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ACCESSORY CAVITATED UTERINE MALFORMATION (ACUM): A RARE AND UNDERDIAGNOSED CAUSE OF SEVERE DYSMENORRHEA.

Obstetrics & Gynaecology

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ABSTRACT

In the absence of any globally accepted definition and specific identifiable characteristics accessory and cavitated uterine malformation (ACUM) is still a rare mullerian anomaly. It is characterised by presence of a mass near the insertion of the round ligament. The mass has an accessory uterine cavity in close proximity with the uterus which is usually non-communicating with its main body. As per the available studies ACUM is usually found in young women. These women mostly present with severe dysmenorrhea. The diagnosis of ACUM is made by the presence of a central cavity in the uterus, lined by functional endometrium, and surrounded by smooth muscle. Medical management options include oral contraceptive pills (OCP), levonorgestrel intrauterine system (LNG-IUS), GnRH agonist therapy, ultrasound-guided sclerotherapy. But they provide temporary and symptomatic relief only. Surgical management remains the main stay of treatment due to severe dysmenorrhea and chronic pelvic pain. We present a case of a 40-year-old female who presented with severe dysmenorrhea for 2 years and heavy menstrual bleeding for 1 year which was not relieved with any form of hormonal treatment. Her USG and MRI findings were inconclusive. Finally, she underwent laparotomy. A uterine mass was found just beneath the insertion of the round ligament on the right side. The rest of the uterus was grossly normal. The mass was excised and sent for histopathological examination. The HPR report showed cystic cavity with endometrial lining surrounded by benign monotonous spindle cells arranged in fascicular pattern. Postoperative period was uneventful. Patient was relieved from all her symptoms after the surgery.

KEYWORDS

Accessory and Cavitated Uterine Malformation, Mullerian Anomaly, Cystic Adenomyosis, Accessory and Cavitated Uterine Mass, Juvenile Cystic Adenomyosis

INTRODUCTION

Accessory and Cavitated Uterine Malformation (ACUM) was first identified in 1912. It contains functional endometrium within the external myometrium and/or in the broad ligament. The uterus is grossly normal [1,2]. The aetiology of ACUM is still undefined but seems likely congenital. The malformation is thought to involve duplication and persistence of Mullerian tissue with possibly dysfunction of the gubernaculum [3]. The exact prevalence of ACUM is still unknown as many terminologies were being used in the past for similar findings like "Accessory cavitated uterine mass", "Adenomyotic cysts", "cystic myometrial lesion", "Juvenile cystic adenomyosis", "Myometrial cyst", "Mullerianosis", "non-communicating uterine horn" and "Uterine-like Mass".

Latest definition is proposed by Naftalin et al., 2021 which defines ACUM as a cavitated lesion with a myometrial mantle and echogenic contents in the anterolateral wall of the myometrium beneath the insertion of the round ligament. Obstructive congenital anomalies, such as a communicating and noncommunicating horns must be ruled out for the diagnosis of ACUM. [4]

Most common clinical features of ACUM are severe dysmenorrhea and cyclic pelvic pain. Other can be chronic pelvic pain and infertility. Dysmenorrhea is usually unresponsive to medical therapy. The management of ACUM mostly involves excision of the mass.

Case Report

A 40-year-old, P2L2A1 female came with complaints of severe dysmenorrhoea since last two years and heavy menses since last one year. On bimanual examination her uterine size was normal and firm bossellated mass was felt on right side in continuity with the uterus. Uterus was freely mobile and nontender. Bilateral adnexa free and non-tender. She was evaluated for the uterine mass. On transvaginal sonography uterus was anteverted, normal in size and bicornuate in shape. There was evidence of heterogenous lesion measuring 28x22 mm with anechoic region showing tiny echogenic content (? clot) seen in the right cornua and the endometrial lining of left cornu was normal (7.8 mm). Both ovaries were normal (Fig 1). A differential diagnosis of non-communicating rudimentary horn of uterus or posterior wall degenerated fibroid was made. For further confirmation she was advised MRI pelvis that reported uterus mildly bulky (9x4.8x4.5cm) and intramural fibroid of size 3.6 x3 cm in posterior wall with degenerative changes. A well-defined cystic lesion measuring 2.8x2.5cm was seen in left ovary was seen, without any septa or solid component (Fig 2). She was given a course of analgesics, as her dysmenorrhea did not respond and rather worsened, she was counselled regarding the need for surgical intervention (removal of the mass/ hysterectomy). Finally, the decision of surgical intervention was taken with the primary aim of removing the mass but with consent of hysterectomy. She underwent laparotomy. Her intra-operative findings show normal size uterus. A mass seen near the right cornual region on posterior-lateral surface extending into broad ligament measuring up to 3x4 cm. There was a haemorrhagic cyst of 2x3 cm in left ovary. The mass was removed with difficulty as no proper plane was found between the mass and the myometrium for dissection. After resection the mass was cut open and was found to be filled with chocolate coloured haemorrhagic fluid (Fig 3). The uterus was repaired successfully, haemostasis was achieved. There was no need for hysterectomy. Specimen was sent for histopathology examination. Histopathology report was suggestive of benign monotonous spindle cells arranged in fascicular pattern with few interspersed thick-walled arteries. Sections through the cystic cavity show partial endometrial lining and partial granulation tissue lining along with many hemosiderin laden macrophages. Occasional endometrial glands also seen adjacent to the cyst wall. Rare mitosis seen. No atypia/malignancy seen (Fig 4). The post operative period was uneventful and patient recovered well. At follow up after her menses, patient had no complaints of dysmenorrhea and heavy bleeding.

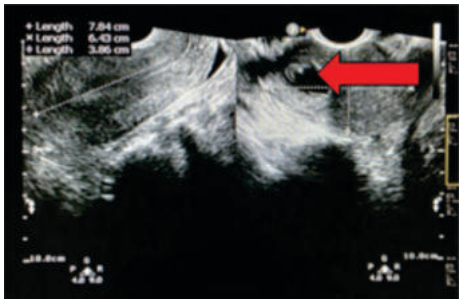

Fig 1 Ultrasound (TVS): mass marked with arrow suggestive

differential diagnosis of non-communicating rudimentary horn of uterus or posterior wall degenerated fibroid.

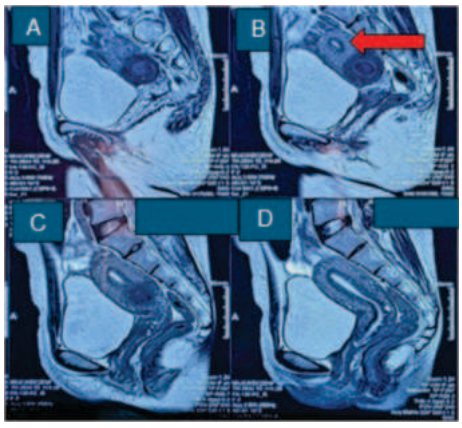

Fig 2 (A, B, C, D) MRI images showing the mass in different planes of sagittal section of the pelvis. Arrow showing the mass interpreted as intramural fibroid of size 3.6x3cm in posterior wall with degenerative changes.

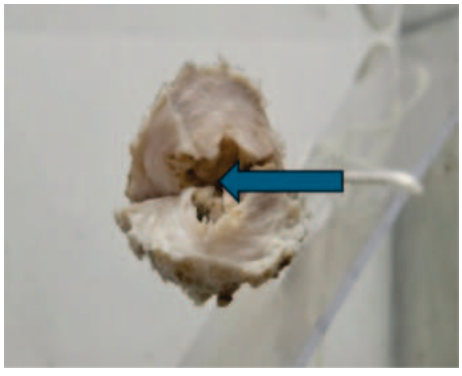

Fig 3 Cut section of the mass showing greyish white whorled appearance with arrow showing empty cystic cavity with brownish discolouration.

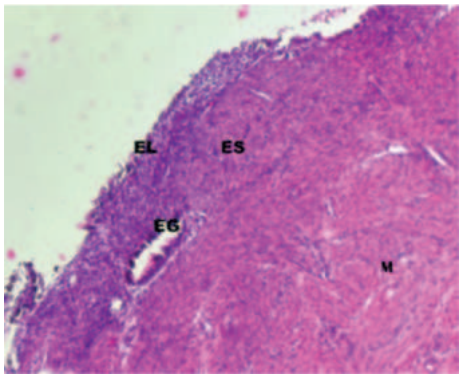

Fig 4 Histopathology image of the specimen.
M= myometrium
EG= Endometrial gland
ES=Endometrial stroma
EL= Endometrial lining

DISCUSSION

ACUM still remains a poorly understood entity even after more than 100 years of its first documentation. Earlier ACUM was called accessory cavitated uterine mass or juvenile cystic adenomyoma. In 2021, the terminology of "accessory cavitated uterine malformation" was recommended based on the description of the cystic lesions in the myometrium with specific location, typical symptoms, and characteristic morphological imaging features [4]. Severe dysmenorrhea, non-responding to analgesics, and recurrent pelvic pain are the major symptoms of ACUM.

As these symptoms are non-specific, they often are ignored by the patients leading to delayed presentation to the gynaecologist. On imaging studies ACUM is difficult to distinguish from uterine fibroid, fibroid with cystic/degenerative change, or adenomyosis further delaying the diagnosis as was reported by Hu Q et al [5], Mollion M et al [6] and was also seen in our case. The diagnosis of ACUM depends on cumulative findings of clinical features, radiological appearance, as well as histopathological findings.

Various management options have been described in studies reporting ACUM including NSAIDs, oral contraceptive pills (OCP), levonorgestrel intrauterine system (LNG-IUS), GnRH agonist therapy, mass excision, ultrasound-guided sclerotherapy and hysterectomy [7, 8]. But surgery is considered the definitive treatment for ACUM as the dysmenorrhea usually recurs after medical management. For young women fertility reservation should be taken into consideration before planning surgery, and minimally invasive surgery is the preferable treatment [9]. In our case also excision of the mass was done, preserving the uterus. Patient recovered well postoperatively.

CONCLUSION

Accessory Cavitated Uterine Malformation is a rare but important differential diagnosis in women presenting with severe, treatment-resistant dysmenorrhea and chronic pelvic pain. Due to its nonspecific clinical features and radiologic resemblance to other uterine pathologies, it is frequently underdiagnosed or misdiagnosed, leading to delays in appropriate treatment. Heightened clinical awareness in patients with characteristic symptoms unresponsive to conventional therapy are essential. As our understanding of this condition continues to evolve, further research is needed to elucidate its pathogenesis, optimize diagnostic pathways, and refine treatment strategies, particularly in reproductive-age women where fertility preservation remains a priority.

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Conflict of Interest: NIL

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