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Case Report

Successful laparoscopic management of accessory cavitated uterine mass: a rare case

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ABSTRACT

Accessory cavitated uterine mass (ACUM) is a rare and unique congenital anomaly. The dysmenorrhea presented by these patients is almost never resolved by any medications but only by surgical line of management. Herein, we are discussing a case of ACUM who had medically refractory dysmenorrhea and treated by laparoscopic excision and repair. ACUM is the newly identified popping up cause of dysmenorrhoea requiring surgical line of management only. Fewer than 40 cases, worldwide, have been reported in the literature with the youngest case being a 13-year-old girl. Laparoscopic excision has shown good postoperative results. Dysmenorrhea is a significant cause of reduced quality of life. High degree of suspicion, early investigations and identification of such cases help in appropriate management and reduce the duration of symptoms and thus the seamy side of life can be made better.

Keywords: ACUM, Dysmenorrhea, Laparoscopic excision

INTRODUCTION

Dysmenorrhea is one of the important cause leading to disability-adjusted life years in women, ranging from 16% to 91% in women of reproductive age.[1] If the etiology of primary dysmenorrhea is precisely not understood, the well-known causes of secondary dysmenorrheal would include leiomyomas, adenomyosis, pelvic inflammatory disease and endometriosis.[2] Accessory cavitating uterine mass refers to a uterine like cavity lined with endometrium, surrounded by myometrium, non communicating with the real endometrial cavity.[3] This is a rare, usually misdiagnosed or under-recognized cause of medically unresponsive dysmenorrhea.

Nowadays, ACUM is also said to be a new variety of Müllerian anomaly that is usually seen at the level of the insertion of the round liga¬ment (either left or right) and is related to dysfunction of the female gubernaculum.[3] As ACUM closely mimic uterine anomalies such as a unicornuate uterus with a rudimentary horn, bicornuate uterus with a noncommunicating horn, and hematometra in uterine didelphys with a transvaginal septum, ruling out these differential diagnosis is not that easy even with the use of pelvic ultrasonography, hysterosalpingography (HSG) and magnetic resonance imaging (MRI).[4] Confirmation of both the ostia is helpful in accurate diagnosis. Herein, we are reporting this rare case of ACUM who underwent laparoscopic surgery successfully.

CASE REPORT

An unmarried girl, aged 20 years, presented to our hospital with severe dysmenorrhea and recurrent abdominal pain. She attained her menarche at 13 years and had regular cycles. The pain was severe, with no relief with analgesics and NSAIDS for several months. The pain was so severe that she had to miss her college several times. Her ultrasound examination revealed a normal uterus and adnexa with a fluid filled lesion with the fluid debris level within the myometrium on right side of the uterus abutting and pushing the endometrium towards left side suggesting

a rudimentary horn on the bicornuate uterus with hematometra.

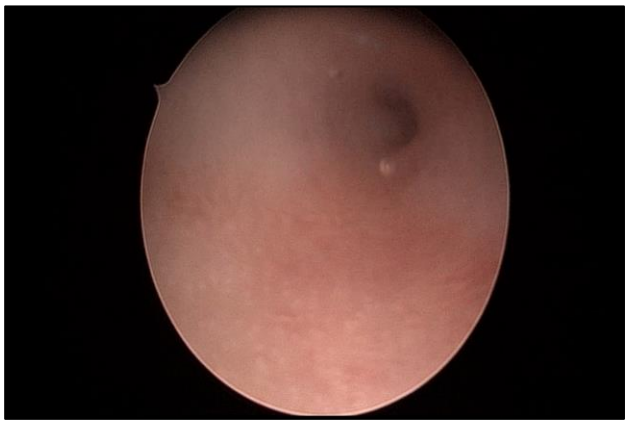

Figure 1: Hysteroscopy showing left ostia.

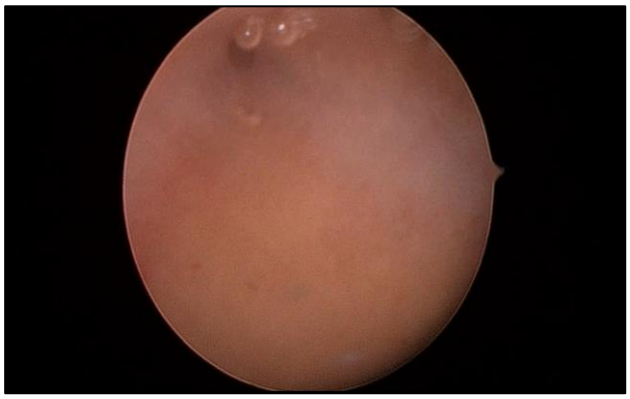

Figure 2: Hysteroscopy showing right ostia.

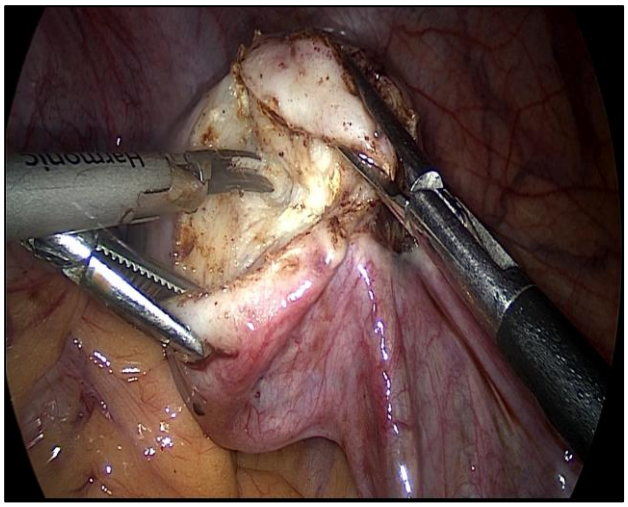

Figure 3: ACUM seen below and medial to the insertion of right round ligament.

Her transrectal ultrasound suggested Anterior myometrial cystic mass,? pus,? blood, ?ACUM. She was posted for laparoscopic management, with prior valid consent Hysteroscopy was performed under general anaesthesia which demonstrated a normal uterine cavity with bilateral ostia seen (Figure 1, 2). On laparoscopy both adnexa were seen normal. A hard nodular mass was felt on palpation by the probe over the anterior myoma. It was present below and medial to the insertion of right round ligament (Figure 3). The cavity of mass was cut open by monopolar cautery and thick chocolate coloured fluid drained out. Accessory endometrium was seen in the cavity suggestive of ACUM (Figure 4). The nodule was excised with harmonic scalpel along with the entire endometrium. The myometrial defect was closed in layers with V loc sutures (Figure 5). The abdominal cavity was thoroughly irrigated with normal saline. The histopathological examination of the specimen showed endometrial glandular epithelium and stroma. We took around 40 minutes to operate and the patient was stable throughout the procedure. The intraoperative blood loss was roughly around 10-20 ml.

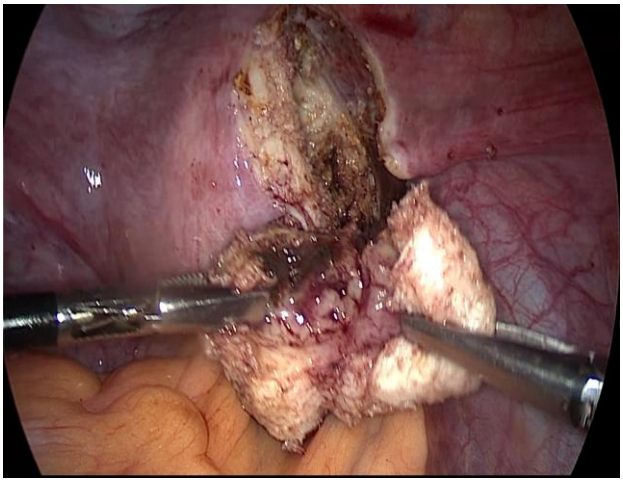

Figure 4: Excision of accessory cavity with functional endometrium.

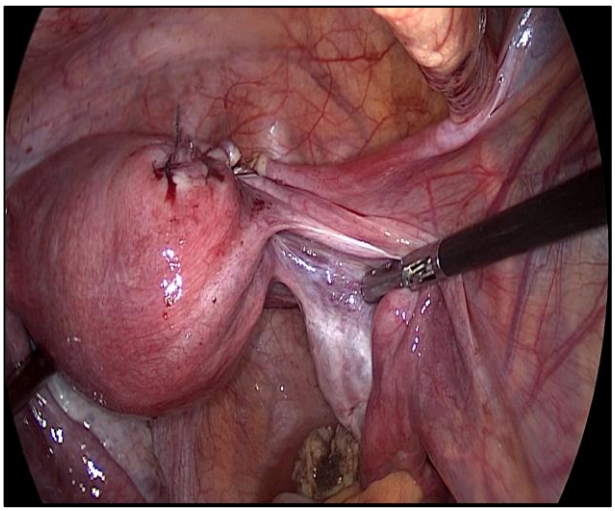

Figure 5: The myometrial defect was closed in layers with V loc sutures.

Her postoperative period was uneventful. She was discharged on 4th postoperative day in a good condition.

The patient is asymptomatic 2 months after surgery with a normal evolution.

DISCUSSION

ACUM is the newly identified popping up cause of dysmenorrhoea requiring surgical line of management only. Fewer than 40 cases, worldwide, have been reported in the literature with the youngest case being a 13-year-old girl.[5]

The differential diagnosis of ACUM goes as unicornuate uterus with a rudimentary horn, bicornuate uterus with a non-communicating horn, and hematometra in uterine didelphys with a transvaginal septum, hematocolpos with obstructing Müllerian anomaly, myomas and adenomyosis. ACUM is a cystic lesion that is independent of the normal endometrium, whereas haematometra and hematocolpos are present in the obstructing Müllerian anomaly.[6] Even with MRI, it can sometimes be difficult to differentiate it from a cavitated noncommunicating rudimentary uterine horn. In this situation, hysterosalpingography and hysteroscopy can be useful in distinguishing it from a uterine anomaly.[7] Acie´n et al. hypothesized that what was earlier described as juvenile cystic adenomyosis was the same entity as accessory cavitating uterine mass.[8] Even though adenomyosis is usually a diffuse solid mass and develops in women in their forties, it can present as an adenomyoma, adenomyomatous polyps, and cystic adenomyomas.[9] Small cystic spaces, less than 0.5 cm in diameter, may be associated with adenomyosis.[10] Juvenile cystic adenomyoma is defined as a solitary myometrial cyst measuring ≥1 cm, which is surrounded by hypertrophic endometrium, independent of the uterine lumen, and is usually present in women less than 30 years of age, in association with severe dysmenorrhea.[8] Additionally, the degenerated myoma and vesicouterine endometrioma are also considered as differential diagnoses.[11]

The diagnostic criteria for ACUM are: an isolated accessory cavitated mass; a normal uterus (endometrial lumen), fallopian tubes, and ovaries; surgical evidence with an excised mass and histopathological finding; an accessory cavity lined with endometrial epithelium with glands and stroma; chocolate-brown coloured fluid content; and noadenomyosis (if the uterus has been removed), although there could be small foci of adenomyosis in the myometrium adjacent to the accessory cavity.[3]

Laparoscopic excision has been attempted and has shown good postoperative results, comparable to those of exploratory laparotomy.[8] Other surgical approaches have been proposed, such as radiofrequency ablation under ultrasonography guidance, single incision with monopolar cautery, or the use of robotic surgery.[12]

CONCLUSION

Dysmenorrhea is a significant cause of reduced quality of life and loss of productivity in women. ACUM is one of the cause of medically refractory dysmenorrhea which can be completely treated through surgical line of management under expert hands. High degree of suspicion, early investigations and identification of such cases help in appropriate management and reduce the duration of symptoms and thus the seamy side of life can be made much better.

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