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Original Research Paper

Gynaecology

A CASE OF FUNCTIONING NON COMMUNICATING RUDIMENTARY HORN WITH UNICORNUATE UTERUS: AN UNUSUAL CAUSE OF SECONDARY DYSMENORRHEA IN A PERIMENOPAUSAL WOMAN.

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ABSTRACT Mullerian duct malformations represent a miscellaneous group of congenital anomalies that result from the arrested development, abnormal formation or incomplete fusion of the paramesonephric ducts. Congenital Mullerian abnormalities usually present at adolescent age with menstrual irregularity, dysmenorrhea, infertility and recurrent pregnancy loss. The Unicornuate uterus is a rare uterine malformation which usually features a rudimentary accessory horn with or without functioning endometrium. We are presenting a case of perimenopausal multiparous patient who had history of secondary dysmenorrhea diagnosed with endometrioma on imaging studies, but on laparotomy incidentally found to have Unicornuate uterus with functioning noncommunicating rudimentary horn leading to hematometra and hematosalpinx. The objective of this clinical case report is to highlight this rare Mullerian malformation as a differential diagnosis of secondary dysmenorrhea, chronic pelvic pain or adnexal mass in perimenopausal age group females.

KEYWORDS : Unicornuate uterus, Rudimentary horn, Dysmenorrhea, Hematosalpinx.

INTRODUCTION:

The incidence of Mullerian duct anomalies range between 0.4 to 10%, [1] in which Unicornuate uterus prevalence fluctuates between 2.4 to 13.7% of all uterine malformations.[2] Although its true incidence is unknown. In more than 75% of the cases of Unicornuate uterus, we appreciate a contralateral rudimentary horn, and most of these horns bear functioning endometrium which is noncommunicating with the other horn. The prevalence of Unicornuate uterus with rudimentary horn (UUWRH) in fertile women is approximately 1 in 100,000.[2] A non-communicating rudimentary horn usually has inactive endometrium but if it contains active endometrium can be associated with complications from menarche to menopause such as endometriosis, primary infertility, hematometra, anomalies of the urinary system and obstetrical problems such as malpresentation, habitual abortion, ectopic pregnancies and premature birth.[3] Generally, it is considered to become the adolescent girl's pathology but here we presented 43-year-old woman with this abnormality who had previous three vaginal deliveries presented with severe secondary dysmenorrhea.

Case History:

43 years P3L3 patient presented to OPD with complaint of vague pain in lower abdomen since past two and half years which was aggravated during menses. Since menarche, she had regular menstrual period with average flow with progressively increasing dysmenorrhea. There was no other menstrual complaint or dyspareunia or bladder bowel symptoms. Her symptoms did not respond completely to analgesics and continuous progesterone for three months. She was known case of Hypertension and hypothyroidism controlled on medication. Her general, systemic and per abdomen examination was normal. On pelvic examination, a cystic tender mass of size 4x5cm was felt in right fornix. Ultrasonography was suggestive of right tubovarian complex mass, possibly endometrioma measuring 3.7x2.9cm size. CECT Pelvis was suggestive of right adnexal complex cystic lesion 5x4.7x4.5cm, right ovary not seen separately. Another heterogenous lesion 2.7x2.6x2cm seen adjacent to the lesion. Left adnexa unremarkable. Bilateral kidneys were normal. Her CA-125 was 74U/ml. Her pap smear was negative for intraepithelial lesion or malignancy. In view of prior diagnosis of endometrioma with severe pelvic pain in perimenopausal case with failed medical management, surgery was planned. On laparotomy, 4x3.5x2cm rudimentary horn was attached to the right side of uterus. The right fallopian tube arose from rudimentary horn, it was dilated, tortuous, measuring 14cm in total length and diameter varying from 1.5cm to 4cms (Figure 1). The left round ligament arose from uterus while right from the rudimentary horn. Cut section of rudimentary horn showed small cavity with endometrial lining, the rudimentary horn and right fallopian tube were filled with dark color blood (Figure 2). Bilateral ovaries were normal and no endometriotic deposits were seen in pelvis. Her postoperative period was uneventful. Histopathology of specimen confirmed the diagnosis of rudimentary horn with functioning endometrium.

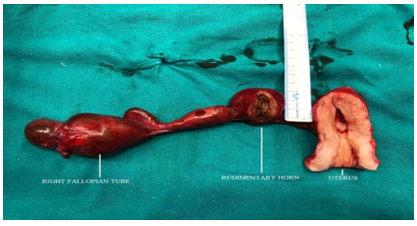

Figure 1: Intraoperative picture showing uterus, rudimentary horn and right dilated and tortuous fallopian tube and Bilateral ovaries.

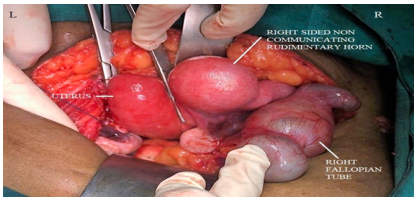

Figure 2: Cut section of specimen showing rudimentary horn with attached elongated tortuous fallopian tube with blood filled lumen.

DISCUSSION:

Mullerian duct malformations delineate a miscellaneous group of congenital anomalies that result from arrested development, abnormal formation, or incomplete fusion of paramesonephric ducts.[1] Unicornuate uterus with non-communicating rudimentary horn (UUWRH) is rare form of Mullerian abnormalities which is also an important cause of dysmenorrhea. Diagnosis of asymptomatic UUWRH is a challenge due to low prevalence, lack of awareness and suspicion among clinicians and radiologists. Often the patient's medical history is completely normal and symptoms depend on the presence of an obstructive anomaly. It causes cyclic pain due to intracavitary retention of menstrual effluent and retrograde menstrual flow. If there was no relationship between rudimentary horn and main functional endometrial cavity, hematometra and maybe hematosalpinx would be observed.[4] Other mechanism of dysmenorrhea in Unicornuate uterus with rudimentary horn is the endometriosis. These patients usually attend to gynecology or emergency departments with severe pain or pelvic mass. Majority of the women with dysmenorrhea are not evaluated in detail during routine examination.[5]

Transvaginal ultrasonography is good for diagnosis but pelvic magnetic resonances may be related with infertility, recurrent pregnancy loss, preterm delivery and ectopic pregnancy. This anomaly is associated with ipsilateral renal agenesis (67%) or ipsilateral pelvic kidney.[6] Cases of non-communicated and cavitated Unicornuate uterus also present a differential diagnosis with didelphys uterus with right cervico-vaginal agenesis and ipsilateral renal agenesis or segmentary atresia in Müllerian malformations.[6] Atresia of lower half of one of the hemi uteri, resulting in a non-communicating cavitated uterine horn with hematometra; in this type of malformation, both kidneys are normal, as in our patient. But in this case, the insertion of the uterosacral ligaments provides the key because if the structure is a uterus didelphys, each uterosacral ligament would be inserted into each uterus, whereas if the structure is Unicornuate, both uterosacral ligaments would be inserted into the principal horn.

In a case reported by Puja Jain et al, a perimenopausal age patient 40-year P3L3 presented with progressive pain in left lower abdomen, diagnosed on MRI as complex/neoplastic ovarian mass on laparotomy it was found to be rudimentary horn with functioning endometrium.[3]

So, our aim of reporting such case is to increase awareness towards these malformations which leads to early diagnosis and management and prevent catastrophic complications in later life.

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