Adenomyosis in a Noncommunicating Rudimentary Horn of Unicornuate Uterus - A Diagnostic Dilemma

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Abstract:
Although asymptomatic cases naturally occur, congenital mullerian abnormalities may present with infertility, menstrual irregularity and recurrent pregnancy loss. Women with normal reproductive outcomes and recurrent pregnancy loss exhibit approximately 2-4% and 5-10% incidence of various congenital mullerian abnormalities. Following septate uterus, bicornuate uterus and arcuate uterus, unicornutat uterus is the fourth most frequent congenital uterine abnormality seen in a combined population of infertile and fertile women. Unicornuate uterus with a non-communicating functional rudimentary horn is a type of mullerian anomaly and is a rare cause of dysmenorrhea. Though endometriosis has been reported, adenomyosis in rudimentary horn is extremely rare. Early diagnosis and treatment with excision of non-communicating horn with salpingectomy is required to prevent obstetric complications.

Key Words: Mullerian anomaly, unicornuate uterus, rudimentary horn, dysmenorrhea.

INTRODUCTION
Unicornuate uterus with a rudimentary horn is a rare type of mullerian duct malformation and results from the defective fusion of the malformed duct with the contra-lateral duct. A fibrous or fibro-muscular band usually connects the horns of the ducts but in 80 - 90% of cases there is no communication. The rudimentary horn may consist of a functional endometrial cavity or it may be a small solid lump of uterine muscle with no functional endometrium. This uterine anomaly covers a wide range of anatomical variability and is divided into four subgroups according to the American Fertility Society classification of Mullerian anomalies: (Ia) rudimentary horn with cavity communicating to unicornuate uterus, (IIb) with cavity non-communicating, (Ic) with no cavity and (Id) with no horn (American Fertility Society, 1988). Type Ib is the most common and clinically significant type. It is also susceptible to many gynaecological and obstetric complications that may be avoided by the removal of the rudimentary horn and its tube (Heimonen, 1997). Nevertheless, this subgroup may encounter fine anatomical variations, particularly in the attachment of the rudimentary horn that may influence surgical treatment.

CASE:
42 year old p212 patient presented with history of congestive dysmenorrhea since 2years. She had regular menstrual cycles with no menstrual irregularities, no dyspareunia or bowel and bladder disturbances. She had received oral antispasmodic medication in the past but no improvement. No history of pelvic inflammatory disorder. She had borne two children vaginally. Both pregnancies were uneventful and underwent postpartum sterilisation. Gynecological examination showed normal size uterus, with b/l fornices free and non tender. Ultrasonography (USG) of the showed anteverted uterus with altered echotexture with small cystic spaces right ovary normal, left adnexal cyst with internal echoes measuring 3.5 x 3.9 cms, s/o hemorrhagic /endometriotic cyst ,there was no abnormality in renal tract. Hysterectomy was planned and on opening abdomen perioperative findings were bicornuate uterus with left tube hydrosalpinx , normal left ovary but the round ligament on left side was obscured probably due to stretch and normal right tube and ovary.

MANAGEMENT:
Total abdominal hysterectomy with bilateral salpingoophorectomy was done. Grossly, specimen consisted of bicornuate uterus with leftsided hydrosalpinx and left ovary. Uterus and cervix measured 9x4x3cms ,another uterus like mass(7horn) measured 6x3x2.5cm attached adnexa with tube measuring 4cms ovary 3x1.4cms The sectioned surface of the uterus show one side of endomyometrium is thickened with trabeculations.Endometrial cavity measures 4cms,
myometrial thickness 3cms. There was no communication between rudimentary horn or endometrial cavity or endocervical canal. Cervix appeared unremarkable done. Grossly, specimen consisted of bicornuate uterus with leftsided hydrosalpinx and left ovary. Uterus and cervix measured 9x4x3cms another uterus like mass(?horn) measured 6x3x2.5cm.attached adnexa with tube measuring 4cms ovary 3x1.4cms.

On histopathological examination, the sections from the uterus showed endometrium in proliferative phase with adenomyosis. Microscopic examination of non communicating rudimentary horn showed proliferative endometrium with adenomyosis. Section from cystically dilated left fallopian tube showed features of hematosalpinx. The cervix showed features of papillary cervicitis.

DISCUSSION:
A unicornuate uterus may lead to various gynecological or obstetric complications and diagnoses are often difficult and delayed till reproductive period or to pregnancy [1]. According to Buttram & Gibbons unicornuate uterus has seven subtypes based on their anatomy [2]. In the present case according to the above classification falls under ‘b’ variant of class II. This constitutes about 7.7 to 42.9% of cases of unicorinate uterus with rudimentary horn [4]. Evaluation of renal tract is necessary owing to its frequent association with this type [1]. The case in discussion presented with only dysmenorrhea. In one study the incidence of endometriosis was as high as 20% [5]. About 50% patients presenting with endometriosis with unicorinate uterus and a cavitary rudimentary horn have communication between the endometrial cavity in the rudimentary horn and tubal lumen [2]. Ultrasonography can pick up anomalies of uterus. MRI & CT scan are other tools [6]. Uterine malformations are associated with late miscarriages and preterm deliveries and high incidence of early pregnancy losses also [3]. Pregnancy in a noncommunicating cavitary rudimentary horn is a rare and life threatening condition and can only be possible by transperitoneal migration of sperms. Our patient was fortunate enough that she did not have any pregnancy related complications. In unicorinate uterus with a rudimentary horn excision of the rudimentary horn is the usual treatment [4]. In the present case as there was hydrosalpinx and patient was 42yrs old we performed abdominal hysterectomy with removal of tubes, ovaries and the adenomyotic rudimentary horn which gave an earlier clinical impression of a solid ovarian lesion. This may also be misdiagnosed as sub-mucous fibroid [1]. Agarwal et al. reported a 18-year-old woman who developed endometriosis and had severe pelvic pain. Generally it is considered to become the adolescent girl’s pathology but here we presented a 42-year-old woman with this abnormality with two vaginal deliveries. Borah et al. presented again an adolescent girl with primary dysmenorrhea with similar Mullerian abnormality [8]. In the literature the laporoscopic excision of the rudimentary horn is offered for possible severe complications [9,10]. Though endometriosis has been reported, adenomyosis in rudimentary horn is extremely rare[7]. Our patient is free from any problem one month following operation. Every effort should be made to diagnose this condition before pregnancy occurs owing to its fatal complications. The rudimentary horn should be excised to prevent obstetric complications

REFERENCES:


