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CASE STUDY

ACUM: A RARITY

***,[2]Mahawar, P., [1]Manchanda, R., [2]Meena, J. and [2]Bhargava, N.**

[1]Manchanda Endoscopy Centre, PSRI Hospital, New Delhi
[2]Gynae Endoscopy at Manchanda Endoscopy Centre, PSRI Hospital, New Delhi

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ABSTRACT

Accessory and Cavitated Uterine Mass is a rare congenital anomaly seen in young females, present with chronic recurrent pelvic pain and progressive dysmenorrhea. It is an haemorrhagic fluid filled cavity lined by endometrium and myometrium, most commonly located below the round ligament and does not communicate with normal uterine cavity. The ultrasound and MRI are two important tool in diagnosing this rare disorder. A 21 year old nulliparous female presented to us in OPD with complains of severe dysmenorrhea and pain in left iliac fossa since 5 months. She was thoroughly investigated by surgeon and gynaecologist for pain abdomen and was prescribed analgesics but her pain was not relieved. During her investigations, CT scan revealed bilateral adnexal cystic lesions with bulky uterus and she was referred to us. After routine workup she was posted for diagnostic and operative laparoscopy which revealed a 4*3 cm mass in left broad ligament simulating broad ligament fibroid. While enucleating the same it showed a cavity draining chocolate color fluid from it. Her uterus, both ovaries and tubes were normal. In her follow up she was symptom free. ACUM, a rare Mullerian anomaly is a treatable cause of severe dysmenorrhea in young females. A high index of suspicion and MRI are helpful in providing the clue to diagnosis but laparoscopy is only confirmatory option for diagnosis as well as treatment of this rare disorder.

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INTRODUCTION

An ACUM (accessory and Cavitated Uterine Mass) is a rare Mullerian amomaly of young women who present with recurrent chronic pelvic pain and severe dysmenorrhea. It is characterized by a cavitated mass which is lined by functional Endometrium surrounded by smooth muscle. The main uterine cavity and myometrium are otherwise normal. The entity needs expertise to diagnose because of unawareness and wide range of differential diagnosis like noncommunicating rudimentary horn adenomyosis with cystic degenerations, degenerated leiomyoma and essential and primary dysmenorrhea. The initial routine pelvic USG can give clue to diagnosis but MRI is highly accurate. The diagnostic criteria (Acién *et al*., 2010) for ACUM are 1) an isolated accessory cavitated mass usually located under round ligament 2). A normal uterus fallopian tube and ovary 3).a surgical case with excised mass and pathological examination 4). An accessory cavity lined by endometrial epithelium with glands and stroma 5) chocolate brown colored fluid content 6) no adenomyosis in uterus (if resected) although there could be tiny foci of of adenomyosis in the myometrium of accessory cavity due to increased intracystic pressure. Most authors accept ACUM as congenital anomaly caused by duplication of ductal Mullerian tissue in critical area at the level of attachment of round ligament, possibly due to gubernaculums dysfunction (Acién *et al*., 2011).

****Corresponding author: Mahawar, P.***
Manchanda Endoscopy Centre, PSRI Hospital, New Delhi

Case report

A 21 yrs old nulliparous female presented to us with complains of pain in left iliac fossa and dysmenorrhea since 5 months. Her menstrual cycles were regular with increased flow. She had consulted many surgeons and gynaecologists and was thoroughly investigated for pain abdomen. In the mean time she was prescribed analgesics but her pain was not relieved. During her investigations CT scan revealed bilateral adnexal cystic lesions with bulky uterus and she was referred to us. After routine investigations she was posted for diagnostic hysteroscopy with diagnostic and operative laparoscopy. Her hysteroscopic findings were normal. On laparoscopy uterus, both fallopian tubes and ovaries were found normal. There was a 4*3 cm mass in left broad ligament situated just below the round ligament simulating broad ligament fibroid (Figure 1). Dilute vasopresin was injected in it and incision given over it with harmonic. While enucleating it showed a cavity draining

chocolate color fluid from it (Figure 2). The histopathology revealed a cavitated mass lined by Endometrium with nodular myometrial hyperplasia. Her post op recovery was good and she was symptom free.

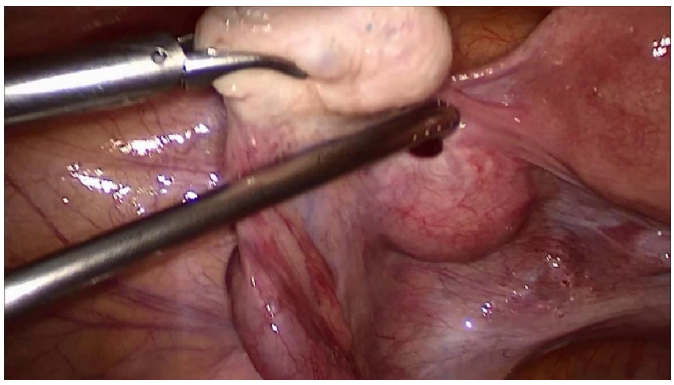

Figure 1. Accessory and cavitated mass just below the round ligament

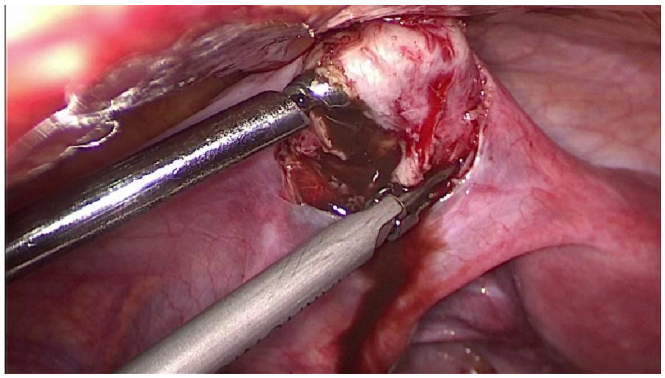

Figure 2. Cavitated mass showing drainage of chocolate color fluid

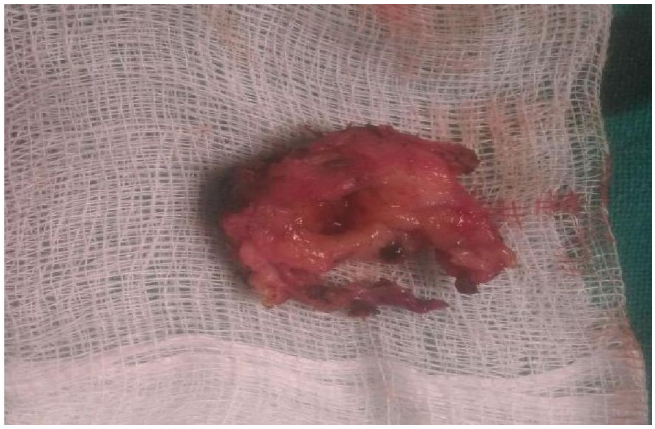

Figure 3. Mass after excision

DISCUSSION

Oliver described a case in 1912 with "an accessory uterus distended with menstrual fluid "in a 34 year old female (Oliver, 1912), Cozzuto reported a case of uterine like mass in 1981 (Cozutto, 1981). Tamura *et al*. (1996) first described juvenile Adenomyotic cyst of the corpus uteri in 16 yrs old girl (Tamura *et al*., 1996). Takeuchi *et al* reported 9 cases with particular emphasis on laparoscopic enucleation (Takeuchi *et al*., 2010). Acien *et al* studied 36 cases in literature, labeled as isolated cystic adenomyoma, uterus like masses, noncommunicating accessory uterine cavities and found that most of these correspond to same pathology: An ACUM. The term ACUM was coined by Acien *et al*. An ACUM is a rare pathology observed in young women and it represent a new variety of Mullerian anomaly that is generally located at the level of insertion of round ligament. But this condition must be differentiated from true cavitated adenomyoma and cystic adenomyosis which are found in older women who develop adenomyosis spread anywhere in the uterine corpus and cyst shows absence of internal epithelial lining as indicated by histopathology. The most common extrauterine site for such masses is ovary, however they have been seen in broad ligament, small bowel, mesentry, colon, uterosacral ligament. Though the clinical suspicion and knowledge of pathology is the key for diagnosis, USG is the initial imaging modality for diagnosis which identifies them as solid isoechoic to predominantly cystic mass. On HSG naturally the mass is not visualized but it rules out other Mullerian anomalies. MRI is highly valuable for diagnostic accuracy if interpreted properly. Inspite of all these imaging facilities most of the cases are misdiagnosed preoperatively as Mullerian anomalies, cystic degeneration in adenomyoma, broad ligament fibroid and leiomyoma. In our case also it was not diagnosed preoperatively and intraop also it was simulating broad ligament fibroid. We have also previously operated a case in our centre where an ACUM was misdiagnosed as uterine bicornis with obstructed rudimentary horn with hematometra and diagnosis was confirmed after histopathology.

In most of the published cases it was treated by laparoscopic excision. The laparoscopy is useful not only in its treatment but also in the diagnosis as they are often misdiagnosed preoperatively.

Conclusion

ACUM, a new type of Mullerian anomaly is less rare than previously considered. The clinical suspicion, USG and MRI facilitate the correct diagnosis. It is a treatable cause of severe dysmenorrhea in young girls. The laparoscopic excision of the mass can relieve the suffering of these young girls.

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