially coexists with malformations of the uterus and/or urinary tract, such as unicornuate uterus, unilateral renal agenesis, and other variations [5].

CLINICAL MANIFESTATIONS

Consequently, some authors support the view that "asymptomatic" torsion cannot occur postnatally [6]. Other authors have suggested that symptoms may be minimal or even absent, especially when torsion occurs during pregnancy [3].

In this case, there was an abrupt interruption of the fallopian tube and absence of any associated ovarion, uterine or other abnormalities. The combination of these findings, along with no indication in the patient's history, suggests that the most likely cause was an asymptomatic ischemic event and resorption during fetal life or after birth.

DIAGNOSTIC EVALUATION

Diagnostic laparoscopy plays a vital role in the diagnosis of these abnormalities. The principle of "three-cycle inspection" is a standard step in all laparoscopic procedures. The first cycle is a clockwise inspection of the upper abdomen before placing the patient in the Trendelenburg position. The second cycle is carried out after insertion of the accessory trocars and preparation of the surgical field and refers mainly to the anatomical structures that enter the pelvis at the level of the pelvic brim. The third cycle involves visualization of the internal genitalia and lower pelvis with the assistance of a uterine manipulator when required and is performed after placing the patient in the maximum Trendelenburg position [7].

It is also important to note that in the clinical setting of evaluating common, acute gynecological emergencies, the suspicion of such a rare abnormality may be raised by preoperative ultrasound imaging findings, drawing the surgeon's attention to actively search for the "missing" ovary. If the ovary cannot be clearly visualized during the ultrasound study, the possibility of UOA should be considered as a differential diagnosis. It is believed that a thorough inspection of the whole abdomen is mandatory whenever laparoscopy is performed despite the adverse conditions that can be encountered during an emergency procedure [7].

EFFECT OF UOA ON CONCEPTION AND FERTILITY

It has been demonstrated, women with UOA and/or fallopian tube absence are fertile, so quite possibly some of these women were fertile and might have raised offspring while unaware of their condition [8]. But in this case women is having right ovarian hypoplasia so it might be difficult to conceive or be fertile.

In 2008, Garret proposed an interesting theory based on his observations of discontinuous fallopian tubes in non-sexually active adolescent girls. This theory stated that the fimbria could have an embryologic origin distinct from that of the rest of the fallopian tube and potentially in common with the ovary [9]. This suggestion is in line with studies involving the fimbria as the likely site of origin of ovarian epithelial neoplasms. The variety of congenital and acquired malformations could offer new insight into the embryologic and developmental processes involved and may prove beneficial for the management of ovarian epithelial neoplasms [10].

MANAGEMENT

There are limited options for management. Oral contraceptive treatment can be given in order to support secondary sexual characteristics and to prevent long term complications. Drugs like metformin and clomiphene citrate can be used to induce ovulation. GnRH agonist IVF-ET, can result intrauterine pregnancy. In-vitro fertilization would be the logical therapy, but commonly associated uterine anomalies may have an undetermined impact on IVF outcome. If the ovarian torsion is there then surgery to untwist the ovary or to remove an ovary damaged by lack of blood supply, typically laparoscopic surgery is used for less severe cases.

In this patient hormonal therapy such as Tab. Progynova 2 mg daily (D1-D21), Tab. Meperate 10 mg OD (D12-D21) along with Tab. Eltroxin 75 μg OD, Tab. D 500 cal 500 mg B.D was given for 3 months to start ovulation from the right ovary and then menstrual cycle started.

A recent case in America reported intrauterine pregnancy achieved by in vitro fertilization using luteal-phase Leuprolide Acetate, follitropin and recombant human chorionic gonadotropin [11].

CONCLUSION

UOA is a very rare condition and can coexist with total or partial absence of the ipsilateral fallopian tube, is associated with many gynecological and reproductive morbidities and can impact a woman's reproductive capabilities. When the entire fallopian tube is missing along with ovary, it is probably the result of a congenital malformation (agenesis or aplasia). In our case, there was no indication in the patient's

history, suggests that the most likely cause was an asymptomatic ischemic event and resorption during fetal life or after birth. Pregnancy in this condition can be considered as high risk or poor reproduction potential.

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