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Case Report
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A Rare Case of Recurrent Dysmenorrhea due to Endometriosis: Müllerian Anomaly in a Female with a Rudimentary Uterus

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Abstract

Introduction: A rudimentary uterus is a rare congenital pathology linked to an abnormality in the development of the Mullerian system. It is closely associated with obstetrical complications such as recurrent abortion, ectopic pregnancy and hysterorrhexis.

Case: A 27-year-old unmarried female was admitted with recurrent dysmenorrhea for 8 months. Gynecological 3D ultrasound revealed uterine malformations and a large irregular complex mass in the left hemipelvis with multiple cystic and solid components. Therefore, a left hysterosalpingectomy and ovarian endometriotic cystectomy were performed for her. No recurrence of dysmenorrhea was observed within 1 year follow-up.

Discussion: Patients suffer from pelvic pain attributable to the increased prevalence of endometriosis, which is caused by retrograde flow of menses through the obstructed horn. Ultrasound is indicated as the first test for rudimentary uterus, while MRI is found to be the optimal choice in the diagnosis as well as for surgical planning. Resection of the rudimentary horn is the definitive treatment modality, regardless of conventional surgery or laparoscopically.

Keywords: Müllerian anomaly; Rudimentary uterus; Dysmenorrhea, endometriosis

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Introduction

Müllerian anomalies are congenital defects of the female genital system that arise from abnormal embryological development of the Müllerian ducts. A rudimentary uterus is a rare Müllerian anomaly [1]. In the female embryo, when one of the Müllerian ducts develops incompletely, an partial accessory horn fuse with the duct of the opposite side, leading to the development of an accessory horn which may have a functional cavity [2]. Rudimentary uterus may lead to symptoms such as pelvic pain, dysmenorrhea, recurrent abortion, ectopic pregnancy and hysterorrhexis.

Majority of the cases remain undiagnosed unless a pregnancy develops in the accessory horn, and most of these patients present when critical rupture has already occurred [3]. Here we report a case of recurrent dysmenorrhea in an unmarried female with Müllerian anomaly; we make a definite diagnosis of rudimentary uterus causing endometriosis, which result in the symptom of recurrent dysmenorrhea.

Case Presentation

A 26-year-old female (G0P0), was presented to our gynecological clinic for recurrent dysmenorrhea. She suffered dysmenorrhea since her menarche 13 years ago, at first; the pain was mild and lasted for 1 day. In the last three years, she has been felt pelvic pain, a scale of 6, and was treated with analgesics. Her dysmenorrhea was significantly worse than before 6 months ago, and analgesics were invalid in relieving her pain. She was diagnosed with renal agenesis during health check-ups 8 years ago.

Gynecological examination indicated wide uterus fundus with irregular shape, left adnexal masses could be touched with poor mobility. Hypogastrium tenderness was present. Gynecological ultrasound showed uterine abnormality, left uterine cavity effusion, left pelvic mass (encapsulated

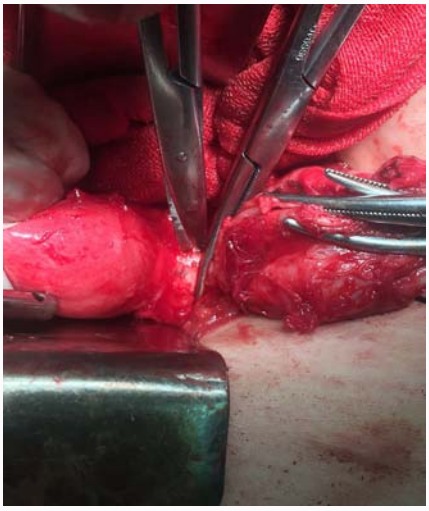

Figure 1: Intraoperative view.

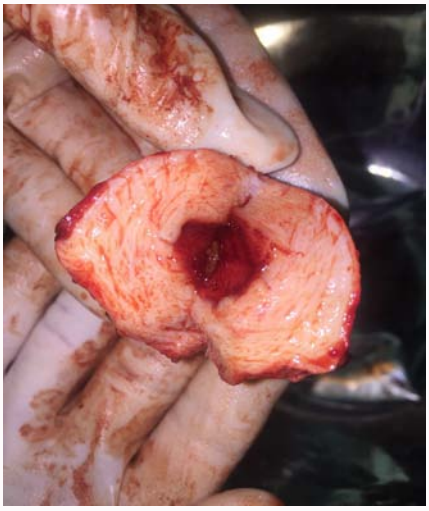

Figure 2: Surgical specimen.

effusion).

Since uterine malformation combined with pelvic endometriosis cyst meet surgical indicator, we performed exploratory laparotomy for her. During surgical process, we further confirmed the diagnosis of "rudimentary uterus and ovarian endometriosis cyst", bilateral uterine bodies were of equal size, and the left uterine body was connected to the right uterine isthmus, the connection was solid with thickness of 0.8 cm (Figure 1). Therefore, we performed "left rudimentary uterus hysterectomy + left ovarian endometriotic cystectomy" for her. When we dissected specimens, we found oviduct opening at the funnel mouth, and the solid connection between left uterine body and right uterine isthmus (Figure 2). Postoperative pathological results were also consistent with clinical diagnosis. She accepted treatment of GnRH and was discharged after 1-week hospitalization without any complications. During the 1-year follow-up, she didn't suffer from dysmenorrhea anymore.

Discussion

Etiology of Müllerian anomaly is unclear, and approximately 4% of the cases are familial with affected siblings. A unicornuate uterus arises from normal development of one Müllerian duct in the presence of arrested development of the contralateral duct. Moreover, up to 67% of rudimentary horns are accompanied by abnormal development of ipsilateral urinary system [4]. In this case, the patient's renal agenesis reminded us possibility of Müllerian anomaly.

The rudimentary horn uterus is classified into type IIA unicornuate uterus, among which the rudimentary horn uterus is divided into the following three types: Type II A-1a (rudimentary horn uterus has a uterine cavity without a cervix, which is connected to the developmental unicornuate uterine cavity), type II A-1b (rudimentary horn uterus has a uterine cavity without cervix, but is isolated with the developmental side), type II A-1c (rudimentary horn uterus doesn't has uterine cavity nor cervix) [5]. The unicornuate uterus in this case belong to type II A-1b, patient got a rudimentary horn with a non-communicating cavity.

Symptoms vary depending on the subtypes. The subtype with a non-communicating functional rudimentary horn (type II A-1b) generally manifests as dysmenorrhea and hematometra at menarche [6]. Since endometrium in the rudimentary horn remains functional and is isolated with developmental uterine cavity, retrograde flow of menses through the obstructed horn may lead to increased prevalence of endometriosis, which gradually aggravates dysmenorrhea. Therefore, patients suffer from periodic pelvic pain during menstruation. The remaining two subtypes are usually diagnosed incidentally or during infertility work-up or obstetric surgery.

Different imaging modalities could be used for early diagnosis of the accessory horn of uterus including transvaginal ultrasound, 3D ultrasonography or MRI. Diagnosis can be confirmed with ultrasonography; however, MRI can better diagnose complex malformations including the rudimentary uterus as well as for surgical planning [7].

Rudimentary uterus may lead to symptoms such as pelvic pain, dysmenorrhea, recurrent pregnancy loss and premature delivery [8]. Once the diagnosis of rudimentary uterus is confirmed, surgical resection is the prior choice [9]. Resection of rudimentary horn with ipsilateral salpinx is conducive to avoid occurrence of endometriosis caused by retrograde flow of menses, while indications of ovarian function preservation were based on needs of the ages and the procreation of patients. The round ligament should be fixed on the developmental uterus horn to prevent the uterus from deviating.

We report a case of recurrent dysmenorrhea due to increased prevalence of endometriosis, which is caused by retrograde flow of menses in a unicornuate uterus, in hope of expanding the knowledge of a rare occurrence. This case also highlights the importance of considering the diagnosis of Müllerian anomaly in patients with a history of other anomalies, and history of early-age secondary dysmenorrhea.

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