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Case Report

Accessory cavitated uterine malformation: a diagnostic challenge of an unclassified rare presentation

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ABSTRACT

Mullerian anomalies are rare anomalies of female genital tract with prevalence of around 5%. Accessory cavitated uterine malformation (ACUM) is a cavitated lesion surrounded by a rim of myometrium, located below the insertion of round ligament and fallopian tubes. A normal uterine cavity with visualisation of both ostia is must to differentiate with non-communicating uterine horn. ACUM presents as severe dysmenorrhoea in females below 30 years of age. It is not included in revised ASRM or ESHRE classification. Diagnostic modalities include 3D ultrasonography and MRI. Medical management including estrogen/progesterone oral contraceptives, nonsteroidal anti-inflammatory drugs (NSAIDs), the progesterone-only pill proves to be ineffective in most of the patients. Most articles mentioned laparoscopic excision of ACUM as the main management. Some papers also mentioned cornual excision or hysterectomy as the treatment modality. Sclerotherapy has also been tried as a conservative management. Fertility has also improved post-surgery of ACUM.

Keywords: Accessory cavitated uterine malformation, Mullerian anomaly, Juvenile cystic adenomyosis

INTRODUCTION

Mullerian anomalies are rare anomalies of the female genital tract. Their prevalence is about 5.5-6.7% in the general population and more in women with recurrent pregnancy losses and infertility.[1] The ASRM Mullerian anomalies classification 2021 is the latest one that classifies mullerian anomalies into nine categories. Other major classifications include European society of human reproduction (ESHRE) and embryology-European society for gynaecological endoscopy (ESGE) who developed a classification based primarily on uterine anatomy, with cervical and vaginal anomalies supplementary subclasses based on clinical significance and degree of deformity.[2,3]

In this case report, we present a rare mullerian anomaly called ACUM. Several authors reported them as juvenile cystic adenomyosis, isolated cystic adenomyoma, non-communicating accessory uterine cavities, adenomyotic cysts, myometrial cyst, and uterine-like mass.[4,5] Revised ASRM classification, ESHRE, ESGE (U6) has not included ACUM.[2,3]

ACUM is a non-communicating accessory cavity with hemorrhagic content lined with endometrial epithelium. Early onset of dysmenorrhoea generally below 20 years of age, close to the origin of round ligament and far from the endometrium with no communication with the cavity, the size ≥1 cm and associated urogenital tract defects.[6]

CASE REPORT

A 20-year-old girl unmarried girl presented to the outpatient department of obstetrics and gynaecology. She complained of pain lower abdomen, more towards the left side, continuous dull aching since 5 to 6 years which got relieved only on medications. It was unrelated to menstruation. However mild dysmenorrhoea was present

during her regular menstrual cycles. She was operated for left ureterocoele 8 months back. Abdomen was soft, non tender, no mass palpable. Per vaginal examination was not done in view of her virgin status. Per rectal examination showed a normal uterine contour with a left sided cystic structure of 2×2 cm. Ultrasonography showed a normal uterus 65×47×26 mm with endometrial thickness of 8 mm with normal adnexa. A well-defined structure of 27×23 mm with central anechoic area measuring 9 mm is seen along the left cornu. Single vagina and cervix present. Diagnosis was possibility of Mullerian anomaly likely rudimentary horn with non-communicating endometrial cavity with hematometra.

MRI pelvis showed a normal uterus with central endometrium. A well-defined T2W hyperintense lesion (26×30×32 mm) surrounded by thick myometrial isointense lining seen in left anterolateral wall at cornu. Intralesional central T1/T2 hyperintensity suggests hemorrhage. No evidence of communication with uterine endometrial lining is seen-likely ACUM and less likely obstructed rudimentary horn. Hysteroscopy showed normal uterine cavity and bilateral ostia. Laparoscopy showed a mass of 3×3.5 cm arising below the left round ligament extending till the left cornu. Left fallopian tube was stretched over it and few adhesions were present. Right tube was normal. 20 IU of vasopressin diluted in 200 ml NS was injected at the base. A transverse incision was given over the most prominent part with cold scissors. ACUM was enucleated and excised. Myometrial space closed with vicryl 1-0. Histopathology showed cavity lined with functional endometrium and surrounded by smooth muscle fibres. Few foci of endometrial gland and stroma interspersed between myometrial tissue (adenomyosis identified). Diagnosis of ACUM was confirmed.

Figure 1 shows MRI axial section of ACUM and Figure 2 shows ACUM on left side.

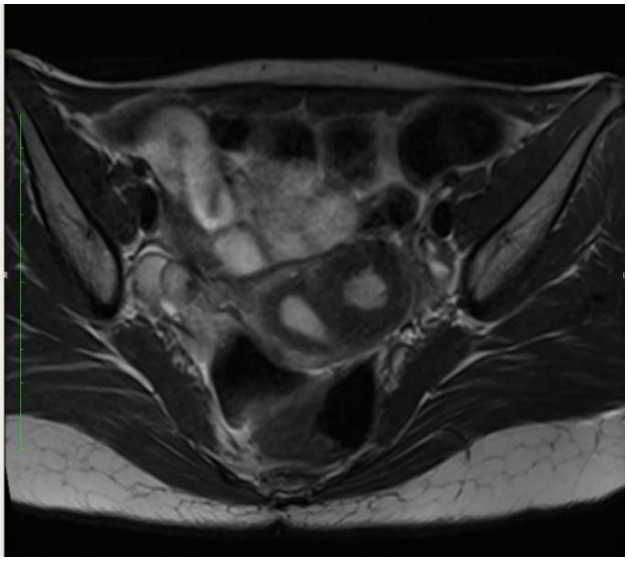

Figure 1: Contrast MRI axial section showing accessory non communicating cavity.

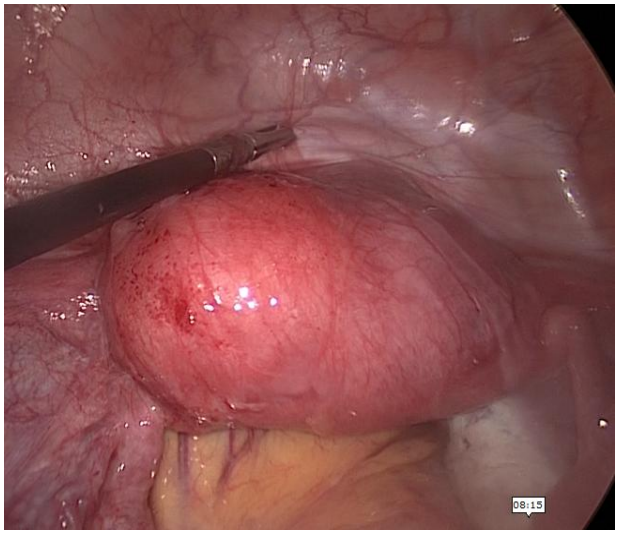

Figure 2: ACUM on left side.

DISCUSSION

According to Timmerman et al ACUM is a cavity surrounded by a myometrial rim continuous with the anterolateral uterine wall, located below the insertion of the round ligament and the interstitial portion of the fallopian tubes.[4] Normal uterine cavity differentiates it from obstructive lesions. Naftalin et al found isolated cavitated lesions in the anterolateral region of the myometrium, below the insertion of the round ligament. They defined it as a fluid-filled cavity surrounded by myometrial mantle. Fluid within the cavity echogenic with a ground-glass appearance signifying altered blood or hyperechoic with avascularity on doppler. Mean size of ACUMs was 22.8 mm. Identification of interstitial portions of both Fallopian tubes to exclude obstructive uterine anomaly was required.[7] There are various descriptions of ACUM by Acien et al, Takeuchi et al and Naftalin et al.[7-9]

It is a benign lesion presents with severe dysmenorrhea since menarche or late onset dysmenorrhea or noncyclic pelvic pain probably due to accumulation of blood in the cavity. Mainly seen at median age of 21.5 years, but may be seen at later.[4] Hu et al described 3 cases of ACUM initially misdiagnosed as fibroid or adenomyoma. Their patients were less than 30 years and nulliparous. Primary symptoms included severe dysmenorrhoea nonresponsive to medications along with pelvic pain and dyspareunia, although it could be asymptomatic in some patients.[10]

Ultrasound shows well-defined cavitated lesions with ground-glass echogenicity and smooth inner lining signifies functional surrounding endometrium. ACUM has a normal uterine contour and interstitial portions of both fallopian tubes. 3D ultrasound shows the coronal display section of the uterus that aids in diagnosis.[4] Shah et al diagnosed ACUM on 3D coronal ultrasound which shows a well-defined, ovoid, noncommunicating cavitated mass along the right lateral uterine wall below right cornu with

normal uterine cavity. They found 3 D to be cost effective and non-invasive in ruling out a common differential diagnosis of obstructed cavitated rudimentary horn with unicornuate uterus where it shows an isolated cavitated mass with absent ipsilateral uterine cornu and a contralateral banana shaped uterine cavity.[11] Non contrast MRI not only provides a precise localization and morphology but characterizes lesion as well. T1 and T2 weighted image shows hypersignal reflecting hemorrhagic content and surrounding tissue on T2-weighted is hypointense reflecting fibrous tissue. MRI differentiates ACUM with a non-communicating uterine horn with hematometra which has an oval or tubular outline without any fibrous surroundings. Though T2 weighted image displays hypersignal for necrosis in uterine myoma also being T2-weighted hypersignal but it is surrounded by a thinner hypointense wall-weighted sequences without enhancement, at the difference of ACUM if an injection is performed.[6]

Mullerian ducts are invagination of the dorsal coelomic epithelium at 6 weeks gestational age.[12] These two ducts fuse at 10 to 12 weeks ducts in craniocaudal direction and then join the urogenital sinus.[13] Deng et al in their article mentioned that there is normal fusion of bilateral mullerian ducts but endometrium does not get absorbed, resulting in formation of abnormal chambers and forming a new Mullerian duct malformation, ACUM. Although the mean lesion size is around 3.35 cm but Deng et al reported a giant ACUM of size 9 cm.[14]

Table 1: Differential diagnosis of ACUM.

Variables	Accessory cavitated uterine mass	Non-communicating rudimentary uterine horn	True cavitated adenomyoma	Degenerating fibroid	Robert's uterus
Pathophysiology	Duplication and persistence of the ductal Mullerian tissue at the insertion of the round ligament.[14]	Failure of one Mullerian duct to elongate towards the urogenital sinus while the contralateral Mullerian duct develops normally.[18]	Invagination of the endometrial layer into myometrium.[19]	Benign monoclonal tumor of smooth muscle cells and fibrous connective tissue.	Resorption defect of Mullerian duct.[1]
Definition	Isolated cavitated mass consistent with normal shape and functional uterus.	An isolated cavity with a horn and fallopian tube attaching on it without communicating with the true cavity.	A focal adenomyosis not in direct continuation with junctional zone with central degeneration and fluid accumulation	Benign tumor of myometrium. May undergo degeneration like hyaline, cystic, myxoid and red.	Uterine cavity is asymmetrically separated by an oblique septum with ipsilateral cervical aplasia.
Histopathological findings	Functional endometrium surrounded by smooth muscle cells. Myometrium adjacent to ACUM may have small foci of adenomyosis.	Thick myometrial wall with cavity lined by endometrial epithelium. No evidence of adenomyosis.	Lack of uterus like smooth muscle organization with absence of epithelial lining of the cyst. Diffusely spread adenomyotic foci in the uterus corpus.	It is well circumscribed with solid rubbery firm texture.[13] Composed of elongated smooth muscle cells with eosinophilic cytoplasm and centrally located cigar-shaped nucleus.	Contralateral blind hemicavity with or without hematometra. No specific histopathologic features.
Fallopian tubes	Normal ostia. Normal fallopian tubes attachment to uterus.	The fallopian tube arises from the accessory uterine horn. One normal fallopian tube arising from true uterine cavity seen through hysteroscope as single tubal ostium.	Fallopian tubes are normal.	Fallopian tubes are normal.	Only one uterine horn and ipsilateral fallopian tube will be seen on hysteroscopy.

Uterine-like mass (ULM) is classified as a cavitated mass lined with functional endometrium made of hormone-sensitive smooth muscle cells arranged irregularly at the periphery. ACUM is a subtype of non-communicating ULM, associated with normal uterus.[15]

Medical management of ACUM includes estrogen/progesterone oral contraceptives, NSAIDs, the progesterone-only pill, gonadotrophin-releasing hormone agonists, the levonorgestrel-releasing intrauterine device (LNG-IUD). 85.7% of the cases treated with medical

treatment, had persistent symptoms and had to undergo surgical intervention.[4] Timmerman et al performed and many authors mentioned laparoscopic excision of ACUM in most cases and cornual excision or a total hysterectomy in few others.[4-6,14] Merviel described first case of ethanol sclerotherapy in ACUM with successful outcome. In their case, the patient conceived after 2 months of sclerotherapy without risk of rupture as opposed to adenomyomectomy which has 1% risk of uterine rupture if patient conceives within 6 to 12 months of surgery.[16] Strug et al concluded that successful excision not only resolve symptoms but also improves fertility.[17]

CONCLUSION

Though ACUM is a rare mullerian anomaly but it should be kept in mind when dealing with young patients with severe dysmenorrhoea or chronic pelvic pain. 3 D ultrasound or MRI aids in diagnosis. Laparoscopic excision is the modality of choice.

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