J Cases Obstet Gynecol, 2017;4(4):87-90

Case Report

Dysmenorrhea in association with non-communicating rudimentary uterine horn of a unicornuate uterus: Case report

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Abstract

One of the most frequent reasons of pelvic pain is dysmenorrhea. It not only decries patients daily of life but also results in activity of restriction. Although a physical examination, including a pelvic examination in patients may reveal the cause, preoperative diagnosis of such cases can be challenging and they also can be miss-diagnosed occasionally and thus could be led into bigger problems in the post period time. In this study, we report the case of an 18-year-old adolescent who had recurrent pelvic pain regularly at the time of menses as well as routine menstrual cycle complaints. The patient underwent ultrasonography and magnetic resonance (MRI) imaging of the pelvis. The diagnosis was non communicating rudimentary uterine horn of a unicornuate uterus with two cervix according to test results. An urgent laparoscopy was performed and the symptoms and complaints were perished after the successful surgery. Diagnosis and management of this congenital anomaly was a challenging issue due to the complexity of the anatomic structures, nonspecific disagreements, and heterogenic presentation. The purpose of this paper is to clarify the diagnosis and therapy choices which could be remedy for this rare condition, moreover, to increase awareness of dysmenorrhea.

Key Words:

Uterus rudimentary horn, müllerian anomalies, dysmenorrhea

Introduction

Dysmenorrhea occurs in painful cramps during menstruation [1]. Reported prevalence is 16.8%-81% in overall, even some reports can be up to 90% while adolescents with severe dysmenorrhea is 42% [2]. Dysmenorrhea can be categorized into two groups; first one is no pelvic pathology before revealed and second one is resulting from identifiable organic diseases. Secondary dysmenorrhea develops with the beginning of pain in 7th- 14th days before menstruation, the pain continues after menstruation as a result of persistence to non-steroidal anti-inflammatory analgesics and oral contraceptives. Seconder dysmenorrhea depends on some crucial causes such as endometriosis, uterine myoma, adenomyosis, uterine and vaginal anomalies causing menstrual outflow obstruction [3]. Congenital abnormalities in the mullerian duct accompany with dysmenorrhea along with transverse septum of the vagina, cervical atresia, imperforated hymen and rudimentary horn besides unicorn unite uterus [4]. The European Society of Human Reproduction and Embryology (ESHRE) and the European Society for Gynaecological Endoscopy (ESGE) represented the classification system based on anatomic uterine anomalies with or without anomalies of vagina, cervix in 2013 [5]. All cases of hemi-uterus along with unilateral development of the uterus and the contralateral part, which could be either incompletely formed or absent are included in class U4 according to classification system of ESHRE and ESGE. Relevant class is subdivided into two sub-classes depending on

Article History:
Received: 22/05/2016
Accepted: 21/09/2017

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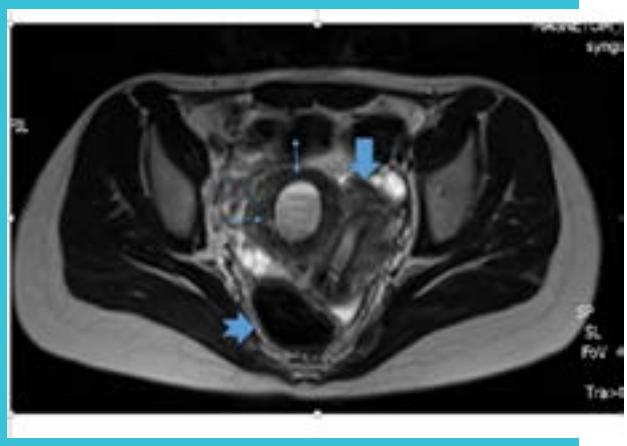
the presence (U4a) or absence (U4b) of a functioning communicating or non-communicating rudimentary horn [5]. The literature revealed that the unicornuate uterus is a case of an irregular lateral fusion fault arising on an incidence of 1/4020 women in the general population, in particular, unicornuate uterus is one of the least prevailing congenital uterine anomalies [6]. Either inadequate development of single side mullerian canal or its absence lead to unicornuate uterus. On the other hand, non-communicating accessory horns in other words dyspareunia or dysmenorrhea with an endometrial cavity (Class 2b) are the most commonly detected types according to American Fertility Society (AFS) classification system [7]. When dysmenorrhea is observed in adolescents, symptomatic treatment is usually attempted. However, one of the causes of dysmenorrhea may take place as a uterine anomaly. The purpose of this paper is to clarify the diagnosis and therapy choices which could be remedy for this rare condition, moreover, to increase awareness of dysmenorrhea.

Figure 1.



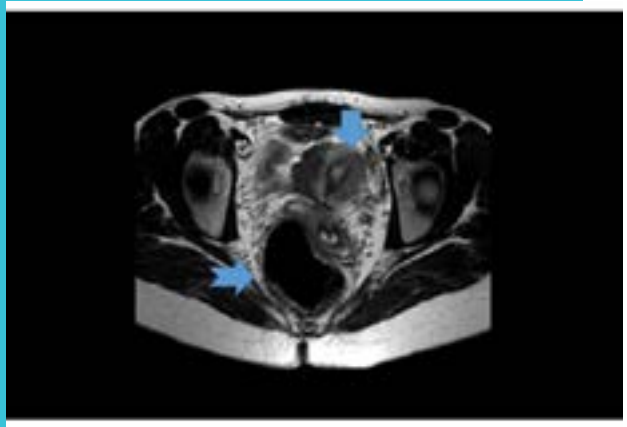
Axial T2-weighted image MRI acquired before the surgery, shows uterine duplication with two separate corpora. Right cavitary noncommunicating rudimentary horn (two sided arrow), uterus (arrow), bladder (open arrow) Endometrial thickening is identified in the right horn of the uterus (two sided arrow).

Case Presentation

An 18-year-old virgin adolescent presented at our department with complaints severe regular, progressive pelvic pain during her menses for the last three years. The patient experienced first menstruation at 14-year-old, while her regular cycle was 30 day intervals, her menstrual period was about 4 to 7 days with a normal amount of bleeding. During her menstruation she suffered from acute and severe right lower quadrant pain. She had no other medical or surgical illness, and per abdominal examination findings were all normal. The patient was taken to emergency room by complaining of lower quadrant pain. Acute abdominal findings with nausea and vomiting were initial conditions in the beginning of her period. Physical examination revealed diffuse tenderness of the lower abdomen in the right side of the pelvis. Tenderness, defense and rebound on the right lower quadrant were disclosed by physical examination indicating that the number of white blood cell (WBC) is 9,100 g/L, hematocrit 33,1 % and her blood human chorionic gonadotropin level was negative respectively. The patient was suspected acute appendicitis and responded to appendectomy where rudimentary horn was not able to detect and her pelvic pain was still existing after the procedure. After four months the patient was admitted to our clinic in the condition of complaining on dysmenorrhea. Gynecological examination shows that the hymen was intact, also the major and minor labia, urethral meatus and vaginal introitus were normal, whereas a small uterus was identified as a result of rectal examination. Ultrasonographic examination was firstly conducted on abdomen to assess the likelihood of unicornuate uterus with right sided hematosalpinx and hematometra. Secondly magnetic resonance imaging (MRI) was performed and proved that silent non-communicating rudimentary horn with a functioning cavity and two cervices with hematometra and hematocolpos exist on the right side (Figure 1). Also, both ovaries were normal in shape and size as well as urinary system screening showed no abnormality. The patient was delivered to laparoscopy in which intraoperatively right sided hematometra and hematosalpinx were identified, however, no endometriotic lesions were detected in the pelvis. Incision via harmonic scalpel into the serosa to myometrial tissue arose an efflux of chocolate-colored viscous fluid. The rudimentary horn was excised laparoscopically using through morcellation and the abdominal decayed area was cleaned and closed after accomplishing entire hemostasis as well. By that time blood loss was minimal, operating

time was 120 min. In addition, the removal specimen was sent for histopathological examination. Histopathology acknowledged a rudimentary horn with tissue composed of smooth muscle bundles. After the surgery she had no pelvic symptoms. Postoperative three months' control in MRI (Figure 2) patient anatomical image was completely normal.

Figure 2.



Postoperative axial T2-weighted image, uterus (arrow), bladder (open arrow)

Discussion

A non-interacting rudimentary horn along with a working endometrial cavity is a rare case , this type can account for ~10% (range 6-13%) of uterine anomalies [8] and beginning from menarche when hormonal activity was started, it may stimulate the endometrium of the rudimentary horn. It usually originates with pelvic pain following the menarche, dysmenorrhea, but the clinical presentations are variable, granted with obstetric complications which are miscarriage, ectopic pregnancy, intrauterine growth restriction, preterm labor or gynecological problems with infertility, ruptured horn, endometriosis, and chronic pelvic pain [9]. A history of pelvic pain following the menarche, intense dysmenorrhea and a unilateral pelvic mass are the main complaints suggesting congenital müllerian duct anomalies. The patient's severe dysmenorrhea predictable to intracavitary retention of menstrual effluent and retrograde menstrual flow. Hematometra causing distension of uterus might be the real reason of pain in these patients. Rudimentary horn pelvic pain has been described by several case report articles. Arab et al. reported on a pelvic pain a 15-year-old woman with a history of severe intermittent pelvic pain presented a 4-5 centimeter mass [10]. The ultrasound revealed high vascular flow suggesting leiomyosarcoma or degenerated myoma. Laparotomy findings confirmed that rudimentary horn and ipsilateral fallopian tube resection of non-communicating functional rudimentary horn were existing. While cyclic pelvic pain along with a mass like lesion sometimes mistakenly can be considered to myoma and in addition to that should Rudimentary horn dyysmenorhea is visually miss-diagnosed, the occurrence of rupture in an emergency case is the likely conclusion. Atmaca et al. also reported on a [11] acute abdomen as seen in their case. Ultrasound revealed a 36x39-mm smooth contoured, homogenous solid mass in her right adnexal region. Minimal fluid was observed in the pouch of Douglas. In the laparotomy procedure, rudimentary horn and fallopian tube resection was done. The rudimentary horn must be excised since the intervention will prevent possible endometriosis development, torsion, distention, acute abdomen and possible infertility also are avoided although some complications might be encountered. Sometimes these patient can be diagnosed by chance. Their history included several surgical interventions and miss diagnosis with appendicitis and other entities. Ultrasonography, conventional and sonohysterosalpingography, magnetic resonance imaging and three-dimensional computed tomography, angiography are effective tools for diagnosing complex Müllerian anomalies [12]. MRI has been considered the best noninvasive means of diagnosing anomalies of the reproductive tract. However Mazouni et al. found that MRI the gold-standard non-invasive diagnostic test, short of laparoscopy [12] . Its imaging allow better visualization of the uterine fundal contour and configuration and excellent soft tissue resolution admited accurate diagnosis of all subtypes of unicornuate uteri. In our case T2-weighted magnetic resonance images of the pelvis at the level of the uterus showing two uterine cavities with no communication of the cavities inferiorly. There is a right unicornuate uterus (arrow) with a right cavitary noncommunicating rudimentary horn (two sided arrow). Endometrial thickening is identified in the right horn of the uterus (two sided arrow). İn front of the uterus bladder is seen (open arrow) (Figure1).

The resection of the rudimentary horn will be more complex, unless the rudimentary horn has not attached firmly to the unicornuate uterus. In our cases, sharp dissection and electrocautery of rudimentary horn that firmly attached the uterus are encountered respectively. Throughout the operation, it was first removed the right rudimentary horn with hemaotmetra, then it was sutured the myometrium in multiple layers and then viseral peritoneum. In the meantime, no hysteroscopy was performed in our patient. Consequently, we accomplished a laparoscopic removal of this uterine anomaly without any complication in the postoperative period. After three months' control in MRI (Figure 2) patient anatomical image was completely normal. Young female patients having dysmenorrhea with adenexal mass evolved into rudimentary horn with functional endometrium should be subjected to differential diagnosis. Management of this complex congenital anomaly through the surgical removal of the anomaly is still support of management due to treatment of dysmenorrhea in order to contain functional endometrium, to prevent endometriosis and pregnancy complications. Though most cases of dysmenorrhea are the primary type and can be managed by symptomatic treatment. But routine ultrasonography should be done to rule out any organic pathology. Therefore, careful examination and imaging must be performed to avoid possible causes. These cases should be managed by expert surgeons since wrong operations could be led into bigger problems in the post period of the operation.

Acknowledgement
None

Declaration of Interest
None

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