

Functional Rudimentary Horn as a Rare Cause of Pelvic Pain: A Case Report

Maliheh Arab^{1,2,*}; Sepideh Mehdighalb¹; Donya Khosravi¹

¹Preventative Gynecology Research Center (PGRC), Imam Hossein Medical Center, Shahid Beheshti University of Medical Sciences, Tehran, IR Iran

²Shahid Beheshti University of Medical Sciences, Tehran, IR Iran

*Corresponding Author: Maliheh Arab, Preventative Gynecology Research Center (PGRC), Imam Hossein Medical Center, Shahid Beheshti University of Medical Sciences, Tehran, IR Iran. Tel: +98-9121593277, Fax: +98-2177543634, E-mail: drmarab@yahoo.com

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Introduction: Pelvic pain results from many causes such as primary dysmenorrhea, uterine anomalies, menstrual outflow obstruction, endometriosis, myoma and adenomyosis. This study reports on a rare case of non-communicating functional rudimentary horn.

Case Presentations: A 15-year-old nulligravida young woman with a history of severe intermittent pelvic pain presented a 4-5 centimeter mass. A surgical procedure for appendicitis was previously performed on this patient. Per-operative diagnosis was myoma and suspicion of leiomyosarcoma. Laparotomy revealed left rudimentary horn, non-communication was confirmed by postoperative hysterosalpingogram (HSG) and magnetic resonance imaging (MRI). Resection of mass and left fallopian tube was done during the second surgery.

Conclusions: Rudimentary horn should be considered in differentiation of pelvic pain and mass in young females. Early diagnosis and horn resection prevents emergency surgery and reliefs pain.

Keywords: Pelvic Pain; Dysmenorrhea; Pregnancy; Rupture; Congenital Abnormalities

1. Introduction

Dysmenorrhea is reported in about 60% of females of the reproductive age. Dysmenorrhea is categorized as primary when no pelvic pathology is revealed and secondary when there is a known pathological source. Characteristics of secondary dysmenorrhea include beginning of pain 7-14 days before menstruation, continuation of pain after menstruation, resistance to non-steroidal anti-inflammatory drugs and contraceptive pills. In case of secondary dysmenorrhea, uterine and vaginal anomalies, menstrual outflow obstruction, endometriosis, adenomyosis and uterine myoma are considered in the differential diagnosis (1). Some of the congenital abnormalities in the mullerian duct present with dysmenorrhea including imperforated hymen, cervical atresia, transverse septum of the vagina and rudimentary horn besides unicorn unite uterus. In imperforated hymen, there is fusion of the sinovaginal bulbs (2). Cervical atresia and hypoplasia occur with diethylstilbestrol (DES) exposure. Complete absence of the cervix might result in pain and obstruction for which hysterectomy is necessary (3). Transverse vaginal septum might be in association with DES exposure. Clinical presentation is similar to imperforated hymen (2). The incidence of mullerian duct malformation in the general population is estimated to be 43% while that of unicornuate uterus is about 0.4% (4). In another study unicornuate uterus is about 2.5-13.2% of uterine malformations (5). Unicornuate uterus resulting from defect of mullerian duct development might coexistent with rudimentary horn. Rudimentary horn preg-

nancy could present a clinical course like ectopic pregnancy. Sometimes chronic pain is observed. Removal of rudimentary horn is indicated (1). In the present study a rare case of non-communicating functional uterine horn is introduced.

2. Case Presentations

A 15-year-old female presented severe pain during her menstruation period. History of her intermittent pain showed initiation of pain with menarche. History of appendectomy belonged to about 9 months ago. Ultrasonography performed two months ago, revealed a mass of 4.5 centimeters in her left adnexa. Characteristics of the mass were in favor of a hemorrhagic cyst. Another sonography described the mass as a 5 cm solid tumor in favor of uterine myoma on the left side. She was admitted to an academic hospital in Tehran, Iran in 2012. Expert sonography of this center and color Doppler velocimetry reported a 5.3 centimeters myoma on the left side with high vascular flow suggesting leiomyosarcoma or degenerated myoma. She was scheduled for laparotomy by a midline incision. Surgical findings revealed bilateral normal appearing ovaries. On the left side there was a solid mass with the fallopian tube connected superior to this mass and in a lower position in comparison to the right side. A rudimentary horn was considered. Thus, a small incision was made on the mass extracting about 2 cm³ of chocolate fluid compatible with old blood. Exploration of the cavity by a narrow metal applicator, showed a small cavity without an outflow pathway. At this stage, the ru-

rudimentary horn was confirmed and its communication to main cavity was under doubt. Thus, dye injection via the cervix was done. Blue dye transit from fallopian tubes occurred without entry to the peritoneal cavity. Due to no bilateral entrance of dye, while passing from tubes, technical problems was not ruled-out. In the operative field non-communication of the rudimentary horn was not definite. Biopsy of rudimentary mass and cavity was done for pathological review and surgical intervention was not taken. The treatment plan was the use of contraception while hysterosalpingography and MRI were performed to achieve definite diagnosis before the next surgery. Unfortunately the patient rejected surgery due to post-surgical pain relief resulting from evacuation of chocolate fluid. Pathological report of mass biopsy suggested a "vascular myoma" and cavity biopsy reported "inactive endometrium". Hysterosalpingography (HSG) report indicated a right unicornuate uterus with normal right fallopian tube and normal peritoneal spillage. The other side of the rudimentary horn and fallopian tube was not shown by the HSG examination. The MRI report about 1.5 month after the operation revealed a 4 cm mass on the left side of the uterus with a small central cystic blood-containing locus of about 1.1 cm. Diagnosis of a non-communicating rudimentary horn was definite. The patient accepted a third operation after menarche and resection of rudimentary horn and left fallopian tube was done about 10 months after the second surgery, due to recurrence of severe pain. Pathological review confirmed the diagnosis (Table 1).

3. Discussion

Uterine anomalies are rare and they are seldom considered in differential diagnosis of patients with clinical presentation of pelvic pain (6). In the present case, uterine myoma was considered as the first diagnosis and uterine anomalies were not mentioned in differential diagnosis. Most rudimentary horns are asymptomatic due to their non-functional and non-communicating pattern (3). In the case of endometrial development in rudimentary horn, menstrual fluid is produced and implantation

of conception product might occur. Thus, rudimentary horn pregnancy and pelvic pain occur. Rudimentary horn pregnancy mimics the clinical presentation of ectopic pregnancy resulting in surgical emergency (7). Rudimentary horn pregnancy has been described by several case report articles. Dhar reported on a rupture of rudimentary horn pregnancy at 22 weeks. The ultrasound revealed that the fetus was dead and intraperitoneal free fluid was observed. Laparotomy findings showed rupture of horn besides a dead fetus. Rudimentary horn and ipsilateral fallopian tube (8). Thakur et al. also reported on a ruptured rudimentary horn pregnancy at 19 weeks. Ultrasound revealed an empty uterus and free fluid in the abdominal cavity. In the laparotomy procedure, rudimentary horn and fallopian tube resection was done (9). Another case report of ruptured rudimentary horn pregnancy at 19 weeks was by Hassan et al. (7). Rudimentary horn pregnancy is visually miss-diagnosed until the occurrence of rupture and emergency. There is a case report on an asymptomatic 33-year-old pregnant woman who was diagnosed by laparoscopy-hysteroscopy following the diagnosis of ectopic pregnancy. In the laparoscopy, both fallopian tubes were normal and diagnosis of rudimentary horn pregnancy was made. Hysteroscopy confirmed the diagnosis and surgical resection was done. This case cooperated due to hysteroscopic and pre-rupture diagnosis. In our case, intraoperating dye injection was done, as well, although definite diagnosis was not done. As mentioned earlier, diagnosis of rudimentary horn is not made until the reproductive age when ruptured rudimentary horn pregnancy or pelvic pain occur. Pain is due to endometrial response to hormonal activities after menarche. In the present case, symptoms of pain started from menarche. Her pain was cyclic and a mass existed besides the uterus. Her history included several surgical interventions and miss diagnosis with appendicitis and other entities. In the rare case of unicornate non-communicating horn, surgical resection is necessary to relieve pain and prevent rudimentary horn pregnancy and resulting emergency situations (10). This case is a rare presentation of uterine anomaly. Surgery was done more than two years after menarche as

Table 1. Summary of Clinical and Paraclinical Findings of the Studied Case

Cyclic pain from menarche
First surgery after menarche due to pain and diagnosis of appendicitis
5 cm solid mass with high vascularity on the left side of the uterus suggesting leiomyosarcoma or degenerated myoma
Second surgery after menarche due to pain and a solid mass on the left side of the uterus with anatomical features of rudimentary horn revealed during the surgery
Intraoperative findings of the second surgery: 1. Anatomical features of rudimentary horn; small chocolate aspirate of the mass. 2. Passing due to bilateral fallopian tubes without peritoneal entrance, which made a non-communicating rudimentary horn indefinite
Hysterosalpingography and MRI after the second surgery allowed the diagnosis of non-communicating functional rudimentary horn
Third surgery: resection of non-communicating functional rudimentary horn

the third surgical intervention. In the first surgery diagnosis was appendicitis due to pain. After the second surgical intervention definite diagnosis was made. Due to transient pain relief after the second surgery, the patient and her family rejected definite surgery until she experienced severe pain again within 10 months. Intraoperative limitation of case management (in the second surgery) was inability to show open tube on the right side, thus definite diagnosis and surgery at that stage was not possible. Hysteroscopy was acceptable, although it was not done due to the inability to diagnose this case. Rudimentary horn should be considered in cyclic pelvic pain starting after menarche besides a mass like lesion, sometimes mistaken by myoma. Early diagnosis is needed to relief pain and prevent surgical emergency by rudimentary horn resection.

Authors' Contributions

All authors contributed to the performance of this project and the preparation of the manuscript, and all approved the final version of the manuscript.

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