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Case Report

Case report on accessary and cavitated uterine mass-a rare form of mullerian anomaly

Chaitali Kalokhe[1]*, Abhijeet Bhosale[2]

[1]Department of Obstetrics and Gynecology, King Edward Memorial Hospital, Parel, Mumbai, Maharashtra, India
[2]Department of Radiology, Dr. Ulhas Patil Medical College and Hospital, Jalgaon kh Maharashtra, India

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***Correspondence:**
Dr. Chaitali Kalokhe,
E-mail: drchaitalikalokhe@gmail.com

ABSTRACT

Accessory and cavitated uterine mass (ACUM) are a rare form of developmental Mullerian anomaly seen in young females with chief complaints of chronic pelvic pain and dysmenorrhea. Imaging modalities, including hysterosalpingography (HSG), ultrasonography (USG) and magnetic resonance imaging (MRI) forms the mainstay for diagnosis of this condition. It is an accessory cavity in otherwise normal uterus. It is lined by normal functional endometrium and surrounded by myometrium- like smooth muscle cells hence, it has macroscopic and microscopic resemblance to the uterus. In most of the cases, surgical treatment is recommended due to relieve the severe dysmenorrhea.

Keywords: Cavitated uterine mass, Dysmenorrhea, Mullerian anomaly

INTRODUCTION

Accessory and cavitated uterine mass (ACUM) are a rare newly described Mullerian anomaly. Patients with this condition is generally presents with chronic recurrent pelvic pain and severe dysmenorrhea in younger than 30 years of age.[1] It is an accessory cavity lined by functional endometrium within an otherwise normal uterine cavity, in contrast to other mullerian anomalies in which uterus is malformed. It is an accessory mass at the insertion of the round ligament and it has dark brown coloured fluid content within it.[2,3]

Differential diagnosis includes unicornuate uterus with non-communicating rudimentary horn, adenomyosis with cystic or degenerated areas, degenerated leiomyomas. Essential and primary dysmenorrhea makes the diagnosis of ACUM more difficult. This entity can be easily picked up in routine pelvic ultrasonography (USG). Magnetic resonance imaging (MRI) is highly accurate in the diagnosis and is confirmatory.

CASE REPORT

An 18-year-old girl presented with complaint of severe dysmenorrhea and chronic pelvic pain since menarche which was aggravated since past 2 years. Her menstrual cycles were regular with normal flow. Her past medical and surgical history was not significant, and she had no previous sexual history. She had history of temporarily pain relief on taking anti-inflammatory medications. Her vitals were normal. She showed normal pubertal development with height of 146 cm and weight of 46 kg and normal thelarche and adrenarche.

External genitalia were normal. There was no definitive palpable mass on per abdominal examination. Ultrasound was done which was showing completely septate uterus with 2 separate endometria seen. Left sided endometrium measures 3 mm. There is 2.8×1.7×2.5 cm (6.4 cc) heterogeneous hypoechoic collection with air fluid level seen in the right endometrial cavity. Two separate cervices not seen. USG findings were suggestive of differential

diagnosis of 1) complete septate uterus with possibility of transverse vaginal septum causing hematometra on right side, 2) non communicating rudimentary horn with unicornuate uterus, 3) Accessory and 4) cavitated uterine mass. Magnetic resonance imaging was advised for confirmation of ultrasound findings. Pelvic MRI was performed, which revealed that there is well defined cavitating structure with blood level seen along the right lateral wall of the uterus which shows endometrial lining with junctional zone. This lesion is not communicating with the uterine cavity.

Differential diagnosis includes ACUM and unicornuate uterus with non-communicating rudimentary horn. All routine investigations were sent and traced-within normal limits. Subsequently patient went laparotomy, a well-defined fleshy mass was seen of approximately 3×3 cm size on right side of the pelvic cavity. The mass was located just below the right round ligament away from right ovary, not adherent to surrounding structures. Both the ovaries and uterus were normal. Uterus has both the fallopian tubes normal.

Diagnostic hysteroscopy done to rule out other mullerian anomalies followed by excision of ACUM and reconstruction of myometrium done. While performing complete excision of mass, a chocolate-coloured fluid flowed out from the mass. Patient had an uneventful postoperative recovery and was discharged on post operative day 5. Gross evaluation of mass revealed a cavitated mass with thick muscular wall and pathological examination showed findings consistent with ACUM.

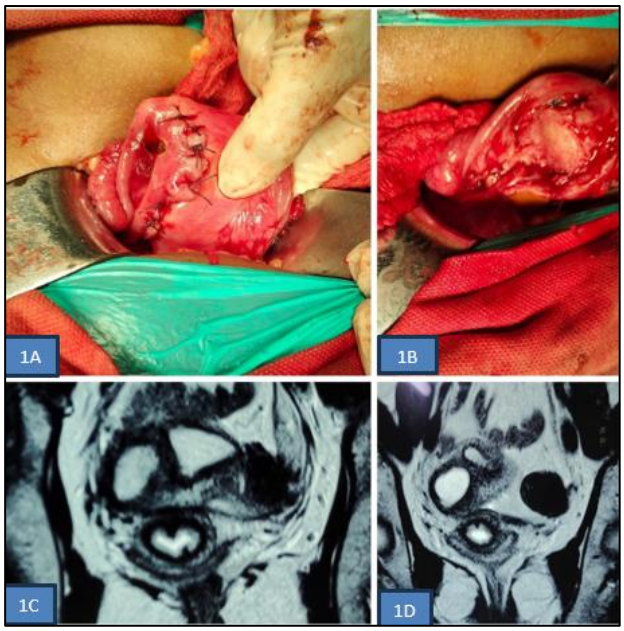

Figure 1: (A) reconstruction of the uterus after excision of ACUM. (B) right lateral wall of the uterus after excision of ACUM. (C and D) MRI findings suggestive of normal uterus on left side with Cavitated mass with hypointense content filled within it on the right side.

DISCUSSION

Uterus develops from embryonic fusion of two mullerian ducts. At around 10 to 12 weeks of gestation, the two mullerian ducts fuses with each other in a craniocaudal direction and then finally fuses with the urogenital sinus, forming the uterus, bilateral fallopian tubes and upper one third of the vagina.[4,5] Different classes of mullerian anomalies have been described.[6] Septate uterus is the most common mullerian anomaly, which is followed by unicornuate uterus. Mullerian anomalies are deviations from normal anatomy resulting from distortion of the complex embryogenesis. Some of these anomalies may lead to significant reproductive problems based on the degree and the type of the development.[5] So, an effective and reliable classification of mullerian anomalies can contribute to the proper diagnosis and the management of affected individuals.[7]

Uterine-like mass (ULM) is a distinct clinical entity defined as a cavitated mass lined with functional endometrium that is composed of sex hormone sensitive smooth muscle cells, which are arranged irregularly at the periphery of the mass. These masses can be located anywhere within the uterus or outside the uterus. ACUM is a non-communicating ULM arising in the uterus itself. This entity needs to be classified separately as the uterine cavity is normal unlike other mullerian anomalies.[8]

There are three theories of development of ACUM, 1) congenital anomaly theory, 2) heterotopias theory, 3) metaplasia theory.[9] Most of the authors accept ACUM as a congenial anomaly.[8] According to this theory, accessory mass could be caused by duplication of ductal Mullerian tissue in the critical area at the level of attachment of the round ligament, possibly due to the gubernaculum dysfunction.[8] It's association with gastrointestinal and genitourinary anomalies have also been explained in some cases.

The same appearing different masses have also been explained in the literature by the different names, including cavitated adenomyoma, accessory cavitated mass, and juvenile cystic adenoma. However, ACUM is different from the cystic adenomyosis, which usually occurs in the middle-aged women, and it constitutes diffusely distributed adenomyotic foci within the uterus. Also, these cystic adenomyosis lacks the normal endometrium and myometrial organization.[10]

The current criteria used for the diagnosis of ACUM includes 1) an isolated accessory Cavitated mass 2) normal uterus (endometrial cavity), tubes ovaries 3) surgical case with excised mass and with pathological examination 4) accessory cavity lined by endometrial epithelium with glands and stroma 5) chocolate- brown-coloured fluid content 6) no adenomyosis (if uterus removed) but there could be tiny foci of adenomyosis in the myometrium adjacent to the accessory cavity.[8] The present case has fulfilled all of the diagnostic criteria of ACUM. Since

Uterus, fallopian tubes and ovaries are normal in this patient, other mullerian anomalies were ruled out. Although most of the reported cases of ACUM are located on the right lateral wall of the uterus near the round ligament, about one- third of cases are adjacent to the left round ligament. USG is the initial imaging modality in the diagnosis. On hysterosalpingography (HSG), mass may not be visualized at all. But the most important role of HSG is to rule out the other Mullerian anomalies. However, MRI is the imaging modality of choice as it is non-invasive and preferred over the HSG in young females. Management of the entity is surgical excision. laparoscopic excision is preferred over the laparotomy, as most of the patients are females younger than 30 years old. But laparoscopic excision requires expertise.

CONCLUSION

In patients with chronic pelvic pain and dysmenorrhea, with otherwise normal uterus (no any mullerian anomaly), with the help of radiological investigations like USG and MRI this condition can be diagnosed easily. Histopathological confirmation also supports the final diagnosis of ACUM. Surgery forms the mainstay of treatment in all the patients with significant complaints. Although medical management can also be tried in some cases, but this condition is found to be resistant to medical treatment.

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