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Case Report

A rare cause of dysmenorrhea-accessory cavitated uterine malformation: a case report

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ABSTRACT

Accessory cavitated uterine malformation is a newly recognized mullerian anomaly where the uterine cavity is normal unlike other Mullerian anomalies and hence needs to be described more accurately. These are non-communicating ULMs that occur contiguously along the wall of the uterus. This case report is about a 31 years old female who came with complaints of acute severe dysmenorrhea with a chronic course for the past 2 years and was evaluated for a similar episode in the past where she was diagnosed as fibroid with cystic degeneration and treated with SPRM and OCP which gave no relief from symptoms. With another exacerbation of symptoms two years later, patient was evaluated with an USG suggesting hemorrhagic degeneration in rudimentary horn and gave two differentials- fibroid with cystic degeneration and adenomyoma, wherein the latter was more likely. As a part of pre-op evaluation, MRI was done. Then came the third differential-ACUM [Accessory cavitated uterine mass]. Patient was counselled about the risk of infertility and uterine rupture in subsequent pregnancy as a post-op complication underwent laparoscopic surgery. Intraoperative and postoperative period was uneventful and HPE of the same confirmed the diagnosis. ACUM is a very treatable cause of severe dysmenorrhea and adequate awareness of this new entity will enable in making the diagnosis early.

Keywords: Uterine cavity, ACUM, Myometrium

INTRODUCTION

Accessory cavitated uterine mass (ACUM) is a newly diagnosed entity in the mullerian duct anomalies. This entity was previously described as juvenile cystic adenomyoma, cavitated adenomyoma and cystic myometrial lesions.[1] ACUM is an unusual form of adenomyosis, characterized by a well-circumscribed cavitated endometrial gland and stroma, ≥1 cm in diameter, located within the myometrium.[2]

CASE REPORT

A 31-year-old, P1L1 female came with complaints of severe dysmenorrhea with left sided pelvic pain on and off for the past 2 years which required multiple admissions and injectable analgesics. She had no history of previous surgeries nor any medical comorbidities.

Two years back, patient was evaluated outside for the same and was diagnosed with an ultrasonogram as fibroid with cystic degeneration and was treated with ulipristal acetate (SPRM) and OCPill with no relief in symptoms.

Two years later, she presented to us during an episode of severe dysmenorrhea and was evaluated with: An ultrasonogram suggesting hemorrhagic degeneration in rudimentary horn and an MRI pelvis that gave a differential diagnosis of a fibroid with cystic degeneration or an adenomyoma.

Since her history and evaluation suggested the possibility of adenomyoma and she didn't respond well to medical

management, she was suggested to undergo adeno-myomectomy. But, patient was not willing for surgery and she understood the possibility of sacrificing one fallopian tube and the chance of rupture in future pregnancy as the lesion was close to the cornua. She went home wanting to continue medical management as she was keen on second pregnancy. Few months later, after an episode of unbearable dysmenorrhea, patient wanted adenomyomectomy. As a part of pre-op evaluation, MRI was repeated for mapping. Then came the third differential diagnosis-ACUM. Patient was counselled about the risk of uterine rupture in subsequent pregnancy after adenomyomectomy. After her consent she was prepared for laparoscopic adenomyomectomy in the post menstrual period.

Intraoperatively- A 3×2 cm mass was seen below the left fallopian tube anteriorly. The mass was separate from the uterine musculature and contained chocolate colored fluid, it was possible to enucleate it without entering into the uterine cavity. It did not have a capsule like a fibroid and there was no need to resect the uterine myometrium as we do in adeno-myomectomy. The mass was enucleated in toto laparoscopically the base was sutured in layers with 2-0 polygalactin. Chromopertubation was done which showed bilateral tubal spill, which also confirmed that uterine cavity was not breached. Surgery and post op recovery went uneventful. Histopathological examination came as adenomyoma which was consistent with our diagnosis of ACUM.

Patient was reviewed 2 months later with nil complaints. She was totally relieved of dysmenorrhea and pelvic pain.

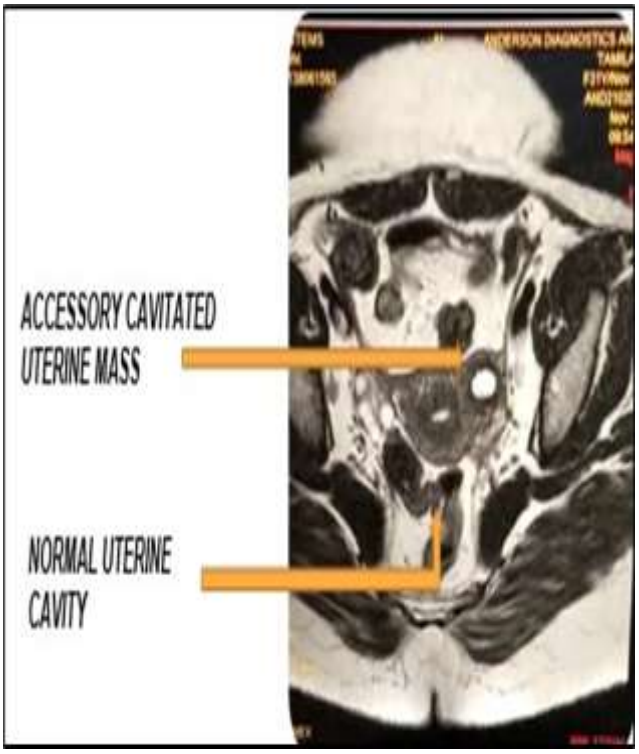

Figure 1: Pre-operative image-MRI pelvis.

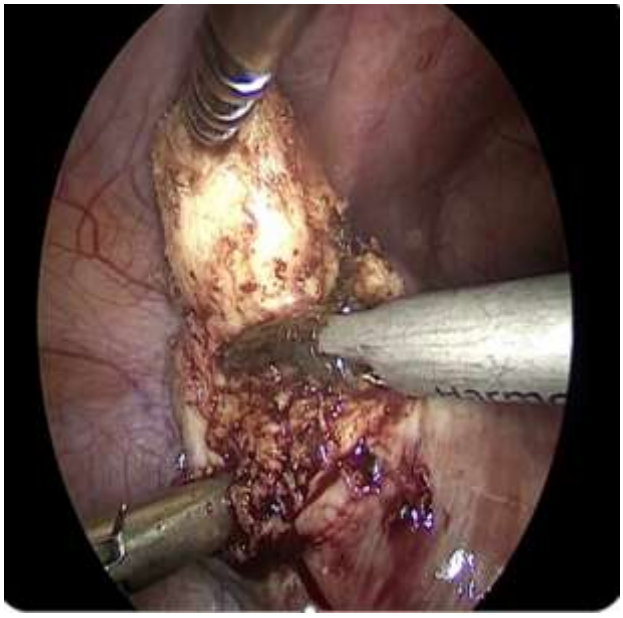

Figure 2: Intra-operative laparoscopic image-ACMA around attachment of round ligament.

DISCUSSION

ACUM is a newly recognized mullerian anomaly with a non-communicating uterus-like mass (ULM) arising in the uterus itself without involving the uterine cavity and uterine myometrium. Most of the authors accept ACUM as a congenital anomaly. The proposed mechanism says that the accessory mass could be caused by duplication of ductal Mullerian tissue in the critical area at the level of attachment of round ligament, possibly related to gubernaculum dysfunction.

The criteria for diagnosing ACUM are[1]

Location: Solitary lesion located in the lateral myometrium or broad ligament, no communication with uterine cavity or fallopian tubes, Morphology: A cavitated lesion containing functional endometrium surrounded by a myometrial mantle. Histology: Cavitated lesions filled with dark brown haemorrhagic content, lined with functional endometrium and myometrial mantle has concentric organisation of smooth muscle.

In our case, visualization of normal size and shape of the uterus and both cornua ruled out Mullerian anomalies. As bilateral ovaries were separate and appeared normal, ovarian tumours were excluded. Cystic degeneration in adenomyoma and fibroid will not show T2-hyperintense endometrial lining and hemorrhagic contents.

Regarding therapeutic management, most publications have included laparoscopic excision of the mass than laparotomy. Most of the cases were misdiagnosed preoperatively as mullerian anomalies, cystic degeneration in adenomyoma and leiomyoma, and broad ligament fibroids.

The MRI findings of an accessory cavitated uterus like mass located below the attachment of round ligament usually with haemorrhagic contents in an otherwise normal-shaped uterus with bilateral normal tubes and ovaries, without any evidence of adenomyosis should suggest the diagnosis of ACUM pre-operatively.

CONCLUSION

ACUM, a rare form of Mullerian anomaly related to dysfunction of gubernaculum seen in young females, is a treatable cause of severe dysmenorrhea in. However, it is not that rare entity as ACUM is quite underdiagnosed.

An MRI finding of an accessory cavity below the attachment of round ligament in an otherwise normally shaped uterus with normal tubes and ovaries should include ACUM among one of its possible diagnoses. Thus, awareness and adequate knowledge of the entity can help us to make an accurate pre-operative diagnosis of ACUM.

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