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Laparoscopic excision of non-communicating accessory uterine horn in an adolescent girl with unicornuate uterus

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Abstract
Dysmenorrhea in is seen in 41-91.5% adolescent girls and is mostly due to primary dysmenorrhea. Endometriosis is the most common cause of secondary dysmenorrhea and very rarely is due to obstructive mullerian anomalies.
An adolescent girl with unrelenting dysmenorrhea with frequent hospital visits, had an obstructed accessory uterine horn on ultrasound and was confirmed on MRI scans. She underwent successful laparoscopic excision and was relieved of her dysmenorrhea.
These lesions are rare but can cause severe dysmenorrhea not responding to medical management and unrecognized lesions can lead to infertility, ectopic pregnancy in accessory horn with catastrophic complications and infertility. They are the most commonly detected during 3rd decade of life, with very few cases being diagnosed and successively treated in adolescent girls. Early recognition and laparoscopic excision can relieve the dysmenorrhea and preserve fertility in these patients.

Keywords: Secondary dysmenorrhea, Accessory communicating uterine malformation (ACUM), Obstructive Mullerian anomalies, Unicornuate uterus, rudimentary uterine horn

Introduction
Mullerian anomalies affect 5.5-7% of women and are increasingly seen in women with infertility and miscarriages. Obstructive lesions can be symptomatic especially during adolescence. Completely obstructive lesions can present with amenorrhea and abdominal pain, while those with partial obstruction, present with cyclical pain and the diagnosis can get delayed. In a large proportion of patients with unicornuate uterus, the accessory horn may be functional and are non-communicating. These patients can present with cyclical pain and are usually diagnosed during 3rd decade of life. Evaluation and management of an adolescent girl with accessory blind ending uterine horn who presented with severe dysmenorrhea is presented here.

Case report
A 13 year old girl, apparently well and healthy presented with worsening dysmenorrhea with each menstrual cycle. She had attained menarche 1 year ago and the cycles were regular but always had some dysmenorrhea. In the current cycle she presented to emergency with severe abdominal pain and vomiting for 4 days. On admission she was mildly dehydrated, in severe pain and had a diffusely tender abdomen. After adequate resuscitation and analgesia, she underwent further evaluation. Her blood investigations were essentially normal. Ultrasound was suspicious of non-communicating left rudimentary horn filled with blood, which was then confirmed on MRI Scan. (Figure 1). She was electively planned for laparoscopy with a plan to resect the accessory horn. After bladder catheterization a 3-port laparoscopy was performed using 10mm Umbilical camera port and 2 infraumbilical 5 mm lateral ports. On laparoscopy, there was no blood in the peritoneal cavity and the uterus appeared bulky with deviation to right. A large accessory horn was evident on the left side with a deep notch on the superior aspect of uterus (Figure 2a). Bilateral tubes were present with normal looking ovaries and fimbria. Marking with hook diathermy delineated the accessory horn with care being taken to avoid entry into the main uterine cavity (Figure 2 b).

Using ligature and hook diathermy the accessory horn was separated from the uterine wall and broad ligament and completely resected laparoscopically (Figure 2 C). The myometrium was repaired with interlocking 2-0 vicryl sutures. The specimen was delivered out through small suprapubic incision (Fig 3). She recovered uneventfully; urinary catheter was removed next day, and she was discharged on full orals after 2 days. The biopsy confirmed rudimentary uterine horn. In the last follow up one year from the procedure, her menstrual cycles are painless and regular.

Discussion

Uterine malformations are known to occur in 4% of infertile women and in 15% of those women, who have experienced recurrent miscarriage, can harbor Mullerian malformation. Obstructive Mullerian anomalies (Uterus didelphys with imperforate hemivagina, a noncommunicating cavitated uterine horn in unicornuate uterus; an accessory cavitated uterine mass (ACUM); a micro perforate transverse vaginal septum) can also present with severe dysmenorrhea [1]. The unicornuate uterus with rudimentary horn is one of the rarest of uterine anomalies affecting 1in 100,000 fertile female population [2]. Seventy five percent of unicornuate uterus cases present with a rudimentary horn, and in 80-90% of cases, there is no communication with the primary uterine cavity [3].

The basis of the defect lies in normal development of one Mullerian duct coupled with failure of the contralateral mullerian duct to elongate or to reach the urogenital sinus (which forms the lower third of the vagina) during the ninth week of gestation [4]. The most common presentation of these anomalies is severe dysmenorrhea dating back to menarche, but mean age at diagnosis is usually in 3rd decade of life. Our index patient presented with dysmenorrhea since menarche and had multiple outpatient consultations for the same, which was initially diagnosed as primary dysmenorrhea.

If dysmenorrhea is not responding to 3 cycles of medical therapy, detailed pelvic imaging and laparoscopy may be indicated to exclude mullerian anomalies, endometriosis, or other pathologies [1]. These rudimentary horns are predominantly right-sided (62%) and in our case it was found on the left side. The clinical presentation is based on the configuration of horn, whether blind or communicating and presence of endometrium. Unilateral renal agenesis are associated with mullerian anomalies in up to 30% of cases [4]. In our patient the imaging modalities showed presence of normal appearing kidneys bilaterally with no other renal tract anomalies.

Though the mean age of presentation is usually in 3rd decade of life when patients are symptomatic, detection before clinical symptoms have also been reported in 14 % of cases [5] and in our patient persistent dysmenorrhea and imaging led to diagnosis, early in her adolescence. The criteria used for the diagnosis are: Presence of an accessory intramyometrial cavitated mass, normal appearance of the uterus, fallopian tubes and ovaries, and the cavity must not communicate with the normal endometrial cavity. More than 90% of these rudimentary horns are non-communicating. There are other modalities (hysteroscopy) described to ensure blind nature of the rudimentary horn in parous women [1, 6]. In our index patient, on laparoscopy the horn was well above the confines of pelvic peritoneal reflection and with the findings on imaging, it was non-communicating variant.

The primary objective of surgery is to alleviate symptoms, prevent complications and preserve future pregnancy. Laparoscopic excision is an established technique in alleviating symptoms and also prevents future complications [7]. Ectopic pregnancy occurring in a communicating uterine horn is a possibility and can lead to serious complications if unrecognized. Successful pregnancy following excision of accessory uterine horns have been reported [8]. The aryl recognisition, especially in adolescent period as in our case, may prevent future complications such as endometriosis, infertility and ectopic pregnany in future. Our index patient is now 14 year old and needs close meticulous follow up by obstetricians during adult hood to go through safe pregnancy.

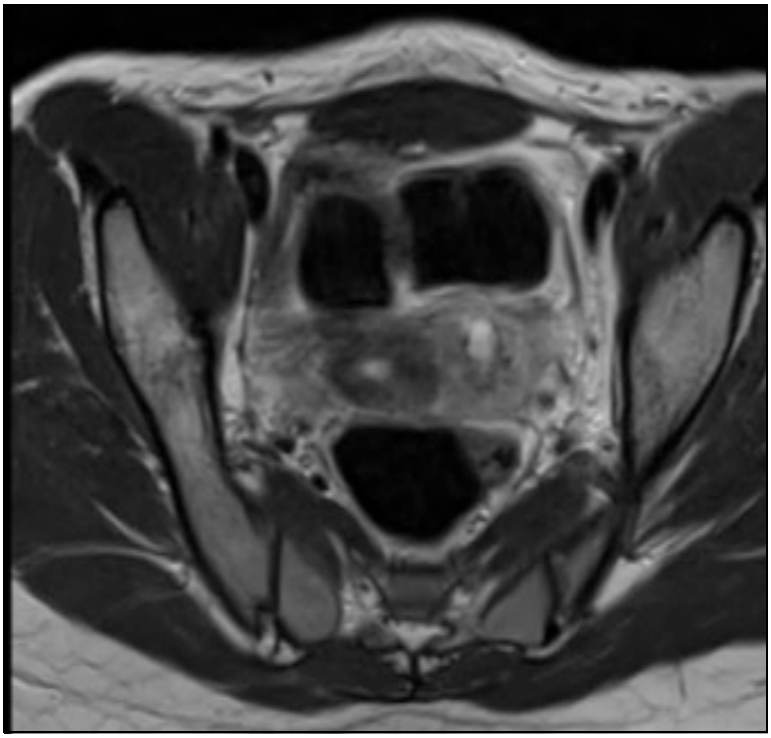

Fig 1: MRI Scan-large left accessory horn.

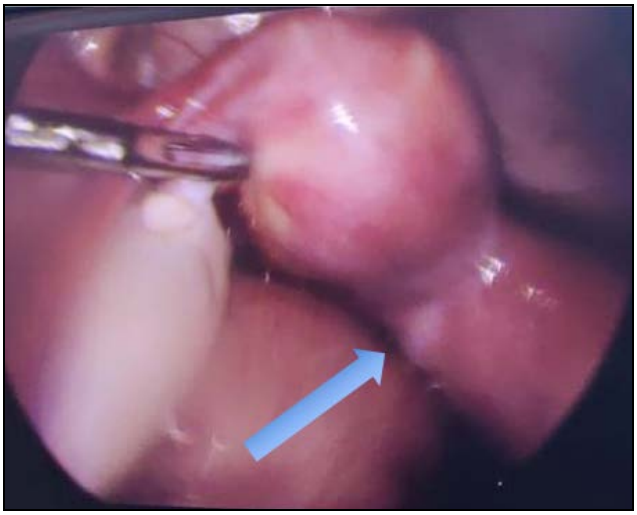

Fig 2 a: Laproscopic appearance of accessary uterine horn. A deep notch (Arrow) demarcates the main uterine body and the horm

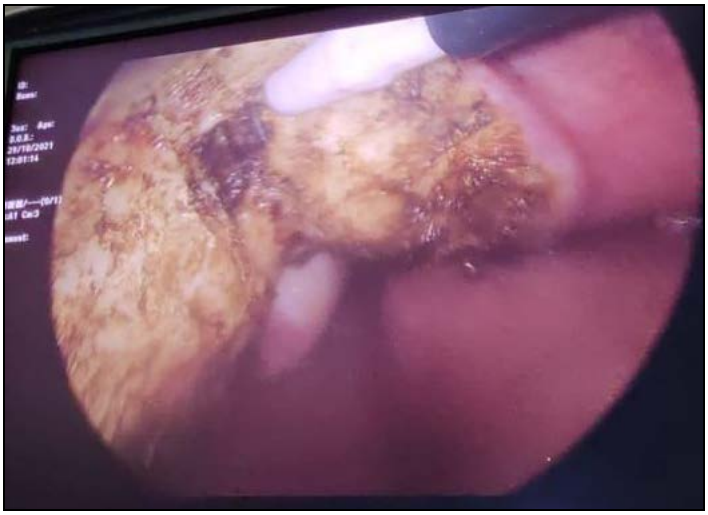

Fig 2 b: Laparoscopic excision using diathermy and ligasure, avoiding entry into main uterine cavity

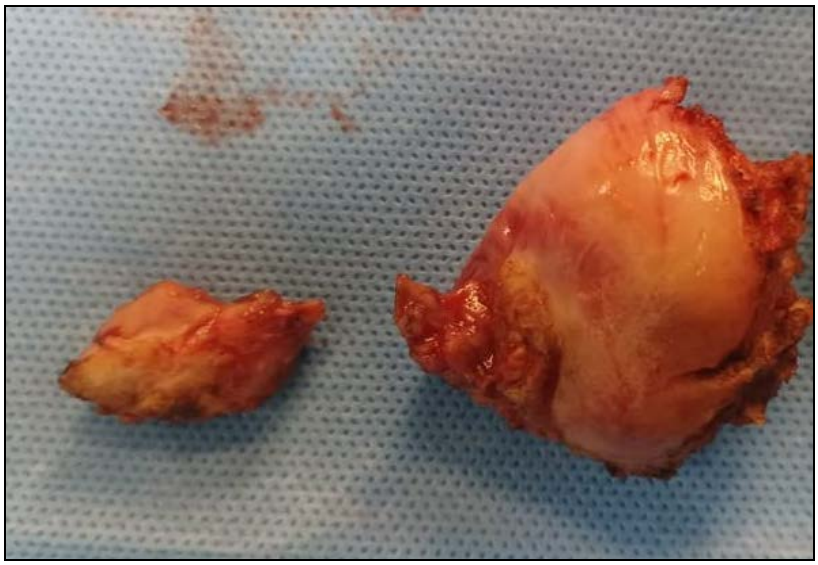

Fig 2c: Resected specimen

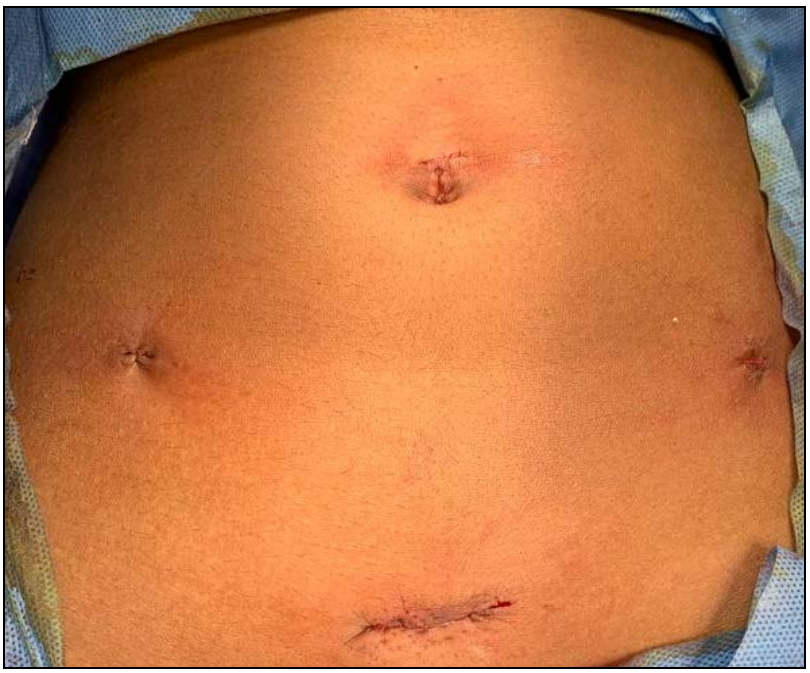

Fig 3: Port sites closed after completion. Suprapubic incision was used to extract the specimen

Conclusions

Significant dysmenorrhea in young adolescent girls can occasionally be due to obstructive mullerian anomalies. Severe dysmerrhoea not responding to repeat cycles of medical management, may need imaging to exclude obstructive uterine lesions. Lpaparoscopy is the most useful tool in evaluating pelvic pathology in unexplained dysmenorrhea. Laparoscopic resection of obstructed accessory uterine horns is feasible in young adults and this will prevent complications and can facilitate successful pregnancy in future.

Conflict of Interest

Not available

Financial Support

Not available

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